Factors Associated with the Psychosocial Well-being Among Parents of Children who are Deaf or Hard of Hearing

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Abstract

Purpose: To identify factors associated with low psychosocial well-being among parents of children who are deaf or hard of hearing.

Method: A cross sectional survey that included instruments to measure psychological distress, functional impairment, and psychological inflexibility. Two hundred and ninety-six parents completed the survey.

Results: Analyses revealed that lower income, presence of additional disabilities, younger child age, and psychological inflexibility were factors associated with low parent psychosocial well-being.

Conclusions: Parents of younger children who are deaf or hard of hearing with low income, have children with additional disabilities, and higher psychological inflexibility may experience lower psychosocial well-being. Clinicians serving families may need to provide additional and/or different support for parents in adapting to and managing their child's hearing loss.

Key words: pediatric hearing loss, parent psychosocial

Acronyms: AAQ-MCHL = Acceptance and Action Questionnaire–Managing Child Hearing Loss; DASS-21 = Depression, Anxiety, and Stress Scale; DHH = deaf or hard of hearing

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The psychosocial well-being of parents of children with disabilities or chronic health conditions has been explored across multiple conditions and contexts for years (e.g., Barlow & Ellard, 2006; Gilson et al., 2018; Moody et al., 2019). For parents of children who are deaf or hard of hearing (DHH) specifically, research has explored a variety of psychosocial well-being factors, such as parents' satisfaction with life (Yiğit et al., 2018), anxiety and stress (Bilsin et al., 2015; Quittner et al., 2010), and the impact of pediatric deafness on the family dynamic (Dammeyer et al., 2019; Jackson & Turnbull, 2004). Identifying factors associated with low psychosocial well-being for parents of children who are DHH is an important step in helping providers and parents develop effective care plans, that include parent needs, during the intervention process.

After hearing loss is identified, parents are tasked with learning new skills and incorporating intervention recommendations into daily routines, and this can be challenging. For example, 10 or more hours of daily hearing aid use is recommended for optimizing spoken language development (Tomblin et al., 2015); however, studies have found low hours of hearing aid use for young children (Muñoz et al., 2016; Walker et al., 2013). This is not surprising, many parents are unfamiliar with hearing

loss as 92% of children who are DHH have hearing parents (Mitchell & Karchmer, 2006). Individualized support is needed to address the range of barriers parents experience. They may experience difficult emotions and parental well-being can pose a barrier to how parents engage in hearing care. For example, Muñoz, Olson, and colleagues (2015) found in a sample of 40 parents of children who are DHH that 40% linked depression to their difficulty in managing consistent hearing aid use.

When parents experience difficult or uncomfortable thoughts and emotions about their child's hearing loss they may avoid these internal experiences (e.g., not put the hearing aids on their child). Experiential avoidance is the inability to handle private events (e.g., thoughts, emotions), and when this occurs, the individual's behaviors support avoidance of these private events (Hayes et al., 1996). Experiential avoidance can interfere with parents making critical health behavior changes (e.g., consistent hearing aid use; checking device function) to support their child's development. Psychological flexibility is a process important for behavioral change and has not been previously explored for parents of children who are DHH. Psychological flexibility can be measured by a recently developed instrument called the Acceptance and Action

Questionnaire–Managing Child Hearing Loss (AAQ-MCHL; Ong et al., 2019). This process describes how parents are influenced by their internal experiences (e.g., thoughts), that is, being in the present moment and taking actions that are consistent with their values, even while feeling uncomfortable with their internal experiences.

Of particular concern are parents who are struggling with the intervention process. Parent psychosocial well-being can be a barrier; however, it is often not explored or addressed as part of the intervention process for children who are DHH. Psychosocial well-being refers to outcomes for how people function (e.g., activities of daily living, social relationships, emotional health), which can play a role in how quality of life is perceived (Burns, 2016). In a recent study, Kasin and colleagues (2020) found that, among 296 parents of children who are DHH, approximately 15 to 34% reported clinical levels on a variety of psychosocial outcomes, including psychological distress, overall well-being, and functional impairment. Although many parents were doing well, the subset of parents who were experiencing distress suggests implications regarding audiology service delivery and the potential for hearing health at home to be compromised. Furthermore, it remains unclear when and/or which parents are at risk for psychosocial challenges, and how they can be identified to ensure appropriate supports are in place for optimal hearing health care.

The current study presents secondary analyses from Kasin et al. (2020) to identify factors that may be associated with low psychosocial well-being by investigating the influence of factors on two dependent variables: (a) psychological distress, and (b) functional impairment. The secondary aim was to investigate whether significant correlations existed between these two psychosocial outcomes and the reported daily amount of time spent using amplification technology.

Method

Participants and Recruitment

The data were collected over a three-month period from June to August 2018 using an online cross-sectional survey. Parents were notified of the opportunity to participate through multiple venues: (a) national organizations that provide technical and other supports to parents of children who are DHH, (b) pediatric audiology clinics, and (c) Facebook groups dedicated to parents of children who are DHH. Parents were eligible to participate if their child had a permanent hearing loss and if the parent was proficient in English; not all instruments were available in languages other than English. Due to the potential overlap of participant recruitment through the multiple venues, it was not possible to calculate the response rate. This study met ethical approval by the Utah State University Institutional Review Board.

Instruments

Demographic data were obtained to understand the make-up of the sample. All items were self-reported (e.g., age, race, relation to child) or reported to the best of

parents' knowledge (e.g., child's degree of hearing loss, child has other disabilities). In addition to the demographic questions, three instruments were used to measure the outcomes of interest: The Depression, Anxiety, and Stress Scale (DASS-21; S. H. Lovibond & Lovibond, 1995); the Work and Social Adjustment Scale (WSAS; Mundt et al., 2002); and the Acceptance and Action Questionnaire–Management of Child Hearing Loss (AAQ-MCHL; Ong et al., 2019).

Psychological Distress

The Depression, Anxiety, and Stress Scale (DASS-21; S. H. Lovibond & Lovibond, 1995) is a self-report questionnaire with three 7-item scales to measure depression, anxiety, and stress, with the total score reliably indicating general psychological distress (Henry & Crawford, 2005). Since its development, it has been widely used to assess for depression, anxiety, and stress in adult populations with consistent results surrounding its validity and reliability (Crawford & Henry, 2003; Le et al., 2017; P. F. Lovibond & Lovibond, 1995, S. H. Lovibond & Lovibond, 1995).The scale has good total reliability (Cronbach's α = .88), and good to excellent item reliability for Depression (Cronbach's α = .82), Anxiety (Cronbach's α = .90) and Stress (Cronbach's α =.93); finally, the instrument reports good convergent and discriminant validity. For the present study, item reliability for the DASS-21 was measured as good for depression (Cronbach's α = .89), anxiety (Cronbach's α = .83), and stress (Cronbach's α = .89).

Functional Impairment

The Work and Social Adjustment Scale (WSAS; Mundt et al., 2002) is a 5-item self-report questionnaire which measures the impact of an individual's functional impairment in the context of employment, home management, social and private leisure, and personal/family relationships. The instrument may be used for comparisons of functional impairment across studies and disorders. For the purpose of this study, it was modified by placing the carrier phrase "Because of my child's hearing loss…" at the start of each item (e.g., "Because of my child's hearing loss, my ability to work is impaired" or "Because of my child's hearing loss, my ability to form and maintain close relationships with others, including those I live with, is impaired"). Scoring is continuous up to a maximum score of 40. The higher the score, the more an individual sees their child's deafness or hearing loss as an impairment to parents' functioning. The scale has acceptable to excellent internal consistencies reported, ranging from Cronbach's α = .70 to Cronbach's α = .94 (Mundt et al., 2002), with good convergent and known-groups validity. For the present study, reliability for the WSAS was excellent (Cronbach's α = .90).

Psychological Inflexibility

The Acceptance and Action Questionnaire–Management of Child Hearing Loss (AAQ-MCHL; Ong et al., 2019) is an 8-item questionnaire designed to measure psychological inflexibility. Psychological inflexibility refers to the extent to which parents of children who are DHH avoid difficult

thoughts and feelings associated with the diagnosis and management of their children's hearing loss. Example questions include "My frustrations with my child's hearing loss have negatively affected my parenting" or "I worry what others think about my child's hearing loss." This questionnaire has shown good reliability (Cronbach's α = 0.85), and concurrent and discriminant validity (Ong et al., 2019).

Analysis

Backward elimination regression analyses were used to identify variables that independently predicted variance in psychological distress and functional impairment. First, all relevant independent variables (IVs) were included as factors in a regression model (IVs included demographic variables presented in Table 1, and psychological inflexibility). Second, IVs that did not significantly contribute to the model were excluded from subsequent models. Third, change in R^2 ($R^2\Delta$) was calculated for these nested regression models. If $R^2\Delta$ was non-significant, the more parsimonious model was retained. From this process, the following IVs were fit for the present multiple regression model:

(1) For psychological distress: Psychological inflexibility and income.

(2) For functional impairment: Psychological inflexibility, presence of other disabilities, and child age.

In addition, residual plots, Q-Q plots, and VIFs (cutoff was < 4) were examined to determine if regression models met statistical assumptions. Whether these assumptions are met reflects reliability of findings derived from these analyses. The plots and VIFs indicated homoscedasticity and no multicollinearity in all reported models. That is, variables had similar scatter and were not correlated so highly with each other as to adversely impact findings. A Spearman's correlation was calculated between each of the variables of interest. Finally, Pearson's correlation was calculated to determine whether significant correlations existed between parent-reported hours of device use and psychological distress and functional impairment.

Results

Data were analyzed for 296 parents (see Table 1). The sample represented primarily mothers who were White and college educated. Seventy-four percent of children had a severe to profound degree of hearing loss, and 78% of children had hearing loss in both ears.

Table 2 presents correlational data for each of the variables explored in this study. Greater psychological distress was weakly associated with lower income (rho = -0.226, $p < 0.001$) and greater psychological inflexibility (rho = 0.345, $p < 0.001$) and moderately associated with greater functional impairment (rho = 0.468, $p < 0.001$). More psychological inflexibility was weakly associated with lower income (rho = -0.138, $p < 0.05$) and moderately associated with greater functional impairment (rho = 0.488, $p < 0.001$). Higher income was weakly associated with less functional impairment (rho = -0.242, $p < 0.001$) and

older child age (rho = 0.138, $p < 0.05$). More functional impairment was weakly associated with fewer co-occuring disabilities (rho = -0.28, $p < 0.001$) and younger child age (rho = -0.166, $p = 0.01$).

Factors Associated with Psychological Distress

Psychological inflexibility had the greatest influence on outcomes for psychological distress (β = 0.30, SE = 0.05, $p < .001$), followed by low income ($21,000–40,000; β = 6.87, SE = 3.12, $p < .05$). Whereas, participants with higher incomes (i.e., $41,000–80,000 and more than $81,000) did not show a significant influence (β = 1.44, SE = 2.74, $p > .05$; β = -1.35, SE = 2.60, $p > .05$, respectively). Psychological inflexibility and income were independently influential, given no significant interaction between the two variables was found (β = 3.56, SE = 2.74, $p > .05$). Thus, in this sample, parents with greater avoidance of difficult thoughts and feelings were more likely to experience greater levels of psychological distress when compared to parents with lesser avoidance. Parents with lower incomes were also more likely to experience greater levels of psychological distress than parents with higher incomes. See Table 3.

Factors Associated with Functional Impairment

All three independent variables in this model significantly influenced impairments to work, home, and social life. Specifically, parents of children who are DHH with no other disabilities showed they were less likely to experience impairment compared to parents of children with multiple disabilities (β = -4.79, SE = 1.02, $p < .001$); parents with high levels of psychological inflexibility were more likely to experience impairment compared to parents with lower levels of psychological inflexibility (β = 0.36, SE = 0.04, $p < .001$); and parents of younger children who are DHH were more likely to experience impairment compared to parents of older children who are DHH (β = -0.32, SE = 0.09, $p < .001$). Finally, a statistically significant relationship was found among the independent variables (β = 5.18, SE = 1.50, $p < .001$). See Table 4.

Correlation of Psychosocial Outcomes and Hearing Device Use

A negative correlation was found between parent-reported hearing device use and both psychological distress and functional impairment, meaning lower device use was correlated with higher levels of impaired well-being as measured by DASS-21 and WSAS. In both cases, however, the correlation was not significant (r = -0.05, r = -0.02 respectively).

Discussion

The purpose of this study was to identify factors that may be associated with low psychosocial well-being for parents of children who are DHH and to explore relationships with hours of hearing aid use. Four variables emerged as having higher risk for negatively impacting parents' psychosocial well-being: lower income, presence of other disabilities, younger child age, and psychological inflexibility.

Table 1
Demographics

Demographic Variables	% (n)	M (SD)	Median	Range
Parent				
Race: White	83 (248)			
Age		39 (8)	38	45
Education: college degree	75 (222)			
Annual Income				
More than $81,000	58 (172)			
$41,000–80,000	26 (78)			
Less than $ 41,000	16 (45)			
Relation to child: mother	94 (277)			
Child				
Race: White	80 (230)			
Current Age in years		7 (6)	6	30
Age Identified in months		20 (30)	3	168
Degree of Hearing Loss				
Mild-moderate	25 (74)			
Severe-profound	74 (219)			
Unsure	1 (3)			
Bilateral hearing loss	78 (232)			
Age fit with technology in months		26 (31)	15	168
Technology Type				
Hearing aid (HA)	43 (127)			
Cochlear implant (CI)	32 (96)			
Bimodal (HA+CI)	8 (24)			
Other	15 (44)			
Does not use technology	2 (5)			
Parent-reported hours of device use		12 (3.5)	12	23
Additional disabilities	32 (95)			

The findings are similar to other research as income has been shown to influence psychosocial well-being among parents of children with disabilities (e.g., Park et al., 2002; Shivers & Resor, 2019) and hearing loss. The presence of additional disabilities has also been shown to influence parent psychosocial well-being, and experiences specific to parents of children who are DHH with other disabilities (Whicker et al., 2019). Dammeyer and colleagues (2019) found that parents of children who are DHH with other disabilities were less engaged in activities and were more likely to report that the child was a burden for the family. There have been mixed findings in the literature for the relationship of parent or maternal stress and child age (e.g., Hintermair, 2004, 2006; Lederberg & Golbach, 2002;

Pipp-Siegel et al., 2002). Pipp-Siegel and colleagues (2002) suggest this inconsistency may be linked to differing sample characteristics and instruments used to measure stress.

Psychological inflexibility for child hearing loss was found to significantly associate with both psychological distress and functional impairment. Higher levels of psychological inflexibility through avoiding thinking about and adapting to the situational demands of a diagnosis is common among chronic conditions (Ong et al., 2019). Outside of audiologic research, studies have shown psychological inflexibility to influence a variety of outcomes, including poor job performance and increased risk for psychopathology (Hayes et al., 2006). In audiology, this is the first research

Table 2
Correlations

	Psychological distress	Psychological inflexibility	Income	WSAS	Other disabilities
Psychological inflexibility	*rho* = 0.345	-			
	p = 0.000***				
Income	*rho* = -0.226	*rho* = -0.138	-		
	p = 0.000***	*p* = 0.024*			
WSAS	*rho* = 0.468	*rho* = 0.488	*rho* = -0.242	-	
	p = 0.000***	*p* =0.000***	*p* = 0.000***		
Other disabilities	*rho* =-0.114	*rho* = -0.113	*rho* = 0.021	*rho* = -0.28	-
	p = 0.079	*p* = 0.065	*p* = 0.719	*p* = 0.000***	
Child age	*rho = 0.002*	*rho* = -0.038	*rho* = 0.138	*rho* = -0.166	*rho* = -0.084
	p = 0.975	*p* = 0.541	*p* = 0.02*	*p* = 0.01**	*p* = 0.162

Note. WSAS = Work and Social Adjustment Scale.
*$p < 0.05$; **$p < 0.01$; ***$p < 0.001$

Table 3
Regression Results for Psychological Distress

	Depression, Anxiety, and Stress Scales (DASS-21)		
	β	SE	*p*
Independent variables			
Psychological flexibility (AAQ-MCHL)	0.30	0.02	< .001
Income[1]			
$21,000–40,000	6.87	3.12	< .05
$41,000–80,000	1.44	2.74	.60
> $81,000	-1.35	2.60	.60

Note. AAQ-MCHL = Acceptance and Action Questionnaire–Managing Child Hearing Loss.
[1]Reference level was < $20,000.

study to explore the effect of psychological inflexibility for parents of children who are DHH. The findings of this study have important clinical implications for understanding barriers that interfere with how parents take action to care for their child's hearing related needs. Early identification of hearing loss provides a developmental advantage for children when early intervention is effectively implemented. It is not surprising that this process can be difficult for parents and providing support for parents who are experiencing difficult thoughts or feelings is a neglected aspect of the overall intervention care plan. McCreery

and Walker (2017) discuss the importance of addressing malleable factors within the intervention process in their model of cumulative auditory experience, and psychological flexibility is a factor that can be addressed.

The AAQ-MCHL can be used as a screening tool for psychological flexibility to help providers identify parents who are struggling. Other audiologic research has inferred a similar message regarding screening audiologic patients for cognitive decline (Shen et al., 2016; Sweetow, 2015) anxiety and depression (Muñoz, MacLeod, et al., 2015; Zöger et al., 2009), and even suicidality (Schwartzer and Parker, 2019; Zitelli & Palmer, 2018). Screening can help

Table 4
Regression Results for Functional Impairment

	Functional Impairment (WSAS)		
	β	SE	*p*
Independent variables			
Psychological flexibility (AAQ-MCHL)	0.36	0.04	< .001
Comorbid disability			
No[1]	-4.79	1.02	< .001
Child age	-0.32	0.09	< .001

Note. AAQ-MCHL = Acceptance and Action Questionnaire–Managing Child Hearing Loss; WSAS = Work and Social Adjustment Scale.
[1]Reference level was yes (comorbid disability was present).

providers as they partner with parents in determining education and support needs. The AAQ-MCHL is a brief questionnaire that can be administered during the hearing aid fitting process and periodically if there are concerns with engagement. The screening tool can serve as a bridge for audiologists to discuss problematic thoughts and perceptions parents are struggling with, and if indicated, a referral for counseling can be offered. Developing a therapeutic relationship through evidenced-based counseling strategies, such as Motivational Interviewing, can help parents to be open and honest about their challenges and may help audiologists understand when referrals to mental health professionals are necessary for parents. Furthermore, by understanding underlying challenges parents are experiencing, audiologists can target parent priorities and needs. For example, parent-to-parent support can be an important element in helping parents adjust (American Academy of Pediatrics, 2021) and support implementation of daily intervention tasks for hearing health care.

Study Limitations

The results of the correlations should be considered when interpreting these data. Spearman's correlations show each of the three measures used (psychological distress, psychological inflexibility, and functional impairment) to have weak to moderate significant correlations, indicating that these variables are not totally independent of one another, that is, to a certain extent they measured similar attributes. The non-statistically significant correlations found between use of technology and both psychological distress and functional impairment should be interpreted with caution. Prior research has found that self-reported device use is often overestimated by parents (Muñoz et al., 2014; Walker et al., 2013). Furthermore, the average age of the children represented by parents in this study is 7 years. Prior research has indicated that device use is typically lower for younger children (Walker et al., 2013). Thus, future research focused on a sample of parents of younger children (e.g., birth to 3 years) could provide further insights. Furthermore, the study design was cross-sectional and reflects only a moment in time. Future research to explore parent psychosocial well-being over time is needed. The sample primarily consisted of White, college-educated mothers, and does not reflect the multicultural demographics that make up the United States. Future research is needed to explore psychosocial well-being for a more diverse parent sample and a broader range for degree of hearing loss as 74% of the children had severe to profound degrees of hearing loss.

Conclusion

Parents of children who are DHH may experience psychosocial impacts related to the diagnosis and care of their children's hearing loss. This study found that psychological distress and functional impairment may be influenced by factors including psychological inflexibility, low income, the presence of other disabilities tangent to hearing loss, and younger child age. Clinicians serving families may need to provide additional and/or different support for parents in adapting to and managing their child's hearing loss. Screening tools can be used to identify when parents are experiencing psychosocial struggles so that underlying issues can be addressed through targeted support within the overall care plan. Parent well-being is important to the intervention process, as parent struggles can negatively influence daily hearing care habits, such as hours of hearing aid use, and this can negatively affect child outcomes.

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Intervention and Outcomes of Children in Different Types of Listening and Spoken Language Programs

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Abstract: This study explores the impact of the type and dosage of listening and spoken language (LSL) services on speech and language outcomes in children with cochlear implants or hearing aids in two LSL programs. Identical demographic variables were collected across the two programs for use in the statistical analyses. Speech and language outcomes were examined at ages 3 and 5 using standardized test measures. At age 3, significant differences in LSL outcomes existed between programs for children using cochlear implants but not for children using binaural hearing aids. However, at age 5, outcomes were similar between the different LSL programs for children with hearing aids and cochlear implants. Total hours of LSL services do not serve as a predictor of LSL outcomes at 5 years of age. However, early identification of hearing loss, early amplification, and early enrollment in an LSL program were highly influential factors affecting LSL outcomes at 3 and 5 years of age. Non-verbal IQ and maternal education levels also influence LSL outcomes. Children with earlier access to hearing technology and LSL intervention may need fewer hours of LSL services to achieve age-appropriate LSL outcomes. Overall, both of these LSL programs supported age-appropriate speech and language outcomes by age 5.

Keywords: Children; Hearing Loss; Cochlear Implants; Hearing Aids; Early Intervention; Listening and Spoken Language

Acronyms: CI = cochlear implant; DHH = deaf and hard of hearing; LME = linear mixed effect; LSL = Listening and Spoken Language; LSLS = Listening and Spoken Language Specialists; PCA = Principle Components Analysis; SES = socio-economic status; WNL = within normal limits

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For children who are deaf or hard of hearing (DHH), listening and spoken language (LSL) services focus on intelligible speech production, auditory comprehension, and receptive and expressive language abilities. Several factors are known to influence LSL outcomes of children who are DHH including age at identification of hearing loss, ages of hearing aid fitting and cochlear implantation, the child's non-verbal IQ, and caregiver socioeconomic status (SES) and education level (Ching et al., 2018; Geers et al., 2011; Leigh et al., 2016; Niparko et al., 2010). LSL intervention is critical to listening and spoken language outcomes; however, only a few studies have explored the impact of LSL intervention dose (i.e., frequency of intervention) on the LSL outcomes of children who are DHH.

Geers and colleagues (2019) evaluated the effect of LSL intervention dosage on LSL outcomes at 4–6 and 8–14 years of age for 50 children who were DHH and received services prior to 36 months of age. Between birth to 18 months, children received one-hour home visits from a LSL provider at least twice a month and a one-hour LSL session at the Moog Center for Deaf Education once a month. The sessions were primarily parent-centered with a focus on coaching the caregiver to facilitate the child's LSL development. Children older than 18 months attended a LSL class at the Moog Center for Deaf Education for 3.5 hours per day from 2 to 5 days a week depending on age. This LSL class included a one-hour individual LSL therapy session with the child, 2.5 hours of LSL group experiences, and weekly, 30-minute individual sessions

with the parent and child. Individual LSL services hours ranged from 0 to 279, and group LSL services hours ranged from 0 to 482. Over half the children achieved LSL outcomes within normal limits by 4 to 6 years of age, and over 70% achieved normal LSL outcomes by 8 to 14 years of age. Children who received more LSL hours between 0 to 36 months achieved higher LSL outcomes at 4 to 6 and 8 to 14 years of age when compared to children with fewer LSL hours, even after accounting for age at hearing aid fitting and intervention, speech perception ability, and non-verbal IQ. In addition, children with poorer speech perception scores were more likely to benefit from greater dosage of LSL services when compared to the LSL peers with better speech perception abilities.

Previous work by Scott and colleagues (2019) examined longitudinal growth of phonological awareness, letter-word identification, and expressive vocabulary skills in 56 children between the ages of 3 and 5 who were DHH. All children in the study were enrolled in DHH preschools and instructed by teachers of the deaf. Results showed significant improvements in literacy and vocabulary skills during the school year but not during summer break. For students with access to auditory cues, significant growth in phonological awareness was only observed during the school year as well. The results support intensive early education for children who are DHH and suggest additional schooling during the summer might be indicated.

In an earlier study, Moog and Geers (2010) examined the effect of age of LSL services and type of intervention on receptive and expressive language, vocabulary, and verbal reasoning at 5 to 6 years of age for 141 children with cochlear implants. Better LSL outcomes were found for earlier-implanted children (i.e., < 24 months) and those enrolled in weekly parent-infant LSL intervention by one year of age. In addition, children who were enrolled in LSL services for at least nine hours a week by two years of age had better LSL outcomes than those enrolled at a later age. Across LSL outcomes, 44% to 65% of children had standard scores within normal limits (WNL is defined as less than or equal to one SD from normative mean) by 5 to 6 years. Moreover, 71% of the children who attended a LSL education program from two to four years of age achieved outcomes WNL when compared to 41% who did not attend a LSL program until 3 years of age. Overall, better outcomes were reported for children with an earlier age at implant and earlier and more frequent LSL services.

In contrast, a recent study by Chu and colleagues (2019) found an inverse relationship between LSL intervention dosage and expressive language outcomes. In their study, they examined the effect of LSL services dosage on LSL outcomes of 42 children who used cochlear implants and received intervention up to 7 years of age. The average age at implantation was 1.9 years, and 14 children received implants before 12 months of age. In the study cohort, some children received home-based LSL services, whereas others received center-based services with individual dosages determined using a family-centered, evidenced-based approach. The results indicated that

children who received fewer LSL intervention hours were more likely to receive a cochlear implant at an earlier age, likely because earlier-implanted children were achieving better outcomes than later-implanted children. In addition, caregivers of children who were achieving age-appropriate LSL skills attended fewer LSL sessions. Overall, the authors report better LSL outcomes for earlier-implanted children (i.e., < 12 months) and the need for fewer LSL hours for earlier-implanted children.

Given the mixed findings and the limited number of studies exploring the dosage and type of LSL services on the outcomes of children who are DHH, additional research is warranted. The current study explores the type and dosage of LSL services received by children from two listening and spoken language programs with different approaches to intervention. The objectives of this study are to: (a) summarize LSL outcomes of the children participating in the two LSL programs, and (b) explore the relationship between type and dosage of LSL services and outcomes measured at 3 and 5 years of age.

Method

Study participants included children who received services from two LSL programs: the Moog Center for Deaf Education and Hearts for Hearing.

Moog Center for Deaf Education Description

The Moog Center for Deaf Education is an independent, not-for-profit audiology and LSL program that provides pediatric audiology and LSL services in an educational setting to children who are deaf or hard of hearing from birth to early elementary years and their families. Pediatric audiologists complete diagnostic assessments (e.g., auditory brainstem response testing, otoacoustic emissions, middle ear measurements, behavioral audiologic assessment) to evaluate auditory function of children who have been identified with hearing loss or referred to the Moog Center for concerns regarding auditory function and/or speech and language delay. Hearing aids are fitted as soon as possible following identification of hearing loss and referral. Recommendation for cochlear implantation is made for children who have severe to profound hearing loss and whose needs are not adequately supported by hearing aid use.

For children who are birth to 18 months of age, the Moog Center provides one-hour home visits or online (tele-intervention) sessions led by a certified teacher of the deaf at least once a month and a center-based session once a month. These sessions include the provision of information to parents/caregivers, coaching of parents/caregivers to facilitate their children's individual speech, listening, and spoken language outcomes, and engagement in activities focused on LSL strategies designed to support listening and spoken language development in their children's daily lives. For children who are 18 months to 3 years of age, the Moog Center provides a center-based LSL program in addition to their home visits or tele-intervention sessions, as described above. Children may attend the center-based program 2 to 5 days a week depending

on age, developmental factors, and family factors. The center-based program includes 60-minute individual sessions which focus on the development of speech, language, and listening skills, and 2.5-hour group sessions which focus on early cognitive, motor, and social skills development. For children who are 3 to 5 years of age, the Moog Center offers a Preschool program. Services in the Preschool are provided by certified teachers of the deaf and speech-language pathologists, all of whom are Listening and Spoken Language Specialists (LSLS) or seeking certification, along with early childhood educators. Children in the Preschool may receive 3 hours of individualized LSL services and 2 hours of small-group instruction daily. Preschool sessions focus on the development of individualized speech, language, and listening skills, as well as math, early literacy, and social skills. In addition, optional weekly parent/caregiver coaching, support group, and parent educational sessions are offered.

Hearts for Hearing Description

Hearts for Hearing is an independent, not-for-profit audiology and LSL program that provides pediatric audiology and LSL therapy for children with hearing loss. Pediatric audiologists complete diagnostic assessments (e.g., auditory brainstem response testing, otoacoustic emissions, middle ear measurements, behavioral audiologic assessment) to evaluate auditory function of children who do not pass newborn hearing screening or are referred for concerns regarding auditory function and/or speech and language delay. In line with the center's mission, hearing aids are fitted within days of the diagnosis of hearing loss, and cochlear implants are provided for children who have severe to profound hearing loss and whose needs are not adequately supported by hearing aid use.

Hearts for Hearing provides weekly or monthly, one-hour LSL therapy sessions led by an LSL clinician (who is either a LSLS or pursuing certification) in person or via tele-intervention sessions. Sessions include information for parents, parent coaching, and activities to facilitate LSL development. A monthly, one-hour, parent-infant group, led by two LSL specialists and a pediatric audiologist, is provided for children birth to 24 months of age. The group provides information on hearing loss and LSL development as well as peer support for caregivers of infants with hearing loss. A two-hour, parent-toddler class, led by a LSLS and an early childhood educator, is provided for children who are 2 to 3 years old. This class includes activities to promote and enrich the child's listening and spoken language. Finally, a 3-year-old class, team-taught by an early childhood educator and a speech-language pathologist pursuing LSLS certification, is offered for children ages 3 to 4 years. The class of 8 to 10 children is offered twice a week for 2.5 hours a day. Most children attend the class for up to one year, but children may participate longer if they have language delays affecting potential success in a mainstream preschool setting.

Study Participants

The enrollment databases and clinical records were reviewed at the Moog Center for Deaf Education and Hearts for Hearing to identify children who had received services at each program. Children who met the following criteria were included in this study.

Inclusion Criteria

- Bilateral hearing loss with a pure tone average (mean air conduction thresholds 500, 1000, and 2000 Hz) poorer than 25 dB HL in the better ear.
- Children with congenital hearing loss or perilinguistic hearing loss identified by 36 months of age.
- Children who received services at one of the two programs and for whom results are available for standardized assessments of LSL aptitude at 3 and/or 5 years of age.
- Children who regularly participated in the LSL programs of the respective study sites as defined by an attendance rate of at least 50% (i.e., attended at least 50% of scheduled appointments).
- Children who use air conduction hearing aids, bone conduction devices, and/or cochlear implants.
- Children who communicate primarily via listening and spoken language and who are native speakers of American English.

Exclusion Criteria

- Children clinically diagnosed with neurocognitive disabilities or other disabilities that would adversely impact LSL development (e.g., autism spectrum disorder, apraxia, dysarthria, selective mutism, etc.).
- English spoken as a second language.
- Non-verbal IQ standard score poorer than 70.
- Unilateral hearing loss.

A total of 218 children met the listed inclusion criteria, with 111 children from the Moog Center, 47 of whom used binaural hearing aids and 64 who used cochlear implants. From Hearts for Hearing, 107 children were included, 61 of whom used binaural hearing aids and 46 who used cochlear implants. Across sites, the cohort of children with cochlear implants included 19 children with a bimodal approach (hearing aid + cochlear implant), 5 children using a unilateral cochlear implant, and 86 children using bilateral cochlear implants.

The study participants' scores from standardized measures of listening and spoken language aptitude administered at 3 and 5 years of age were obtained from their personal files at the study programs and from the OPTION Schools, Inc. Listening and Spoken Language Data Repository (LSL-DR; i.e., REDCap database; Bradham et al., 2018). The Western Institutional Review Board provided regulatory approval for this study. The following standardized measures were used to evaluate the LSL outcomes of the children in this study.

Language Assessment

- Clinical Evaluation of Language Fundamentals Preschool-2 (CELF P-2; Semel et al., 2004).
  - The First Edition of this assessment was used in some early data.
- Clinical Evaluation of Language Fundamentals–Fifth Edition (CELF-5; Wiig et al., 2013).
  - The Fourth Edition of this assessment was used in some early data.
- Preschool Language Scales–Fifth Edition (PLS-5; Zimmerman et al., 2011).

Vocabulary Assessment

- Expressive Vocabulary Test–Third Edition (EVT-3; Williams, 2018).
  - The First and Second Editions of this assessment were used in some early data.
- Expressive One-Word Picture Vocabulary Test–Fourth Edition (EOWPVT-4; Brownell, 2010a).
- Receptive One-Word Picture Vocabulary Test–Fourth Edition (ROWPVT-4; Brownell, 2010b).
- Peabody Picture Vocabulary Test–Fourth Edition (PPVT-4; Dunn & Dunn, 2007).
  - The Third Edition of this assessment was used in some early data.

Speech Production/Articulation

- Goldman-Fristoe Test of Articulation 3 (GFTA-3; Goldman & Fristoe, 2015).
  - The Second Edition of this assessment was used in some early data.
- Clinical Assessment of Articulation and Phonology–Second Edition (CAAP-2; Secord & Donohue, 2013).

Of note, the children who were evaluated at 5 years of age also were evaluated at 3 years of age. However, not all the children who were evaluated at 3 years of age were evaluated at 5 years of age (i.e., some children were no longer enrolled in intervention at 5 years of age, and as a result, were not evaluated).

For each of the standardized vocabulary and language measures, test items increase in difficulty throughout the test, and assessment continues until the child encounters a ceiling score determined by a specified sequence of incorrect responses. Each measure yields a standard score based on normative data obtained from a group of age-matched, typically-developing peers with normal hearing. The group mean obtained from the normative data is set to 100, and each standard deviation (SD) from that mean is represented by +/-15 points (i.e., 85 and 115 are +/- 1 SD from the mean, respectively). For additional information pertaining to a description of the measures used to evaluate LSL outcomes in this study, the reader is referred to the citations associated with each test listed above.

The children's non-verbal intelligence quotients (IQs) were evaluated with the Central Institute for the Deaf Preschool Performance Scale (CID-PPS; Geers & Lane, 1984), Kaufman Brief Intelligence Test (K-BIT; Kaufman & Kaufman, 1990), Kaufman Brief Intelligence Test–2nd Edition (KBIT-2; Kaufman & Kaufman, 2004), Primary Test of Nonverbal Intelligence (PTONI; Ehrler & McGhee, 2008), Weschler Intelligence Scale for Children–5th Edition (WISC-V; Wechsler, 2014), Weschler Preschool and Primary Scale of Intelligence–3rd Edition (WPPSI-III; Wechsler, 2002), and Weschler Preschool and Primary Scale of Intelligence–4th Edition (WPPSI-IV; Wechsler, 2012). As with the standardized measures of LSL outcomes, the non-verbal IQ assessments administered in this study were norm-referenced with a mean of 100 and +/-1 SD corresponding to 15 points.

Statistical Analysis

Similar to a previous study of LSL outcomes (e.g., Ching et al., 2018), separate statistical analyses were conducted for children who used binaural hearing aids and those who used cochlear implants for at least one ear. Principal Components Analysis (PCA) was used to compute eigenvalues for the two different test measures and confirmed the CELF and PLS loaded onto the same expressive language factor (only the first principal component exceeded 1), ensuring equivalence of the different measures. To reduce Type I errors, PCA was also used to create a composite score for expressive language (PLS/CELF, EOWVT) outcomes (Davidson et al., 2019; Strube, 2003; Tomblin et al., 2015). The expressive language composite score had a mean of 100 and a standard deviation of 15. Similar to the expressive language measures, PCA confirmed scores from the PLS/CELF and PPVT loaded onto the same factor, and a composite receptive language score was computed for each child (mean 100, standard deviation of 15).

Separate linear mixed-effect (LME) regression analyses were performed to examine expressive and receptive language, core language, and articulation outcomes in (a) children with cochlear implants at 3 and 5 years of age, and (b) children with hearing aids at 3 and 5 years of age. In the cochlear implant (CI) analyses, *Cochlear Implant Recipient* was treated as a random effect to control for baseline differences across pediatric patients. *Mother's Education Level* (high school, some college, college); *Nonverbal IQ; Age at Hearing Aid* (months); and *Age at 1st CI* (months) were included in the models to control for important demographic and audiological characteristics. To assess the effects of LSL intervention on language outcomes, *LSL Program* (Moog Center vs Hearts for Hearing); *Age of Enrollment in LSL Program* (months*); Intervention Hours from 0–3 Years of Age* (when applicable)*; Intervention Hours from 0–5 Years of Age* (when applicable) and two and three-way interactions between intervention variables were also included in the models as fixed effects.

In the hearing aid analyses, *LSL Participant* was treated as a random effect to control for baseline differences across pediatric patients. *Mother's Education Level* (high school, some college, college); *Nonverbal IQ; Age at Hearing*

Aid (months); and *Degree of Hearing Loss* were included in the models to control for important demographic and audiological characteristics. To assess the effects of LSL intervention on language outcomes, *Program* (Moog Center vs Hearts for Hearing); *Age of Enrollment in LSL program* (months); *Intervention Hours from 0–3 Years of Age* (when applicable); *Intervention Hours from 0–5* (when applicable) and interactions between intervention variables were also included in the models as fixed effects. For the cochlear implant and hearing aid analyses, full models were run with all fixed effects and interactions. If the interactions were not significant, they were removed from the model. Fixed effects were assessed using a significance α = 0.05. Regression diagnostics were performed for each analysis and all assumptions were met.

Results

Comparison Demographic Characteristics

The demographics for the study participants are provided in Table 1. Items in bolded font indicate a statistically significant difference in demographic variables between children from the Moog Center and Hearts for Hearing.

As shown in Table 1, the Moog Center group contained a greater percentage of children with severe to profound hearing loss who were using hearing aids. Additionally, children using cochlear implants were fitted with hearing aids at significantly earlier ages at Hearts for Hearing.

A summary of the age of enrollment and hours of LSL services received by the children in the Moog Center and Hearts for Hearing programs is provided in Table 2. Items in bolded font indicate statistically significant differences in the LSL services received by children from the Moog Center and Hearts for Hearing. As shown in Table 2, for cochlear implant recipients, children enrolled in the LSL program started earlier at Hearts for Hearing relative to their counterparts at the Moog Center. There was no difference in the age of enrollment at Hearts for Hearing and the Moog Center for children who were using binaural hearing aids. Moreover, children at the Moog Center received significantly more LSL hours from birth to 3 years of age and from birth through 5 years of age than their Hearts for Hearing counterparts, which was true for both those with binaural hearing aids and those who received cochlear implants.

Table 1
Demographic Information for the Study Participants with Hearing Aids (HA) and Cochlear Implants (CI)

Intervention	Hearing Aids		Cochlear Implants	
Treatment Group	Hearts for Hearing (*n* = 61)	Moog Center (*n* = 47)	Hearts for Hearing (*n* = 46)	Moog Center (*n* = 64)
Maternal Education				
High school	23.3%	2.9%	17.4%	15.0%
Some College	11.7%	34.3%	10.9%	20.0%
College	65.0%	62.8%	71.7%	65.0%
SES				
<$25,000	0%	4.5%	0%	6.3%
$25,000–$49,999	21.3%	18.2%	15.2%	18.8%
$50,000–$74,999	31.1%	9.1%	26.1%	15.6%
$75,000–$99,999	23%	18.2%	23.9%	15.6%
$100,000+	24.6%	50.0%	34.8%	43.8%
Mean Nonverbal IQ	106.6 (13.3)	109.6 (13.1)	106.2 (10.3)	110.4 (11.5)
Mean Age HA (months)	10.7 (12.9)	10.3 (10.2)	**6.2 (8.3)**	**8.9 (7.6)**
Mean Age 1st CI (months)	.	.	22.0 (16.5)	22.9 (16.5)
Degree of Hearing Loss			.	.
Mild	24.6%	8.5%	.	.
Moderate	41.0%	19.1%	.	.
Moderate–Severe	29.5%	14.9%	.	.
Severe–Profound	4.9%	57.5%		

Note. IQ = intelligence quotient; SES = socioeconomic status. Numbers in bold represent significant program differences according to *t*-test (*p* < 0.05). Numbers in parentheses represent standard deviation.

Table 2

Summary of Early Intervention Ages and Hours by Program

Intervention	Hearing Aids		Cochlear Implants	
	Hearts for Hearing	Moog Center	Hearts for Hearing	Moog Center
Mean Age of Enrollment (Months)	12.9 (14.0)	13.5 (12.7)	7.0 (8.6)	20.0 (13.9)
Mean Total Hours Per Child from 0–3 Years	49.6 (39.5)	364.2 (198.6)	75.9 (49.2)	356.0 (245.8)
Mean Total Hours Per Child from 0–5 Years	103.4 (76.7)	1350.9 (532.5)	163.9 (105.1)	1547.9 (529.7)

Note. Numbers in bold represent significant program differences according to *t*-test ($p < 0.05$). Numbers in parentheses represent standard deviation.

Children Using Cochlear Implants

The following results are for children using cochlear implants. LME regression analyses were used to analyze how LSL intervention factors contributed to expressive language scores of children at Hearts for Hearing and the Moog Center when controlling for important demographic and audiological variables for children using cochlear implants. Table 3 shows the regression weights and the associated significance values for predicting expressive language outcomes at 3 years and 5 years of age. At 3 years of age, earlier age of hearing aid fit, higher maternal education, and greater amount of LSL intervention hours were associated with a significant increase in expressive

language outcomes (Table 3). At age 3, children receiving intervention at the Moog Center were predicted to have expressive language scores 12.7 points lower than children at Hearts for Hearing. However, at 5 years of age, none of the demographic, program, or intervention factors were predictive of expressive language outcomes, meaning children at both programs were predicted to have similar expressive language outcomes at age 5. Figure 1 shows the distribution of the expressive language scores for the Moog Center and Hearts for Hearing groups at 3 and 5 years of age for children using cochlear implants. Between programs, expressive language outcomes were significantly different at age 3, but not at age 5.

Table 3

Regression Analysis Results of Cochlear Implant (CI) Expressive Language Outcomes at 3 and 5 Years of Age

Expressive Language	3 years of age ($R^2 = 0.37$)			5 years of age ($R^2 = 0.37$)		
Effect	Parameter	*F* Value	*p* Level	Parameter	*F* Value	*p* Level
Intercept	85.9	4216.7	< .0001	70.8	4075.2	< .0001
Maternal Education		4.44	.02		0.22	.81
High School	-10.9			-6.24		
Some college	-12.5			-4.62		
College graduate	0			0		
Nonverbal IQ	0.19	1.6	.21	0.32	2.45	.13
Age HA (months)	-0.5	9.4	.003	-0.3	2.9	.1
Age 1st CI (months)	0.02	0.04	.84	0.02	0.03	.86
Treatment Group		3.7	.06		0.04	.84
Hearts for Hearing	0			0		
Moog Center	-12.7			4.61		
Age enrollment LSL	-0.03	3.7	.06	-0.27	2.2	.15
LSL Intervention Hours	0.02	4.5	.04	-0.001	.07	0.79

Note. HA = hearing aid; IQ = intelligence quotient; LSL = listening and spoken language program. Parameter represents the Beta coefficient from the analysis.

Figure 1

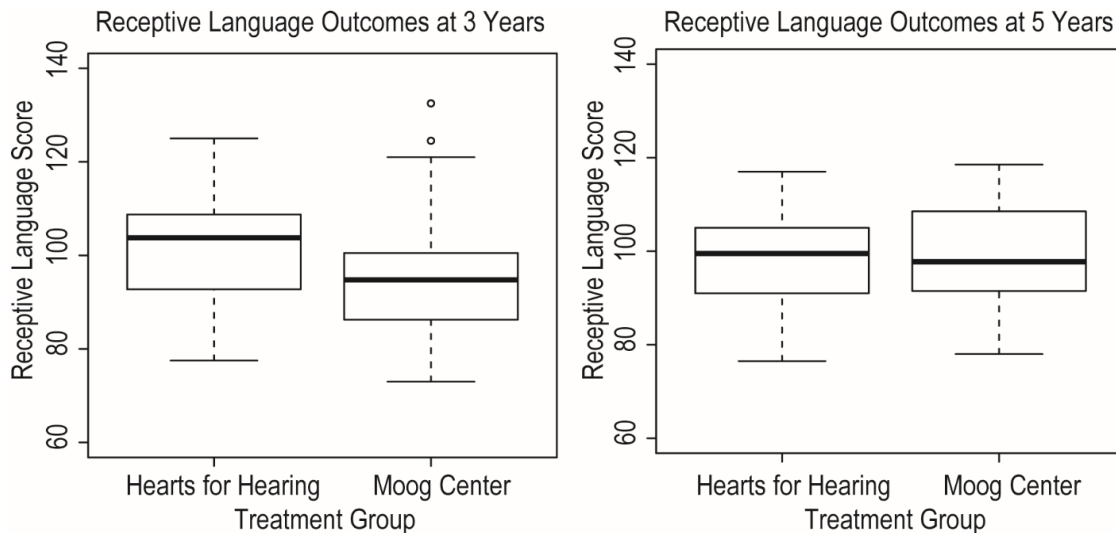*Expressive Language Scores for Children with Cochlear Implants at 3 and 5 Years Old*
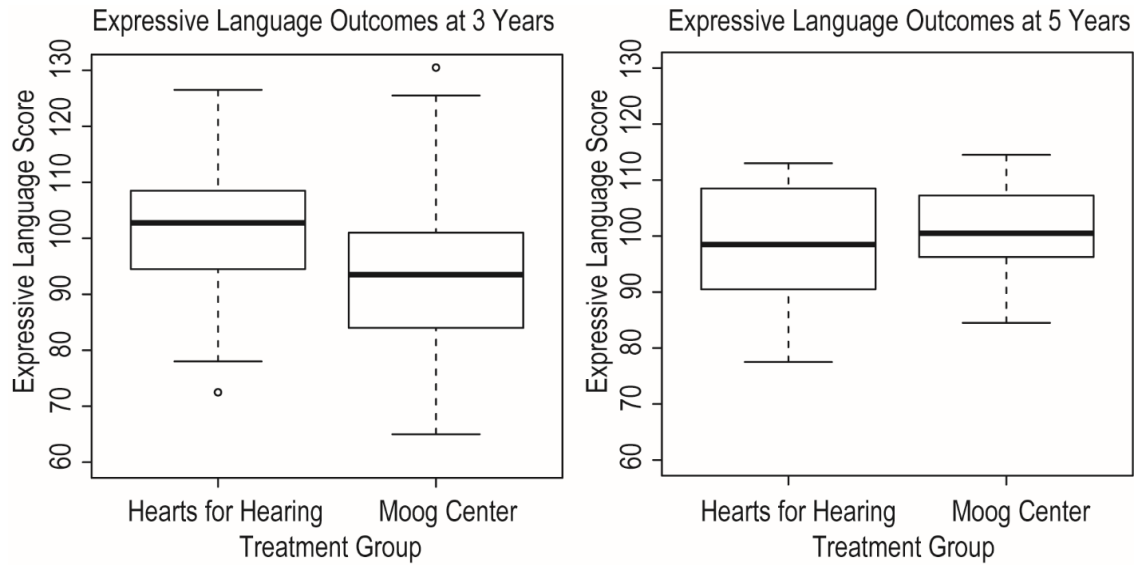
Note. The median of the distribution is denoted by the horizontal black line in the box, and the edges of each box represent the 25th and 75th percentiles of the distribution. The whiskers extend to the minimum and maximum individual data points that are not outliers. Outliers, denoted as circles, are values greater than 1.5 times the interquartile range.

LME regression analyses were used to analyze how LSL intervention factors contributed to receptive language scores in children at Hearts for Hearing and the Moog Center. The regression weights and associated significance values for predicting receptive language outcomes at 3 years and 5 years of age for children using cochlear implants are displayed in Table 4. Higher maternal education years, higher nonverbal IQ, and earlier age of hearing aid fit were significant predictors of receptive language outcomes at 3 years of age (Table 4). Earlier age of enrollment in LSL intervention, and higher number of LSL intervention hours were associated with better receptive language outcomes at age 3, but these effects just failed to reach significance ($p = 0.06$). Similar to expressive language outcomes, none of the factors that were significant at 3 years of age were significant predictors of receptive language outcomes at 5 years of age. Figure 2 shows the distribution of the receptive language scores for the Moog Center and Hearts for Hearing groups at 3 and 5 years of age for children with cochlear implants. Between programs, receptive language outcomes were not significantly different at age 3 or age 5.

Figure 2

Receptive Language Scores for Children with Cochlear Implants at 3 and 5 Years Old

Note. The median of the distribution is denoted by the horizontal black line in the box, and the edges of each box represent the 25th and 75th percentiles of the distribution. The whiskers extend to the minimum and maximum individual data points that are not outliers. Outliers, denoted as circles, are values greater than 1.5 times the interquartile range.

Table 4

Regression Analysis Results of Cochlear Implant (CI) Receptive Language Outcomes at 3 and 5 Years of Age

Receptive Language	3 years of age ($R^2 = 0.43$)			5 years of age ($R^2 = 0.09$)		
Effect	Parameter	F Value	p Level	Parameter	F Value	p Level
Intercept	81.02	6672.7	< .0001	94.03	3279.2	< .0001
Maternal Education		9.3	.0003		0.54	.59
High School	-10.8			-4.2		
Some college	-12.6			-0.36		
College graduate	0			0		
Nonverbal IQ	0.26	4.7	.04	0.08	0.19	.67
Age HA (months)	-0.43	7.13	.009	-0.2	0.38	.54
Age 1st CI (months)	-0.09	2.4	.13	-0.04	0.07	.79
Treatment Group		3.2	.08		0.22	.64
Hearts for Hearing	0			0		
Moog Center	-8.9			-9.6		
Age enrollment LSL	-0.06	3.7	.06	0.04	0.07	.8
LSL Intervention Hours	0.02	3.5	.06	0.005	0.8	.38

Note. HA = hearing aid; IQ = intelligence quotient; LSL = listening and spoken language program. Parameter represents the Beta coefficient from the analysis.

LME regression analyses were used to analyze how LSL intervention factors contributed to core language outcomes in children at Hearts for Hearing and the Moog Center at age 3 and age 5 for children using cochlear implants. Table 5 shows the regression weights and the associated significance values for predicting core language outcomes at 3 years and 5 years of age. Higher maternal education years and earlier age of hearing aid fit were significant predictors of language core outcomes at 3 years of age. Children receiving intervention at Hearts for Hearing were predicted to have language core scores 15.3 points higher than children at the Moog Center at age 3. However, the program was not a significant predictor of language core scores at age 5, suggesting children at the Moog Center and Hearts for Hearing performed similarly at age 5. Figure 3 shows the distribution of the core language scores for the Moog Center and Hearts for Hearing groups at 3 and 5 years of age for children using cochlear implants. Between programs, core language outcomes were significantly different at age 3, but not at age 5.

Table 6 displays the regression coefficients and associated *p* values for the fixed effects for predicting articulation outcomes at 3 and 5 years of age for children using cochlear implants. At age 3, earlier age of hearing aid fit, higher nonverbal IQ, and program were significant predictors of higher articulation outcomes. Children receiving intervention at Hearts for Hearing were predicted to have articulation outcomes 17.4 points higher than children receiving intervention at the Moog Center at age

3. However, at 5 years of age, there were no significant predictors of articulation outcomes. Figure 4 shows the distribution of the articulation scores for the Moog Center and Hearts for Hearing groups at 3 and 5 years of age for children using cochlear implants. Between programs, articulation outcomes were significantly different at age 3, but not at age 5.

Children Using Binaural Hearing Aids

The following results are for children using hearing aids. LME regression analyses were used to analyze how LSL intervention factors contributed to expressive language scores in children at Hearts for Hearing and the Moog Center when controlling for important demographic and audiological variables for children using binaural hearing aids. Table 7 shows the regression weights and the associated significance values for predicting expressive language outcomes at 3 years and 5 years of age. At 3 years of age, higher number of LSL intervention hours was associated with higher expressive language outcomes. Higher nonverbal IQ and better hearing thresholds were associated with higher expressive language outcomes at age 3 as well (Table 7). Similarly, higher nonverbal IQ was associated with higher expressive language outcomes at age 5. Figure 5 shows the distribution of the expressive language scores for the Moog Center and Hearts for Hearing groups with binaural hearing aids at 3 and 5 years of age. Figure 6 shows a scatterplot of the expressive language scores as a function of hours of LSL services received by 3 years of age. As shown in Figure 6, a statistically significant

Table 5

Regression Analysis Results of Cochlear Implant (CI) Language Core Outcomes at 3 and 5 Years of Age

Language Core	3 years of age ($R^2 = 0.35$)			5 years of age ($R^2 = 0.2$)		
Effect	Parameter	*F* Value	*p* Level	Parameter	*F* Value	*p* Level
Intercept	81.7	3424.9	< .0001	119.5	1723.5	< .0001
Maternal Education		5.0	.009		0.7	.51
High School	-10.75			-8.13		
Some college	-10.7			-2.4		
College graduate	0			0		
Nonverbal IQ	0.21	2.3	.13	-0.09	0.35	.51
Age HA (months)	-0.52	7.6	.008	-0.16	1.2	.29
Age 1st CI (months)	-0.05	0.5	.47	-0.22	1.6	.21
Treatment Group		10.8	.002		0.73	.39
Hearts for Hearing	0			0		
Moog Center	-15.3			-12.7		
Age enrollment LSL	-0.02	1.47	.22	-0.21	1.2	.29
LSL Intervention Hours	0.02	1.95	.17	0.006	0.7	.4

Note. HA = hearing aid; IQ = intelligence quotient; LSL = listening and spoken language program. Parameter represents the Beta coefficient from the analysis.

Figure 3

Core Language Scores for Children with Cochlear Implants at 3 and 5 Years Old
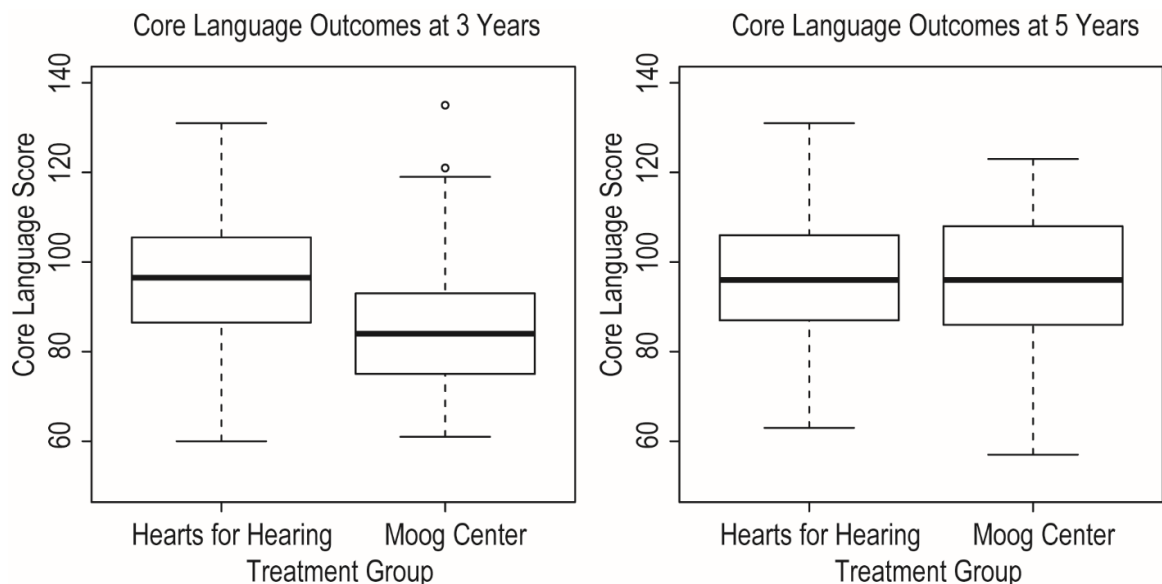
Note. The median of the distribution is denoted by the horizontal black line in the box, and the edges of each box represent the 25th and 75th percentiles of the distribution. The whiskers extend to the minimum and maximum individual data points that are not outliers. Outliers, denoted as circles, are values greater than 1.5 times the interquartile range.

Table 6

Regression Analysis Results of Cochlear Implant (CI) Articulation Outcomes at 3 and 5 Years of Age

Articulation	3 years of age $(R^2 = 0.40)$			5 years of age $(R^2 = 0.13)$		
Effect	Parameter	*F* Value	*p* Level	Parameter	*F* Value	*p* Level
Intercept	58.9	2542.9	< .0001	80.08	1213.5	< .0001
Maternal Education		2.16	.12		1.07	.36
High School	-7.7			8.5		
Some college	-8.2			5.7		
College graduate	0			0		
Nonverbal IQ	0.34	3.99	.05	0.09	0.05	.82
Age HA (months)	-0.5	7.9	.007	0.14	0.001	.97
Age 1st CI (months)	0.04	.03	.86	-0.05	0.05	.83
Treatment Group		16.8	.0002		1.3	.26
Hearts for Hearing	0			0		
Moog Center	-17.4			-18.5		
Age enrollment LSL	-0.04	0.04	.84	0.08	0.08	.77
LSL Intervention Hours	0.02	0.01	.9	0.008	0.93	.34

Note. HA = hearing aid; IQ = intelligence quotient; LSL = listening and spoken language program. Parameter represents the Beta coefficient from the analysis.

Figure 4

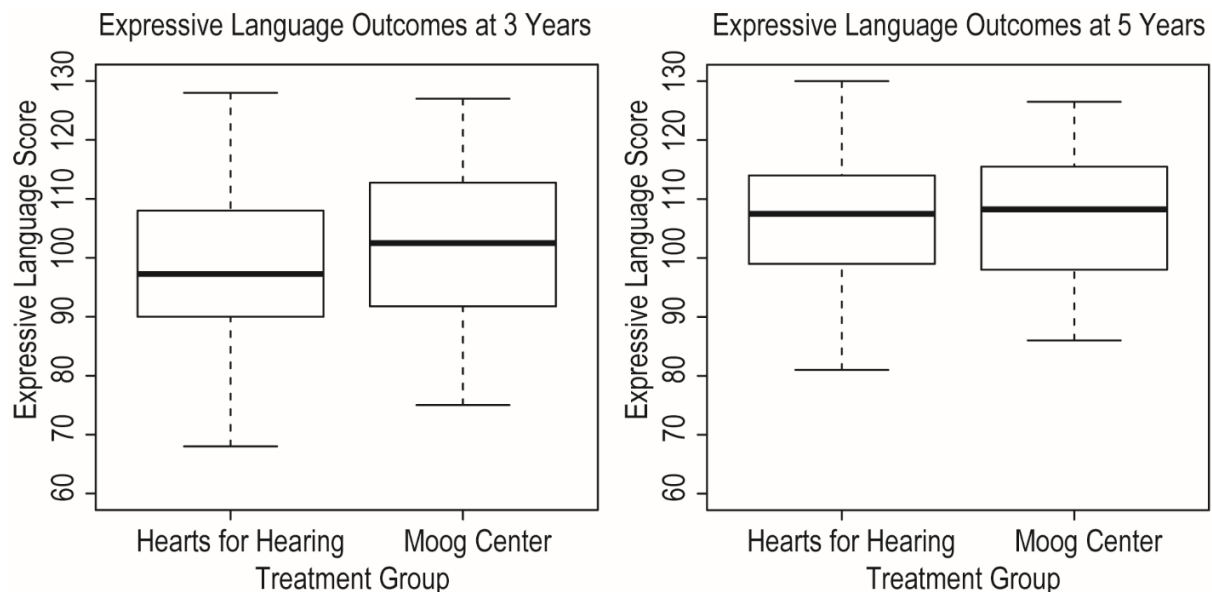*Articulation Scores for Children with Cochlear Implants at 3 and 5 Years Old*
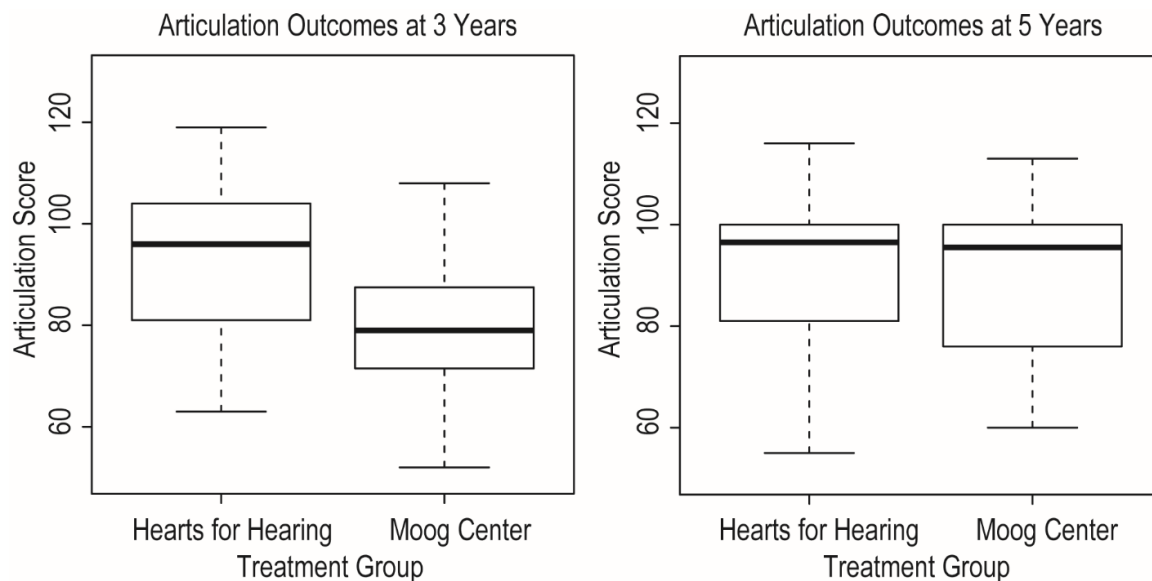
Note. The median of the distribution is denoted by the horizontal black line in the box, and the edges of each box represent the 25th and 75th percentiles of the distribution. The whiskers extend to the minimum and maximum individual data points that are not outliers. Outliers are values greater than 1.5 times the interquartile range.

Table 7

Regression Analysis Results of Hearing Aid (HA) Expressive Language Outcomes at 3 and 5 Years of Age

| Expressive Language	3 years of age ($R^2 = 0.28$)			5 years of age ($R^2 = 0.26$)		
Effect	Parameter	*F* Value	*p* Level	Parameter	*F* Value	*p* Level
Intercept	84.3	740.2	< .0001	98.6	5768.5	< .0001
Maternal Education		2.5	.08		2.4	.09
High School	-6.2			-6.1		
Some college	-5.0			-8.7		
College graduate	0			0		
Nonverbal IQ	0.14	5.3	.02	0.13	4.9	.03
Age HA (months)	-0.07	0.73	.39	-0.09	1.4	.25
Degree Hearing Loss		3.4	.02		0.47	.7
Mild	0			0		
Moderate	-3.1			-2.6		
Moderate-Severe	-11.12			-5.2		
Severe-Profound	-6.5			-0.8		
Treatment Group		0.05	.82		0.0001	.99
Hearts for Hearing	0			0		
Moog Center	-1.45			-14.3		
Age enrollment LSL	0.06	0.12	.72	-0.04	0.13	.72
LSL Intervention Hours	0.02	6.4	.01	0.009	2.2	.15

Note. IQ = intelligence quotient; LSL = listening and spoken language program. Parameter represents the Beta coefficient from the analysis.

Figure 5

Expressive Language Scores for Children with Hearing Aids at 3 and 5 Years Old

Note. The median of the distribution is denoted by the horizontal black line in the box, and the edges of each box represent the 25th and 75th percentiles of the distribution. The whiskers extend to the minimum and maximum individual data points that are not outliers. Outliers are values greater than 1.5 times the interquartile range.

Figure 6

Expressive Language Scores as a Function of Hours of LSL Services Received by Children with Hearing Aids by 3 Years of Age
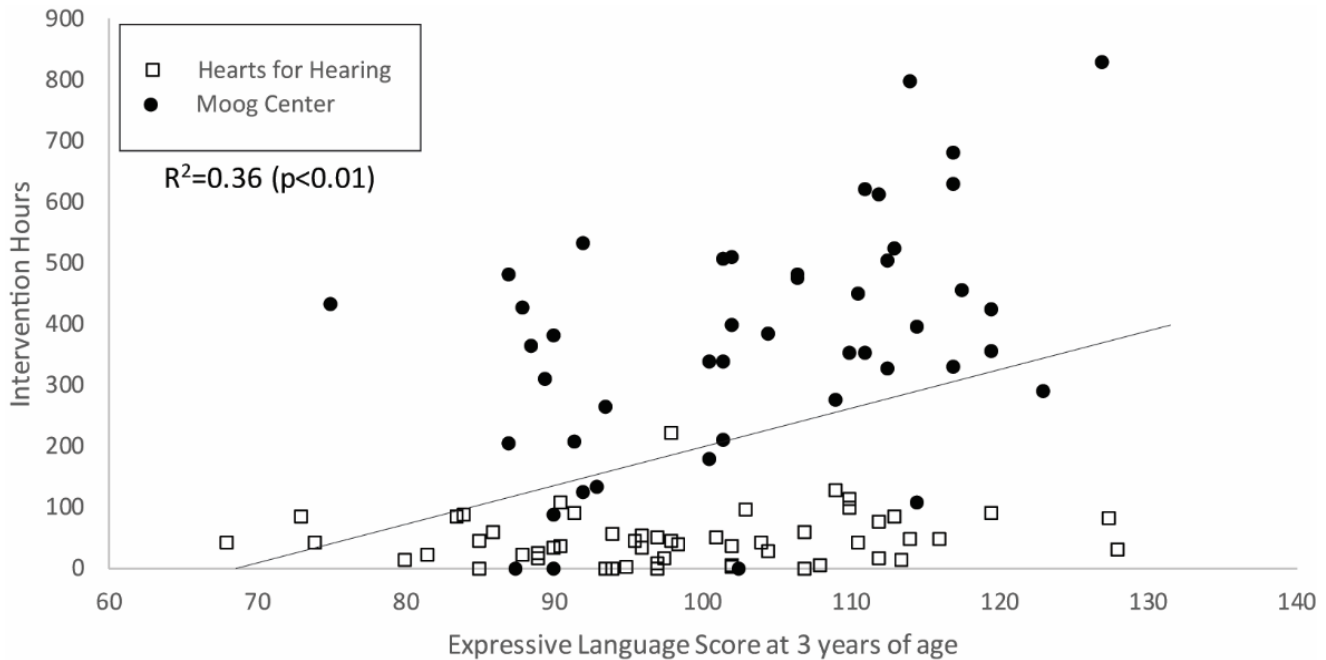

Note. R^2 represents the correlation between intervention hours and expressive language scores across both treatment groups. LSL = Listening and Spoken Language.

but weak positive correlation exists between expressive language at 3 years of age and number of LSL hours from birth to 3 years of age for children using binaural hearing aids across treatment groups. However, this relationship is likely driven by the Moog Center group as the correlation between LSL hours and expressive language increases when only children from the Moog Center are included in the analysis (Figure 6). Figure 7 shows the number of LSL intervention hours by degree of hearing loss. As shown, children with severe to profound hearing loss received significantly more hours of LSL intervention than their peers with lesser degrees of hearing loss. Between programs, expressive language outcomes were not significantly different at age 3 or age 5.

LME regression analyses were used to analyze how LSL intervention factors contributed to receptive language scores in children at Hearts for Hearing and the Moog Center. The regression weights and associated significance values associated with receptive language outcomes at 3 years and 5 years of age for children using binaural hearing aids are displayed in Table 8. LSL services, maternal education and nonverbal IQ were the only significant predictors of receptive language at age 3, and nonverbal IQ was the only significant predictor of receptive language at age 5 (Table 8). Figure 8 shows the distribution of the receptive language scores for the Moog Center and Hearts for Hearing groups with hearing aids at 3 and 5 years of age for children using binaural hearing aids. Between programs, receptive language outcomes were not significantly different at age 3 or age 5.

Figure 7

LSL Intervention Hours Received by 3 Years of Age as a Function of Degree of Hearing Loss for Children with Hearing Aids
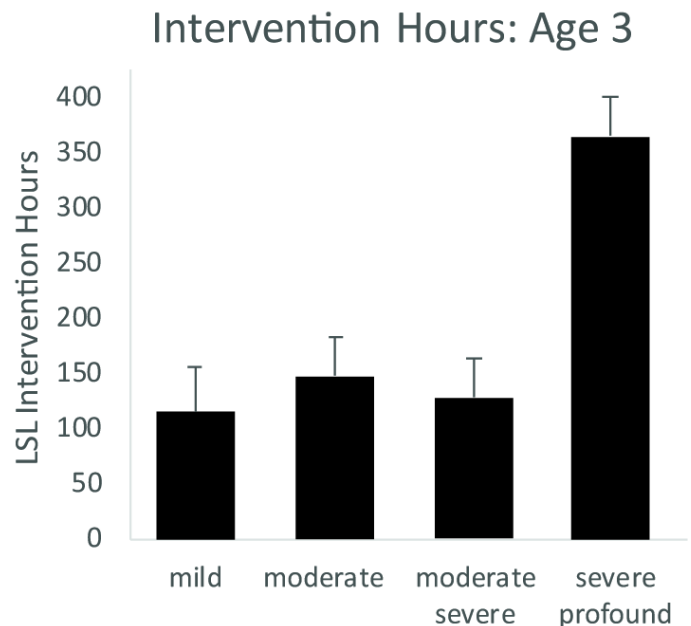

Note. Error bars represent standard error of the mean. LSL = Listening and Spoken Language.

Table 8

Regression Analysis Results of Hearing Aid (HA) Receptive Language Outcomes at 3 and 5 Years of Age

Receptive Language	3 years of age ($R^2 = 0.23$)			5 years of age ($R^2 = 0.25$)		
Effect	Parameter	*F* Value	*p* Level	Parameter	*F* Value	*p* Level
Intercept	94.01	7453.6	< .0001	94.8	6325.7	< .0001
Maternal Education		3.9	.02		0.48	.62
High School	-8.3			-2.3		
Some college	-5.2			-5.0		
College graduate	0			0		
Nonverbal IQ	0.13	4.4	.04	0.17	5.62	.02
Age HA (months)	-0.13	1.1	.31	-0.004	0.46	.49
Degree Hearing Loss		1.4	.26		2.17	.10
Mild	0			0		
Moderate	-4.6			-8.1		
Moderate-Severe	-8.04			-9.2		
Severe-Profound	-7.5			-11.5		
Treatment Group		0.13	.72		0.55	.46
Hearts for Hearing	0			0		
Moog Center	-5.8			-4.2		
Age enrollment LSL	0.11	0.02	.89	-0.07	0.25	.62
LSL Intervention Hours	0.02	3.5	.06	0.004	0.59	.44

Note. IQ = intelligence quotient; LSL = listening and spoken language program. Parameter represents the Beta coefficient from the analysis.

Figure 8

Receptive Language Scores for Children with Hearing Aids at 3 and 5 Years Old.
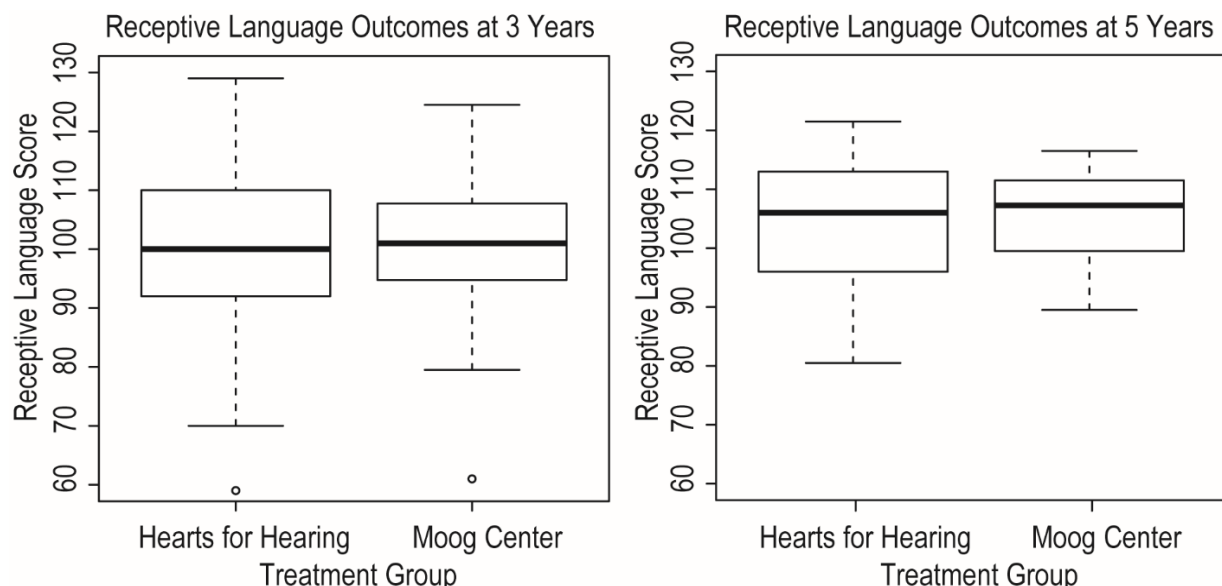
Note. The median of the distribution is denoted by the horizontal black line in the box, and the edges of each box represent the 25th and 75th percentiles of the distribution. The whiskers extend to the minimum and maximum individual data points that are not outliers. Outliers, denoted as circles, are values greater than 1.5 times the interquartile range.

LME regression analyses were used to analyze how LSL intervention factors contributed to core language outcomes in children at Hearts for Hearing and the Moog Center at age 3 and age 5 for children using binaural hearing aids. Table 9 shows the regression weights and the associated significance values for predicting core language outcomes at 3 years and 5 years of age. At age 3, higher maternal education was associated with higher core language outcomes, whereas greater degrees of hearing loss were associated with significantly poorer core language outcomes. At age 5, earlier age of hearing aid fitting and higher nonverbal IQ were associated with better core language outcomes (Table 9). Figure 9 shows the distribution of core language scores for the Moog Center and Hearts for Hearing groups with hearing aids at 3 and 5 years of age. Between programs, core language outcomes were not significantly different at age 3 or age 5.

Table 10 displays the regression coefficients and associated p values for the fixed effects for predicting articulation outcomes at 3 and 5 years of age for children using binaural hearing aids. LSL services at age 3, earlier age at hearing aid fitting was associated with significantly better articulation outcomes. At age 5, higher nonverbal IQ was associated with better articulation outcomes (Table 10). Figure 10 shows the distribution of the articulation scores for the Moog Center and Hearts for Hearing groups with hearing aids at 3 and 5 years of age for children using binaural hearing aids. Between programs, articulation outcomes were not significantly different at age 3 or age 5.

Table 9

Regression Analysis Results of Hearing Aid (HA) Core Language Outcomes at 3 and 5 Years of Age

Core Language	3 years of age ($R^2 = 0.25$)			5 years of age ($R^2 = 0.43$)		
Effect	Parameter	F Value	p Level	Parameter	F Value	p Level
Intercept	99.9	4763.4	< .0001	100.9	3947.8	< .0001
Maternal Education		3.4	.04		1.45	.25
High School	-6.6			-5.4		
Some college	-7.7			-11.4		
College graduate	0			0		
Nonverbal IQ	0.08	1.7	.20	0.15	9.8	.003
Age HA (months)	-0.19	0.8	.38	-0.32	7.4	.01
Degree Hearing Loss		3.6	.02		1.2	.33
Mild	0			0		
Moderate	-5.2			-3.2		
Moderate-Severe	-14.2			-12.34		
Severe-Profound	-9.14			-10.4		
Treatment Group		1.1	.29		0.3	.58
Hearts for Hearing	0			0		
Moog Center	-10.4			-15.9		
Age enrollment LSL	0.17	0.22	.64	-0.05	0.12	.73
LSL Intervention Hours	0.02	2.2	.14	0.01	2.7	.11

Note. IQ = intelligence quotient; LSL = listening and spoken language program. Parameter represents the Beta coefficient from the analysis.

Figure 9
Core Language Scores for Children with Hearing Aids at 3 and 5 Years Old
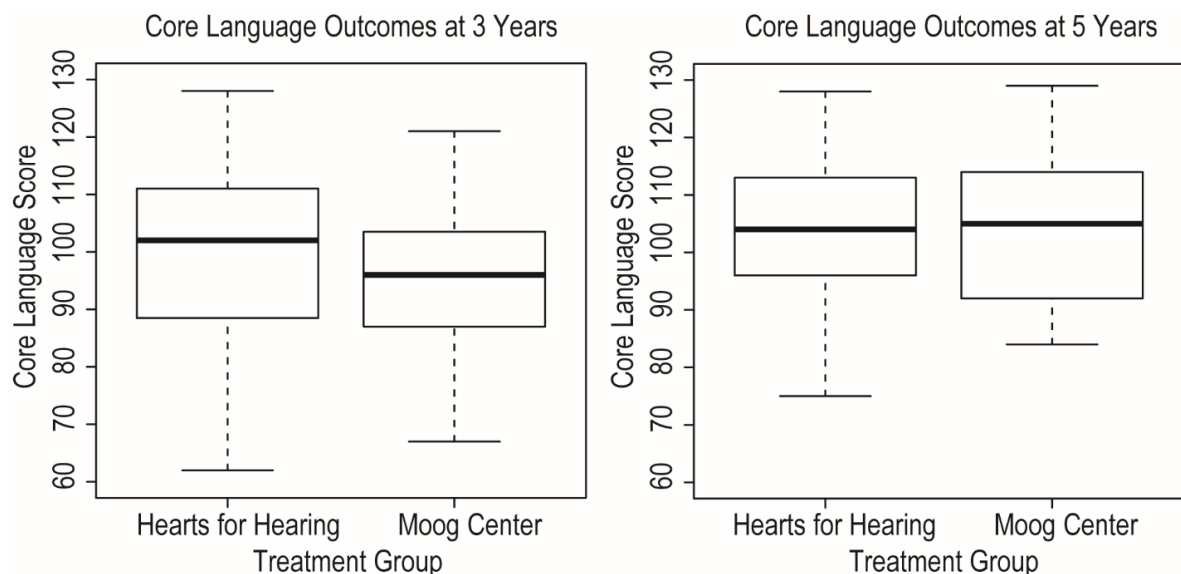
Note. The median of the distribution is denoted by the horizontal black line in the box, and the edges of each box represent the 25th and 75th percentiles of the distribution. The whiskers extend to the minimum and maximum individual data points that are not outliers. Outliers are values greater than 1.5 times the interquartile range.

Table 10
Regression Analysis Results of Hearing Aid (HA) Articulation Outcomes at 3 and 5 Years of Age

Articulation	3 years of age ($R^2 = 0.21$)			5 years of age ($R^2 = 0.32$)		
Effect	Parameter	F Value	p Level	Parameter	F Value	p Level
Intercept	101.5	1965.6	< .0001	54.5	1403.0	< .0001
Maternal Education		2.25	.11		1.6	0.21
High School	1.13			0.78		
Some college	-7.9			15.6		
College graduate	0			0		
Nonverbal IQ	-0.03	0.19	.65	0.57	8.8	.005
Age HA (months)	-0.47	4.8	.03	-0.29	2.13	.15
Degree Hearing Loss		0.81	.49		0.12	.95
Mild	0			0		
Moderate	7.04			0.89		
Moderate-Severe	-0.47			-0.14		
Severe-Profound	2.92			6.9		
Treatment Group		2.2	.15		2.2	.15
Hearts for Hearing	0			0		
Moog Center	-5.8			-11.4		
Age enrollment LSL	0.02	0.03	.87	-0.004	0.0002	.99
LSL Intervention Hours	-0.006	0.10	.75	0.0006	0.004	.95

Note. IQ = intelligence quotient; LSL = listening and spoken language program. Parameter represents the Beta coefficient from the analysis.

Figure 10
Articulation Scores for Children with Hearing Aids at 3 and 5 Years Old

Note. The median of the distribution is denoted by the horizontal black line in the box, and the edges of each box represent the 25th and 75th percentiles of the distribution. The whiskers extend to the minimum and maximum individual data points that are not outliers. Outliers are values greater than 1.5 times the interquartile range.
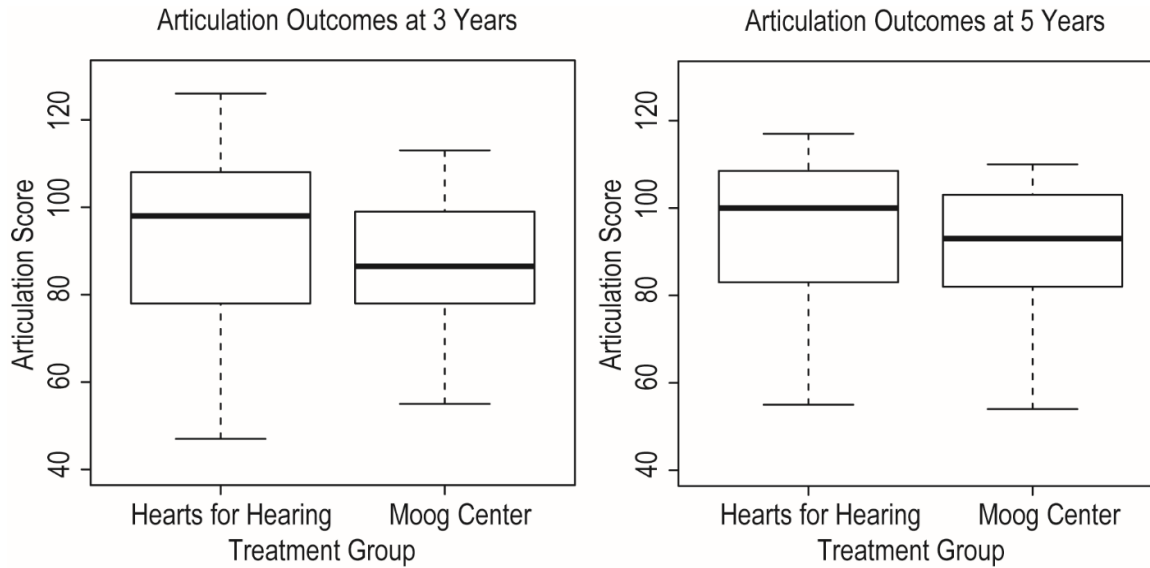
Discussion

This is the first study to show age-appropriate listening and spoken language (LSL) outcomes by 5 years of age for children who received LSL services at two different programs focused on parent and child-centered LSL services and early audiologic intervention. However, service provision between the two programs differs in referral processes, setting, amount of child-directed services provided, and amount of parent coaching offered.

Following is a discussion of the outcomes and factors influencing those outcomes for children using cochlear implants and binaural hearing aids from two different LSL programs.

Children Using Cochlear Implants

For children using cochlear implants, 3 primary differences existed between the participants in the two programs. First, at Hearts for Hearing, children began receiving LSL services at an average age of 7 months, whereas children from the Moog Center began receiving LSL services beginning at an average age of 20 months. Second, the children from Hearts for Hearing were fitted with hearing aids at an earlier age than children from the Moog Center. Third, children from Hearts for Hearing received fewer hours of LSL intervention by 3 years of age (mean of 75.9 hours) and 5 years of age (mean of 163.9 hours) as compared to their counterparts at the Moog Center (356.04 and 1547.9 hours at 3 and 5 years, respectively).

For children using cochlear implants, those attending Hearts for Hearing typically achieved better LSL outcomes at 3 years of age compared to children from the Moog Center, but by 5 years of age, there were no

differences in LSL outcomes between the two programs. As a result, the advantages of early amplification and early entry into LSL programs are illustrated in the relatively better outcomes obtained by the children from Hearts for Hearing at 3 years of age. Fewer LSL hours may be necessary to achieve age-appropriate listening and spoken language outcomes when LSL intervention is initiated and hearing aids are fitted at an early age. Moreover, the benefits of intensive LSL intervention are illustrated in the accelerated progress made by the children from the Moog Center between 3 and 5 years of age. A greater number of LSL intervention hours at a later age may allow children who have later access to LSL services and later-fit hearing aids to achieve age-appropriate LSL outcomes by school-age entry. Given that the present study did not include children with neurocognitive disabilities, the results may not be representative of the entire population of children using cochlear implants. Some children may need additional LSL services to optimize listening and spoken language outcomes, regardless of the age at which LSL intervention is initiated or when hearing aids are fitted.

For children using cochlear implants at 3 years of age, on average, better LSL outcomes were obtained by children who had been fitted with hearing aids at an earlier age. The benefits of early amplification have been clearly established in the literature (Ching et al., 2018; Moeller et al., 2015). Maternal education and nonverbal IQ also were associated with better LSL outcomes at 3 years of age. Again, previous research has shown each of these factors to be associated with better LSL outcomes (Ching et al. 2018; Moog & Geers, 2003; Niparko et al., 2010). Additionally, a greater number of LSL intervention hours was predictive of better expressive language outcomes

at 3 years of age, a finding that is consistent with that of Geers and colleagues (2019).

Of interest, none of the independent variables under study, including hours of LSL intervention, were predictors of LSL outcomes for children with cochlear implants at 5 years of age. The finding that total number of LSL hours did not predict LSL outcomes differs from the finding of Geers and colleagues (2019) but is similar to the findings of Chu and colleagues (2019). Although 5-year outcomes did not differ between programs, children at the Moog Center had higher average LSL intervention hours. Children at Hearts for Hearing may have achieved age-appropriate LSL outcomes because they were identified with hearing loss at an earlier age, fitted with hearing aids earlier, and their parents were coached to create a language-rich listening environment at an earlier age. These steps may have allowed children from Hearts for Hearing greater access to an enriching LSL model throughout a longer portion of the critical period of language development.

Children from the Moog Center showed impressive improvement in LSL abilities from ages 3 to 5 years. This finding is consistent with Ching et al. (2018) where improvements in LSL development were measured from 3 to 5 years of age. Together, the current study and the Ching et al. (2018) study indicate intensive LSL intervention can mitigate delays in LSL outcomes that occur at early ages. Of note, the variance in the standardized language scores of the children who participated in this study was similar to the variance observed in these measures for children with typical hearing. Additional research is needed to determine the dosage of LSL services required to obtain age-appropriate listening and spoken language outcomes for children who receive LSL services at later ages.

Children Using Binaural Hearing Aids

The results of this study suggest that, on average, children who use binaural hearing aids, receive LSL intervention at a program specializing in listening and spoken language development, and have no neurocognitive disabilities achieve age-appropriate LSL outcomes by 3 or 5 years of age. Unlike the findings for children using cochlear implants, there were no differences in LSL outcomes at 3 years of age between the two programs. Because the mean age of hearing aid fitting and program enrollment were similar between the two programs, it is probable that early access to spoken language via hearing aids positively influenced LSL outcomes for children in both programs. However, there were some demographic and audiologic differences present for the children from the two LSL programs. Children from Hearts for Hearing had lower non-verbal IQ, mothers with lower education levels, and families with lower SES, whereas a greater percentage of children from the Moog Center fitted with hearing aids had severe to profound hearing loss.

As with the children using cochlear implants, the number of LSL intervention hours provided to children with hearing aids was not largely predictive of the LSL outcomes, with the lone exception of greater LSL hours associated with better expressive language at 3 years of age. Despite the similar outcomes between programs, LSL intervention hours differed substantially with averages at 5 years of 1350.9 hours at the Moog Center and 103.4 hours at Hearts for Hearing. Of note, higher non-verbal IQ, greater levels of maternal education, earlier age at hearing aid fitting, and better unaided pure tone thresholds were associated with better LSL outcomes for children with hearing aids, findings which are consistent with previous research (Ching et al., 2018; Moeller et al., 2015). Also of note, the variance in the standardized language scores of the children who participated in this study was similar to the variance observed in these measures for children with typical hearing.

Study Limitations

As previously discussed, the current study did not include children with neurocognitive disabilities. Cupples et al. (2018) reported the presence of an additional disability other than hearing loss in 39% of the children participating in the Longitudinal Outcomes of Children with Hearing Impairment (LOCHI) study. Consequently, the results of the current study cannot be applied to all children who are deaf or hard of hearing. Additional research is needed to better understand the role of LSL intervention dosage on listening and spoken language outcomes of children with neurocognitive disabilities.

Moreover, the children in the current study were all active participants in one of the two LSL programs from which the children were recruited to be included in this research. Children who are deaf or hard of hearing may achieve poorer LSL outcomes if their families do not have the same level of access and/or demonstrate a commitment to LSL services that is similar to the access and commitment made by the families of the children in the current study. Additional research is needed to explore LSL outcomes of children whose families do not have a consistent access or commit to services at a specialized LSL program.

Additionally, as noted in the Method section of this paper, not every child who was evaluated at 3 years of age also was evaluated at 5 years of age. It is impossible to know how the study results would have been affected if all children in the study also were evaluated at 5 years of age. It is possible that some of the children who were not enrolled in intervention at 5 years of age had ceased services because they had developed excellent listening and spoken language skills. If this is true, then inclusion of the test scores for those children at 5 years of age may increase the mean scores. Once again, however, it is impossible to speculate on the effect that participant attrition at 5 years of age has on the study results evaluated at 5 years of age.

Furthermore, information pertaining to audiologic intervention was not included in the current study. For

instance, complete hearing aid and cochlear implant datalogging records (i.e., usage time) were not available. Also, there were too many discrepancies regarding the manner in which speech perception scores were obtained across participants (e.g., types of speech perception tests that were administered, presentation level, recorded versus monitored live voice, quiet vs. noise, etc.) to allow for speech perception abilities to be included as a factor in the prediction of LSL outcomes. Additional research is needed to determine the relationship between LSL intervention dosage, audiologic variables, and LSL outcomes. In addition, future work will need to examine effects of service delivery dosage on children implanted at less than 12 months compared to those implanted at 12–18 months of age.

Conclusions

The results of this study indicate age-appropriate LSL outcomes are probable for children who have typical neurocognitive abilities and whose families have access and actively commit to LSL services from a specialized LSL program. Non-verbal IQ and maternal education levels also influence LSL outcomes. Total hours of LSL intervention do not serve as a predictor of LSL outcomes at 5 years of age. However, when poorer-than-expected outcomes are measured at 3 years of age, it may be possible to achieve age-appropriate LSL outcomes by age 5 with intense LSL intervention from 3 to 5 years of age. Children who have earlier access to hearing technology and LSL intervention may need fewer LSL hours to achieve age-appropriate LSL outcomes; however, those who are later identified and later enrolled in LSL intervention may require more hours of services to achieve the same age-appropriate LSL outcomes. Early identification of hearing loss, early amplification, and early intervention are highly influential factors affecting LSL

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Advancing Clinical Practice through Integration of Congenital Cytomegalovirus (cCMV) Testing with Newborn Hearing Screening at Mayo Clinic

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Abstract

Although congenital cytomegalovirus (cCMV) is the leading non-genetic cause of childhood hearing loss in the United States, neither targeted nor universal screening protocols have been well established to identify cCMV in newborns. Moreover, until cCMV testing is universal, clinical protocols need to account for the complexities of individualized care in partnership with interprofessional care teams. This work addressed an immediate clinical practice need to identify cCMV with subsequent hearing monitoring of babies who test positive for cCMV. This effort focused on three primary objectives to: (a) define interprofessional, team-based approach to facilitate care pathways; (b) develop a clinical workflow for all babies who refer on inpatient hearing screening to be tested for cCMV by 21 days of age; and (c) develop a hearing monitoring plan for all babies who test positive for cCMV. The article describes the development and integration of our interprofessional, team-based approach to institute cCMV testing by 21 days of age on all babies who refer. Description of the inpatient newborn hearing screening and subsequent monitoring is also included. Our observed referral rate was lower than predicted (2.7%) from existing literature with only one positive cCMV outcome noted in the two-year span. This study demonstrates the feasibility of a hearing-targeted cCMV testing paradigm in our clinic practice.

Key Words: newborn hearing screening, CMV, cCMV, targeted screening, interprofessional collaborative care

Acronyms: AABR = automated auditory brainstem response; ABR = auditory brainstem response; ASSR = auditory steady state response; AUD = audiology; CBC = complete blood count; cCMV = congenital cytomegalovirus; CMV = cytomegalovirus; DPOAEs = distortion product otoacoustic emissions; EHDI = Early Hearing Detection and Intervention; GCE = genetics, IFD = infectious disease; JCIH = Joint Committee on Infant Hearing; LFT = liver function test; MDH = Minnesota Department of Health; MRI = magnetic resonance imaging; OAE = otoacoustic emissions; ORL = Otolaryngology; PCR = Polymerase Chain Reaction; TEOAEs = transient otoacoustic emissions

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Babies born with congenital cytomegalovirus (cCMV) infection may present with immediate and long-term health problems, one of which is hearing loss detectable at birth or developing later in childhood (reviewed by Goderis et al., 2014 and WHO, 2021). cCMV is common in the United States presenting in 1 out of 200 babies (~0.6%; Centers for Disease Control and Prevention [CDC], 2020; Fowler et al., 2018). Of babies infected with cCMV, about 10% are symptomatic at birth, 10 to 15% are asymptomatic at birth yet develop hearing loss or other neurological impairments at a later onset, and the remaining majority (75–80%) are asymptomatic (Boppana et al., 2013; CDC, 2020; Goderis et al., 2014; Kenneson & Cannon, 2007). Symptomatic babies may exhibit multiple system conditions because

of cCMV which may include thrombocytopenia, hyperbilirubinemia, or central nervous system involvement such as microcephaly with significant neonatal morbidity and mortality (e.g., Rawlinson et al., 2017).

Sensorineural hearing loss is the most common diagnosis for a baby infected with cCMV, whether symptomatic or asymptomatic for other systems' involvement (Naing et al., 2016). Estimates suggest that cCMV accounts for 25 to 40% of total hearing loss in children (Goderis et al., 2014). Sensorineural hearing loss occurs in approximately 20 to 40% of babies with multisystem involvement and is a single system finding in 5 to 10% of cCMV cases (Dollard et al., 2007; Fowler & Boppana, 2006, 2018; Goderis et al., 2014; Rawlinson et al., 2017). Although this is counter-intuitive, hearing loss has not been included in the definition of symptomatic cCMV; asymptomatic cCMV is a distinct category and can include hearing loss (Petersen et al., 2020; Rawlinson et al., 2017). Therefore, asymptomatic cCMV cases may present with hearing loss as the only clinical finding (Fowler & Boppana, 2006, 2018; Goderis et al., 2014).

Universal newborn hearing screening successfully detects congenital hearing impairment at birth; however, concerns for delayed onset or progressive hearing loss require longer term monitoring (Joint Committee on Infant Hearing [JCIH], 2019; World Health Organization [WHO], 2021). At this time, screening for cCMV is not performed for all newborns. The debate over universal newborn cCMV screening versus targeted screening is ongoing. However, growing evidence and clinical practice goals of universal or extended neonatal cCMV screening aimed at detection of cCMV at the earliest are progressing (e.g., Krishna et al., 2020). Hearing-targeted screening for cCMV can be one step in advancing toward the goal of universal cCMV testing for all newborns and for promoting earlier detection of delayed onset or progressive hearing loss (e.g., JCIH, 2019). That said, such targeted approaches are imperfect as they are biased toward missing cCMV positive cases with passed newborn hearing screening results (see review of considerations by Haller et al., 2020 and Krishna et al., 2020). Evidence in the realm of early detection has resulted in recommendations for inclusion of cCMV testing if sensorineural hearing loss is detected as a result of newborn hearing screening (Choi et al., 2009; Haller et al., 2020; Korver et al., 2017; Park & Shoup, 2018). Hearing-targeted cCMV screening and outcomes data (Diener et al., 2017; Fowler et al., 2017) support legislative efforts to mandate cCMV testing based on newborn hearing screening outcomes and potentially beyond (National CMV Foundation, 2021). Moreover, accounting for delayed-onset or progressive hearing loss over the first years of life is of growing importance (e.g., Cannon et al., 2014; WHO, 2021).

Early cCMV testing is critical as this is the only means to differentiate between congenital and postnatally acquired infection. Detection of cCMV can be made within the first weeks of life by detecting the virus from a culture or polymerase chain reaction (PCR) of body fluids such as urine or saliva (Boppanna et al., 2011; CDC, 2020). This is most helpful for timely detection of asymptomatic cCMV cases as positive cCMV tests within the first 14 to 21 days of life help distinguish congenital from acquired CMV (Revello & Gerna, 2002). Testing of the newborn screening card dried blood spots may permit later diagnosis of cCMV as such samples are collected in the desired timeframe and may retrospectively help to distinguish between congenital and acquired CMV (e.g., Choi et al., 2009). Only congenital CMV causes hearing loss or symptomatic disease; whereas, postnatally acquired infections are not associated with disease (e.g., Boppana et al., 2010; Choi et al., 2009; Meyer et al., 2017). In the case of cCMV, newborns who are symptomatic or asymptomatic will continue to shed and transmit the virus through bodily fluids for 18 to 30 months (Pati et al., 2016). This knowledge is critical for treatment planning and consideration for antiviral therapy (Rawlinson et al., 2017). Timely diagnosis leads to timely intervention; specifically, all treatments began before 30 days of life in initial trials validated antiviral medications (Kimberlin et al., 2003, 2015; Rawlinson et al., 2017). Similarly, continued monitoring for later onset of hearing loss in children who were positive for cCMV can help with earlier diagnosis of hearing loss, facilitate fitting of assistive hearing devices, and support earlier access to speech and language interventions (Boppana & Fowler, 2017; JCIH, 2019; Kennedy et al., 2006).

The work presented here developed from the immediate clinical practice need for Mayo Clinic Rochester and Mayo Clinic Health System (outreach clinical sites throughout Minnesota and Wisconsin) to converge on clinical practice approaches to identify cCMV and subsequently monitor babies who tested positive for cCMV in the newborn population. This effort focused on three primary study objectives that we developed (based on baseline program review from 2015–2017), deployed (January 2018), and reviewed over a two-year span (2018–2019). This article describes the development and integration of our interprofessional, team-based approach to quality improvement efforts to conduct cCMV testing by 21 days of age on all babies who refer (in at least one ear) on inpatient newborn hearing screening. It also describes the follow up process developed for ongoing hearing monitoring of this population. Specific project objectives include: (a) define interprofessional, team-based approach to facilitate care pathways; (b) develop a quality improvement strategy where all babies who refer on inpatient hearing screening get cCMV testing by 21 days of age; and (c) develop a hearing monitoring plan for all babies who test positive for cCMV, keeping in mind that some may be found to have normal hearing.

Method

Participants

All newborns at Mayo Clinic undergo newborn hearing screening according to Minnesota state guidelines (Minnesota Department of Health [MDH], 2021). The population at Mayo Clinic includes newborns who stay on a postpartum/newborn nursery unit (well child) with their

mothers and patients who stay on a Level III neonatal intensive care unit (NICU) and a Level IV NICU. Mayo Clinic Rochester has been conducting inpatient hearing screening on all newborns since 1999. The practice screens approximately 2335 newborns per year (5-year average) on the newborn nursery unit and two NICUs. Overview of the birth cohort focus for this evaluation pre- and post-screening for cCMV is detailed in Table 1.

Table 1
Overview of Birth Cohort from the Newborn Nursery Population Defined as Number of Patients Screened

		Mayo Clinic Rochester, MN Newborn Nursery Population				
Year		2015	2016	2017	2018	2019
Birth Cohort		1899	1839	1903	1882	1921
Hearing Screenings	Pass	1850	1774	1800	1747	1803
	Refer/Fail	49	65	103	135	118

Note. Screening results of pass in both ears and the target for this investigation of referred in one or both ears also depicted for 2015 to 2019. The thick vertical line delineates baseline (2015–2017) prior to implementation of the congenital cytomegalovirus (cCMV) screening triggered by refer/fail on newborn hearing screening (active since 2018).

Newborn Hearing Screening Protocol

Audiology staff, including audiology assistants and audiologists, conduct the newborn hearing screenings at Mayo Clinic Rochester. On the newborn nursery unit, screening is conducted using a two-step method (See Figure 1). Otoacoustic emissions (OAE) are used as a first level screening. Screening is considered complete if there is a passing result for both ears using OAE. Automated auditory brainstem response (AABR) is conducted if there is a refer result on the first OAE screening. Screening is considered complete if there is a passing result for both ears using the AABR technology. In the NICU settings, screening is conducted using only the AABR technology. Screening is considered complete if there is a passing result for both ears. A maximum of two screening attempts are conducted during the inpatient stay. For patients on all units, risk factors for early childhood hearing loss (JCIH, 2007; 2019) are reviewed and documented.

Location

The focus of this evaluation is on the newborn nursery unit, where screening is conducted by one audiologist coordinator, seven audiology assistants, and two audiology doctoral student externs. The location of screening varies and is prioritized for family-centered care. Screening is initially offered to be conducted at the mother's bedside and with parent(s) present whenever possible. This aspect of the program lends a family-centered care emphasis and is the most common location for screening in our practice.

Figure 1
Overview of Two Step Protocol in the Newborn Hearing Screening Workflow
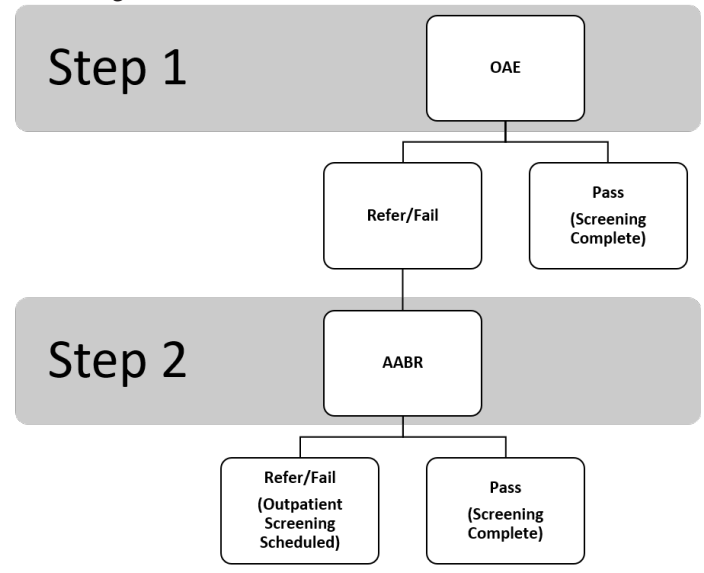
Note. The first level of screening (Step 1) uses otoacoustic emission (OAE) technology and the second step uses automated auditory brainstem response (AABR) technology. Refer/Fail outcomes prompt next level of screening.

Timing

Newborns on the nursery unit are typically seen for hearing screening on the first full day of life. Screening is typically completed late morning or early afternoon on the first day, so the patients may be in the range of 12 to 36 hours old when screening is completed. In either of the NICU settings, screening is conducted as the patient is getting closer to dismissal (typically within 1–2 weeks of dismissal).

Equipment

Screening is conducted using the Otodynamics Otoport TE+DP OAE+AABR equipment in all units. This is a handheld piece of equipment that plugs in to a computer kept on a cart and is transported to the newborn's location for screening. This allows the screener to be mobile and permits timely access to the electronic health record and reporting applications for maximum efficiency in the clinical practice. The handheld equipment can be transported into the mother's room without the full cart for a less obtrusive experience for the family and to maximize bedside screening opportunities.

AABR and OAE screening equipment settings are consistent across all units. Of note, the equipment that was in use from 2015 to 2018 included the ALGO 5 Newborn Hearing Screener for AABR and Otodynamics Echoport 292 for transient otoacoustic emissions (TEOAE). The Otodynamics Otoport TE+DP OAE+AABR was in use from 2018 to 2019.

Data Management

Results and risk factors are entered into an internally created database called the Early Hearing Detection and Intervention (EHDI) application (developed at Mayo Clinic Rochester, 2009). This application was designed to track

inpatient and outpatient screening as well as follow up results for newborn hearing screening. The audiologist or audiology assistant creates a note in the electronic health record as well. Results are also sent electronically to the MDH. Data is exported from the handheld units to minimize errors due to manual entry. A program coordinator maintains the internal EHDI database, oversees review of risk factors for early childhood hearing loss, monitors follow up, and directs the ongoing quality improvement initiatives.

Patient Education

Newborn hearing screening results are delivered by the examining audiologist or audiology assistant to the parent(s) at the time of the screening. Results are delivered verbally and in writing as standard practice. In person, tablet, or telephone language interpreters are used when appropriate. Two patient education brochures are offered to families. The first brochure describes the screening process and explains why hearing screening is being done for a newborn. This brochure also has a checkbox for a pass or refer result so that the family has a record of the results before hospital dismissal. The family also has access to the screening results in the electronic health record patient portal. The second brochure has a list of typical developmental milestones for speech and language abilities up to age 5 years and is intended as a reference for parents to use while monitoring their child's speech and language development.

When a baby is leaving the hospital with a refer result, the person who conducts the hearing screening documents this in the internally created EHDI database and in the note in the electronic health record and sends a message (also through the electronic health record) to the audiology scheduling team. The scheduling team contacts the family directly to schedule an outpatient rescreen appointment in 1 to 2 weeks.

Congenital Cytomegalovirus (cCMV) Testing Protocol

For this evaluation, patients leaving the hospital with a refer result on newborn hearing screening in one or both ears were offered cCMV testing. This included patients with a refer after one attempt if a second attempt is not possible before discharge. cCMV testing was performed as urine PCR (polymerase chain reaction) or saliva swab depending on what was feasible at the time of collection prior to hospital dismissal (e.g., Rawlinson et al., 2017; JCIH, 2019). Refer to Figure 2 for workflow of cCMV testing prompted by hearing screening outcome. This was implemented in January 2018 and this article reviews the outcomes over a two-year period (2018 to the end of 2019).

In addition to the previously described process for newborn hearing screening above, the person who conducts the hearing screening also notifies the nurse caring for the patient that the patient will be leaving with a refer result (one ear or two) for hearing screening. This prompts the nurse to activate the cCMV collection order set in the electronic health record (and workflow in Figure 2). The nurse informs the family that cCMV testing is completed whenever a patient is leaving the hospital with a refer result on newborn hearing screening. The nurse collects a sample for this test (as appropriate). Urine is the preferred specimen, but saliva (buccal swab) is considered appropriate if urine cannot be collected in a timely manner.

Figure 2
Overview of cCMV Testing Workflow for Newborns Triggered by Refer/Fail On Newborn Hearing Screening by 21 Days of Life
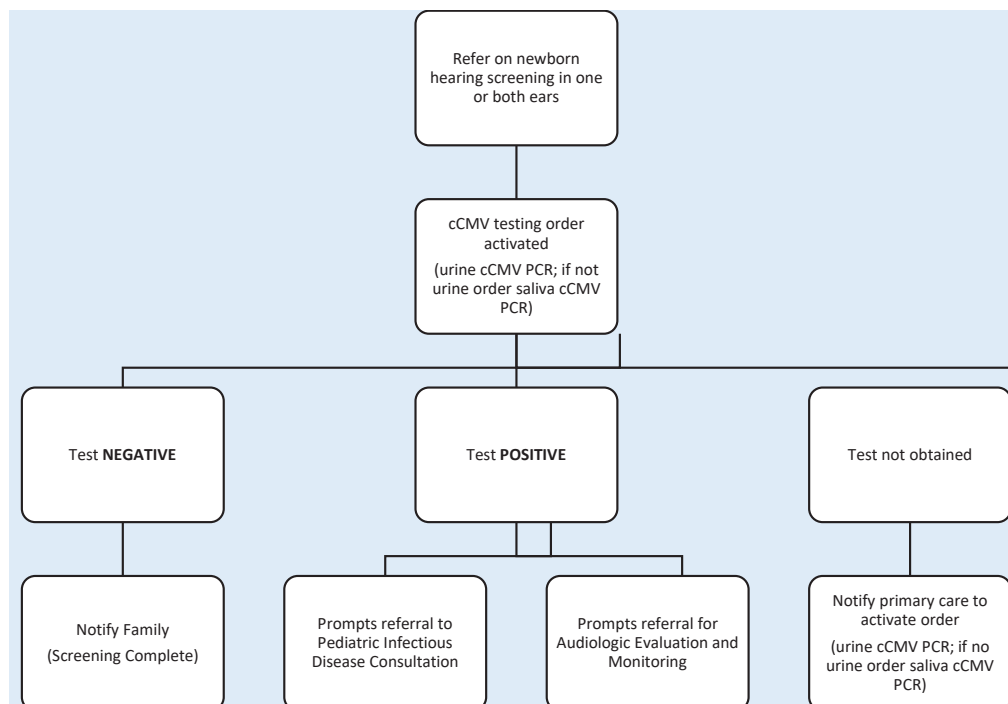
Note. Congenital cytomegalovirus(cCMV) testing ordered and sample collected prior to hospital dismissal. PCR = polymerase chain reaction.

PCR testing is completed at the Mayo Clinic Laboratories with results reported in the electronic health record in 1 to 2 days. A PCR positive result triggers an immediate referral to the Pediatric Infectious Diseases Outpatient Clinic or a Pediatric Infectious Diseases inpatient consultation if the patient remains in the hospital. The Pediatric Infectious Disease provider then initiates an evaluation for other evidence of all organ involvement while awaiting final audiology results. If the evaluation indicates the infant has symptomatic cCMV, treatment is discussed with the parents or guardian.

Patient Education Development

cCMV testing results are delivered to the parent(s) by the managing primary care team. In a coordinated effort by the interprofessional care team, a patient education piece was created to assist the primary care providers in educating their patients and their families about cCMV and its connection to hearing loss. The piece, entitled "*Congenital Cytomegalovirus (cCMV),*" is a 12-page brochure created by Mayo Clinic Health Education and Content Services led by Audiology in collaboration with colleagues in Pediatric Infectious Disease, Genetics, Pediatric Otolaryngology, and Primary Care. Topics include a description of cCMV and causes, the relationship between cCMV and hearing loss, an overview of testing for cCMV, as well as treatment and prevention of cCMV.

Results

Interprofessional Collaborative Care Team Approach

An interprofessional collaborative care team was established to address this clinical practice need with the goal of initiating cCMV testing prior to dismissal from the hospital on all babies in the newborn nursery with a refer result on their newborn hearing screening. Interprofessional collaborative practice occurs when multiple health workers from different professions provide comprehensive services by working with patients, caregivers, and communities to deliver the highest quality of care across settings (WHO, 2010). The Interprofessional Education Collaborative (IPEC, 2016) further defined several competencies for teams of professionals working toward this type of practice. These include a climate of mutual respect and shared values, clearly defined roles and responsibilities, responsive and responsible communications with patients and their families as well as other professionals, and application of the principles of effective team dynamics.

For the purposes of this project, the interprofessional collaborative care team (See Figure 3) included representatives from the following specialty areas: Audiology, Genetics, Pediatric Infectious Disease, Pediatric Otolaryngology, Nursing, Primary Care (Family Medicine; Community Pediatrics), Neonatology, Hospital Desk Operations, Patient Appointment Services, and Patient Education. Representatives from the various groups worked together in smaller groups to accomplish portions of the project. For instance, pediatric expert representatives from audiology, infectious disease, genetics, pediatrics, and otolaryngology collaborated to

Figure 3
Overview of Interprofessional Collaborative Care Team Approach
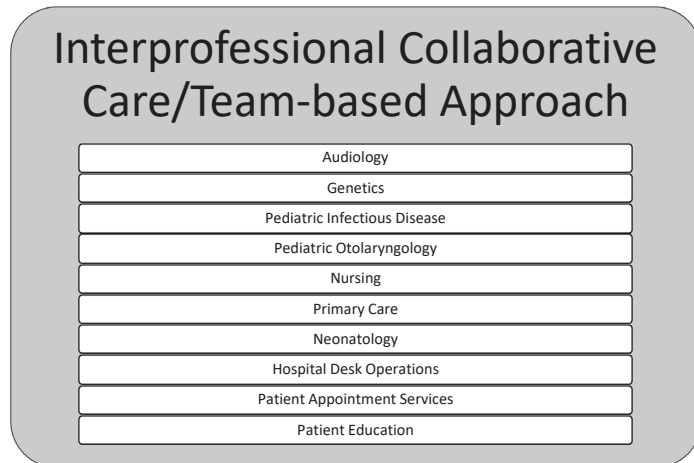
Note. There are many other collaborating providers; however, this reflects the initial launch team described in this study.

create a patient education piece (described in Patient Education Development) designed to support primary care providers when discussing the concept of cCMV testing with parents.

Defined Care Team Pathway for Audiology (Outpatient)

To facilitate the goal that all babies who do not pass hearing screening will undergo cCMV testing by 21 days of age, care pathways within the inpatient setting (see Figure 2 above) as well as in the outpatient setting (Figure 4) were defined as part of this project. Our objective was to develop a follow up plan for all babies who test positive for cCMV, keeping in mind that some may be found to have normal hearing. Figure 4 provides an overview of the care team coordination and plan for additional monitoring and management.

With implementation of a cCMV testing program into the clinical practice, an audiological monitoring pathway and protocol needed to be defined to account for individuals testing positive for cCMV and based on hearing status. The testing within this protocol will vary slightly depending on the patient population and individualized patient needs. Primary populations following this protocol will be patients who test positive for cCMV and who (a) Refer, miss, or refuse on newborn hearing screening and subsequently have confirmed sensorineural hearing loss, conductive hearing loss, or mixed hearing loss; and (b) Refer, miss, or refuse on newborn hearing screening and subsequently have confirmed hearing sensitivity within normal limits.

An interprofessional collaborative care team-based approach is critical when serving patients with cCMV with or without hearing loss. Clearly defined clinical pathways for the identification and management of cCMV can facilitate early intervention options. The care team needed for an effective monitoring program is one in which team

Figure 4

Care Team Coordination and Management Plan for Individuals Testing Positive for cCMV

Care Team Coordination and Management Plan

Audiology (AUD)

Referred inpatient screening:
- Outpatient rescreen within 1–2 weeks

Diagnosed with typical hearing:
- Diagnostic evaluation with audiologist to obtain baseline ABR, OAE, and immittance data by 6–8 weeks of life
- Monitor (prioritize OAE and immittance)
  - Every 3 months until age 1 year
  - Every 6 months until age 3 years
  - Annually until age 19 years

Referred inpatient screening:
- Outpatient rescreen within 1–2 weeks

Diagnosed with hearing loss:
- Diagnostic evaluation with audiologist to obtain baseline ABR, OAE, and immittance data by 6–8 weeks of life
- Monitor (full measures as appropriate)
  - Every 3 months until age 1 year
  - Every 6 months until age 3 years
  - Annually until age 19 years

Otolaryngology (ORL)

Diagnosed with hearing loss:
- Medical evaluation of hearing loss (referral to additional specialties as needed)
- Brain imaging for medical evaluation to help predict neurodevelopmental outcomes
- Early medical clearance for amplification and re/habilitation trials
- Consideration of early cochlear implant candidacy as appropriate
- Medical management of conductive hearing loss as needed

Infectious Disease (IFD)

cCMV positive – Identification of symptomatic disease:
- Physical examination
- Imaging (preferably MRI)
- Lab studies
- Developmental evaluation beginning the first year for those with symptomatic cCMV and as needed

Symptomatic cCMV positive – Treatment:
- Offer oral valganciclovir*
- Monthly physical examination
- Lab studies regularly (e.g., weekly for 6 weeks, then at week 8, then monthly if no abnormalities)
- Liver function tests and creatinine regularly (e.g., monthly)
- Advise additional monitoring based on treatment and outcomes

Genetics (CGE)

- Utilization of evaluations obtained from care team
- Referral to Pediatric Ophthalmology (additional specialties as needed)
- Detailed family history and genetic counseling as appropriate
- Offer genetic testing panels specific to hearing loss

Note. Care team coordination and management plan defined prospectively to define care pathway for individuals testing positive for congenital cytomegalovirus (cCMV) with specific focus on differences between care plan for individuals with hearing loss versus those with typical hearing. Please note this is an overview rather than exhaustive care plan in which individualized patient needs are also addressed. ABR = auditory brainstem response; OAE = otoacoustic emissions. *If initiated, valganciclovir treatment is for 6 months unless adverse effects prevent the full course of treatment.

members from different professions work in collaboration for timely identification, monitoring, and intervention as appropriate. Four of the professions (see Figure 3) are described in detail below for an overview of this approach.

Audiology

For Audiology, when babies refer on inpatient newborn hearing screening (refer to Figure 1), patients are immediately scheduled for outpatient rescreen within 1 to 2 weeks. If there is a refer result on the outpatient rescreen of hearing, then patients proceed with a scheduled diagnostic audiologic evaluation with an audiologist as soon as possible (typically, within 1–2 weeks). Audiologic evaluation may include frequency-specific threshold auditory brainstem response (ABR), auditory steady state response (ASSR), otoacoustic emissions (may include TEOAEs and distortion product otoacoustic emissions [DPOAEs]), and immittance measurements (including tympanometry and acoustic reflex testing). Evaluation and confirmation of hearing status may occur over multiple visits.

When audiologic evaluation leads to diagnosis of hearing loss, the next steps (refer to Figure 4; AUD section) are to monitor every 3 months until age 1, every 6 months until age 3, and annually until age 19. When audiologic evaluation leads to diagnosis of hearing sensitivity within normal limits, the next steps are to monitor every 3 months until age 1, every 6 months until age 3, and annually until age 19. Although the timing of the monitoring visits is the same for the two populations, the specific monitoring tools are different. The testing for the population with a diagnosis of hearing sensitivity within normal limits will focus on and prioritize objective screenings, such as OAE and tympanometry unless change in hearing is more highly suspect (modified from Figure 2 in Foulon et al., 2015).

Pediatric Otolaryngology

For Pediatric Otolaryngology, there is a close partnership with Audiology when audiologic evaluation leads to diagnosis of hearing loss (refer to Figure 4; ORL section). Additional medical evaluation of hearing loss may include detailed review of medical history, brain imaging for medical evaluation to help predict neurodevelopmental outcomes, referrals for speech and language development evaluation, et cetera. Efforts are led by otolaryngology for medical management of conductive hearing loss, which may include partnership with primary care teams. Pediatric Otolaryngology will often serve as the lead for early medical clearance for amplification or rehabilitation trials as well as the entry point for referrals for consideration of early cochlear implantation as appropriate. Specific to cCMV positive cases, additional management may include brain imaging for medical evaluation to help predict neurodevelopmental outcomes, early medical clearance for amplification and (re)habilitation trials, and consideration of early cochlear implantation as appropriate.

Genetics

For genetics, the identification of cCMV positive individuals in combination with hearing status helps guide next steps for the care plan (refer to Figure 4; CGE section). Additional work-up may include: utilization of evaluations obtained from care team, referral to pediatric ophthalmology (and other specialties as needed), and detailed family history and genetic counseling as appropriate. Targeted genetic testing is warranted, particularly, if there is a family history of hearing loss suggesting that there may be coincident cCMV as well as a genetic condition. Negative targeted genetic testing in the face of positive cCMV testing provides supporting evidence that cCMV alone would be the underlying cause of hearing loss.

Pediatric Infectious Disease

For Pediatric Infectious Disease, evaluation for evidence of symptomatic cCMV disease and treatment for symptomatic infants are the key considerations and components to manage (refer to Figure 4; IFD section). Next steps following a cCMV positive test result include physical examination; lab studies such as Complete Blood Count (CBC) with differential, liver function tests (LFT), creatinine; and brain imaging (preferably MRI). Developmental evaluation should begin at the first year for children with symptomatic cCMV on a case by case basis. Practitioners will also want to review audiology in at least 6 month intervals through age 3 years and align them with the prospective monitoring (defined in Figure 4; AUD section).

Treatment of symptomatic cCMV disease is led by Pediatric Infectious Disease in partnership with the broader care team. Specific to cCMV treatment, infants are examined at least monthly with dose adjustments of valganciclovir based on weight gain and monitoring for adverse effects of oral valganciclovir treatment including CBC with differential, liver function tests, and creatinine. Monitoring of hearing helps support the treatment which may be conducted through age 19 years based on need (as described above).

Hearing-Targeted cCMV Testing

Table 2 provides an overview of the numbers of newborns who did not pass hearing screening in both ears and cCMV testing. cCMV testing was implemented in January 2018 and here we review outcomes over a two-year period (2018 to the end of 2019) with baseline data reviewed from January 2015 to December 2017. During this time, 1882 newborns were screened in 2018 and 1920 screened in 2019 (refer to Table 1). The referral rate from newborn hearing screening is displayed by year in Table 2 with the 5-year average of 5% of newborns screened referred for hearing. Of those

Table 2

Overview of cCMV Testing Results from the Newborn Nursery Population

		Mayo Clinic Rochester, MN Newborn Nursery Population				
Year		**2015**	**2016**	**2017**	**2018**	**2019**
Hearing Screenings	Refer Rate (refer total/ birth cohort in %)	2.58%	3.53%	5.41%	7.17%	6.14%
	Refer/Fail	49	65	103	135	118
cCMV Testing	*Hypothesized*					
	Test Positive	1	2	3	4	3
	Actual					
	Test Complete				112	115
	Test Positive				0	1
	Test Negative				112	114

Note. Screening results of pass in both ears and the target for this investigation of referred in one or both ears also depicted for 2015 to 2019. The thick vertical line delineates baseline (2015–2017) prior to implementation of the congenital cytomegalovirus (cCMV) testing triggered by refer/fail on newborn hearing screening (active since 2018). The gray shading denotes hypothesized values or intentionally blank cells prior to the initiation of cCMV testing.

that refer on hearing screening, we anticipated to find about 2 to 3 babies per year from our birth cohort that would refer for our monitoring protocol based on our program data from 2015 to 2017 (see Table 2). This referral rate for cCMV was predicted based on prior evaluations at similar institutions (2.7% refer rate; Choi et al., 2009). Based on these predictions, we planned a monitoring protocol that we expected would be manageable within the framework of our existing clinical practice (see Figure 2).

Most of those individuals that referred on newborn hearing screening were tested for cCMV (83.0% in 2018 and 97.5% in 2019). No newborns were identified via targeted testing for cCMV in 2018 and only one patient was identified in 2019. This is less than our hypothesized cCMV refer rate of 4 in 2018 and 3 in 2019 (Table 2).

Discussion

Congenital cytomegalovirus (cCMV) is a cause of neurodevelopmental delay in children and a common cause of nonhereditary sensorineural hearing loss (CDC, 2020; Goderis et al., 2014; Kenneson & Cannon, 2007; Kimberlin et al., 2015). Although prevalent, cCMV has gone largely undetected because most babies that are cCMV positive are asymptomatic. The early detection of hearing loss may help identify cCMV as well as promote early intervention for hearing loss. In our practice, exploration of hearing-targeted screening for cCMV was an initial step in advancing toward the goal of universal cCMV testing for all newborns and for promoting earlier detection of delayed onset or progressive hearing loss. In this article, we described our efforts focused on defining the care pathway for the identification and audiologic monitoring of individuals who refer on newborn hearing screening and subsequently test positive for cCMV by 21 days of age in the newborn nursery population. Three primary study objectives were to (a) define the interprofessional, team-based approach to facilitate care pathways; (b) develop the clinical workflow for all babies who refer on inpatient hearing screening to get cCMV testing by 21 days of age; and (c) develop a hearing monitoring plan for all babies who test positive for cCMV.

Family-Centered Interprofessional Collaborative Care

The first objective was to establish an interprofessional collaborative care team (Figure 3) to address the immediate clinical practice need with the goal of initiating cCMV testing prior to dismissal from the hospital on all babies with a refer (in one or both ears) on their newborn hearing screening. As described above, many considerations and care team components were explored. Future considerations around defining pathways for inpatient versus outpatient screening and monitoring, internal versus external patient entry options, as well as hearing loss risk (based on degree and progression concern) will be explored. Moreover, longer term monitoring aspects warrant continued exploration given the small population and need for longer-term data (beyond the age of 19 years as described above).

Throughout the work on this project, the interprofessional care team also recognized that the child and their family are arguably the most important members of the overall team caring for the child. Family-centered care has always been a focus of the Mayo Clinic newborn hearing screening program. From the decision to offer to screen in the mother's room as well as with parent(s) present to the scheduling of outpatient appointments before dismissal whenever possible, every decision is made with the experience of each family in mind. Family-centered care means working toward a respectful partnership between the family and the professionals. It also focuses on the principles of honoring and respecting the strengths, cultures, and expertise that families and professionals each bring to the health care interaction (Family Voices, 2021; Kuo et al., 2012; American Academy of Pediatrics, 2012). The principles of family-centered care were considered during all of the work on this project. It is well documented in the literature that parent and medical professional knowledge about cCMV is quite limited. Others are working on increasing awareness of cCMV among pregnant women, those who may become pregnant, and medical professionals (see resources in the National CMV Foundation, 2021; Park et al., 2020). During our project, the creation of the patient education brochure was part of our team's efforts to increase awareness among providers and parents. One way the team could enhance these efforts in the future is to incorporate patient experience feedback and refinement of the materials.

Linking Newborn Hearing Screening and cCMV Testing

Development of a care pathway for a targeted approach to cCMV screening was the primary focus and one of the key objectives of this collaboration. Based on review of available literature, we anticipated that the newborn hearing screening program would identify more individuals with cCMV using this targeted approach to testing based on hearing screening outcomes. Specifically, we predicted a referral rate by year based on prior evaluation at a similar institution (e.g., Choi et al., 2009) which estimated about 2.7%. Our predicted estimates for the newborn nursery population is displayed by year in Table 2. As can be seen from two years of this targeted approach, we anticipated identification of approximately seven patients with cCMV. Instead, we identified only one newborn during the two years with the targeted screening (equating to a referral rate of 0.85% in 2019 and 0% in 2018). Outcomes from this study demonstrate the feasibility of a hearing-targeted cCMV testing paradigm in our clinic and establishes the framework for expanded neonatal cCMV screening or universal screening for cCMV in the future. This aligns with prior published efforts (Diener et al., 2017; Fowler et al., 2017; Haller et al., 2020; Krishna et al., 2020).

It is expected that more cCMV positive cases would be identified if a universal approach to cCMV testing were implemented. Because hearing loss that is secondary to cCMV is often progressive or later-onset in nature, a universal approach to cCMV testing and monitoring would help to identify those patients who receive a pass result on their newborn hearing screening and should be monitored for hearing changes (e.g., Haller et al., 2020). A universal cCMV testing approach would further the critical goals

of timely detection and hearing loss prevention, while promoting accessibility and affordability of care (e.g., Choi et al., 2009).

Audiologic Monitoring of Patients with cCMV Positive Outcomes

Before cCMV testing was implemented (see Figure 2), the audiologists worked to define a protocol for monitoring the newborns who test positive for cCMV. The defined protocol (see Figure 4), described earlier is similar in approach for those who have a higher degree of suspicion for or are known to have hearing loss as it is for those who continue to have results suggesting hearing that is within normal limits. The main difference is a focus on streamlined screening using more objective measures for those who continue to exhibit typical hearing in the context of a larger diagnostic assessment and monitoring plan as appropriate (e.g., Foulon et al., 2015; Park et al., 2014). There are ongoing multi-site studies from leading research centers in this realm (e.g., Choi et al., 2009; 2013; Haller et al., 2020; Park et al., 2014) validating various types of testing for cCMV and looking at the efficacy of treatment on hearing loss prevention. Findings from these continued efforts will be key for informing future changes in clinical practices and prioritizing global efforts for early detection and monitoring of hearing loss across the life span (e.g., JCIH, 2019; WHO, 2021).

Lessons Learned and Future Considerations

There are several aspects to consider when testing for cCMV, from the perspectives of the patient and their family as well as the interprofessional care teams in relation to hearing monitoring. Several of these considerations were gathered as part of the development of this clinical practice initiative and represent several areas needing further and larger scale exploration. Here we highlight considerations for social, clinical practice, and longer-term/life-span care approach.

Social considerations may include such items as the patient's birth hospital may not be their managing hospital, requiring internal and external care pathways to be clearly defined. Patient and family ability to return for frequent monitoring (e.g., travel/financial burden) may also be a factor to address for the clinical program. Patient education materials may be overwhelming or not specific to the needs of the individual. Moreover, variability may exist in expected outcomes based on elected treatment and management options. Affordability and accessibility of care also warrants future consideration.

Clinical practice considerations may include the fact that infectious disease treatment options for symptomatic cCMV symptoms require close monitoring and may have varied outcomes. The individualized treatment approach supported through the interprofessional collaborative care team is deemed to be important for this and warrants future investigation. The complexity of audiologic evaluation in young children as well as in those who are neurologically complex is also a consideration

given screening tool limitations and balancing affordability of preventative monitoring. There may be limitations to appointment availability and coordinating between audiology and associated sub-specialties (e.g., Pediatric Infectious Disease, Genetics, Pediatric Otolaryngology, etc.). The timeline for cCMV testing and confirming hearing status is short to maximize treatment and management options. Until cCMV testing is universal, allowing early intervention for sensorineural hearing loss and developmental delay where appropriate (Rawlinson et al., 2017), current workflows require ordering of the test and this can increase the risk of missed tests.

Considerations across the lifespan and long-term for meeting the needs of the patient are worth immediate consideration as well as continued refinement as evidence emerges. Long term audiologic monitoring is recommended for those identified with cCMV regardless of newborn hearing screening outcome given potential risk for later onset hearing loss. The details of how often and what tools are rapidly emerging with increasing exploration as balancing accessibility, affordability, and quality of care continues to be weighed. It is our recommendation that clinical care pathways should be tailored to the individual needs of the patient and based on treatment options elected.

Conclusion

Although cCMV is the leading non-genetic cause of childhood hearing loss in the United States, there are not widespread established practices for cCMV testing universally at birth or on babies who refer on newborn hearing screening. Development of this interprofessional quality improvement project has greatly enhanced Mayo Clinic protocols and care plans for working with patients with cCMV and their families. It has also enhanced our ability to make recommendations for patients later identified with CMV. It has strengthened the interprofessional collaborative care relationships that audiology has with primary care and various specialties. Although this targeted screening project has identified fewer patients testing positive for cCMV than anticipated, it could be expected that the implementation of a program such as this could have positive implications for practices that have the resources to manage necessary referrals and follow up. Moreover, until cCMV testing is universal, clinical protocols need to account for the complexities of individualized care in partnership with interprofessional care team coordination.

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Readability of Online Hearing-Based Early Intervention Materials

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Abstract

Purpose: A quantitative readability assessment of currently accessible online materials for parents of children who are deaf or hard of hearing (DHH).

Design: Consistent with current recommendations discussing grade-level of materials, Flesch-Kincaid Grade Level (FKGL) analysis, along with five other related measures, was conducted for each website. These analyses provide a readability score for each of the websites analyzed.

Study sample: The first five pages of results from a Google search of "early intervention deaf" and "early intervention hear" were compiled for readability assessment.

Results: Sixty-three websites were included in the analysis. Following article modification, inter- and intra-rater reliability were excellent ($p < .002$). All websites were analyzed based on FKGL, intended audience, page displayed on, and producer. All but one of the websites ($n = 62$) were written at a higher level than the recommended 6th-grade reading level ($m = 12.62$, $SD = 2.65$). There was no significant impact of the search page, intended audience, or producer on FKGL ($p > .1$).

Conclusion: Currently accessible online resources for parents looking at early intervention for children who are deaf or hard of hearing (DHH) are written at a level that may not be accessible. Materials may benefit from being revised and edited with readability and health literacy recommendations in mind.

Keywords: readability, early intervention

Acronyms: ARI = Automated Readability Index; CLI = Coleman-Liau Index; DHH = deaf or hard of hearing; EHDI = early hearing detection and intervention; FKGL = Flesch-Kincaid Grade Level; FRES = Flesch Reading Ease Score; IFSP = Individualized Family Service Plan GFI = Gunning-Fog Index; LFUD = lost to follow-up/documentation; SMOG = Simple Measure of Gobbledygook

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In 2017, Early Hearing Detection and deaf or hard of hearing (DHH) (EHDI) programs across the United States identified 6,537 children as having hearing thresholds outside of the typical range (CDC, 2019a). EHDI is a public health service that applies screening and follow-up care to the general population to maintain and improve the community's overall health. The first goal of EHDI is to ensure that all children, regardless of risk factors, receive a hearing screening, ideally before one month of age (Joint Committee on Infant Hearing, 2019; White, 2019). For children who refer on their hearing screening, the goal is to schedule diagnostic evaluations by three months of age. Following the identification of children as deaf or hard of hearing (DHH), early intervention services are initiated as indicated.

At any point in this system of referrals and service providers, a child can be *lost* and not make it to the next clinically indicated step. These children who are lost to follow-up/documentation (LFUD) can contribute to the number of individuals who have delayed access to early intervention services. One way a child is LFUD is that they have been identified as being at risk for hearing differences via traditional screening measures, yet hearing levels have not been confirmed. This population can consist of children who are DHH and children who, for idiopathic or transient reasons, are referred for further testing after their initial screenings while they have hearing levels in the typical range. Alternatively, a child can be LFUD when they have been identified as DHH and have not enrolled in early intervention services. Children being LFUD after identification and before early intervention may be the most troubling element of LFUD.

Despite the federal mandate to provide hearing-related intervention services, children continue to go without support services and early intervention. Specifically, 2,837 (34.5%) children identified as DHH through EHDI programs across the United States did not receive early intervention services in 2017 and may be at risk for language deprivation (CDC, 2019b). Individuals who experience language deprivation may encounter challenges in learning languages, employment, and social-emotional development (Hall, 2017). In 2017, the most commonly reported reason for children not to get early intervention services, aside from "unknown," is parents declined the service altogether (CDC, 2019b). The 2,837 members of this population have a developmental risk factor, and their families refuse developmental support.

Within the Early Hearing Detection and Intervention Act of 2017, the information made accessible to parents is explicitly discussed in terms of being "accurate, comprehensive, and, where appropriate, evidence-based, allowing families to make important decisions for their children in a timely way…." This statement is in stark contrast to the idea that one potential source of this disengagement with early intervention might be a lack of information about the role of early intervention. Within the literature, there are reports that parents of children who are DHH are looking for and/or need more information on early intervention topics such as what early intervention is (Khoza-Shangase, 2019; Larsen et al., 2012), the EHDI process (Krishnan et al., 2019; Pendersen & Olthoff, 2019), hearing aids and assistive technology (Haddad et al., 2019; Van der Spuy & Pottas, 2008), and parental support services (Haddad et al., 2019; Van der Spuy & Pottas, 2008). This expressed need for information may be indicative of a lack of accessible information to support decision making.

Aside from the call for appropriate information within the Early Hearing Detection and Intervention Act of 2017, various governmental and non-governmental organizations have made recommendations on how to ensure materials are accessible to the general public and available to be used in decision making. It is recommended that all health information, such as EHDI-based websites, be written at no greater than a 6th-grade reading level (Safeer & Keenan, 2005; Sax et al., 2019; U.S. Department of Health and Human Services, n.d.; Weiss, 2006). The purpose of this study was to assess the readability of commonly accessible information for parents on early intervention for children who are DHH using a similar methodology to Sax et al. (2019). From here, there can be future work to look at the necessity of editing online materials for increased readability and may serve as a mechanism for addressing lost to follow up. The potential compounding of inaccessible informational materials with a need for more information for parents of children who are DHH merits evaluation.

The concept of literacy is linked to health literacy. Health literacy is the skill of taking in, processing, and understanding health-related content such as information and needed services (American Speech-Language-Hearing Association, n.d.). The type of language used when providing information must be consistent with an individual's literacy level. The literature has noted that audiologists tend to speak at a level of complexity that is not accessible or is vastly different from what an individual or family may be able to understand (Donald & Kelly-Campbell, 2016; Nair & Cienkowski, 2010). Given that health literacy is already a concern across the medical field, EHDI-based information for families is not exempt from this weakness or the need for a global overhaul in the documentation and general communication provided to families (Sax et al., 2019).

The accessibility of written materials in EHDI has been approached in terms of referral for diagnostic services following hearing screening. Sax et al. (2019) evaluated the readability of the top 55 links derived from a Google search of "failed newborn hearing screening" and materials from top medical institutions on the same topic. Their study concluded that "online patient education materials about the newborn hearing screen may be too difficult for the average reader" and serves as a call for material revision to be more inclusive of potential readers (p. 168). This information provides insight into potential risk factors for children not following through for hearing evaluations after referral on their screening. It also begs the question of how accessible materials for the next step in the EHDI system, early intervention, are to the average reader. This study continues this line of inquiry to assess if materials found online about early intervention for children who are DHH conform to readability recommendations.

Materials and Method

Data Collection

Google searches for "early intervention deaf" and "early intervention hear" were performed on February 4, 2020, and the first five pages of English language results were compiled. Google was selected as the search engine to be used based on the precedent in the literature to use this as the primary search tool and is supported by recent publications of the use of "conventional search engines" inclusive of Google (Ahmadian et al., 2020; Sax et al., 2019; Ting & Hu, 2014. The search term "early intervention" was selected to be broad enough to include services that families of children who are DHH can access, including those governed by Part C of the Individuals with Disabilities Education Act of 2004 and services that practitioners provide outside of that system. Both "deaf" and "hear" were selected to be used in conjunction with early intervention to represent the various terms that parents may have experience with or heard, including deaf, hearing loss, hearing impairment, hard of hearing, and hearing levels (Joint Committee on Infant Hearing [JCIH], 2019). This procedure led to 53 links in response to "early intervention deaf" and 50 links in response to "early intervention hear."

Article Modification

Article modification is the process of preparing materials for readability analysis. The methods used here represent various readability analyses within the healthcare domain. Each link was opened and its contents copied entirely. The content was pasted as plain text into a Microsoft Word document. Documents were then modified by removing extraneous text as delineated by related studies (Badarudeen & Sabharwal, 2008, 2010; Flesch, 1948; Kong & Hu, 2015; Sax et al., 2019; Ting & Hu, 2014; Wong & Levi, 2016, 2017). Extraneous text includes navigation links, author information, dates, headers, titles, subheaders, acknowledgments, copyright notices, references, disclaimers, citations, feedback questionnaires, URLs, numbers, decimal points, bullets, abbreviations, paragraph breaks, colons, semicolons, dashes, captions, percentages, and charts/figures.

Readability Analysis

Implementing the methodology of Sax and colleague's (2019) evaluation of newborn screening materials, this study has six assessment tools that create a rich data set. These include Flesch-Kincaid Grade Level (FKGL), Flesch Reading Ease Score (FRES), Gunning-Fog Index (GFI), Simple Measure of Gobbledygook (SMOG), Coleman-Liau Index (CLI), and Automated Readability Index (ARI). One online readability calculator was used for short samples (https://www.webpagefx.com/tools/read-able/) with a second calculator used for longer samples (https://www.readable.com/).

Statistical analysis was only conducted on FKGL. Current recommendations on the use of reliability calculations are to keep the grade level required to understand the material at a 6th-grade level, equivalent with it being below 7th grade as the average adult reads at a level consistent with an 8th-grade education (Weiss, 2006; U.S. Department of Health and Human Services, n.d.). With this, the use of the FKGL to determine the grade level of a resource fits well with providing actionable steps to assess what materials are most accessible and is a consistent measure in the literature (Kong & Hu, 2015; Sax et al., 2019; Ting & Hu, 2014; Wong & Levi, 2016, 2017). The FKGL is based on the length of a sentence in words and the number of syllables in the words that make up the sentences (Flesch, 1948; Sax et al., 2019; Weiss, 2006). The formula provides the approximate grade level of education that is required to understand the text. All other reliability calculations are included to support claims from FKGL about the grade level needed to read a text as FKGL should vary with these other measures. In addition, these measures are additional metrics to determine correlation both between and within observers.

Two raters also reviewed materials to determine the intended audience. Materials were deemed to be parent-oriented when using the possessive tense in writing about the child, rights, or expectations (e.g., You and your child will work with the early intervention team to decide what services to access), or when the information was framed as an introduction to the topic of early intervention. Provider-oriented materials did not use the possessive or were framed as practice guidelines. The determination of the intended audience was conducted by the first author and a research assistant. Materials determined to be provider-oriented were included in the analysis as they are accessible and presented within the search results alongside parent-oriented materials. Thus, parents looking for parent-oriented materials could find these and review them as a part of their search. To determine the material source, each web page was reviewed for the group that held the copyright, provided updates, or hosted the web page.

Statistical Analysis

Statistical analysis was completed using SPSS version 27. Descriptive statistics were calculated for all nominal categories. Statistical significance was set at .05 and calculated across groups using independent samples t-tests or ANOVAs.

Results

Before the statistical analysis of the readability scores was started, the reliability of the article modifications was assessed. This step was done to ensure that while keeping with the procedure of article modification, there was replicability in the methods. The first author repeated the editing process on 30 randomly selected links over one week after their original modification to provide intra-observer reliability data. To determine inter-observer reliability of the article modification process, a research assistant performed the editing process on another 30 randomly selected links and then calculated readability scores. Intra- and inter-rater reliability were calculated using intraclass correlation coefficients. Intra-rater and inter-rater reliability were significant for each measure of readability ($p < .002$).

Of the 103 links collected, 40 (38.8%) were excluded from analysis for being a video only ($n = 1$), being a dead link ($n = 1$), being contact information only that would have been erased during article modification for analysis ($n = 2$), being a list of links only that would have been erased during article modification for analysis ($n = 2$), primarily selling something ($n = 3$), being a job ad only ($n = 4$), being a duplicate of a link that was already accepted for analysis ($n = 8$), or being a journal article ($n = 19$).

This left 63 links for evaluation, with 24 (38.10%) derived from the search term "early intervention deaf," 32 (50.79%) derived from the search term "early intervention hear," and 7 (11.11%) links appearing in both searches.

All but one of the webpages reviewed were written at a reading level above 6th-grade and thus not in line with literacy recommendations. The average FKGL of all documents ($n = 63$) was 12.62 ($SD=2.65$), with a range from 4.4 to 18.1 (see Figure 1).

Of those links included in the evaluation, 49 (77.78%) targeted parents/the general public and 14 (22.22%) targeted professionals. Between raters, there was no

Figure 1

Flesch Kincaid Grade Level (FKGL) Scores Across Populations and Search Pages
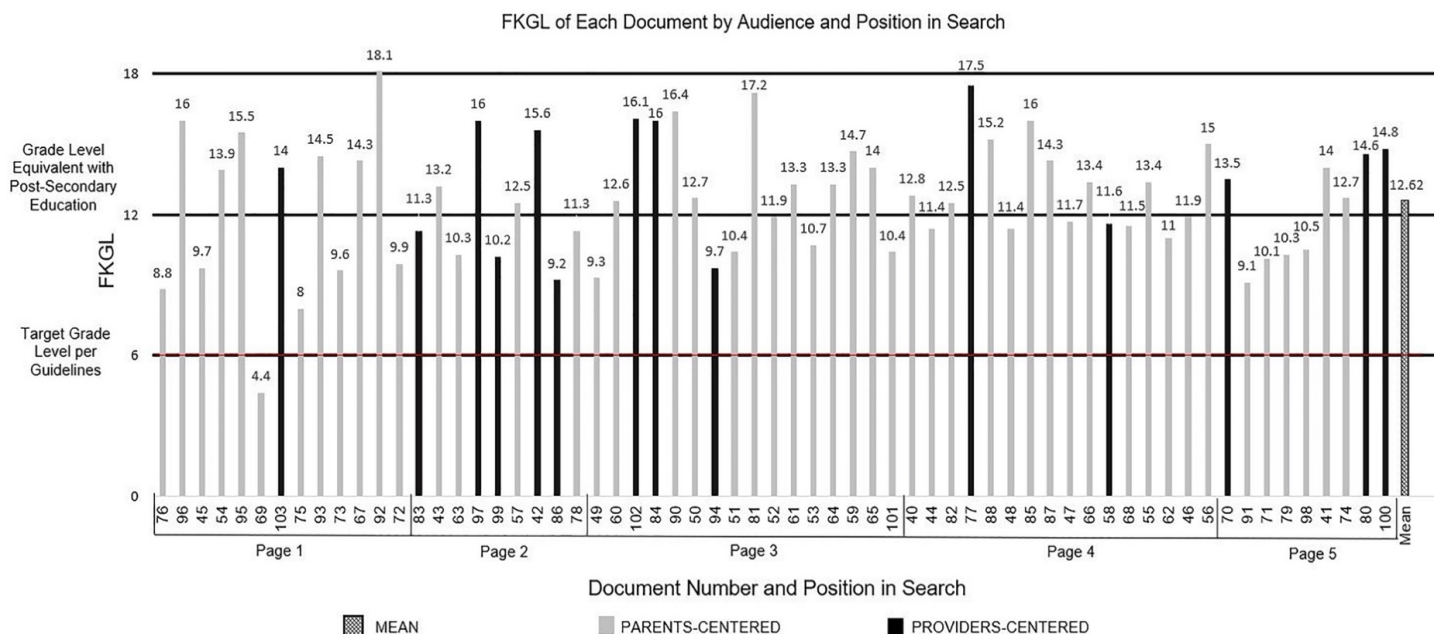
disagreement across any of the resources in terms of the intended audience. Forty-nine resources were deemed parent-oriented with a mean FKGL of 12.41 ($SD = 2.74$) and 14 were determined to be provider-oriented with a mean FKGL of 13.357 ($SD = 2.26$). Using an independent samples t-test, results indicate no significant difference in FKGL based on the intended audience ($p > .17$).

A majority of these links came from large reputable sources such as government websites ($n = 22$), advocacy groups ($n = 14$), educational systems ($n = 12$), hospitals and other healthcare providers ($n = 7$), or professional groups ($n = 3$). Only five results (7.93%) were from general media outlets. With a one-way ANOVA, there was not a significant impact of the information's source on the FKGL of the document ($p > .1$).

The search result page that resources were present on was also considered for analysis. Resources were grouped by the page on which the result was found, ranging from the first to the fifth page (see Figure 1). A one-way ANOVA revealed no significant impact of the display page on FKGL ($p > .8$).

Discussion

At the heart of pediatric audiology and early intervention services for children who are DHH is the family. Families shape a child's trajectory by working with professionals to set developmental goals with the Individualized Family Service Plan (IFSP; Individuals with Disabilities Education Act of 2004). Before the IFSP, parents serve as gatekeepers who decide whether or not to enroll in early intervention services. As they make this decision, they may be referred to, find, or be provided information from friends, family, professionals, and the internet. The support that parents find must be at a level that is accessible to them and meets their literacy needs while scaffolding their health literacy and decision-making skills.

Accessibility of information is a critical component to meeting parents' and caregivers' educational needs around hearing-related topics such as early intervention. Overall, current online materials related to early intervention for children who are DHH are not written in a manner that is accessible according to health literacy guidelines (see Figure 1). Some of the most apparent drivers of high FKGL scores are long sentences and multisyllabic words (Flesch, 1948). Audiology-specific recommendations to address access concerns also call for the reduction of jargon, among other components. The intersection of long sentences, jargon, and many multisyllabic words can be seen in this sentence from a parent-oriented material used in the study with a FKGL of 18.1.

> The U.S. Department of Education recently published IDEA and FERPA Confidentiality Provisions [PDF] (June 2014), a side-by-side comparison of the primary legal provisions and definitions in the Individuals with Disabilities Education Act (IDEA) Parts B and C and the Family Educational Rights and Privacy Act (FERPA) that relate to the confidentiality of personally identifiable information of children served under the IDEA. (NCHAM, 2020)

Although all of this information is critical for families to know and understand, the sentence length, vocabulary, and use of jargon may be challenging for a first-time reader or new parent to understand without support. Within early intervention, there may be situations where specific vocabulary and sentence structure is required. However, to work toward accessibility, there is a need for scaffolding to support understanding in these situations. These materials may not be accessible and thus are not working to address the stated and hypothesized lack of education that impacts

parents and, thus, children who are DHH as they enter early intervention.

This study, combined with other works on health literacy, patient education, and accessible materials, suggests that all sources of information, including government-sponsored sites, educational systems, and the general media, could benefit from making materials more accessible. Given that these results are consistent with Sax et al. (2019), it leads to the conclusion that both hearing screening and hearing-related early intervention could benefit from improved accessibility. Increased accessibility could be attained by implementing readability strategies as described by several national groups, including the U. S. Department of Health and Human Services (2020) and the American Speech-Language-Hearing Association (n.d.). When looking at readability scores, the grade level is impacted by a number of features, including syllables used in words and the length of sentences (Flesch, 1948; Weiss, 2006).

Although authors have been cautioned not to write with a readability formula in mind, potential strategies to support readability and lower required FKGL do exist. Remediation for current materials to improve readability and thus accessibility include the use of short paragraphs that implement active voice, using one and two-syllable words, prioritizing information and considering the relative importance of information to be presented, reducing jargon, using simple pictures/graphics, and encouraging the potential use of audience assessment measures to determine if the material is accessible (American Speech-Language-Hearing Association, n.d.; U.S. Department of Health and Human Services, 2020, n.d.).

Of the webpages that parents and caregivers may access to learn about early intervention for children who are deaf or hard of hearing (DHH), the average readability score is higher than is recommended. Thus, parents and caregivers who turn to the internet as a source of information to help them make early intervention decisions may find inaccessible information. These results suggest that those who develop and maintain web-accessible content on early intervention for children who are DHH need to examine the role of readability in their materials. However, increased readability measures of English language materials do not explicitly address the needs for culturally- and linguistically-diverse materials on the same topics. This work can be considered a reminder to consider the current recommendations and strategies from reputable national resources to ensure accessibility of information.

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Hearing Loss Diagnosis Provision of Information and Support: Audiologist and Parent Perspectives

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Abstract: When a child is diagnosed with hearing loss, the parents are faced with many decisions that will impact their child's future. This study aimed to obtain data to determine viewpoints on information being provided to parents of children with hearing loss from both audiologist and parent perspectives. Topics of information surveyed included information on modes of communication, Deaf culture, cochlear implants, emotional support, and state resources and laws. A survey was created and shared in Facebook groups for audiologists and parents of Deaf or hard of hearing children. The survey was completed by 91 audiologists and 111 parents. Audiologist and parent survey data were analyzed and compared to look for similarities, differences, and possible biases. The data obtained in the study showed that overall, information on the topics investigated is being provided to parents when their child is initially diagnosed with hearing loss. However, many parents felt that the information that was provided was insufficient; therefore, they did not feel confident in their decision-making process. Audiologists have the responsibility to provide parents with unbiased, extensive information for parents to successfully make informed decisions for their child. Provision of practical, comprehensive information and recommendations may lead to improved parent knowledge and confidence.

Keywords: Children; Hearing Loss; Cochlear Implants; Hearing Aids; Early Intervention; Listening and Spoken Language

Acronyms: ASL = American Sign Language; AV = Auditory Verbal; LSL = Listening and Spoken Language; SEE = Signed Exact English; UNHS = Universal Newborn Hearing Screening

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The introduction of Universal Newborn Hearing Screening (UNHS) protocols drastically improved early detection of hearing loss in infants. Prior to the implementation of these protocols, children were identified with hearing loss at 2 ½ to 3 years old on average, which is a critical period for speech and language development (Eiserman et al., 2007; Walker et al., 2014). When an infant or child is diagnosed with a hearing loss, parents have many decisions to make, including what mode of communication they will choose for their child. Since more than 90% of children who are born Deaf or Hard of Hearing (DHH) are born to hearing parents, many find themselves in a world previously unknown to them (Mitchell & Karchmer, 2004). Decisions on whether to have their child become auditory-verbal via the use of a cochlear implant or implement American Sign Language (ASL) and become immersed in Deaf culture often need to be made sooner rather than later due to the critical age of language development (Chute & Nevins, 2002; Humphries et al., 2012; Li et al., 2003). This early deadline and need to begin early intervention for language development may make parents feel rushed in their decision making (Hyde et al., 2010). Because it

is a choice that will determine their child's path in life, pressure can be felt by parents to make the right decision for their child (Chute & Nevins, 2002). Making effective decisions requires a certain level of confidence (Stewart, 2014). For a parent to be confident in their decision, they must acquire information from a variety of sources and consider all possible modes of communication options and outcomes for their child (Incesulu et al., 2003).

Upon a child's diagnosis of hearing loss, parents are faced with the choice of how they want their child to communicate. Fitzpatrick et al., (2008) identified several areas that parents deemed important following a diagnosis of hearing loss. These included audiological screening and therapy, social support, coordinated services, group support with other parents, and access to pertinent information. There is no right decision on which form of communication the child should use, as it varies from case to case and is based on what works well for the family. The main goal is to provide the child with a form of communication (Li et al., 2003). Research shows there is a critical period for language development, and it is recommended to intervene early so the child can meet

appropriate language milestones. Prolonging the decision of communication modality may result in language delays in children with hearing loss (Hayes et al., 2009). There are five main modes of communication, these include: Auditory Verbal (AV) or Listening and Spoken Language (LSL), Cued Speech, ASL, Signed Exact English (SEE), and Total Communication (Gravel & O'Gara, 2003).

One of the most utilized approaches is AV/LSL. AV is a communication approach that equips parents with the skills to maximize their child's speech and language development. This approach focuses on using the child's residual hearing along with having the child wear their amplification devices on a daily basis (Kaipa, 2016). Cued Speech is a communication strategy that uses manual phoneme-based handshapes in combination with mouth movements for speech. The purpose of Cued Speech is to promote understanding using speechreading along with visual cues for the phonemes being verbalized. ASL is often favored among deaf and hard of hearing populations in the United States. ASL is a form of manual communication with unique grammar and syntax, where hand movements and facial expressions play an important role in conveying information (National Institute on Deafness and Other Communication Disorders, 2019). SEE is a form of manual communication that is modeled after the English language. It is the visual form of English, representative of all English vocabulary and grammar. Total communication encompasses all modalities of communication to educate and optimize language development for the deaf and hard of hearing. This includes the use of gestures, fingerspelling, formal signing, body language, listening, lipreading, speech, and facial expressions (Hands & Voices, n. d.). Total communication capitalizes on the unique strengths and needs of each child to find the best modalities for language acquisition. All previously listed communication strategies should be discussed in detail with the child's parents so they can make informed decisions based on what they think will best serve their child and personal family dynamics.

It is critical that parents know their child's audiologist is doing everything possible to provide all of the support and information available to best help their child. Gilliver et al., (2013) analyzed 40 parental reports of experiences when their child was diagnosed with hearing loss. The study specifically examined emotional and informational support provided by their child's audiologist. They found that approximately half of the parents reported "a perceived lack of information provision" (Gilliver et al., 2013). Many parents reported that a single booklet was provided for them by the audiologist, forcing them to seek out additional early intervention and communication options for their child through the internet. Overall, "parents expressed a desire for more information than they have received" (Gilliver et al., 2013).

During their graduate studies, audiologists are trained in counseling; however, research has revealed counseling deficits in audiology. Training variations among different graduate programs can affect the degree to which students are prepared to effectively counsel patients (Muñoz et al., 2017). Research shows that during encounters with patients, audiologists have been found to dominate conversations, which reduces the opportunity for patients and caretakers to voice concerns and ask questions. Audiologists are using counseling skills such as reflection, assessment of psychological factors, and development of an action plan minimally (Muñoz et al., 2017). Audiologists, especially those that specialize in pediatric audiology, should implement evidence-based counseling services that are patient-centered. This includes providing parents with continuous support and information.

It is the parent's right to be provided uninfluenced information when they are deciding about their child's hearing loss. After their child's diagnosis, parents may be given or need to seek out a vast amount of information regarding the educational, communication, and technological options for their child (Hyde et al., 2010). It is critical that parents are provided a comprehensive understanding of their child's diagnosis and their options to make decisions that are free from the influence of opinion. Previous research has evaluated the parental decision-making process when choosing modes of communication (Li et al., 2003), and explored the extent to which audiologists provided training and skill support to parents seeking to integrate strategies for communication into their child's intervention (Munoz et al., 2015). Differentiating the current study from previous research is the surveying of both the audiologist and the parent of the child with hearing loss to look for discrepancies in the information reportedly given by the audiologist and received by the parent. Understanding what information is provided to parents by clinicians may provide insight into how to improve services and counseling, promoting successful patient outcomes.

The purpose of this study was to survey perspectives of parents of children diagnosed as Deaf or Hard of Hearing (DHH) as well as survey perspectives of audiologists who diagnose children as DHH. Information from the survey obtained data views on what information is being provided to parents from the audiologist perspective as well as the parent perspective regarding their child's hearing loss, Deaf culture, and available communication options. The hypothesis proposed that audiologists would overestimate the number of resources they provided to parents, while parents would report not feeling like they were provided with enough information and support from their audiologist.

Method

Participants

Audiologists and parents of children who are DHH were surveyed about their respective experiences involving information about modes of communication. These participants were recruited from active members in social media groups for audiologists and parents of children who are DHH. Specific social media groups on Facebook where the survey was shared included: Audiology Happy Hour; Audiology Antics and Anecdotes–for All Hearing Professionals, Pediatric Educational Audiologists: AKA–Freakin' Miracle Workers; National Hands and Voices Chapter; Alabama Hands and Voices Chapter; and

Support Group for Parents of Kids with Hearing Loss. Participant inclusion criteria was broad and included any parent of a child diagnosed as DHH or an audiologist who diagnosed children as DHH.

Survey

A survey was prepared using Qualtrics software (Qualtrics, Provo, Utah). The invitation to complete the survey included a brief introduction about the study, consent information, and a link to the online study. The survey was voluntary, anonymous, took approximately 10 minutes to complete, and no Personal Health Information (PHI) data was obtained in the process. At any time before submission of the survey, participants were able to withdraw from participation without penalty. The total number of surveys completed by audiologists was 91 and the total number of surveys completed by parents was 111.

The data collected pertained to the information being provided by audiologists to parents concerning various modes of communication and other educational information regarding hearing loss and options for amplification. Both audiologists and parents' experiences were measured by the survey. Several questions were administered to pinpoint the type and extent of information that was provided at the time of diagnosis.

Audiologist Survey

At the beginning of the survey, the participant was asked to identify as an audiologist or a parent. The answer to this question determined which set of questions the participant would be asked. If the individual selected *Audiologist*, they would be led to a survey of eight questions. Five of the questions allowed the audiologist to choose more than one answer (unlimited choice), and three questions required the audiologist to choose one answer (multiple choice). The unlimited choice questions all had an *Other* option and were followed by a comment section to explain and elaborate on the survey data. The multiple-choice questions gave a comment section for the audiologist to elaborate if they responded *No* or *Other* to the question. For reference, a complete listing of the audiologist survey questions can be found in Appendix A.

Parent Survey

If the individual selected *Parent*, they would be led to a set of thirteen questions. Five of the questions allowed the parent to choose more than one answer (unlimited choice). The unlimited choice questions all had an *Other* option and were followed by a comment section to explain and elaborate on the survey data. Six of the questions allowed the parent to choose one answer (multiple choice), and two of the questions were rating scales. The first rating scale had parents rate the information they received about each communication option listed by selecting either *Excellent*, *Good*, *Fair*, or *Poor*. The second rating scale had six individual statements concerning their personal experience with information on early intervention, communication modes, emotional support, and audiologist bias/pressure. The parent was asked to rate each statement with *Strongly Agree*, *Agree*, *Somewhat Agree*,

Neither Agree or Disagree, *Somewhat Disagree*, *Disagree*, or *Strongly Disagree*. For reference, a complete listing of the parent survey questions can be found in Appendix B.

Results

Demographics

Out of the 91 audiologists who completed the survey, 39.13% of participants worked at a hospital, 10.14% worked at a university, 8.70% at a private practice, and 8.70% for an Ear, Nose, and Throat (ENT) practice. One third of the participants (33.33%) worked at other settings such as educational audiologists, non-profit, and state agencies (see Figure 1 for results and Table 1 for comments). Audiologists that had been practicing for less than 5 years accounted for 31.88%, 17.39% had been practicing 5 to 10 years, 23.19% for 10 to 15 years, 7.25% for 15 to 20 years, and 20.29% for more than 20 years. See Figure 2 for results.

Figure 1
Workplace Setting: Audiologist
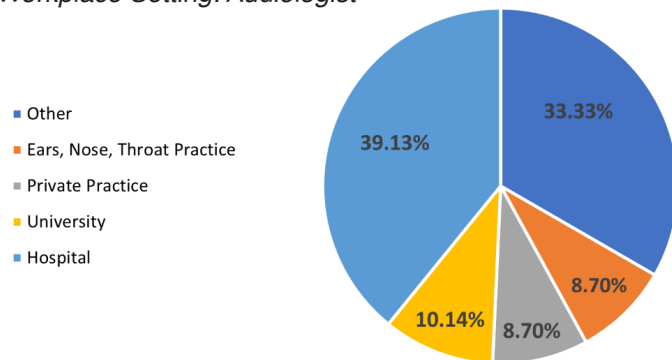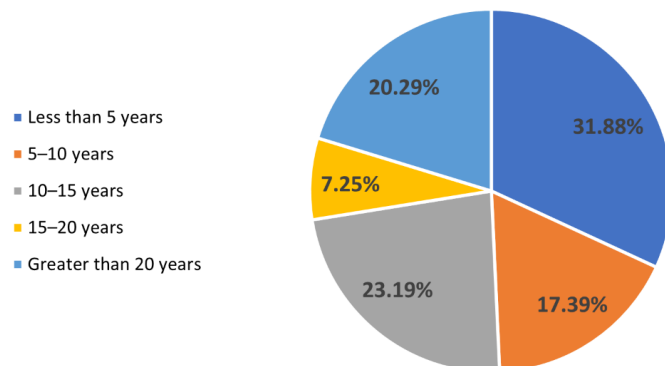

Table 1
Workplace Setting: Audiologist

Q1: What best describes your work setting?	Audiologist Comments
	• State Agency
	• Private Non-Profit
	• Early Interventionist
	• Educational Audiology
	• Multispecialty Pediatric Clinic
	• Contract Audiologist

Figure 2
Years Practicing: Audiologists

Out of the 111 parents who completed the survey, none of the participants responded that they were deaf, 6.25% indicated that they were hard of hearing, and 93.75% were individuals with normal hearing. Parents that reported their child was diagnosed as DHH from birth to 3 months of age accounted for 47.56%, 2.44% from 4 to 6 months of age, 1.22% from 7 to 12 months of age, 7.32% from 1 to 2 years old, 10.98% at 2 to 3 years old, and 30.49% over the age of 3 (see Figure 3). According to parental participant report, 24.69% of children were part of the Deaf Culture and 75.31% were not. Parents reported their child was fit with hearing aids in 77.46% of cases, and 22.54% reported their child was fit with a cochlear implant.

Figure 3
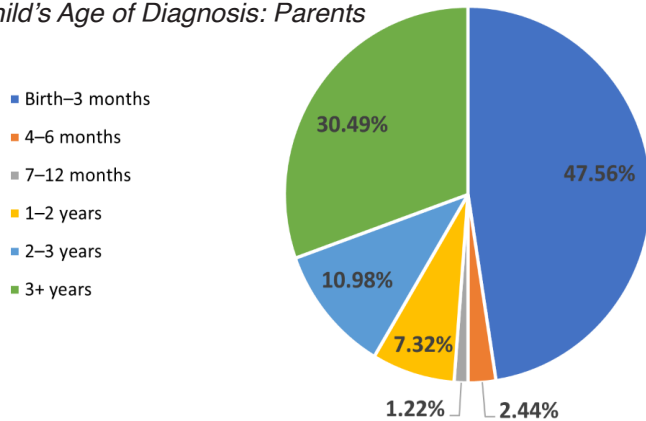*Child's Age of Diagnosis: Parents*

Legend:
- Birth–3 months
- 4–6 months
- 7–12 months
- 1–2 years
- 2–3 years
- 3+ years

30.49%
47.56%
10.98%
7.32%
1.22%
2.44%

Sources of Information and Materials

Audiologists were asked to report what materials they used to explain the hearing loss to parents. Audiologists were given the option to select multiple answers and the reported percentages reflect the total responses selected for that category. The most frequently reported was Verbal Explanation (25.48%), then Visual Aids (24.71%), Pamphlets (17.87%), Hearing Loss Simulation (14.83%), Outside Reading Materials (14.45%), and Other (2.66%). Further analysis of the responses found that 24.00% of audiologists chose not to answer this question, while the other audiologists (76.00%) reported they used a combination of these materials when explaining hearing loss to parents. The comment section was used by respondents to express other responses and more detailed information. Results can be found in Figure 4a, and comments can be found in Table 2a.

Parents were asked what materials were provided by an audiologist to further explain their child's hearing loss. The parent was able to select multiple answers and the reported percentages reflect the total responses selected for that category. The most frequently reported was Verbal Explanation (38.27%), then Pamphlets (22.45%), Visual Aids (14.8%), Outside Reading Materials (11.22%), Hearing Loss Simulation (6.12%), and Other (7.14%). Further analysis of the responses found that 27.00% of the parents chose not to answer this question,

Figure 4a
Sources of Information/Materials: Audiologists

Legend:
- Verbal Explanation
- Visual Aids
- Pamphlets
- Hearing Loss Simulation
- Outside Reading Material
- Other

2.66%
14.45%
25.48%
14.83%
17.87%
24.71%

Figure 4b
Sources of Information/Materials: Parents

Legend:
- Verbal Explanation
- Visual Aids
- Pamphlets
- Hearing Loss Simulation
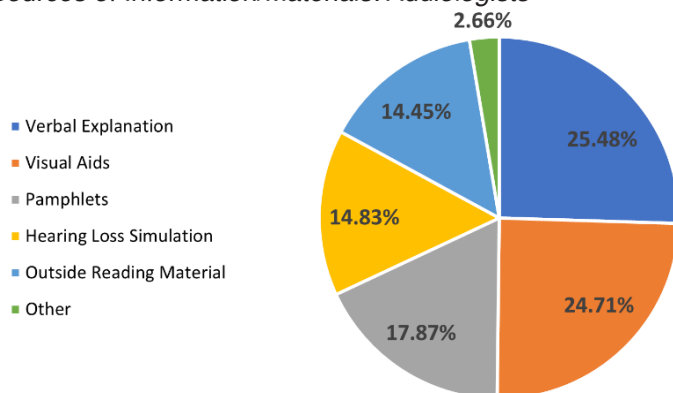- Outside Reading Material
- Other

7.14%
11.22%
6.12%
38.27%
22.45%
14.80%

Table 2a
Sources of Information/Materials: Audiologists

Q3: What materials did you use in order to explain the child's hearing loss to the parents?	Audiologist Comments
	• Informational videos, social media, etc. • Audiogram, speech banana, other parents, etc. • Hands and Voices Resource Guide • I ask parents how they would like the information presented; everyone has different learning styles. • Materials from Beginnings are excellent.

Table 2b
Sources of Information/Materials: Parents

Q5: What materials were you provided by an audiologist in order to explain your child's hearing loss?	Parents Comments
	• Contacts with School for the Deaf and families with similar experiences • Information on FM systems • Resource binder on deaf issues, culture and assorted information explaining topics • Just test results (Auditory Brainstem Response [ABR], audiogram, etc.) • None • A resource binder on deaf issues, culture and assorted information explaining different things. Also had different organizations contact info.

while 21.60% of parents reported that only one type of material was used in explaining their child's hearing loss with Verbal Explanation (16.20%) being the most frequent method. The other parents (51.32%) reported that the results were explained with two or more types of materials. Results can be found in Figure 4b, and comments can be found in Table 2b.

Cochlear Implant and Deaf Culture Information

Audiologists were asked if they provided parents with information on both cochlear implants and Deaf Culture. It was found that 77.94% of audiologists reported providing both information on cochlear implants and Deaf culture, while 22.06% of audiologists reported not providing this information. Results can be found in Figure 5a, and comments on why they did not provide both can be found in Table 3a. When parents were asked if they were provided information on both cochlear implants and Deaf Culture, 71.95% of parents reported *No*, 12.20% reported *Yes*, and 15.85% reported *Other*. Results can be found in Figure 5b, and comments can be found in Table 3b. A stark contrast is seen in the materials reportedly given by the audiologist and those reported being received by the parent. Although this numerical difference does represent a breakdown in relaying of information from the professional to the parent, it has also been influenced by factors such as (a) setting of the audiologist, (b) parental inquiry and wants for their child, and (c) the severity of the child's hearing loss not necessitating these conversations (see Table 3a and 3b).

Figure 5a
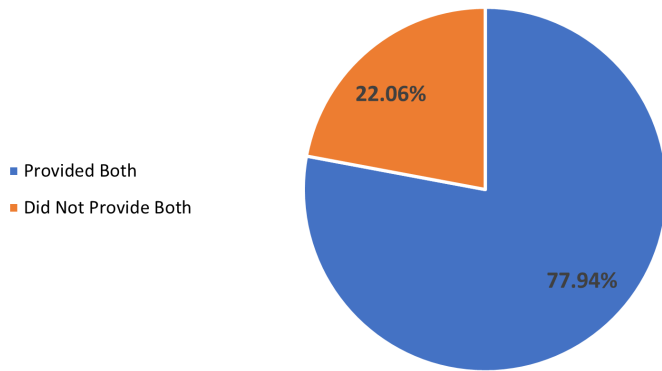*Cochlear Implant and Deaf Culture Information: Audiologists*

Figure 5b
Cochlear Implant and Deaf Culture Information: Parents
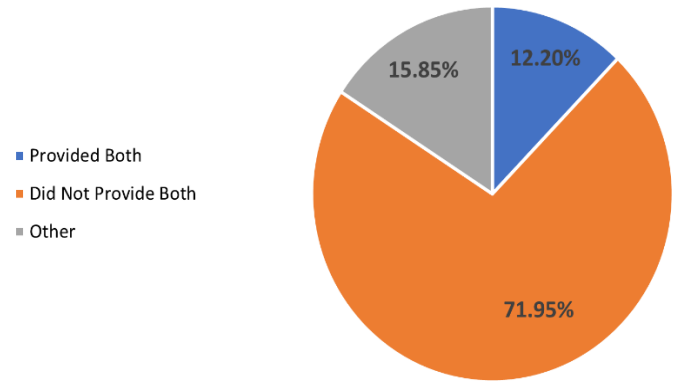
Table 3a
Cochlear Implants and Deaf Culture Information: Audiologists

Q4: Did you provide the parents with information on both cochlear implants AND Deaf Culture?	Audiologist Comments
	● All parents I've seen want their child to be hearing
	● I do not do cochlear implants, so I generally do not discuss them at length. I refer ALL newly diagnosed children and their parents to Parent Support services, which include Guide By Your Side, unbiased support from parents of both aided/implanted children and children using ASL. I do not feel I am enough of an expert on either to provide an opinion.
	● We provide information on ALL communication options. So, yes, but this question could be expanded on. Also, we only discuss CIs if applicable. CI candidates referred to another AuD
	● I would not choose either yes or no, but rather, "it depends." I allow the parents'/family's questions to guide the information provided, especially in the early stages. In general, I think audiologists talk too much and listen too little. What I do consistently encourage is that the family develop an action plan- as quickly as their unique circumstances permit.
	● In our team if we newly identify a profound hearing loss we send it to audiologists that work either CIs for confirmation and discussion of hearing aids CI and Deaf culture. I usually ask about whether the parents' goal is for their child to use hearing/speech to communicate and only provide info on Deaf Culture if parents ask about other options.
	● We are an LSL preschool. By the time families reach us they are implanted (or on the way) and have already chosen communication method
	● If I see a child who has significant hearing impairment, I refer them to the ENTs who perform cochlear implants and have audiologists who deal almost exclusively with kids. They are plugged in to all the local resources for these kids.
	● I provide info on learning ASL but not on Deaf culture. They are not the same, so I chose no.

Note. ASL = American Sign Language; CI = cochlear implant; ENT = Ear, Nose, and Throat doctor; LSL = listening and spoken language

Table 3b

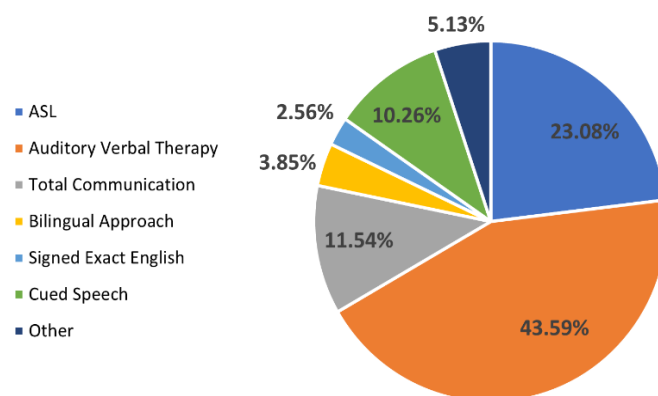*Cochlear Implants and Deaf Culture Information: Parents*

Q6: Were you provided with information on both cochlear implants AND Deaf culture by an Audiologist?	Parents Comments • CI but told that child would be a good candidate if needed later • Little information • "They told me cochlear implants were the only thing that could help my son learn. They completely ignored that there should always be a trial of hearing aids first and they were incredibly rude about deaf culture" • Neither • No, but only because we did not need these

Note. CI = cochlear implant.

Modes of Communication

Audiologists were asked to indicate what modes of communication they used to provide information to parents. They could respond by selecting all modes that they have provided, and the reported percentages reflect the total responses selected for that mode of communication. The most frequently reported was American Sign Language (25.11%), Auditory Verbal (24.67%), Total Communication (23.35%), Bilingual Approach (14.54%), Signed Exact English (6.17%), Cued Speech (4.41%), and Other (1.76%). Additional analysis found that 31.87% of audiologists chose not to answer this question, while 50.55% of audiologists reported giving parents information on three or more modes of communication with auditory verbal and ASL being the two most often recommended approaches. Results can be found in Figure 6a, and comments can be found in Table 4a. Parents were asked what information on modes of communication they were provided and could select multiple answers. The reported percentages reflect the total responses selected for that mode of communication. The majority reported receiving information on Auditory Verbal (43.59%). Information for other modes of communication were ASL (23.08%), Total Communication (11.54%), Cued Speech (10.26%), Other (5.13%), Bilingual Approach (3.85%), and Signed Exact English (2.56%). Additional analysis found that 63.96% of parents chose not to answer this question, while 21.62% of parents reported getting on one mode of communication with auditory verbal being the most often recommended approach. Parents reported 9.00% of the time they were given information on two modes of communication and 5.41% of the time they were given information on three or more modes of communication with auditory verbal and ASL being the most often recommended approaches. Results can be found in Figure 6b, and comments can be found in Table 4b.

Figure 6a

Modes of Communication: Audiologists

Note. ASL = American Sign Language.
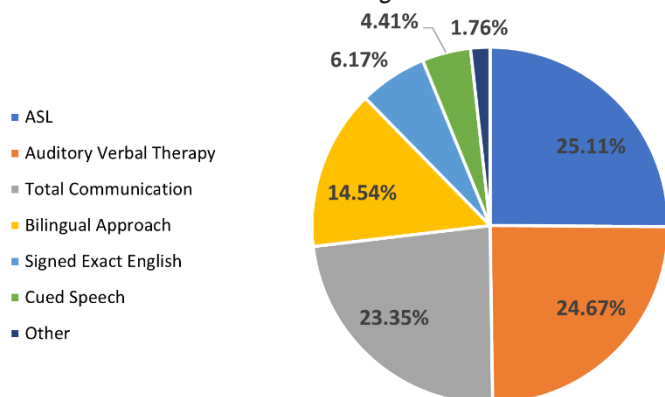
Figure 6b

Modes of Communication: Parents

Table 4a

Modes of Communication: Audiologists

Q5: Which of the following did you provide information regarding modes of communication?	Audiologists Comments • I would argue that AVT and TC are not modes of communication but are teaching methods • As I said above, I do not feel I am enough of an expert on any of these methods of communication to guide the parent of a profoundly hearing-impaired child. When I initially diagnose a child with a profound hearing impairment, I refer these parents to agencies in our state who provide excellent, unbiased information on many of these methods of communication. Again, really depends on the degree of hearing loss. • Parents are provided with information that allows them to access information about all treatment options so they can be make informed decisions for their child and take the lead.

Note. AVT = auditory verbal therapy; TC = total communication.

Table 4b

Modes of Communication: Parents

Q7: Did an audiologist provide you with any of the following information regarding modes of communication?	Parents Comments ● Information came more from EI ● Not our initial audiologist ● Child already speaking

Note. EI = early intervention.

Emotional Support Information

Provision of emotional support materials was also evaluated. Audiologists were given the option to select multiple answers. The reported percentages reflect the total responses selected for that category. The most frequently reported were Family Support Groups (36.77%), Blogs/Social Media Groups (25.16%), Deaf Mentors (16.77%), Family Counseling Services (16.77%), and Other (4.52%). Additional analysis found that 30.77% of audiologists chose not to answer this question, while 31.87% of audiologists reported giving three or more supports with the same frequently reported categories listed above. Information on one support was reportedly given by 13.19% of audiologists, while 24.18% reported giving information of two supports. Results can be found in Figure 7a, and comments can be found in Table 5a. Parents were asked what information the audiologist provided on emotional support materials and were also given the option to select multiple answers. The reported percentages reflect the total responses selected for that category. The most frequently reported answers for parents were Family Support Groups (46.67%), Blogs/Social Media Groups (18.33%), Deaf Mentors (16.67%), Family Counseling Services (8.33%), and Other (10%). Additional analysis found that 63.10% of parents chose not to answer this question, while 24.32% of parents reported getting information on one support, 5.40% on two supports, and 5.41% on three or more supports with the same frequently reported categories listed above. Results can be found in Figure 7b, and comments can be found in Table 5b.

Figure 7a

Emotional Support Information Provided: Audiologists
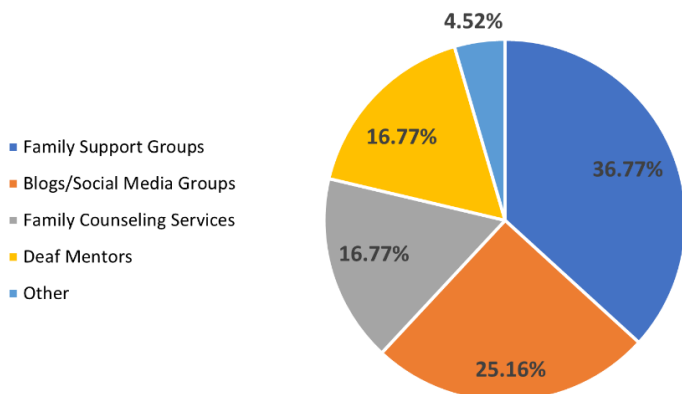
Figure 7b

Emotional Support Information Provided: Parents
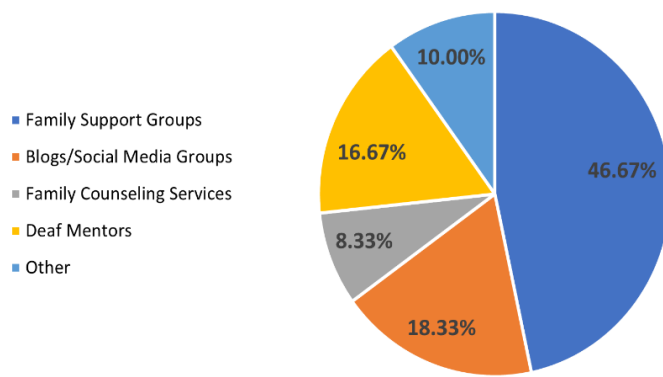
Table 5a

Emotional Support Information Provided: Audiologists

Q6: Which of the following did you provide information on regarding emotional support?	Audiologists Comments ● Hands & Voices, CRS Parent Consultant, Family Voices ● Summer camps in our state, semi-annual family events available through the school district ● Again, this is unique to each family, but on the whole, Hands and Voices Guide by Your Side program is an excellent resource as is making family to family links where appropriate. Each family's circumstances are unique. Every effort is made to support parent choice at every stage. ● Other parents who have children with hearing loss. ● EI/Beginnings ● Educational Support Groups

Note. CRS = Children's Rehabilitation Services; EI = early intervention.

Table 5b

Emotional Support Information Provided: Parents

Q8: Did an audiologist provide you with any of the following information concerning emotional support?	Parents Comments ● Early Intervention ● Beginnings ● Phone number of a family in our town who's now grown kids have CIs and spoken language ● Referral to early intervention and schools for the deaf who connected us to families, deaf adults, and support groups

Note. CI = cochlear implant.

State Resources

Audiologists were asked to indicate what state resources they recommended to parents and were given the option to select multiple answers. The reported percentages reflect the total responses selected for that category. The most frequently provided responses were Early Intervention (52.42%), Hands and Voices Chapter (38.71%) and Other (8.87%). Additional analysis found that 25.27% of audiologists chose not to answer this question, while 28.57% of audiologists gave information to parents on both Early Intervention and Hands and Voices Chapter. Audiologists gave parents information on Early Intervention in 6.59% of cases and in 8.79% of cases a combination of state resources was given. Results can be found in Figure

8a, and comments can be found in Table 6a. Parents were asked what information they received from audiologists on state resources, and they were given the option to select multiple answers. The reported percentages reflect the total responses selected for that category. The most frequently reported responses were Early Intervention (55.56%), Hands and Voices Chapter (23.33%), and Other (21.11%). Additional analysis found that 36.03% of parents chose not to answer this question, while 32.43% of parents were given information on only one state resource (most often Early Intervention and Hands and Voices) and 17.12% of parents reported receiving information on two state resources (most often Early Intervention). Results can be found in Figure 8b, and comments can be found in Table 6b.

Figure 8a
State Resources: Audiologists

- Early Intervention
- Hands and Voices Chapter
- Other

Figure 8b
State Resouces: Parents

- Early Intervention
- Hands and Voices Chapter
- Other

Table 6a
State Resources: Audiologists

Q7: Which of the following state resources did you recommend to the parents?	Audiologists Comments
	• Public health nursing • Offer to sponsor online sign classes and John Tracy clinic • Children's Rehabilitation Services for hearing aids (Medicaid) • Colorado Home Intervention Program (CHIP) providing home-based early intervention support and services from birth to age 3 years. • I have reservations about our state EI services. The intensity and frequency of intervention is insufficient to support positive outcomes. • Referrals to private providers or the school for the deaf (depending on chosen communication mode) is always required. In our state, Hands and Voices is not a "state resource," but I think it would be beneficial if it was. • Guide by your side program • AG Bell Chapter

Note. EI = Early Intervention.

Table 6b
State Resources: Parents

Q9: Which of the following state resources were recommended by your audiologist?	Parents Comments
	• None • Really nothing like this was offered/available • Our audiologist never recommended any resources. I had to research them myself. • Eventually we connected to Hands and Voices, but not until a few years after diagnosis. • Already in EI • School for Deaf and Blind

Note. EI = Early Intervention.

Legislative Materials

Provision of information on legislation regarding the Deaf and hard of hearing was also evaluated. Given the option to select multiple answers, the reported percentages reflect the total responses selected for that category. Audiologists most frequently reported providing information on the Individualized Family Service Plan/Individualized Education Plan (27.94%), Individuals with Disabilities Act (22.06%), Section 504 of the Rehabilitation Act of 1973 (21.32%), American Disabilities Act of 1990 (8.09%), and Other (3.68%). Additional analysis found that 29.67% of audiologists chose not to answer this question, while 26.37% indicated they did not give information on any of

the choices listed or they gave information indicated as Other. Most of the audiologists (37.36%) reported giving information on a combination of the listed laws. Results can be found in Figure 9a, and comments can be found in Table 7a. Parents were given the choice to select multiple answers and the reported percentages reflect the total responses selected for that category. Most frequently reported responses were Family Service Plan/Individualized Education Plan (36.36%), Individuals with Disabilities

Education Act (27.73%), Section 504 of the Rehabilitation Act of 1973 (15.91%), American Disabilities Act of 1990 (13.64%), and Other (9.09%). Additional analysis found that 25.23% of parents chose not to answer this question, while 13.51% of parents were given information on only one law, 4.50% of parents reported receiving information on two laws, and 4.50% of parents reported receiving information on three or more laws. Results can be found in Figure 9b, and comments can be found in Table 7b.

Figure 9a
Legislative Materials: Audiologists

Legend:
- Family Service Plan/Individualized Education Plan
- Individuals with Disabilities Education Act
- Section 504 of the Rehabilitation Act of 1973
- American Disabilities Act of 1990
- Other

Values: 27.94%, 22.06%, 21.32%, 8.09%, 3.68%
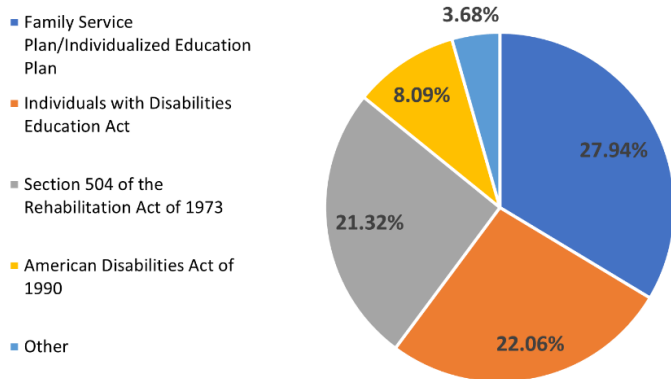
Figure 9b
Legislative Materials: Parents

Legend:
- Family Service Plan/Individualized Education Plan
- Individuals with Disabilities Education Act
- Section 504 of the Rehabilitation Act of 1973
- American Disabilities Act of 1990
- Other

Values: 36.36%, 27.73%, 15.91%, 13.64%, 9.09%
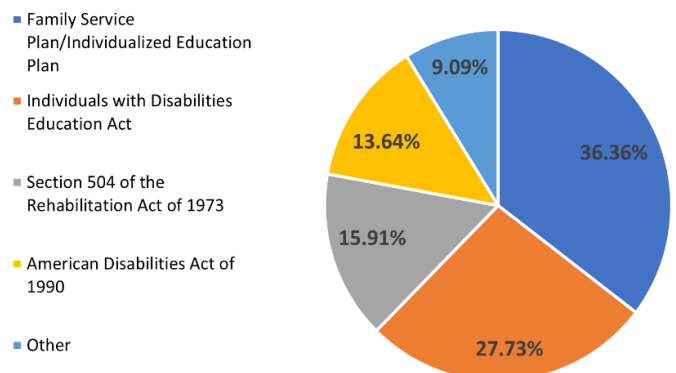
Table 7a
Legislative Materials: Audiologists

Q8: Which of the following laws did you provide information on to the parents?	Audiologists Comments • There is SO MUCH information being provided when a child is initially diagnosed with hearing loss, I don't feel that particular time is the best to discuss laws and advocacy for the child. I generally provide age appropriate information at follow-up appointments and guide the parents with the appropriate channels to contact re: obtaining an IFSP/IEP or 504. Unfortunately, in the city I live in, many of the parents are not very motivated despite being educated about their child's hearing impairment to advocate for their child and many, many of the schools do not comply with IDEA, so the children do not get the accommodations they need. It is extremely difficult to empower some parents while they are grieving the loss of their "perfect" child. Then you have others who take it and run with it. It is the nature of our business :-) • I talk about school laws when children get close to school age. Also, IEPs are part of IDEA • ...but not at initial diagnosis! Families need space and time to process. This information can come later. • Our EI/Beginnings does this

Note. EI = Early Intervention; IDEA = Individuals with Disabilities Education Act; IEP = Individualized Education Plan; IFSP = Individual and Family Service Plan; 504 = Section 504 of the Rehabilitation Act of 1973.

Table 7b
Legislative Materials: Parents

Q10: Which of the following laws were you provided information on by the audiologist?	Parents Comments • We live abroad • None (4 responses)

Parent Perspective

When asked if they felt their audiologist provided them with unbiased, extensive information on intervention for their child's hearing loss, 47.56% of parents said *Yes*, 43.90% said *No*, and 8.54% said *Other*. See Table 8 for

parent comments. Parents were then asked to rate the information (using *Excellent, Good, Fair, or Poor*) they received about each of the communication options when their child was diagnosed. See Table 9 for results. Finally, they were given a list of questions and asked how much they agree or disagree with each of the statements (i.e., *Strongly Agree, Agree, Somewhat Agree, Neither Agree or Disagree, Somewhat Disagree, Disagree, or Strongly Disagree*). The majority of parents agreed with the following statements:

- "It was easy for me to get information regarding early intervention options for my child."
- "It was easy for me to obtain information regarding different modes of communication for my child."

- "I feel that my audiologist's goal was to provide me with unbiased options in order for me to make the best decisions for my child."
- "I understand and utilize the public laws that seek to help the Deaf population."

On the other hand, most parents surveyed disagreed with the following statements:

- "I was provided with extensive information regarding emotional support after my child's diagnosis."
- "I felt pressured by my audiologist to choose one communication option over others."
- "I feel as though I do not understand my child's hearing loss and its effects."
- "I felt as though my audiologist did not have enough time to explain important concepts thoroughly."

Nearly equal numbers of parents agreed and disagreed with the statement, "I often felt frustrated and confused regarding what decisions to make." See Table 10 for detailed results.

Discussion

The findings of the survey demonstrated that the information that audiologists reported providing was consistent with what parents reported receiving. Although the intended goal of the study was to compare audiologist and parent perspectives on the information and support provided when a child is deaf or hard of hearing, the study also analyzed the parent's opinion of their overall experience in working with an audiologist and the quality of the information they received about their child's hearing loss. When comparing what audiologists reported providing to what parents reported receiving, survey data revealed much consensus concerning types of information provided. This includes types of materials provided, emotional support, state resources, and related legislation. The responses indicated that audiologists are providing this information to parents; however, there is questionable value and ability to apply the information for parents, as seen in the parent perspective rating questions.

Table 8

Comments on Quality of Information about Hearing Loss Intervention

Q11: Do you feel that your audiologist provided you with unbiased, extensive information regarding intervention for your child's hearing loss?	Parents Comments • Unbiased, but not extensive • Somewhat • "I'm not sure if it was unbiased but we planned to choose spoken language for our child and the audiologist may have perceived that" • "She was definitely biased towards implants and one size fits all for individuals with hearing loss"

Table 9

Communication Option Information Intervention

	Excellent	Good	Fair	Poor
Auditory Verbal Therapy	26.76%	14.08%	16.90%	42.25%
Cued Speech	2.84%	8.82%	17.65%	70.59%
American Sign Language	9.09%	12.12%	19.70%	59.09%
Signed Exact English	0%	6.15%	15.38%	78.46%
Total Communication	9.09%	13.64%	18.18%	59.09%
Bilingual Approach	1.54%	7.69%	10.77%	80%

For the rating scale questions, a discrepancy was found between reported provision of Deaf culture and cochlear implant information. This could be attributed to the chance that the child did not meet candidacy requirements, and the audiologist did not present this information because they knew the child was not a candidate. Therefore, the discrepancy does not mean that audiologists are not providing information on both because of their personal biases, but possibly that the choices are not applicable for the child's specific loss. However, one parent participant left a comment that their audiologist told them, "Cochlear implants were the only thing that could help my son learn. They completely ignored that there should always be a trial of hearing aids first, and they were incredibly rude about Deaf culture." One audiologist participant commented, "I usually ask about whether the parents' goal is for their child to use hearing/speech to communicate and only provide info on Deaf culture if parents ask about other options." When parents were asked if they felt that their audiologist provided them with unbiased, extensive information regarding intervention for their child's hearing loss, nearly half of respondents reported that they did not believe that they received unbiased, extensive information. Participants could elaborate further on this question in the comment section. One participant stated, "The information was unbiased, but not extensive." Another participant stated, "She was definitely biased towards implants and a one size fits all for individuals with hearing loss." Based on evidence from the survey data obtained, there is an indication that biased information could be being presented to parents. According to American Speech-Language-Hearing Association (ASHA), audiologists help facilitate decision making with families regarding their child's hearing loss by the information they provide at the time of diagnosis. When counseling families, it is the clinician's responsibility to remove their own biased opinions from their professional delivery of up-to-date, relevant information. When the family is ready to make an

Table 10

Statements Regarding Information Given to Parents

	Strongly Agree	Agree	Somewhat Agree	Neither Agree nor Disagree	Somewhat Disagree	Disagree	Strongly Disagree
It was easy for me to get information regarding early intervention options for my child.	19.75%	19.75%	18.52%	9.88%	12.35%	9.88%	9.88%
It was easy for me to obtain information regarding different modes of communication for my child.	7.50%	23.75%	21.25%	12.50%	12.50%	12.50%	10.00%
I was provided with extensive information regarding emotional support after my child's diagnosis.	4.88%	12.20%	10.98%	4.88%	13.41%	28.05%	25.61%
I feel that my audiologist's goal was to provide me with unbiased options in order for me to make the best decisions for my child.	14.63%	19.51%	15.85%	23.17%	15.85%	4.88%	6.10%
I felt pressured by my audiologist to choose one communication option over others.	9.88%	9.88%	7.41%	11.11%	8.64%	30.86%	22.22%
I understand and utilize the public laws that seek to help the Deaf population.	8.64%	18.52%	18.52%	18.52%	7.41%	14.81%	13.58%
I feel as though I do not understand my child's hearing loss and its effects.	2.47%	9.88%	9.88%	9.88%	13.58%	24.69%	29.63%
I often felt frustrated and confused regarding what decisions to make.	13.58%	17.28%	14.81%	8.64%	12.35%	22.22%	11.11%
I felt as though my audiologist did not have enough time to explain important concepts thoroughly.	10%	8.85%	16.25%	12.5%	6.25%	22.5%	23.75%

informed decision about the desired outcomes for their child, audiologists are then obligated to advise families on how best to achieve those outcomes (ASHA, 2008).

Many parents reported confidence in the ability to obtain information on early intervention services for their child and information on modes of communication. In addition, parents felt they were not pressured by their audiologist to choose a certain mode of communication and that their audiologist's goal was to provide them with unbiased options for their child. Approximately one half of parent respondents reported feeling frustrated and confused on what decision to make about their child's hearing loss. Furthermore, ASHA reports that it is the audiologist's responsibility to provide sufficient and concise information to assist families in their decision making. Audiologists should refrain from using terminology that confuses parents and recognize that every family does not process or accept new information in the same manner and pace (ASHA, 2008).

Parent perspective survey questions yielded evidence that many parents felt they were not provided with extensive information regarding emotional support after their child's diagnosis. Stress and grief may occur in parents due to the diagnosis of their child's hearing loss, which can slow down the intervention process. Once the parent's emotional needs are addressed, they become more receptive to new information for making informed decisions concerning their child. It is critical that audiologists are making the appropriate recommendations for emotional support including family counseling, support groups, and connecting with other families with similar experiences (ASHA, 2008).

Data analysis also found most parents felt that they did not fully understand their child's hearing loss and its effects. Only a few audiologists and parents reported using and/or observing hearing loss simulation to further understand hearing loss and its ramifications to the understanding of speech. Research shows that hearing loss simulation is an excellent tool to provide parents with a realistic demonstration of the communicative and psychosocial effects of their child's hearing loss. Through hearing loss simulation, family members can recognize the importance of effective communication strategies, such as lipreading and speaking clearly. Moreover, hearing loss simulation gives parents realistic expectations concerning amplification (Zurek & Desloge, 2007).

Workplace variations can place certain limitations on the audiologist's ability to counsel parents effectively. These limitations may include, but are not limited to, the amount of time the audiologist has available to spend with patients,

how often they see the pediatric population, if their setting provides various amplification options, and limitations to accessing necessary supplies for parent education. More than one-third of audiologists that participated in the survey reported working in a hospital. According to Severn and colleagues (2012), audiologists that reported the highest stress levels were working in public hospitals. This is a notably difficult work environment due to busy caseloads that may limit quality interaction with patients and dealing with grief reactions of patients, parents, or family members (Severn et al., 2012). Although this theory cannot be proven by the current study, audiologists may not have been able to provide as extensive information to parents as they would have liked due to workplace limitations. This possibility could influence the results obtained on audiologist provision of information. Another possible factor that could have influenced results was the number of years the audiologist had practiced. Nearly one-third of audiologist respondents reported only working clinically for less than 5 years. Work experience, self-confidence, and clinical maturity can play a large role in knowledge and the ability to effectively counsel and provide appropriate and extensive recommendations.

Nearly half of the parent respondents reported their child was diagnosed anywhere from birth to 3 months of age. As previous research shows, children that are fit with amplification earlier are more likely to have better language and learning outcomes than children who are fit later in life (Moeller & Tomblin, 2015). Since nearly half of the respondents reported early diagnosis, this could have yielded a possible positive effect on the survey data. An early diagnosis could have led to better parent perspectives and overall ratings on the information they received. Those with early diagnoses may have had more time to make decisions and experienced less stress when it came to the timeline of their child's acquisition of language. Nearly one-third of parents reported their child was diagnosed over 3 years of age, which may have yielded a more negative experience with their audiologist; however, there is no evidence as to how older ages of diagnoses affected the survey data obtained.

Population Considerations

Although most parents reported their child was fit with hearing aids, several parents reported their child was fit with a cochlear implant. Additionally, there was a reasonable number of parents whose child was a part of Deaf Culture. The survey obtained information from a diverse population, with participants in both hearing and Deaf Culture along with considerable variations in amplification use. This provided a wide range of responses for data analysis. Overall, there were 91 audiologists and 111 parents who participated in the survey. The large and comparable audiologist and parent sample sizes allowed the authors to analyze population data and receive a wide array of participant comments. Of the 91 audiologists and 111 parents who participated in this study, there were surveys from both parties that were not fully completed. Therefore, these surveys

were only considered in part of the data analysis, which affected the sample size. As the dissemination approach allowed participants to self-select, the authors suspect that rather than the length of the survey, possible reasons for incompletes may have included loss of cell phone power or service, outside distractions (e.g., work duties, obligations in the home environment), accidental closing of the browser, and/or compatibility across mobile devices for the charts.

Limitations

There are a few factors that limit the application of results from this study. First, it must be considered that audiologists who participated in this survey did not work directly with the parent participants within the survey. Therefore, this survey data does not reflect direct clinician to patient comparisons on counseling, services, and quality of information and recommendations provided. Additional limitations include the possibility that the audiologists surveyed may not have a large pediatric caseload at their workplace and may have depended more on previous experience and education rather than on current patient encounters to answer survey questions. In addition, an audiologist participant commented, "We rarely see children, and if we do, we typically get mild to moderate hearing losses." The intended target population may not have been fully obtained due to limited author control over specific pediatric clinical experience of the audiologists that participated in the survey. Extensive clinical experience with the pediatric population can affect the audiologists' confidence level and ability to effectively counsel parents and provide appropriate and extensive recommendations. Additionally, it is possible that parents who were surveyed may not have been counseled or given information on "cochlear implants and Deaf culture" (Parent Survey-Question 6) due to the degree of their child's hearing loss obviating the need for those conversations with their audiologist. Therefore, this may have directly affected the survey responses obtained. More specific instructions could have been given to the parents to assist them in thinking about their personal experiences and how that could potentially influence their answers.

Lastly, the dissemination approach of using social media platforms and the lack of ability to adequately quantify the response rates from individuals in the survey was problematic. Despite this, the results revealed clear patterns related to the information provided to parents from audiologists in a variety of work settings. Since the survey was exclusively distributed through social media platforms, the survey was not able to accommodate participants from populations who are not on social media and/or do not have access to computers, smartphones, internet access, et cetera. The data showed that nearly one third of the audiologists surveyed had practiced for less than 5 years. This population trend of younger and less experienced audiologists may be because younger generation audiologists may have more access and comfort with the use of social media platforms.

Conclusion

The data showed that in many aspects, adequate information is being provided to parents when their child is diagnosed with hearing loss; however, it also showed that information in many areas was insufficient, and parents did not feel confident in their ability to make decisions for their child. Within the parent survey, 93.75% of parent respondents reported that they were hearing individuals with a child with hearing loss. Research shows this is a common occurrence, and parents who have no previous experience in this realm will need more guidance and information about making decisions for their child. Due to the short time window for intervention, the decisions made at this time are of high priority. It is critical to provide parents with reliable guidance and support during this time to make properly informed decisions for their child (Kushalnagar et al., 2010).

This makes the decision-making process considerably more difficult, as they are navigating unknown waters. Audiologists have the responsibility to provide parents with unbiased, extensive information for parents to successfully make informed decisions for their child. Provision of practical, comprehensive information and recommendations may lead to improved parent knowledge and confidence. Ample time should be allotted by audiologists when scheduling appointments with parents of children who are deaf or hard of hearing; this will allow time for the audiologist to fully educate, counsel, and support the parents who are also processing their emotions. Audiologists also should be vigilant in providing evidence-based practice and in maintaining education of current state and local resources, as well as emotional support available to help families after a diagnosis of hearing loss. To expand on this research, a study could examine parent perspectives on what information and audiological services would have been or were most beneficial while making decisions for their child. This may, in turn, provide clearer information on what audiologists can do to further assist parents during a demanding, yet rewarding time.

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Appendix A
Audiologist Survey Questions

1. What best describes your work setting?
    a.   Hospital
    b.   University
    c.   Private Practice
    d.   ENT
    e.   Other—comments were accepted

2. How many years have you been practicing?
    a.   Less than 5 years
    b.   5–10 years
    c.   10–15 years
    d.   15–20 years
    e.   Greater than 20 years

3. What materials did you use in order to explain the child's hearing loss to the parents? Check all that apply
    a.   Pamphlets
    b.   Verbal Explanation
    c.   Visual Aids
    d.   Hearing Loss Simulation
    e.   Outside Reading Materials (i.e., online articles, books, etc.)
    f.   Other—comments were accepted

4. Did you provide parents with information on both cochlear implants AND Deaf culture?
    a.   Yes
    b.   No (if no, why?)- comments were accepted

5. Which of the following did you provide information on regarding modes of communication? Please select all that apply.
    a.   Auditory Verbal
    b.   Cued Speech
    c.   American Sign Language
    d.   Signed Exact English
    e.   Total Communication
    f.   Bilingual Approach
    g.   None of the above
    h.   Other—comments were accepted

6. Which of the following did you provide information regarding emotional support? Please select all that apply.
    a.   Deaf Mentors
    b.   Family Support Groups
    c.   Family Counseling
    d.   Blogs/Social Media Groups
    e.   None of the above
    f.   Other—comments were accepted

7. Which of the following state resources did you recommend to the parents? Please select all that apply.
    a.   Early Intervention
    b.   Hands and Voices Chapter
    c.   Other—comments were accepted

8. Which of the following laws did you provide information on to the parents? Please select all that apply.
    a.   Individuals with Disabilities Education Act
    b.   Individualized Family Service Plan/Individualized Education Plan
    c.   Section 504 of the Rehabilitation Act of 1974
    d.   American Disabilities Act of 1990
    e.   Other—comments were accepted

Appendix B
Parent Survey Questions

1. Are you
   a.   Deaf
   b.   Hard of Hearing
   c.   Hearing

2. How old was your child when they were diagnosed as Deaf/Hard of Hearing?
   a.   Birth–3 months
   b.   4–6 months
   c.   7–12 months
   d.   1–2 years
   e.   2–3 years
   f.   Over 3 years

3. Is your child a part of Deaf Culture?
   a.   Yes
   b.   No

4. Does your child wear
   a.   Hearing Aids
   b.   Cochlear Implants
   c.   None of the above

5. What materials were you provided by an audiologist in order to explain your child's hearing loss? Please select all that apply.
   a.   Pamphlets
   b.   Verbal Explanation
   c.   Visual Aids
   d.   Hearing Loss Simulation
   e.   Outside Reading Materials (i.e. online articles, books, etc.)
   f.   Other—comments accepted

6. Were you provided with information on both cochlear implants AND Deaf culture by an Audiologist?
   a.   Yes
   b.   No
   c.   Other—comments accepted

7. Did an audiologist provide you with any of the following information regarding modes of communication? Please select all that apply.
   a.   Auditory Verbal Therapy
   b.   Cued Speech
   c.   American Sign Language
   d.   Signed Exact English
   e.   Total Communication
   f.   Bilingual Approach
   g.   None of the above
   h.   Other—comments accepted

8. Did an audiologist provide you with any of the following information concerning emotional support? Please select all that apply.
   a.   Deaf Mentors
   b.   Family Support Groups
   c.   Family Counseling
   d.   Blogs/Social Media Support Groups
   e.   None of the above
   f.   Other—comments accepted

9. Which of the following state resources were recommended by your audiologist? Please select all that apply.
   a.   Early Intervention
   b.   Hands and Voices Chapter
   c.   Other—comments accepted

10. Which of the following laws were you provided information on by the audiologist? Please select all that apply.
   a.   Individuals with Disabilities Education Act
   b.   Individualized Family Service Plan/Individualized Education Plan
   c.   Section 504 of the Rehabilitation Act of 1974
   d.   American Disabilities Act of 1990
   e.   Other—comments accepted

11. Do you feel that your audiologist provided you with unbiased, extensive information regarding intervention for your child's hearing loss?
   a.   Yes
   b.   No
   c.   Other—comments accepted

12. Rate the information you received about each of the following communication options when your child was first diagnosed. Parents were asked to choose one of the following for each communication option listed below: *Excellent, Good, Fair*, and *Poor*.
   ● Auditory Verbal
   ● Cued Speech
   ● American Sign Language
   ● Signed Exact English
   ● Total Communication
   ● Bilingual Approach

13. How much do you agree with the following statements? Parents were asked to rate each statement below as one of the following: *Strongly Agree, Agree, Somewhat Agree, Neither Agree or Disagree, Somewhat Disagree, Disagree*, and *Strongly Disagree*.
   ● It was easy for me to get information regarding early intervention options for my child.
   ● It was easy for me to obtain information regarding different modes of communication for my child.
   ● I was provided with extensive information regarding emotional support after my child's diagnosis.
   ● I feel that my audiologist's goal was to provide me with unbiased options in order for me to make the best decisions for my child.
   ● I felt pressured by my audiologist to choose one communication option over others.
   ● I feel as though I do not understand my child's hearing loss and its effects.
   ● I often felt frustrated and confused regarding what decisions to make.
   ● I felt as though my audiologist did not have enough time to explain important concepts thoroughly.

The Impact of the COVID-19 Pandemic on Newborn Hearing Screening Programs in Western States

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Abstract

The primary objective of this study was to determine the impact of the COVID-19 pandemic on newborn hearing screening guideline adherence and the respective rates of screening, diagnosis, and intervention. This was a review of newborn hearing screening data compiled from the Departments of Health in six states for the time periods of March 2019–September 2019 and March 2020–September 2020. Endpoints included the numbers of live births as well as the numbers and timeframes of screening, diagnostic, and intervention events. Two-tailed paired t-tests were performed to determine statistical significance. Data included assessment of 181,662 births in six states. Compared to March 2019–September 2019, March 2020–September 2020 had a significantly lower mean rate of screening before 1 month of age (97.3% vs. 96.2%, $p < 0.001$) and mean screen rate overall (98.9% vs. 98.0%, $p < 0.001$). Additionally, the 2020 time period had a significantly higher mean rate of patients lost to follow up for referral to early intervention (14.7% vs. 28.9%, $p = 0.005$). The COVID-19 pandemic has had a significant impact on the newborn hearing screening programs of several states in the Western United States. This information holds significant implications for the current evaluation of these newborn hearing screening programs.

Keywords: EHDI, COVID-19 pandemic, newborn hearing screening, early intervention, guideline adherence

Acronyms: CDC = Centers for Disease Control and Prevention; CI = Confidence Interval; EHDI = Early Hearing Detection and Intervention; EI = Early Intervention; JCIH = Joint Committee on Infant Hearing

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The detriments of permanent childhood hearing impairment are well known and include negative impacts on language, speech, and behavioral development, all of which may influence a child's social and academic outcomes (Pimperton & Kennedy, 2012; The Joint Committee on Infant Hearing, 2019). To combat this, all states, territories, and Washington D.C. have developed early hearing detection and intervention (EHDI) programs, which use audiological and medical practitioners to screen infants for hearing loss and provide patients and families with the appropriate treatments and guidance to minimize developmental impact. Following guidance from the Joint Committee on Infant Hearing (JCIH), these programs aim to provide hearing screening no later than one month

of age, comprehensive audiological evaluation no later than three months of age for those that do not pass their hearing screening, and appropriate intervention initiated no later than six months of age for infants with confirmed hearing loss (JCIH, 2019). Due to circumstances surrounding the global COVID-19 pandemic, these recommended timelines may not be achieved, and future patient outcomes could be affected.

Although the importance of EHDI guidelines is well-established and the impact of the COVID-19 pandemic has been widespread, there has been no previous study investigating the role of the current pandemic in delaying hearing screenings and interventions in infants. This study

seeks to determine the effect of the COVID-19 pandemic on EHDI program guideline adherence and screening rates in several rural Western states using a retrospective review of data from their respective State Departments of Health. This information holds significant implications for the current evaluation of the EHDI program in these states and provides insight that could be used to prepare for future, major disruptive events.

Method

The Institutional Review Board at the University of South Dakota granted exemption to this project for purposes of program evaluation and improvement.

Program Structure

EHDI programs consist of many essential team members, including the birth hospitals, primary health care providers, otolaryngologists, audiologists, and speech-language pathologists, among others. The birth hospital is essential for providing initial newborn hearing screening and ensuring that parents and other healthcare providers receive and understand the hearing screening results as well as follow-up instructions. Audiologists play a large role, contributing to the development, management, and coordination of hearing screening programs. Additionally, audiologists conduct the comprehensive diagnostic assessment that determines the presence of hearing loss or normal hearing. Specifically, pediatric audiologists are uniquely skilled to work with infants, children, and their families. The audiologist also refers the family to other services, including early intervention programs to support the infant and family through early childhood development or medical evaluation of the hearing loss to assist in determination of etiology of loss, receipt of medical clearance for amplification (if the family chooses to pursue that option), and building the support team that is necessary for the family.

The EHDI programs in the states included in this study exist within their respective State Departments of Health (Nebraska DHHS, n.d.; North Dakota Center for Persons with Disabilities, n.d.; Idaho Department of Health and Welfare, n.d.; South Dakota Department of Health, n.d.; Kansas Department of Health and Environment, n.d.; Utah Department of Health, n.d.). In North Dakota and South Dakota, these programs collaborate with Minot State University and the University of South Dakota, respectively, for purposes of program assessment and improvement.

EHDI programs throughout the nation report data to the Centers for Disease Control and Prevention (CDC) on a yearly basis via the Hearing Screening and Follow-up Survey (CDC, 2017). This reporting, although voluntary, is usually completed by nearly all EHDI programs and allows for CDC collaboration and assistance with program improvement (Alam et al., 2016).

Study Population and Outcome Variables

This study evaluated EHDI program data acquired from the Departments of Health of six Western states: South Dakota, North Dakota, Utah, Kansas, Nebraska, and Idaho. These states were chosen due to their unique rural setting, their varying levels of pandemic-related restrictions and mandates, and their readily available 2020 EHDI data due mostly to their smaller populations.

All the residents of these states born between March 1 and September 30 of 2019 and 2020 were included in the study population (Figure 1). The data collected included the number of births, infant deaths, and parental refusals of screening services. The number and timing of screening, diagnostic, and early intervention (EI) events and referrals were also obtained. From these measures, several outcome variables were calculated (Table 1). These outcome variables included screen rate by one month, screen rate overall, diagnosis rate by three months, lost to follow up rate for diagnostic evaluation, and lost to follow up rate for referral to EI services. Children were considered lost to follow up for diagnostic evaluation if they did not pass the initial hearing screening and subsequent attempts to contact their parents to schedule a diagnostic evaluation resulted in failure to make contact or lack of response from the parents; this category also included children who were lost to follow up for unknown reasons. Children were considered lost to follow up for referral to EI services if they were determined to be deaf or hard of hearing upon diagnostic evaluation and were not subsequently referred to EI services.

Statistical Analysis

Comparisons of 2019 and 2020 outcome variables were statistically analyzed using two-tailed paired *t*-tests, and 95% confidence intervals were calculated. State results were analyzed in a blinded fashion and will be presented as such.

Results

During the studied time periods, there were a total of 181,662 births across the six states included in this study.

Outcome Variables

Screen Rate Overall

Overall screen rates were near 100% in most of the studied states during the designated time period in 2019, with an overall mean of 98.5% (Figure 2A; Table 2). In the 2020 time period, three states recorded significantly decreased overall screen rates of 93.3% ($p = 0.001$), 99.8% ($p = 0.04$), and 99.1% ($p = 0.03$), respectively. Overall, the 2020 mean screen rate was 98.0%, demonstrating a significant decrease compared to the same time period in 2019 ($p < 0.001$).

Screen Rate by One Month

The rate of screening by one month of age averaged 97.3% across all the studied states in the 2019 period (Figure 2B; Table 2). During March 2019–September 2020, all the studied states recorded decreased rates of screening by one month of age, with three states demonstrating a significant decrease ($p = 0.004$, $p = 0.005$, $p = 0.01$, respectively). Altogether, the average rate of screening by one month of age decreased during the 2020 time period to an average of 96.2% ($p < 0.001$).

Figure 1

Study Population and Program Flow


```
        State cohort resident births March-Sept. 2019 &
                            2020
                              |
                              |------------------------------>  Infant Deaths
                              |                                 Family Declined Services
                              v
                 Infants Eligible for Screening
                              |
          --------------------|--------------------
          |                   |                    |
     Missed            Did Not Pass           Passed
     Screening           Screening           Screening
                            |
                            |--------------------------------> Family Contacted But Unresponsive
                            |                                   Unable to Contact
                            |                                   Unknown Lost to Follow Up
               -------------|-------------
               |                         |
       Hearing Loss on            No Hearing Loss
         Diagnostic               on Diagnostic
         Evaluation                 Evaluation
               |
       --------|--------
       |               |
   Referred to EI   Not Referred to
     Services         EI Services
```

Table 1

Outcome Variables Definitions

Outcome Variable	Description	Operational Definition
Screen Rate Overall	Percentage of infants screened at any age	$\dfrac{\text{Total screened }(n)}{\text{Eligible for screening }(n)}$
Screen Rate By 1 Month of Age	Percentage of infants screened before 1 month of age	$\dfrac{\text{Screened before 1 month of age }(n)}{\text{Eligible for screening }(n)}$
Diagnosis Rate By 3 Months of Age	Percentage of infants completing diagnostic testing by 3 months of age	$\dfrac{\text{Diagnosed by 3 months of age }(n)}{\text{Failed screening }(n)}$
Lost to Follow Up for Diagnosis	Percentage of infants who referred on the initial hearing screening and did not receive diagnostic evaluation	$\dfrac{\text{Family contacted but unresponsive }(n) + \text{Unable to contact }(n) + \text{Unknown lost to follow up }(n)}{\text{Failed screening }(n)}$
Lost to Follow Up for Referral for EI	Percentage of infants who were diagnosed with hearing loss but did not receive referrals to EI	$\dfrac{\text{Not referred to EI }(n)}{\text{Diagnosed with hearing loss }(n)}$

Note. EI = Early Intervention.

Figure 2

Outcome Variables Compared Between States And Years
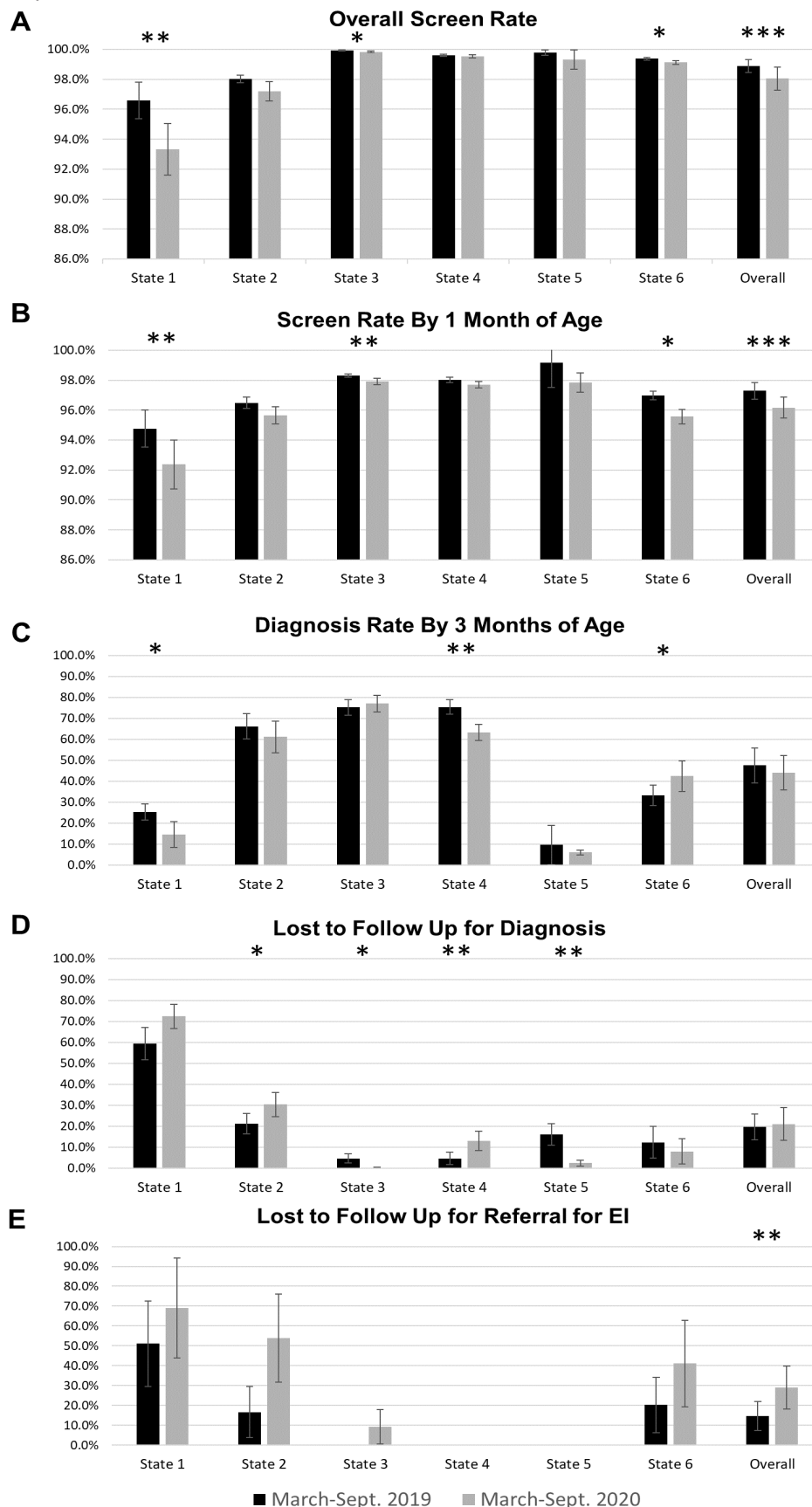
Note. A comparison of (A) overall screening rate, (B) screening rate by 1 month of age, (C) diagnosis rate by 3 months of age, (D) the proportion of children lost to follow up for diagnosis, and (E) the proportion of children lost to follow up for referral to early identification (EI). All data displayed as means +/- 95% CI. Statistical significance determined via two-tailed paired *t*-test. *$p < 0.05$, **$p < 0.01$, ***$p < 0.001$.

Table 2

Outcome Variables Differences Between Years

Measure	March 2019–Sept. 2019	95% CI	March 2020–Sept. 2020	95% CI	p value
Total Births (n)	94,039	-	91,555	-	-
Screen Rate (%)	98.9	98.5–99.3	98.0	97.2–98.8	< 0.001
Screen Rate Before 1 Month of Age (%)	97.3	96.7–97.9	96.2	95.5–96.9	< 0.001
Diagnosis Rate by 3 Months of Age (%)	47.5	39.2–55.8	44.1	35.8–52.4	0.06
Lost to Follow Up Rate for Diagnosis (%)	19.8	13.6–26.0	21.1	13.2–29.0	0.54
Lost to Follow Up Rate for Referral to EI (%)	14.7	7.3–22.1	28.9	18.1–39.7	0.005

Note. EI = Early Intervention; CI = Confidence Interval.

Diagnosis Rate by Three Months

The rate of diagnosis by three months of age varied from 25.4% to 74.4% during the 2019 period, with an overall mean of 60.6% (Figure 2C, Table 2). During the 2020 period, four of the six studied states reported decreases in their rate of diagnosis by three months of age, two of which were statistically significant (p = 0.04, p = 0.006, respectively). Interestingly, two of the six states reported increases in their mean rate of diagnosis by three months of age, one significantly so (p = 0.03). Overall, the mean rate of diagnosis by three months of age decreased in the 2020 period to 54.0%, although this difference was not statistically significant (p = 0.07).

Lost to Follow Up for Diagnosis

The proportion of infants who were lost to follow up for diagnostic evaluation varied in 2019 from 4.7% to 59.5% with an overall mean of 19.8% (Figure 2D; Table 2). In March 2020–September 2020, three of the studied states reported increases in their mean proportion of infants lost to follow up for diagnosis, with two of those states reporting significant increases (p = 0.04, p = 0.004, respectively). Conversely, three of the studied states demonstrated a decrease in the proportion of infants lost to follow up for diagnosis, with two of those states reporting significant decreases (p = 0.01, p = 0.004, respectively). As a whole, the mean percentage of infants lost to follow up for diagnosis during the 2020 period increased to 21.1% in the studied group of states, but this difference was not statistically significant (p = 0.54).

Lost to Follow Up for Referral to Early Intervention (EI)

The percentage of infants lost to follow up for referral to EI averaged 14.7% in the studied states during March 2019–September 2019 (Figure 2E; Table 2). Three of the studied states reported a rate of 0% for this outcome variable during this time period. In the 2020 time period, four of the studied states reported increases in this metric, but none were statistically significant. Two states again reported rates of 0% lost to follow up for referral to EI during the

2020 period. Overall, the mean proportion of infants lost to follow up for referral to EI averaged 28.9% in March 2020–September 2020, demonstrating a significant increase compared to the prior year (p = 0.005).

Discussion

Prior to the widespread implementation of EHDI programs, children with severe-to-profound hearing loss, on average, completed their education at age 18 with reading and language levels equivalent to that of a 10-year-old child with normal hearing (Traxler, 2000). Due to the lack of widespread screening programs, these children were typically not identified and diagnosed until two to three years of age (Hoffman & Beauchaine, 2007). Conversely, the widespread adoption of EHDI programs has resulted in the average age of confirmed hearing loss decreasing to two to three months of age (Harrison et al., 2003). Children with hearing loss who receive appropriate diagnosis and intervention within the first six months of life achieve improvements in receptive and expressive language, vocabulary development, and educational attainment (Pimperton & Kennedy, 2012; Yoshinaga-Itano et al., 2017, 2018). Additionally, some studies have shown that early intervention may enable children who are deaf or hard of hearing to achieve normal levels of language development by five years of age (Calderon et al., 1998; Kennedy et al., 2005). Due to the demonstrable benefits of EHDI programs, all 50 states and many countries around the world continually work to implement and improve their infant hearing screening programs (Grosse et al., 2018; Moodley & Storbeck, 2015; White, 2003; Wroblewska-Seniuk et al., 2017).

The impact of the COVID-19 pandemic on early childhood health screening programs has not been previously reported. However, examples of delayed childhood screening as a result of major disruptive events do exist, including the influx of Syrian refugees to European and Asian nations due to the Syrian civil war, which began in 2011. This mass movement of refugees and collapse of the Syrian healthcare system resulted in large populations

of children who did not receive timely health screenings for a variety of conditions including congenital hypothyroidism, inborn metabolic diseases, and cleft lip and palate (Boynuyogun et al., 2020; Saoud et al., 2019; Schiergens et al., 2018). These gaps in healthcare led to severe, preventable sequelae including neurological dysfunction, delayed neuropsychomotor development, growth failure, and worsened surgical outcomes (Boynuyogun et al., 2020; Saoud et al., 2019; Schiergens et al., 2018). This major event also impacted newborn hearing screening. Studies performed at sites in Turkey reported many Syrian refugee children had not previously passed through hearing screening programs, and the rates of hearing loss were significantly higher in Syrian children compared to their Turkish counterparts (Çıkrıkçı et al., 2020; Kaplama & Ak, 2020; Yücel et al., 2019). Major events may contribute to delayed childhood health screening by disrupting both the program itself and the ability of individuals to pass through the given program.

Due to business restrictions as well as many patients choosing to defer and delay non-COVID-19-related healthcare, many healthcare practices saw reduced patient loads and clinic visits, with some data reporting reductions in outpatient visits by 60% (Commonwealth Fund, n.d.). Despite these restrictions and the shifting healthcare landscape, the American Academy of Pediatrics has strongly recommended that states continue to adhere to the established 1-3-6 EHDI guidelines (American Academy of Pediatrics, n.d.).

Our study demonstrates that the COVID-19 pandemic has significantly affected several aspects of newborn hearing screening programs in South Dakota, North Dakota, Nebraska, Idaho, Kansas, and Utah. With regards to screening, these states reported lower rates of screening overall and by one month of age. Both findings may be partially explained by the changing labor and delivery unit policies during the COVID-19 pandemic, including shortened post-partum hospital stays for mothers and newborns. Some reports describe the rate of newborns who were discharged after one night in the hospital increasing by roughly 25% (Greene et al., 2020). These shortened stays provide less opportunity for initial hearing screening to take place.

Several states' data revealed an impact on the rate of diagnostic evaluation following an abnormal hearing screening. Some states reported significantly decreased rates of diagnosis by three months of age as well as significantly increased rates of children who were lost to follow up for diagnostic evaluation. These results may be due to an increased aversion for healthcare settings as a result of the COVID-19 pandemic, resulting in families choosing not to return to a pediatric audiologist for further diagnostic evaluation.

An analysis of the reported data also revealed an increase in the proportion of children who were lost to follow up for referral to EI. These were children who, upon being diagnosed with hearing loss, were not subsequently referred to EI services. Four states in the studied cohort reported increased rates of loss to follow up for referral to EI, although none of the states' differences were found to be statistically significant alone. When all the states' data is compiled and analyzed as a whole, a significant increase in loss to follow up for referral to EI is revealed. Interestingly, several states reported rates of 0% for this outcome variable for both 2019 and 2020. These findings may be due to multiple important factors. The states included in this study differ in the mandates present for their newborn hearing screening programs. In some states, such as South Dakota, North Dakota, and Idaho, newborn hearing screening is not mandated by law, creating more difficulty for the state Department of Health to collect diagnostic and EI data (National Center for Hearing Assessment and Management, n.d.). This lack of a mandate may result in less funding and fewer positions dedicated to newborn hearing screening programs. These difficulties were likely compounded during the COVID-19 pandemic.

Results of this study may be influenced by several factors unique to the geographic region under research. Rurality is one factor that might influence states' outcome variables. The six states being studied have an average population density ranked lower than 80% of all states' population densities (USA.com, n.d.). This rurality, combined with long driving distances and detrimental weather conditions, creates physical barriers between patients and healthcare providers, including pediatric audiologists (Krumm et al., 2018). In addition to population density, poverty levels of each state were compared to the national average. According to the most recent data reported by the United States Census Bureau, all six states' poverty levels are below the national average (United States Census Bureau, n.d.). Finally, each states' COVID-19 data was analyzed. As of February 2, 2021, the total COVID-19 cases per 100,000 individuals in each state was higher than the national average (CDC, 2020). These factors may have had an impact on the states' newborn hearing screening programs both before and during the COVID-19 pandemic. Other factors, such as states' lockdown measures during the pandemic, might have also impacted outcome variables.

Although the COVID-19 pandemic continues to present new challenges, important lessons have been learned over the past year. One such lesson is the importance of remaining vigilant and taking a proactive stance during an international crisis. Though it is likely that certain healthcare protocols and procedures take less precedence, lack of adherence to these protocols may create unintended ramifications when the crisis subsides. More specifically, lack of adherence to the EHDI 1-3-6 guidelines has affected several states' screening, diagnostic, and EI enrollment rates.

The pandemic has also highlighted the importance of telehealth. Even before the pandemic, several of the states under study had barriers that separated patients from healthcare providers, possibly due to the states' rurality. Telehealth allows patients to circumvent barriers created by both pre-existing factors and the COVID-19 pandemic. The feasibility of using remote control options to connect

patients and providers has changed the healthcare landscape, and it has been advantageous to several healthcare fields during the pandemic.

Some limitations should be considered when reviewing the results of this study. First, the geographic region under research may prevent generalization of data to other states and/or countries beyond the United States. Factors unique to these six Western American states could have impacted outcome variables, and further research must be done to confirm or refute these trends in other geographic regions. In addition, important demographic and socioeconomic factors were not considered when comparing outcome variables across the six states. For example, it is possible that the impact of COVID-19 on the EHDI 1-3-6 benchmarks could have differed among minority communities in each state.

Conclusion

The COVID-19 pandemic has had a significant impact on the newborn hearing screening programs of several Western states. Most notably, these states reported significantly decreased rates of screening by one month of age, screening overall, and referral to early intervention services. This data provides valuable information for the evaluation of these programs as well as insight for future major disruptive events. This disruption in early childhood hearing screening may have far-reaching consequences for future health outcomes, and further research will be needed to fully assess the scope and magnitude of these potential detriments.

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Impact of Face Masks on Audiovisual Word Recognition in Young Children with Hearing Loss During the Covid-19 Pandemic

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Abstract

Objective: To investigate effects of surgical and transparent face masks on audiovisual speech recognition of words for deaf and hard of hearing children.

Design: Recorded Word Intelligibility by Picture Identification test (WIPI) was presented via a computer monitor to children in a quiet test room. The acoustic power spectra of each mask type was compared to the baseline no mask condition. Percent correct word recognition was recorded for four mask conditions (no mask, surgical mask, transparent apron mask, and ClearMask) in counterbalanced order. Repeated measures ANOVA was used to test for significant differences in word recognition scores across mask types.

Study Sample: Thirteen children (3 to 7 years) in a private auditory oral school wearing hearing aids, bone-anchored hearing aids, or cochlear implants. Children were excluded if English was not their primary language or if they had a severe speech-language delay, uncorrected vision loss, or developmental disorder that would affect the results. No children had been exposed to or had contracted the Covid-19 virus.

Results: Acoustic spectra showed a decrease in the 2000–8000 Hz region for the transparent apron mask. The surgical mask and ClearMask showed fewer acoustic effects. Children with hearing aids performed similarly to children with cochlear implants. Word recognition was significantly poorer for surgical masks and transparent apron masks. The ClearMask condition was not significantly worse than the no mask condition for words in quiet.

Conclusions: Standard surgical and custom apron shield masks significantly hampered word recognition, even in quiet conditions. The commercially available ClearMask did not significantly affect scores in quiet for young deaf and hard of hearing children, but scores were highly variable.

Keywords: Covid-19, speech perception, hearing loss, deafness, face mask

Acronyms: BAHA = bone anchored hearing aids; BKB-SiN = Bamford-Kowal-Bench Speech-in-Noise Test; CI = cochlear implants; DHH = deaf or hard of hearing; HA = hearing aids; WIPI = Word Intelligibility by Picture Identification test

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The Covid-19 pandemic has unleashed a plethora of new and difficult situations to manage; among these are the communication difficulties imposed by mask wearing. For infants and young children who are learning communication skills, mask wearing by their parents, teachers, and peers presents both a visual and an auditory barrier to spoken communication and emotional cues. Children who are deaf or hard of hearing (DHH) are especially vulnerable, as they have developing auditory and language skills, and are more reliant upon visual information. Speech perception is inherently a multimodal task that integrates visual and auditory information to aid understanding, especially in noisy environments, where visual cues become more important as the signal-to-noise ratio decreases (von Kriegstein, 2012). Adults use visual timing cues to process and recall speech in noisy

environments with greater accuracy than in auditory-only conditions (Lalonde & Holt, 2016). Normal hearing adults process lip movements by first modulating neuronal activity in the visual cortices at frequencies that match articulatory lip movements. Slower features of lip movements are then mapped onto the corresponding speech sound features and delivered to auditory areas, facilitating speech sound mapping. Visual timing thus facilitates auditory comprehension with cues that are specific to speech sounds (Bourguignon et al., 2020).

Noise is well recognized as a barrier to communication for children learning in classrooms and other acoustically challenging environments, but many other factors are important, including development, language proficiency, hearing status, and auditory experience (Leibold, 2017). As a result, children require a better signal-to-noise ratio to understand speech as well as adults do. When processing speech in low signal-to-noise environments, infants benefit from visual cues timed to the onset and offset of auditory speech, but they are not mature in their use of full visual speech cues, compared to adults (Lalonde & Werner, 2019). Preschool children increase their use of visual cues to support speech perception between 3 and 4 years of age, an important developmental shift (Lalonde and Holt, 2015). As young as 4 years of age, children with typical hearing are able to use knowledge of phonetic cues to aid speech perception in noise (Lalonde & Holt, 2015). Older children (6–8 yrs.) and adults demonstrate advantages in auditory speech detection, discrimination, and recognition when visual speech is available, although adults show more benefit for speech recognition, compared to simpler detection and discrimination tasks (Lalonde & Holt, 2016). Children who are DHH also benefit from audiovisual cues. Interestingly, children who are DHH are better than children with normal hearing at extracting phonetic information from audiovisual signals (Lalonde & McCreery, 2020).

Children who are DHH may be more impacted by the loss of visual cues due to the introduction of personal protective equipment such as masks and shields in the school setting. Solid facial coverings, such as cloth and surgical masks that cover the lips and lower part of the face, inhibit listeners from using the visual cues that facilitate greater accuracy in speech recognition, and masks also decrease auditory cues (Atcherson et al., 2017). In quiet, surgical masks do not appear to negatively impact speech understanding for adults with normal hearing or hearing loss, but in noise, there is a deleterious effect (Mendel et al., 2008). Significant negative impacts on speech perception in noise have been demonstrated with speakers wearing surgical masks (Atcherson et al., 2017; Hampton et al., 2020; Thibodeau et al., 2021). The study by Atcherson et al. (2017) included 30 adults, with 10 in each of three groups (normal hearing, moderate hearing loss, and severe-profound hearing loss) and three mask conditions (no mask, standard paper surgical mask, and transparent surgical mask). A connected speech test, the Bamford-Kowal-Bench Speech-in-Noise Test (BKB-SiN) with background speech babble showed that both groups of DHH adults had better scores in the transparent surgical

mask condition, with the greatest improvement among the profound hearing loss group. The study by Thibodeau et al. (2021) evaluated audiovisual recognition of sentences recorded in background noise with custom made 2-layer cloth masks, with a transparent window that was covered to create an opaque condition. Their study showed that performance was higher for the transparent masks, with subjective ratings of confidence and concentration also better for transparent masks. Acoustic recordings of auditory-only presentation suggested that the benefits were not attributable to an acoustic advantage, but rather to the addition of visual cues. In fact, performance in the auditory-only mode was lower with the transparent mask than with an opaque mask, likely due to decreased sound transmission with the plastic window. Bottalico et al. (2020) studied the effects of wearing face masks on classroom communication in college students and found that fabric masks yielded a significantly greater reduction in speech intelligibility in noise compared to surgical or N95 masks, likely due to greater loss of acoustic cues. Therefore, they recommended the use of medical grade masks in teaching environments. Transparent masks were not examined in that study. Other recent studies found that all masks attenuate frequencies above 1000 Hz to 3000 Hz (Corey et al., 2020; Magee et al., 2020) with higher levels of attenuation observed for masks with plastic barriers (Vos et al., 2021). Acoustic attenuation caused by reflection from hard barriers, such as transparent masks, reduces low frequency transmission less than high frequencies, so is especially problematic for individuals with hearing loss, who tend to have poorer audibility and spectral resolution in the high frequencies.

Understanding the impact of mask type on audiovisual perception is important, as the National Association of the Deaf (NAD) and opinion pieces have recommended use of transparent face masks to allow access of visual cues during both spoken and manual communication (Campagne, 2021; NAD, 2020). The clear mask manufactured by ClearMask™ (ClearMask LLC, Baltimore, MD, U.S.A.) was approved by the FDA in August 2020 for use during the COVID-19 pandemic to improve visual cues in the medical environment, but is more expensive than standard surgical masks. An alternative reusable mask that combines a face shield and washable fabric cover to prevent discomfort around the ears and movement problems is the "apron mask". It is intended to prevent virus transmission that can occur around clear face shields that are worn alone without masks.

We designed this study to determine if young children who are DHH benefit from visual cues provided by transparent masks (ClearMask and transparent apron mask), compared to no masks or standard surgical masks. We hypothesized that all face masks would significantly degrade acoustic quality and word recognition in young listeners, thus a no mask condition would present the highest level of accuracy understanding speech in noise. The ClearMask and a custom transparent apron mask, which provide the added benefit of visual cues, were expected to present a higher percentage of accuracy

than the surgical mask condition. Because young children who are DHH rely more on visual cues than their peers with lesser degrees of hearing impairment, they may demonstrate greater accuracy on the ClearMask and transparent apron mask conditions, and poorer accuracy in the surgical mask condition.

Method

Children aged 3 to 7 years, with varying degrees of hearing loss, who attend school in a private auditory oral program were included in the study. All participants are oral language users of hearing aids (HA), bone anchored hearing aids (BAHA), or cochlear implants (CI). All receive daily intensive speech and language intervention using the Listening and Spoken Language approach. Children were assigned to groups based on the degree of hearing loss in the better ear (*profound* using CI versus *severe or less* using HA or BAHA), detailed in Table 1. Children were excluded if they did not use English as their primary language, had visual impairment not remedied by corrective lenses, or had severe speech-language or developmental delay that precluded their ability to respond verbally to the word recognition task. All children included in the study had routine speech-language and hearing assessments at the school, and data logging of their amplification devices to ensure regular device use. The study was reviewed and approved by the research committee and executive director at the school, and an approved written consent form was sent to parents, who provided informed consent. The Institutional Review Board at Cincinnati Children's was consulted, and the study was not required to be externally reviewed, as research conducted in accepted educational settings, that involves normal educational practices, including most research on special education instruction strategies are exempt according to 45 CFR 46.104. All data were de-identified using a unique numerical identifier prior to statistical analysis.

Table 1
Demographic and Clinical Data for Children Included in the Study

Group		Age at HA or CI (years)	Age at Enrollment (years)	Age at Test (years)	Aided Avg dB HL (.25-8 kHz)
HA or BAHA	Mean	1.64	2.83	5.16	20.50
	Std Dev	1.15	1.42	1.07	9.27
CI	Mean	1.34	1.43	4.47	27.43
	Std Dev	0.56	0.78	0.78	3.80
Student *t*-test (2 sample, heteroscedastic)	*p*-value	0.6473	0.0852	0.2719	0.1697

Note. BAHA = Bone-anchored hearing aid; CI = cochlear implant; HA = Hearing aid; HL = hearing level.

Procedures

The Word Intelligibility by Picture Identification (WIPI) test (Ross & Lerman, 1970) was selected for word recognition testing. Although it has a specified language age between five and eleven years, it has been used routinely at the school with younger children. It is a closed set format and has multiple test lists equalized for difficulty. The WIPI is effective in evaluating ability to identify words on the basis of their spectral characteristics in young children with congenital deafness (Schindler et al., 2003). In this test, the listener hears the phrase "point to," followed by a target word. A set of six pictures is shown, and the listener is asked to identify the picture corresponding to the target word. We adapted and recorded the WIPI test for audiovisual presentation via computer, with pictures displayed on the standard test book. Four 25-item lists, one per mask condition were spoken by a female adult native, Midwestern English speaker (Erin Lipps, educational audiologist). The outcome variable was percent correct recognition of words in quiet for three face mask conditions as shown in Figure 1, in counterbalanced order with the no mask condition as the control. The apron mask was custom designed by the school, while the other masks were purchased from commercial suppliers.

The WIPI lists were audio-visually recorded on an iPad with an internal camera and an external Blue-Yeti microphone in a double-walled sound booth (Industrial Acoustics Company, Inc. Model 120A). The video recording was focused on the speaker's face showing her entire head and shoulders while wearing the different masks, and the speaker was facing the video camera. A Larson-Davis system 824 sound level meter (Depew, New York) with a Brüel & Kjær half-inch free field microphone (type 4189, Nærum, Denmark) was used to ensure the long-term average level was at 65 dBA ± 2 dB sound pressure level (SPL) for all conditions. The speaker was seated three feet from the microphone and instructed to speak each word with a constant effort across the mask conditions. The words were spoken with a 10 second inter-word interval to provide time for responses.

Masks Used in the Study

In the test setting, the child participant sat at a table in a quiet office, with the educational audiologist as the tester. The word lists and mask conditions were presented in a pre-set, counterbalanced order across the participants, to avoid order effects for both word list and mask condition. The simultaneous audio- and video-recorded word lists were presented via a desktop computer and external monitor in a quiet room in the school setting. The computer speaker volume was set at 85% and the video player volume was set at 100%. Using these settings, the stimuli were measured using a Larson-Davis sound level meter (System 824) with a Brüel & Kjær half inch free field microphone (Type 4189). The equivalent continuous sound level (Leq) was 55 dB SPL, ranging from 51 to 60 dB SPL. Peak SPL was 85 dB, ranging from 63 to 90 dB SPL.

The child was instructed to watch the computer monitor that showed the presenter, with or without a mask, and listen to the word lists spoken by the presenter at face level, at a standard distance of three feet, presented binaurally through the computer speaker. The tester showed the participant the standard WIPI test book of six pictures on each page, and the participant chose the picture that matched the word they heard and scored the response on the corresponding word list. Having one person administering and scoring the assessments minimized the effects of interrater reliability, but the scorer was not blinded to the degree of hearing loss or type of amplification device. The percent of correct words identified for each condition and each group (HA vs. CI) was analyzed for significance using a two-way Repeated Measures Analysis of Variance (RMANOVA; mask

condition as the repeated measure). Post-hoc tests were performed if the RMANOVA was significant for each pair of mask conditions.

Results

Children who enrolled and completed testing (*N* = 14) were divided into two groups based on the degree of hearing loss in the better hearing ear and device type. One child with HAs had highly irregular scores across conditions and appeared to have variable attention. That child was subsequently diagnosed with autism, so was excluded from the final analysis. The remaining sample of 13 children included: (a) Bilateral HA or BAHA group (*n* = 6; 5 males and 1 female; 4.0 to 6.9 years) with normal sloping to profound sensorineural or conductive hearing loss, and (b) Bilateral CI group (*n* = 7, 3 males and 4 females; 3.3 to 5.7 years). Children were tested using their devices set to their typical settings. Table 1 provides comparisons for clinical data for both groups. The sample was 79% Caucasian, 14% African American, and 7% Asian. Most of the etiologies were congenital cytomegalovirus (CMV, 38%) or unknown (38%); of the others, 15% had craniofacial anomalies, and 8% had Usher syndrome.

Real ear validation was completed on every child with a hearing aid. Additionally, every child received LING 6 checks twice daily to ensure they had access to the full speech spectrum. Individual aided audiograms are shown in Figure 2 for the left and right ears, and for HA and CI users separately. One child with a BAHA is not included in the aided audiogram figure since the mode

Figure 2

Individual Aided Audiograms for Right and Left ears, for Hearing Aid (HA) and Cochlear Implant (CI) Users
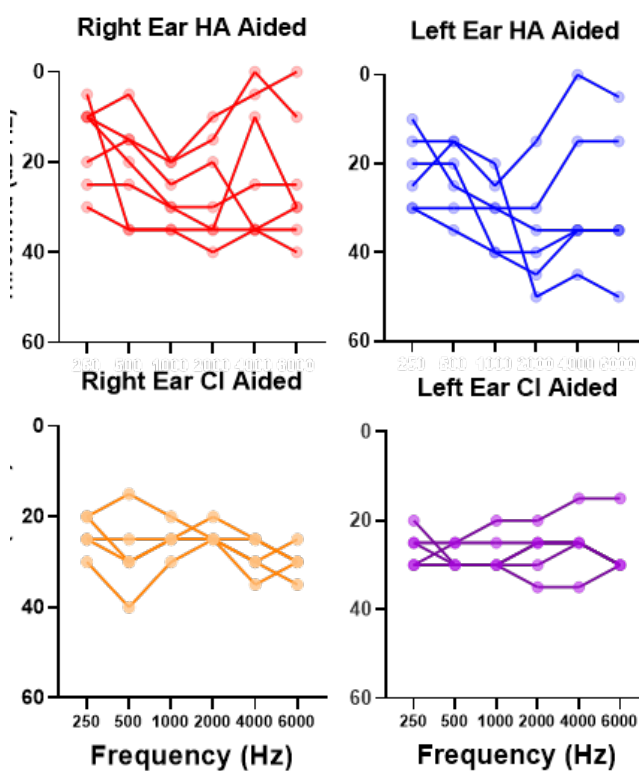
was vibrotactile, and therefore the ear stimulated is unknown. These figures illustrate variability in access to sound, especially for children wearing HAs in the high frequencies. Average aided thresholds for children wearing HAs fell into the 8 to 35 dB HL (hearing level) range, while aided thresholds for children wearing CIs fell in the 21 to 35 dB HL range.

The first 10 words from the WIPI word list were recorded and analyzed for spectral content across the four mask conditions, spoken by the same speaker. Figure 3a shows the spectrograms for the 10 words averaged across each mask condition. The average spectrograms showed that, compared to the no mask condition, the surgical mask had the smallest reduction in high frequencies (> 2 kHz). The ClearMask had a resonant enhancement at 2800 Hz, but slightly less energy overall in the higher frequency range, especially between 3000–4000 Hz. The apron mask had

the largest overall attenuation, especially from 2000 to 8000 Hz. The average difference in band energy between the no mask condition (baseline) compared to the face mask conditions across the 10 words is shown in Figure 3b. All three mask conditions showed an enhanced level of 6–10 dB, relative to no mask, at 500 Hz (Figure 3b), but variable decreases at higher frequencies. Overall, the surgical mask had the least effect, the ClearMask was attenuated uniformly at 1000 Hz and above, and the Apron mask had the largest enhancement at 500–1000 Hz, and the largest decrease above 2000 Hz. Figure 3c shows the spectrograms for six words selected across the range of lower and higher frequency initial consonants, and for different vowels (ball, egg, school, fox, hat, and smoke). These spectrograms demonstrate a similar pattern as the overall patterns for each mask type, indicating that the effects were due to mask differences rather than differences among the words between lists.

Figure 3

Recording and Analysis of Words for Spectral Content
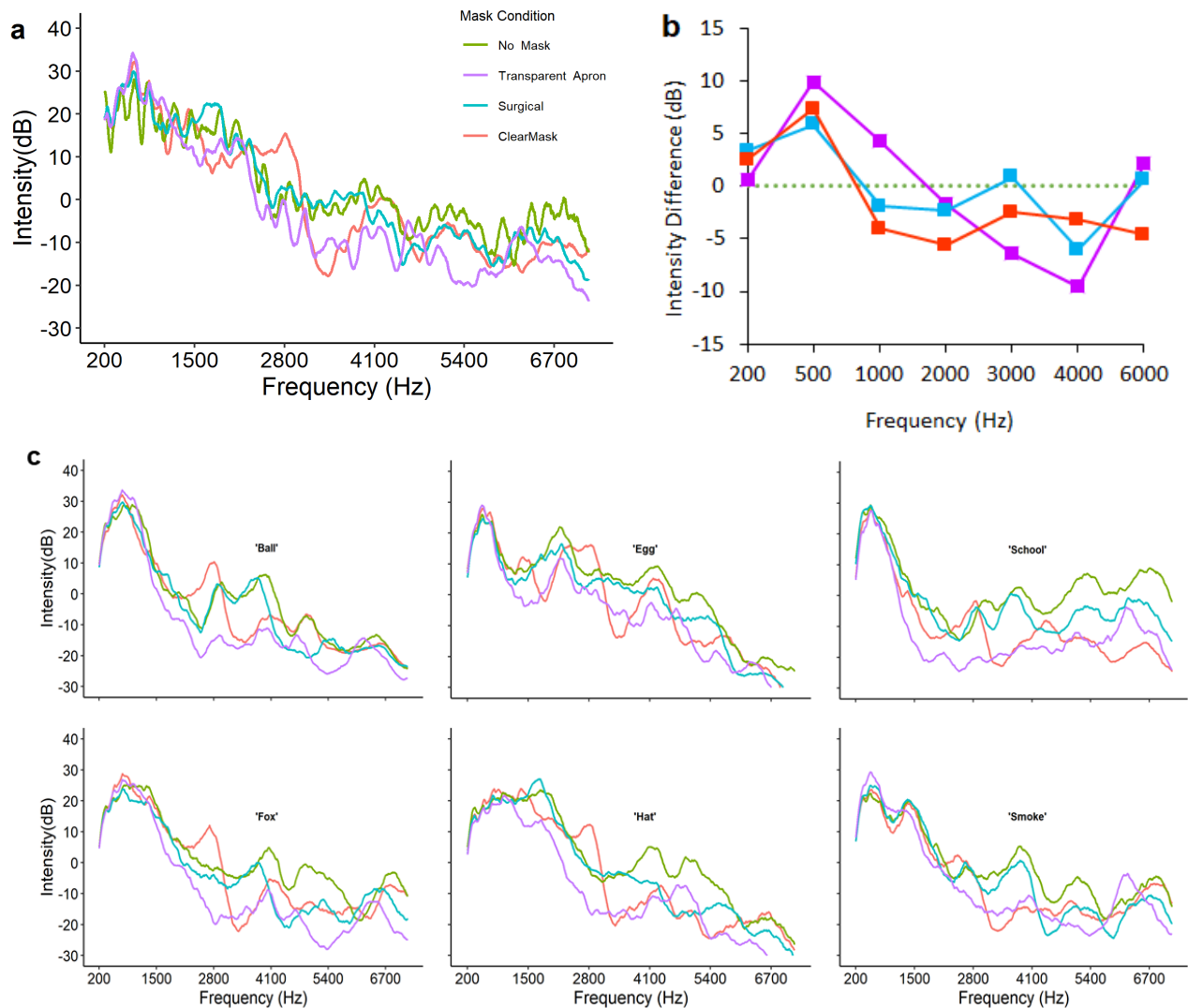
Note. (a) Power spectra of the 10 words averaged across each mask type. (b) Difference in band energy between the three mask conditions in reference to the no mask condition. (c) Power spectra of six example words selected across the range of lower and higher frequency consonants, and different vowels (ball, egg, fox, hat, school, and smoke).

Individual children's performance across the four mask conditions is shown in Figure 4 for HA and CI groups separately. There was substantial variability in each condition in both groups, and the HA group overlapped the scores of the CI group. There were no ceiling or floor effects in the word recognition scores, so the WIPI test was well suited to the children's language ages and their aided speech perception skills. The two-way RMANOVA (Table 2) showed no overall difference in the scores of the HA group compared to the CI group. Since there was not a significant group difference, combined data for both groups across the conditions is shown in violin plots (Figure 5). There was a significant main effect of mask type on word recognition ($p < 0.004$). Post-hoc pairwise comparisons (Holm-Šídák correction) showed that the no mask condition was significantly better compared to the apron mask ($p = 0.017$) and the surgical mask ($p = 0.004$), but the ClearMask was not significantly different from the no mask condition ($p = 0.178$). The range of scores was smaller and generally poorer for the surgical mask, which suggested that loss of visual cues was important, but there was not a statistically significant difference between the mask types.

Table 2
Two-way Repeated Measures ANOVA Results

Within Subjects Effects				
Cases		**df**	**F**	**p**
Mask Condition		3	5.458	*0.004
Mask Condition ✳ Group		3	0.700	0.559
Between Subjects Effects		**df**	**F**	**p**
Group		1	2.543	0.139

Note. Type III Sum of Squares

Post Hoc Comparisons - RM Factor 1					
Comparison		**Mean Difference**	**SE**	**t**	**p** holm
None v.	Apron	10.905	3.466	3.146	*0.017
	Surgical	13.048	3.466	3.765	*0.004
	Clear	7.238	3.466	2.088	0.178
Apron v.	Surgical	2.143	3.466	0.618	0.596
	Clear	-3.667	3.466	-1.058	0.596
Surgical v.	Clear	-5.810	3.466	-1.676	0.309

Note. *p*-value adjusted for comparing a family of 6 using Holm-Šídák method. Results are averaged over the levels of Group. Significant comparisons are noted with an asterisk ($p < 0.05$).

Discussion and Conclusions

In this sample of children enrolled in an oral school setting, we found that both the standard surgical and transparent apron mask presented a significant barrier to audiovisual communication in young children who are DHH. The spectral analysis showed that the surgical mask had a small effect on the acoustics of speech, thus the observed decrease in word recognition is likely due to loss of visual cues. The ClearMask had an interesting effect on the acoustics of the speech signal, with an apparent increase, or resonance in the frequency range around 2800 Hz that may partially offset the loss of cues at higher frequency regions, but a decrease in the range just above 3000 Hz. Even though the surgical and ClearMask had relatively similar impacts on acoustics, the ClearMask was not significantly poorer than the no mask condition on recognition of words in quiet. This may be due to visual cues preserved by the ClearMask compared to the surgical mask. The ClearMask produced the most variable scores, although 9 of 13 children maintained similar scores in this condition, compared to their unmasked performance. The transparent apron mask had a greater impact on acoustics of speech. The size and placement of the apron mask on the face also appears to obscure some visual cues due to greater glaring, and adversely affects transmission of acoustic energy. All three types of mask had a resonant peak at about 500 Hz compared to the no mask condition. This increased level at low frequencies could make speech sounds muffled and less intelligible. Consistent with this finding, studies in adults have consistently found negative effects on speech communication with surgical masks in quiet (Bandaru et al., 2020) and for words and sentences in noise (Atcherson et al., 2017; Bottalico et al., 2020; Hampton et al., 2020; Toscano & Toscano, 2021; Wittum et al., 2013). Studies in adults have found a benefit of transparent masks, especially in noisy backgrounds, even in adults with normal hearing (Atcherson et al., 2017; Thibodeau et al., 2021). A recent study in adults with cochlear implants showed the greatest attenuation of high frequency acoustics and sentence perception in noise with an N95 mask plus a face shield, compared to an N95 mask or no mask (Vos et al., 2021). A survey of impacts on communication with mask wearing in adults reported that face coverings negatively impact hearing, understanding, engagement, and feelings of connection with the speaker, especially when communicating in medical situations (Saunders et al., 2020). People with hearing loss were more impacted than those without hearing loss.

The only other study on communication with masks we are aware of in children who are DHH was recently reported by Lalonde et al. (2021). That study compared auditory alone and audiovisual speech perception of consonant-vowel phonemes in speech-spectrum noise in children who are DHH aged 7–18 years to their siblings with normal hearing and to parents with normal hearing. The no mask condition was compared to a surgical mask, cloth mask, ClearMask, and transparent Communicator brand mask. Similar to our findings, the ClearMask had greater attenuation in the high frequencies than the surgical mask. Results showed

Figure 4

Individual Percent Correct for Each Mask Condition by Group

Note. Left panel: Cases with normal-severe hearing loss using hearing aids (HA) or bone anchored hearing aids (BAHA). Right panel: Cases with profound hearing loss using cochlear implants (CI).

that children with hearing loss performed worse than normal hearing adults or siblings. Children who are DHH benefitted more from visual cues with clear masks, and audiovisual speech perception was the least affected by transparent masks.

Limitations of the current study are a relatively small and restricted sample size at one oral school with a single familiar speaker, and performance on a single monosyllabic word recognition task in quiet. Impacts of noise in the classroom and effects of less familiar speakers or rapid running speech would undoubtedly exacerbate the effects shown here, but were not assessed in this study. We may have had insufficient power to detect small differences among the mask conditions, especially with the large variability among mask types. Strengths of the study include the diversity of hearing loss type, range, and type of devices, as well as etiologies of congenital hearing loss. Because the children were in an auditory-oral educational setting, they rely heavily on acoustic as well as visual cues for communication. Normal hearing children, or children educated with sign language may have different results.

Benefits of the transparent apron or ClearMask may include emotional connections and ability to see facial expressions, in addition to speech reading cues. Facial recognition is an important social and psychological input for children and for adults (Freire & Lee, 2001). Facial cues are important for sign language users, thus non-transparent face masks would be expected to impact their communication accessibility (Campagne, 2021). Additionally, face masks obscure reading of emotion, an important skill for communication development in young children (Carbon, 2020). Facial recognition may also provide a greater advantage in noisy classroom conditions that we were not able to study in the classroom environment due to pandemic restrictions. This would be a valuable area to study in the future since mask wearing may become routine in school settings with continued Covid-19 restrictions or new infectious outbreaks.

Figure 5

Violin Plots for Each Mask Condition for Both Groups Combined
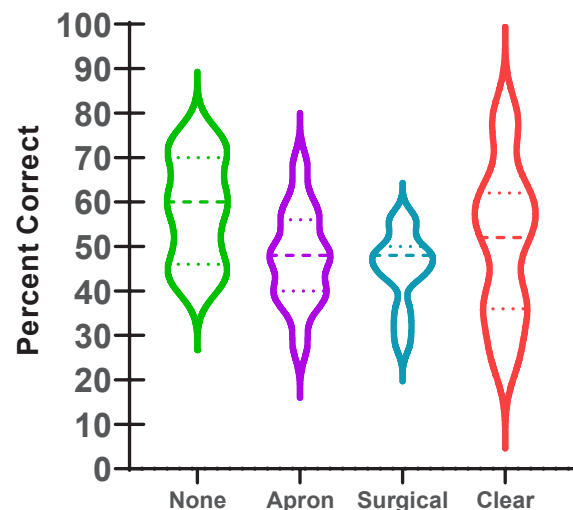
Note. Mean scores are shown by the middle dashed line, dotted lines represent interquartile intervals, and stems show ranges.

Educators using the transparent apron mask at this school reported improvement in ease of communication with children who use visual cues for speech understanding. They reported that the transparent apron mask is particularly useful during speech tasks which require the child to see the educator's mouth for visual cues. They were not using the ClearMask in the classroom, so we do not know how it works in practice in the classroom. Educators did report that the ClearMask was not preferred due to fit issues and shifting around the face when talking. There was concern that this led to increased touching of the face and potential for increased risk of viral transmission. Additionally, the disposable nature of the ClearMask makes it a more expensive option. However, based on speech perception benefits demonstrated in this study, it is a viable, commercially available choice to provide audiovisual cues whenever

audiovisual communication is important and thus deserves further study.

Another option that is readily available in schools for children who are DHH are remote microphone technologies to overcome acoustic degradation, especially in noise. Corey et al. (2020) found that masks have little effect on lapel microphones, suggesting that existing sound reinforcement and assistive listening systems may be effective for verbal communication with masks. Thus, use of existing remote microphone technologies with children who are DHH in combination with transparent masks would allow both auditory and visual cues to be maximized, and provide the emotional connection that children need, especially during stressful times as children and their families experienced during the Covid-19 pandemic. This combined option would be the best choice if masks must continue to be worn by teachers and other personnel in classrooms settings in the future.

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EHDInfo   [Click graphic to see complete Guide.]

Early Hearing Detection and Intervention (EHDI) Frequently Asked Question (FAQ) Guide for Pediatricians

The EHDI FAQ Guide, developed by the American Academy of Pediatrics (AAP) EHDI program, is an introductory resource for pediatricians serving families with children who are deaf or hard of hearing (D/HH). This resource provides important resources about the EHDI program in states, discusses how best to partner with families, and offers tips for billing and coding.

Audiologist Perception on Reporting Hearing Assessment Results to State Early Hearing Detection and Intervention Programs

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Abstract

Purpose: To describe an evaluation conducted by 39 state Early Hearing Detection and Intervention (EHDI) programs on the reporting process and system usability for audiologists when reporting the hearing test results to the EHDI program and the barriers encountered during reporting.

Method: Each author independently extracted numbers, percentages, and texts from the evaluation reports into an Excel spreadsheet, which then became the dataset. Authors then compared and cross-checked the datasets before coding. Texts conveying similar concepts were coded with the same name and organized into categories. Finally, thematic identification and analysis were performed when a theme(s) or concept(s) that pertained to similar challenges encountered by audiologists was identified and organized under a higher-order domain.

Results: Some audiologists reported no barriers when reporting hearing test results to the state EHDI programs. Among those audiologists who reported barriers, the most recurrent barrier was a non-user-friendly data system design. The second most recurrent barrier was not having adequate administrative time to report data as a busy clinician. The third most recurrent barrier was an incomplete understanding of the state EHDI reporting requirements. Finally, the method audiologists were required to use when reporting results also posed some challenges, such as no internet connection in rural areas when required to report via an internet portal.

Conclusion: Because of the wide variety of barriers faced by audiologists, multiple strategies to improve the reporting process would likely be beneficial.

Keywords: reporting hearing result, EHDI program, barriers to reporting, audiologist

Acronyms: AAP = American Academy of Pediatrics; CDC = Centers for Disease Control and Prevention; DHH = deaf or hard of hearing EHDI = Early Hearing Detection and Intervention; NCHAM = National Center for Hearing Assessment and Management;

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All U.S. states and territories have an Early Hearing Detection and Intervention (EHDI) program to help ensure all infants are screened for hearing loss and receive recommended follow-up diagnostic testing and intervention services (National Center for Hearing Assessment and Management [NCHAM], 2020). EHDI programs track and, in some states, coordinate follow-up services for infants who may be deaf or hard of hearing (DHH). Newborns who do not pass their hearing screen are often referred to an audiologist (a licensed provider of hearing evaluation and services) for diagnostic testing by hospital staff or by the state EHDI programs. Audiologists are one of the crucial links in the EHDI surveillance effort because they have information on the hearing status of newborns whom they have tested. Without the audiologists reporting the hearing test results to the state EHDI program timely, service coordination and enrollment into Early Intervention for children who are DHH

may be delayed or not completed. It is equally important for audiologists to report normal hearing results to the state EHDI program as state EHDI program staff cannot accurately determine which cases no longer require follow-up and coordination without these results. The non-reported data gap may result in staff time dedicated to tracking a newborn who does not require service coordination, as well as a downstream effect that leads to an inaccurate estimate of the number of newborns who are DHH.

The importance of clinical providers reporting hearing test results to their state EHDI programs in a timely manner is reflected in statutes enacted by several states (Division of State Government Affairs, American Academy of Pediatrics [AAP], 2014; NCHAM, 2019). Detailed requirements for providers can include how, what, and when to report results to the program responsible for tracking newborns who

have not passed their newborn hearing screen. Despite statutes and regulations, not all audiologists may routinely comply. In the only known published study on audiologists' willingness and compliance in reporting hearing assessment results to the EHDI programs in the United States, of the 1,024 audiology facilities surveyed, 8.6% did not report results to their state EHDI program (Chung, Beauchaine, Grimes, et al., 2017). To date, there are no additional published studies that have attempted to identify barriers encountered by audiologists when reporting hearing assessment results to state EHDI programs.

From 2017 to 2020, the Centers for Disease Control and Prevention (CDC) provided funding to U.S. states and territories to identify and implement approaches to strengthen their program's capacity to capture complete and accurate data on all infants in need of recommended hearing evaluation and intervention services. Not all states applied for the funding. Funded states and U.S. territories were required to evaluate how acceptable the established reporting process and system was to the users when they reported test results to their state's EHDI program and any barriers they might have encountered. This article describes the evaluations conducted and their findings.

Method

Evaluation Framework and the Data Source

In September 2017, CDC provided guidelines on the key concept definition and type of evaluation questions that funded states should use in their process and system evaluation. The key concept, *How acceptable is the EHDI reporting process?* is defined as the willingness of persons or organizations to participate or use an established reporting method (the process) and the interface portal or reporting form (the data system) when reporting a hearing assessment result. The evaluation questions were standardized as follows: (a) To what extent do audiologists in the state know about reporting and are using the established reporting portal or method? (b) Are the reporting portal or other established methods user-friendly? (c) What barriers have prevented audiologists from reporting hearing assessment results? and (d) What are the audiologists' perceptions on the reporting process and system design?

Standardizing how state EHDI programs should evaluate program and system barriers to reporting and at the same time allowing each program room to modify the approach were important. The former allowed us to aggregate the evaluation data across multiple states and the latter allowed the program to adapt the approach to suit their unique process. Although process guidance was also provided to states to help reduce variation in the evaluation process, each state could choose a data collection method, such as survey or interview, that best suited their need and internal process. Process guidance included a requirement to (a) engage key stakeholders in the state to assist in the evaluation, (b) choose an evaluation method(s) that can adequately answer the four evaluation/study questions listed above, and (c) disseminate findings as lessons learned to key stakeholders, in addition to reporting evaluation data and results to CDC.

To ensure all key evaluation elements were reported to the CDC, states and territories used a CDC-designed report template. The following information was requested in the template: (a) the key stakeholders engaged and their role in the evaluation, (b) a description of the statutes and regulation on reporting hearing assessment results to the appropriate program, if applicable, (c) a description of the reporting process audiologists should use, (d) the data collection method(s), and (e) the challenges and barriers encountered by audiologists.

By December 2018, 42 funded EHDI programs successfully completed the process and system evaluation. We excluded three evaluation reports from the analysis, as they were from U.S. territories with either no audiologists or only one audiologist to serve an entire community's hearing care needs. This left 39 evaluation reports for qualitative data coding, thematic identification, and domain analysis.

Qualitative Data Coding and Analysis

We applied an inductive approach to derive explanations from the collected qualitative data, as opposed to a deductive approach, which is used when a hypothesis is developed prior to data collection (Williams, 2019). The grounded theory framework for analyzing and organizing qualitative data was developed by Glaser and Strauss (1967). For this framework, (a) concepts, not data, are the basic units of analysis, and (b) concepts that pertain to the same phenomenon may be grouped to form categories. Coding is a process of classifying and categorizing text data segments into concepts and categories or constructs. Strauss and Corbin developed various ways to code qualitative data (Corbin & Strauss, 1990; Strauss & Corbin, 1990). Analysis and interpretations are grounded solely on collected data representing the observed phenomenon to reduce biases.

No computer-aided qualitative data analysis software was used. Each author independently extracted numbers, percentages, and texts from the evaluation reports and entered them in an Excel spreadsheet, forming our dataset for analysis. The numbers and percentages reflected number of audiologists who had participated in the evaluation and who had encountered barriers when reporting hearing assessment results. Texts described stakeholders who assisted with the evaluation, the evaluation method used, and the audiologists' perception of the challenges and barriers when reporting hearing assessment result to the EHDI program. Both authors compared the datasets to ensure the data were the same before proceeding to open coding, a process to identify concepts related to the phenomenon of interest expressed in a text (Medelyan, 2019). Words, phrases, and sentences that conveyed the same meaning or concepts were coded or tagged as the same (Guest & McLellan, 2003). For example, comments such as "busy," "no time," and "no time for administrative tasks" were coded as "no time" because they all conveyed the same meaning. Coding comments that conveyed the same meaning with a code or label, such as "no time," "password reset issue," "non-user-friendly design", and "internet connection issue,"

also facilitated counting the times a comment recurred. The coded comments were organized into categories. The categories were stakeholder type, stakeholder role, the reporting process created by the EHDI program, type of evaluation method used, survey response rate, and type of barriers reported by audiologists. Each author conducted the coding independently and the results were compared; differences were discussed and resolved before moving to thematic identification and analysis.

The intent of a thematic analysis was to identify concepts that come up repeatedly in a qualitative dataset (Nowell et al., 2017). Each author independently reviewed the meaning of each audiologist's comments to identify a theme(s) that could connect certain comments together. Since all audiologists' comments were already labeled with a code, such as "no time," "password reset issue," "non-user-friendly design", or "internet connection issue," the code also helped to identify a theme. For example, some audiologists reported "system sign-in very cumbersome," "have to sign in twice to access the system," or "takes state IT too long to reset expired password," all of which points to the recurrent theme that system access was a barrier to reporting. Since the number of times certain

types of comments recurred was quantified during the previous step, it helped inform the authors of the frequency of certain themes. Both authors compared and resolved any difference in the themes identified before moving to the final phase, selective coding, where themes were further unified around a *core*. Selective coding usually occurs in the later phase of a qualitative data analysis (Corbin & Strauss, 1990; Williams, 2019). The first author analyzed the 10 themes identified in the previous step to find a higher order domain, or core, that the themes could be subsumed under. For example, the following four themes: system access issue, system reliability, issues locating the right patient file, and non-user-friendly designs could be subsumed under system design domain. See Table 1 for the qualitative data review process and results.

Results

Reporting Process and System Evaluation

When conducting their evaluation, state EHDI programs engaged diverse stakeholders. The number of stakeholders who assisted ranged from 3 to 12 overall, and included staff from other departments, such as the state licensure board or epidemiologists. When designing

Table 1

Thematic Analysis and Coding Process on Audiologists' Perception on the Challenges in Reporting Hearing Assessment Data to State EHDI Programs

First Step: Coding and Counting Comment Frequency	Second Step: Thematic Analysis	Final Step: Theme Consolidation under a Domain
Coding qualitative data and computing frequency of certain type of comments	Identify concepts that come up repeatedly in a qualitative dataset	Subsume related thematic categories under a higher order domain
▪ Comments such as "no time" or "busy" were coded as busy because both terms conveyed the same meaning. ▪ Each comment that reflected having no time to report was counted as 1 ▪ Although "unaware of reporting," "unaware that I need to report normal result," and "don't know how to report" reflected knowledge lack, type of knowledge lack was different in each comment. ▪ Therefore, comments were kept separate but placed in the same category: knowledge lack. ▪ Again, each comment that reflects a lack of knowledge from a responder was counted as 1.	10 themes identified from the coded qualitative comments: 1) Difficulty accessing system 2) System reliability 3) Difficulty locating patient in the system 4) Non-user-friendly design 5) Work demand 6) Assumptions about reporting in a fractured healthcare environment 7) Incomplete knowledge on reporting requirement 8) Lack resource/tool 9) Process issue 10) Perception that reporting is a duplicate effort	 Theme 1-4: System design domain Theme 5-6: Work demands & healthcare environment domain Theme 7-8: Incomplete knowledge and resource domain Theme 9-10: Process-barrier domain

their evaluation, many EHDI programs also engaged community stakeholders, such as audiologists from their own state. State EHDI programs and stakeholders worked collaboratively to design questions for a survey, focus group, or structured interview.

Audiologists were the target population, and EHDI programs compiled a list of audiologists from different sources. Some programs targeted audiologists who had previously reported to the EHDI program. Several programs targeted those audiologists to whom they routinely referred newborns for audiologic assessment, while other programs obtained a list of audiologists from the EHDI-Pediatric Audiology Links to Services website (http://ehdipals.org); Chung, Beauchaine, Hoffman, et al., 2017) or from their state's licensure board. Only two programs targeted audiologists attending local conferences.

Data collection methods implemented by state EHDI programs also varied. Slightly more than half (56%, n = 22) of the EHDI programs used one method to collect audiologists' experiences, while the remaining 44%

used multiple methods (Table 2). When multiple data collection methods were used, a survey was typically done first, followed by a structured phone interview or an in-depth focus group. Most of the state programs (66%, n = 26) used surveys to collect audiologists' experiences and perceptions. In the survey, EHDI programs used a combination of open text fields and a multiple-choice format to capture audiologists' comments. A majority of the state EHDI programs posted their surveys online and contacted audiologists via e-mail to complete the survey. Survey responsiveness ranged from 10% to 100% (median 55%, mean 54%; Table 3); a higher response rate was achieved by surveying regional audiology conference attendees.

Reporting Methods Audiologists Can Use

Most of the state EHDI programs (64%, n = 25) implemented a secure, password-protected online portal or interface for audiologists to report hearing assessment results. To report hearing assessment results via the portal, each audiologist must request system access from the EHDI program. In 19 (48%) states, the EHDI

Table 2
Data Collection Methods Used by State EHDI Programs When Evaluating Audiologists' Perception on the Reporting Process

Number of state EHDI programs	N = 39
Used only one method	22 (56%)
Survey (online, by phone, or onsite at audiology conference)	21
Focus group (in-person)	1
Used multiple methods	17 (44%)
Online survey followed by structured phone interview	8
Survey (online, phone, or onsite at audiology conference) followed by a focus group	5
Structured phone interview followed by an in-person focus group	1
Online survey followed by structured phone interview and an in-person focus group	3

Table 3
Survey Response Rate of Audiologists and Number of States where Audiologists Reported No Barriers to Reporting

Survey Response Rate of Audiologists	
Number of EHDI programs N = 26	**Response rate of audiologists**
10	60–100%*
9	40–59%
7	< 40%
*When survey was conducted in-person at a conference or when there was only a small number of audiologist (less than 20) to serve children in the state, the response rate was higher (80–100%)	
Percent of Audiologists who Reported No Barriers to Reporting	
Number of states where audiologists reported no barriers n = 13	**Percent of audiologists reporting no barriers**
3	81–100%
3	61–80%
5	41–60%
1	21–40%
1	0–20%
Range 19 to 100%, median 50%, mean 58%	

programs requested audiologists fax a hearing result form to the program. Two EHDI programs implemented other less labor-intensive reporting alternatives for audiologists. Both programs signed a data sharing agreement with the hospital so program staff could access only a limited area of the electronic medical record to extract hearing assessment data. Additionally, one of the programs also allowed audiologists to upload their diagnostic reports to the online portal.

Audiologist Perception on Reporting Hearing Results to State EHDI Programs

The number of audiologists reporting barriers versus no barriers varied across participating states. In 13 states there was a percentage of audiologists who reported no barriers at all (Table 3). In these 13 states, only 6 states had a large percentage of audiologists (> 60%) who reported having encountered no barriers (range 19–100%, median = 50%, mean = 58%). Among those audiologists who encountered barriers when reporting hearing results, 10 themes emerged from our qualitative data analysis (Table 4). The 10 themes could be further condensed into four domains. The number one barrier reported most often (58 times) was a non-user-friendly system design. The second most reported barrier (36 times) was related to the demands on a clinician. The audiologists were busy, often commenting that they did not have adequate time to report hearing results. The third most reported barrier (32 times) was a lack of knowledge on, or incomplete understanding of, state reporting requirements. Finally, and to a lesser extent, issues with the reporting method, such as fax not going through or no internet connection to access the online reporting portal, were reported 13 times by the audiologists.

Discussion

Each state has its own unique EHDI data reporting system, some more user-friendly than others. The wide range of audiologists reporting no challenges (19–100%; Table 3) may be a result of this variation in the uniqueness of the reporting system in each state. The most recurrent barrier (reported 58 times) was a non-user-friendly reporting system. The non-user-friendly design covered all areas of the reporting system such as logging on, finding the right child record, and entering and saving data. The following comments from respondents illustrated the different kinds of system design issue:

- Neonatal intensive care unit and well-baby in 2 systems. Have to log into two systems to report

- Poor search function, so difficult to find child

- Difficulty in navigating the reporting tabs

- Diagnosis codes audiologists required to use difficult and non-intuitive

- Takes too long to enter all required fields

- Certain data could not be entered accurately

- System unreliable, reported results not saved

Some of these difficulties could be encountered by audiologists who were not frequent users, but some challenges truly reflected a system design issue irrespective of user comfort level (e.g., "order of reporting tabs not logical," "unsure how to input certain data," "certain data could not be entered accurately," "takes too long to enter all required fields," and "child can have three separate profiles in three different databases. Do not have access to all databases to locate child;" see Table 4).

The second most recurrent barrier (reported 36 times) was related to the demands on a clinician. The primary duty of an audiologist is patient care. Besides patient care, there were other non-direct patient care duties requiring a clinician's time, such as dictating an evaluation report to the referring physicians, returning patient phone calls, obtaining healthcare insurance authorization for hearing aids on behalf of the patient, and ordering hearing aids or earmolds, etc. These non-direct patient care duties were usually done at the end of the day or when a patient did not show for their appointment. Given limited or no time allocated during a workday for non-patient care tasks, audiologists must prioritize. We hypothesize that tasks that directly impact patient care will rise to the top, exclusive of other duties. Reporting hearing assessment results to the EHDI program is not a patient care task. It could be beneficial for EHDI programs to demonstrate to audiologists how reporting may improve patient care.

Another barrier related to the patient care environment was a lack of communication among clinicians from different clinics. Due to this lack of communication, clinicians likely make certain assumptions. Several audiologists commented that the "Patient has been seen by other audiologists. I assume others have reported." This assumption was also reported by Chung, Beauchaine, Grimes, et al. (2017). It was not unusual for parents to seek a second opinion by visiting more than one clinic. Chung and colleagues reported that 5.4% of the surveyed clinics stated that not all hearing assessment results were reported to the EHDI program. One reason was that audiologists assumed the clinicians who completed the initial assessment had already reported results to the EHDI program.

In the Chung, Beauchaine, Grimes, et al. (2017) study, authors found 8.6% of the surveyed clinics did not know how to report. We also found this lack of knowledge on the reporting requirement and process, causing it to be the third most recurrent theme. Audiologists reported that they were not aware that there was a requirement to report, and were unsure when, what, and how to do so, as evident in the following comments: "did not know I need to report normal hearing results," "unsure which case and what to report," and "don't know how to report." Audiologists also commented on a lack of helpful resources or tools that would assist them in reporting hearing assessment results, as evident in the following comments: "The law mandates reporting only infants that don't pass hearing screens. Lack access to the knowledge of which infant has not passed," and "no hearing

Table 4

Results of the Thematic and Domain Analysis on Audiologist Perception When Reporting Hearing Assessment Results to State Early Hearing Detection and Intervention (EHDI) Programs

Domains and Themes	*Frequency of comment*
Domain I Barrier: Inherent to the system design domain	*n = 58*
Theme 1 — Reporting system access issue Sample comments: Sign in process cumbersome; Must sign in twice; Takes state IT too long to reset expired password	11
Theme 2 — System reliability/stability Sample comments: Data were not saved properly; Fax not going through or fax not receiving	7
Theme 3 — Locating the right patient in the reporting system Sample comments: Poor search function so finding the right child is difficult; Child's name often changes after hospital discharge and reporting system requires exact name and date of birth match and I don't have the birth name	10
Theme 4 — Non-user-friendly design Sample comments: Navigation tab very complicated; Reporting form or reporting page too complicated; Neonatal intensive care and well-baby child records are located in two separate systems	30
Domain II Barrier: Related to work demands on a clinician and the healthcare environment domain	*n = 36*
Theme 5 — Work demands Sample comments: Too busy; No time to report because no time was set aside for paperwork; Short staffed; No financial incentive- reporting reduces time to generate income	31
Theme 6 — Assumptions about the need to report related to the care environment Sample comments: Assume other audiologists have reported because patient has visited another clinic; Patients were seen by different audiologists so likely others have reported	5
Domain III Barrier: Related to incomplete knowledge on the reporting requirement and a lack of helpful tool domain	*n = 32*
Theme 7 — Incomplete knowledge on the requirement and the process Sample comments: Did not know I need to report normal hearing result; Unaware that a reporting requirement exists; Don't know when or how to report	27
Theme 8 — Lack helpful tool Sample comments: No access to EHDI data system to determine which patients require reporting; Law requires me to report only infants who failed; No access to database to find out which infant has failed	5
Domain IV Barrier: Inherent to the reporting process domain	*n = 13*
Theme 9 — Access to a workable process Sample comments: No computer/internet access because no internet coverage; Clinic computer not compatible with the reporting portal.	6
Theme 10 —Duplicate effort/task Sample comments: Must enter data in patient's chart and also for the EHDI program; Have to enter data in 3 separate databases–confusing and increase workload.	7

screening result to help me decide if reporting is required." These barriers all pointed to the need to strengthen training and provide audiologists with access to critical data that would facilitate them reporting hearing results to the EHDI program.

Some audiologists also encountered barriers with the reporting process they were required to follow when reporting a hearing assessment result. This process-related barrier was reported only 13 times by audiologists. For online reporting, audiologists commented that some clinics in rural areas had no internet coverage, their computer was not compatible with the reporting portal, or they had no access to a computer. In states where audiologists were required to report by fax only, audiologists commented that the fax often did not go through. Another process-barrier domain theme was duplication of an effort or task. In addition to notating the patient encounter and results in their medical record and dictating an assessment report for the referring physicians on a daily basis, audiologists also had to enter the same kind of information yet again in the EHDI reporting portal or complete a result form and fax to the program. Besides being perceived as a duplicate effort, reporting results was also perceived as a labor-intensive task by some audiologists who are required to use an online portal to report. The following comment illustrated this perception: "Reporting online could only be done by an audiologist. It would have been helpful if faxing an assessment report was permitted because a support staff could assist."

Since the barriers encountered by audiologists spanned multiple domains, a multi-prong approach to improve the reporting process would be most efficacious. Foremost, working to reduce the burden of data entry on audiologists and minimizing duplicate efforts would likely be beneficial. Improving the online reporting portal should also be considered and, ideally, include feedback from audiologists through user testing to help ensure that the reporting system is intuitive and friendly. Allowing audiologists access to other child health data that benefit patient care could improve audiologists' participation in the EHDI process. Finally, recurrent training should be offered, and should cover who, when, what, and how to report hearing assessment results, regardless of whether the audiologists have been previously trained.

There are several limitations with this study. The qualitative data collected by the EHDI programs might be overrepresented by audiologists whose caseloads were predominately children. Audiologists who saw children less frequently might have different challenges. However, barriers reported by audiologists whose caseloads were predominately children should carry greater weight when EHDI programs want to improve the reporting process, since these audiologists would be frequent users. Although we standardized the evaluation questions, it was possible EHDI programs might have interpreted the questions differently, which could have influenced how the questions were posed to audiologists. To help mitigate this possibility, CDC provided definitions for key terms, such

as acceptability, and reviewing their evaluation plan before the program executed the evaluation.

Another limitation was the various ways EHDI programs used to collect the evaluation data and determine the pool of audiologists to target for the evaluation. Slightly more than half (56%, $n = 22$) of the EHDI programs used one method to collect audiologists' experiences, while the remaining 44% used multiple methods (Table 1). Some programs used licensure board information to determine the pool of audiologists to target, while others targeted audiologists who had previously reported to the EHDI program. This variability created a weakness, as the results might not be generalizable to represent all audiologists. On the other hand, allowing the EHDI programs some flexibility in how the evaluation should be conducted was considered important. For example, some EHDI programs vetted clinics to ensure the clinics had the equipment and capable personnel to evaluate newborns, toddlers, and young children since the equipment needed to evaluate the different age groups varies. If the funding evaluation guidance required states to target all licensed audiologists in the state for the evaluation, it would not be appropriate for states that only require vetted clinics to report and if we required states to use only one data collection method, such as a focus group format, it would be impractical for the EHDI program to collect feedback from audiologists located in rural or frontier areas. Despite this variability in evaluation method used by the state EHDI programs, we found convergence of key themes and issues encountered by audiologists across 39 states.

Despite the above limitations, there were several strengths. First, when the EHDI program chose to use a survey to collect audiologists' perception, the response rate was generally high; only seven EHDI programs received less than 40% returned surveys. Secondly, there was high degree of convergence in the qualitative data regarding key themes and issues encountered by audiologists from 39 states, in addition to convergence of findings with the Chung, Beauchaine, Grimes, et al. (2017) study. Although the reporting system varies across each state, the barriers and challenges encountered by audiologists were similar across states; we did not encounter any barrier that was unique to only one state. Independent data coding by each author and repeatedly comparing and resolving differences before moving to the next stage of data analysis was used to help improve the consistency in data interpretation and analysis.

Conclusion

Audiologists described barriers to reporting results. Even though the reporting system varies across each state, the identified barriers were similar across states. A non-user-friendly design was the major challenge reported by participating audiologists. In addition, audiologists noted in their survey response that reporting hearing results was not a direct patient care task; it was, instead, perceived as labor-intensive and a duplication of effort. In a busy clinical environment, many audiologists found prioritizing public health reporting of hearing assessment data difficult. In

addition, parents often sought second opinions by visiting more than one clinic. Audiologists from different clinics did not routinely communicate with each other. When parents told the audiologist that their child was previously seen by another audiologist from another clinic, some audiologists assumed the hearing results had already been reported. Furthermore, some audiologists were also unaware of the procedures to report hearing assessment results in their state. Assumptions and lack of awareness could be remedied by training, as well as clarifying when and how to report results. Due to the wide spectrum of barriers, a multi-pronged improvement strategy that includes soliciting audiologist feedback for improving the online reporting portal, working with audiologists to address identified reporting barriers, and providing additional training to audiologists may be helpful for state EHDI programs looking to improve their reporting process.

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Exploring Speech and Language Intervention for Preschoolers who are Deaf and Hard of Hearing: A Scoping Review

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Abstract

Purpose: The objective of this study was to summarize the extant literature on the effectiveness of speech and spoken language interventions for young children who are deaf or hard of hearing (DHH) to determine which types of speech-language interventions might be most effective, for which hearing levels and types of hearing losses, and at which dosage.

Methods: Using a scoping review methodology, a database search identified 10,360 studies of which 16 met the requirements for inclusion. Data was extracted from each for analysis.

Results: Due to the limited number of studies available, high variability in the nature of the studies, and insufficient details about the interventions and sample in many of the papers, fully addressing the study objectives was difficult. However, common themes included the positive effect of caregiver-centered approaches on language outcomes, the equal effectiveness of virtual versus in person intervention, the addition of other speech and language intervention techniques to Auditory-Verbal Therapy may improve outcomes, and the effect of speech and language therapy on auditory skills is unclear.

Conclusions: This scoping review offers an initial step in analyzing and implementing evidence-based speech and language treatment protocols for children who are DHH.

Keywords: Early intervention; auditory-verbal therapy; auditory-oral therapy; scoping review

Acronyms: AVT = Auditory-Verbal Therapy; AVTs = auditory-verbal therapists; BAHAs = bone anchored hearing aids; CI = cochlear implant; DHH = deaf or hard of hearing; HA = hearing aid; MLU = mean length utterance; PCIT = Parent-Child Interaction Therapy; SLPs = speech-language pathologists; ToDs = teachers of the deaf

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Between 0.5 and 5 in 1,000 children are born deaf or hard of hearing (DHH) in high income countries and that number increases in low- and middle-income countries (World Health Organization, 2010). Hearing loss can have negative effects on speech and language development, academic outcomes, and socioemotional skills (Carney & Moeller, 1998; Geers et al., 2009; Hintermair, 2006; Qi & Mitchell, 2012). To meet the developmental needs of these children, the Joint Committee on Infant Hearing (JCIH) recommends access to universal newborn hearing screenings by one month of age and immediate provision of optimal hearing technology (JCIH, 2019). Children identified with hearing loss at a very early age typically have better communication outcomes compared to peers identified later (Nelson, 2008) as do children who receive their hearing technology earlier (Ching, 2015).

The JCIH also stipulates that language intervention by six months of age is vital for children who are DHH to meet their highest communication potential (JCIH, 2019). They recommend family-centered, culturally responsive, unbiased, developmental, inclusive, accessible, and

naturalistic communication intervention for all children who are DHH provided by knowledgeable and well-trained clinicians (JCIH, 2019). For children who are DHH and learning a spoken language, one way of monitoring the capabilities of clinicians is through Nanette Thompson's *Listening and Language Self-Checklist for Colorado Home Intervention Program (CHIP) Facilitators*, which is presented in JCIH's 2013 Supplement (Muse et al., 2013). It lays out specific techniques that clinicians should use during spoken language intervention with children who are DHH to ensure fidelity of implementation. These include developing listening skills by checking for consistent listening ability, incorporating music and nursery rhymes, maximizing the home listening environment, and holding high expectations for listening in a variety of activities and settings. Thompson also provides recommendations for language development such as including literacy activities in sessions, modeling and expanding child language, rewarding communication attempts, and developing spoken language through audition. Speech sound techniques include expecting, eliciting, and encouraging verbal responses; using acoustic highlighting

techniques; and noting speech errors. Specific strategies for spoken language development cited by the JCIH (Muse et al., 2013) include informing caregivers of the session objectives, scaffolding techniques, pause time, incorporating intervention strategies into daily life, communicating with all of the professionals supporting the family, and ensuring that the family leaves each session with a feeling of empowerment (Muse et al., 2013). Although the JCIH concludes that well-trained, competent clinicians can meet the needs of families of children who are DHH by monitoring their use of these strategies, they do concede that no literature exists linking fidelity of implementation of these strategies with children who are DHH and successful outcomes (Muse et al., 2013).

The language intervention literature investigating communication in children who are DHH primarily focuses on communication modality (Geers et al., 2017; Thomas & Zwolan, 2019), often to great debate (Napoli et al., 2015). Communication options for children who are DHH are on a spectrum from primarily manual, in which families communicate solely in a sign language, like American Sign Language, to Auditory-Verbal Therapy (AVT; Ganek et al., 2012). AVT follows 10 principles that support caregivers who are teaching their children to listen and talk through audition alone (AG Bell Academy for Listening and Spoken Language, 2007). Clinicians can become certified in AVT through an intense three-year training program. More than 90% of children who are DHH are born to families with typical hearing who do not use sign language as their family communication (Mitchell & Karchmer, 2004). In combination with newborn hearing screenings and early access to audition with modern hearing technology, 90% of them choose the listening and spoken language side of the communication spectrum (Fitzpatrick et al., 2013).

AVT, however, is not the only method of spoken language communication intervention available. Other listening and spoken language options include auditory-oral intervention, in which listening and spoken language is the goal but visual and tactile cues may be incorporated during language learning, and cued speech, a system of hand gestures used to augment lip reading. In addition, these methods can be used in combination, as can forms of speech-language intervention that were not specifically designed for children who are DHH, such as drilling, which is effective for children with developmental language delay regardless of hearing status (Shriberg & Kwiatkowski, 1982). It is very rare, however, that these modalities are investigated against one another in high quality randomized control studies (Eriks-Brophy et al., 2020). Although AVT is governed by distinct principles of practice that oversee the consistency of the treatment across clinicians (AG Bell Academy for Listening and Spoken Language, 2007), other speech-language treatment approaches used with children who are DHH typically do not have prescribed protocols that can be precisely implemented by clinicians in the field.

The present study was a scoping review of research on speech and spoken language interventions for preschoolers who are DHH. We aimed to evaluate whether the evidence supports the effectiveness of speech and spoken language interventions for children who are DHH (and if so, for whom), to determine whether certain speech and spoken language interventions led to better outcomes than others, and to identify essential ingredients for the most effective interventions for children who are DHH. We hypothesized that (a) speech and language interventions would positively affect the communication outcomes of children who are DHH, (b) different speech and language intervention protocols would differentially affect the communication outcomes of children who were DHH, and (c) intervention effectiveness would be influenced by hearing status and dosage.
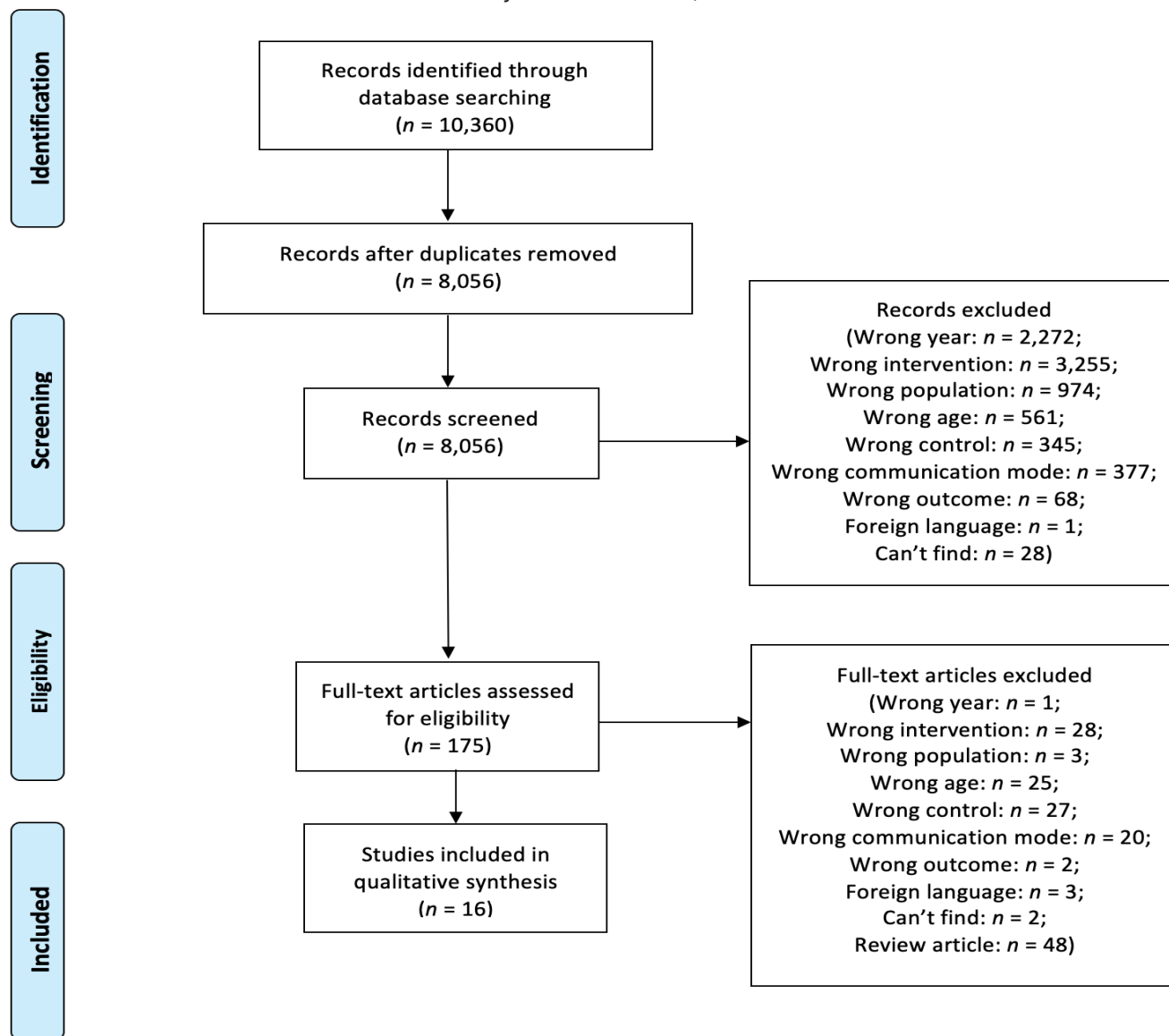
Method

We conducted a systematic search of the literature using seven databases: CINHL, Education, EMBASE, MEDLINE, Nursing & Allied Health, PsychInfo, and SCOPUS. The search was comprised of publications from before June 2021. Search terms appear in Table 1. Broad search terms were chosen to ensure capture of all speech and language related intervention studies for children who are DHH.

Table 1
Search Terms

Participants	Intervention	Hearing loss
Preschooler/s	Speech therapy	Hearing loss
Toddler/s	Language therapy	Hearing impairment/ed
Baby/s	Aural re/habilitation	Hearing disorder/s
Infant/s	Deaf education	Deaf/ness
Newborn		Auditory neuropathy spectrum disorder/ANSD
Child/Children		

As shown in Figure 1, studies were excluded if they were published prior to 2002. This review was initiated as part of a quality improvement project for the Ontario Infant Hearing Program, which implemented its provincial newborn hearing screening program in 2002 (Hyde et al., 2004). Children identified before this period had fundamentally different intervention needs (Yoshinaga-Itano, 2003). Studies were also removed if participants were over five years old and/or did not have a hearing loss. In addition, studies that investigated interventions focused on manual communication methods, did not have an appropriate control group (e.g., a control group with hearing loss), and/or measured outcomes that were not directly related to the child (e.g., caregiver perceptions of intervention) were excluded. Review studies were also excluded. Finally, studies were not included if they did not report an intervention or the intervention was not speech and language related (e.g., provision of a cochlear implant). Papers were excluded if they were not published in English or French or were unavailable through the Western University library service or other online resources.

Figure 1
Included and Excluded Studies Presented in the Style of Moher et al., 2009

Identification

Records identified through database searching
(*n* = 10,360)

Records after duplicates removed
(*n* = 8,056)

Screening

Records screened
(*n* = 8,056)

Records excluded
(Wrong year: *n* = 2,272;
Wrong intervention: *n* = 3,255;
Wrong population: *n* = 974;
Wrong age: *n* = 561;
Wrong control: *n* = 345;
Wrong communication mode: *n* = 377;
Wrong outcome: *n* = 68;
Foreign language: *n* = 1;
Can't find: *n* = 28)

Eligibility

Full-text articles assessed
for eligibility
(*n* = 175)

Full-text articles excluded
(Wrong year: *n* = 1;
Wrong intervention: *n* = 28;
Wrong population: *n* = 3;
Wrong age: *n* = 25;
Wrong control: *n* = 27;
Wrong communication mode: *n* = 20;
Wrong outcome: *n* = 2;
Foreign language: *n* = 3;
Can't find: *n* = 2;
Review article: *n* = 48)

Included

Studies included in
qualitative synthesis
(*n* = 16)

The first author screened all identified records by title and abstract. A second coder independently made judgements based on title and abstract for 10% of the identified studies. Point-by-point comparison was conducted to determine interrater reliability. The first author then reviewed the remaining full-text articles for inclusion. For each included study, the authors agreed to collect information in the following categories: (a) study information (author, year, title, design, journal), (b) participant characteristics (sample size, age, gender, socioeconomic status, language of intervention, culture/race), (c) hearing status (hearing level, hearing type, hearing technology), and (d) intervention (treatment type, service provider, length, dosage, outcome measure, outcome). Effect size was also collected from each study and was calculated manually when not provided. All effect sizes were converted to Cohen's *d* for comparability when reported by a different measure. Effect sizes of *d* = .2 were considered small, *d* = .5 were considered moderate, *d* = .8 large, and *d* = 1.2 very large (Sawilowsky, 2009).

Results

After removing duplicates, 8,056 articles were identified in our search. Sixteen papers met our inclusion criteria and were included in this analysis. They are listed in the reference section of this paper with an asterisk. There was 95% agreement between coders. Two of the 16 studies were randomized control trials (Monshizadeh et al., 2019; Zamani et al., 2016), one was a retrospective nested case-control study (Moog & Geers, 2010), six were prospective cohort studies (Behl et al., 2017; Brooks, 2017; Costa et al., 2019; Nanjundaswamy et al., 2017; Talebi et al., 2015; Zhou et al., 2013), and the remaining seven were retrospective cohort studies (Arumugam et al., 2021; Bunta et al., 2016; Chen & Liu, 2017; Constantinescu et al., 2014; Davidson et al., 2021; Percy-Smith et al., 2018; Yanbay et al., 2014).

Participant Demographics

The intervention studies reviewed here were relatively diverse, representing programs from seven countries on four continents (United States [6], Iran [3], Australia

[2], India [2], China [1], Denmark [1], & Taiwan [1]) and seven languages (English, Danish, Kannada, Mandarin, Persian, Spanish, & Tamil). Only one study (Costa et al., 2019) reported demographic information related to culture or race. Nine of the studies reported maternal education as a proxy for socioeconomic status (SES; Arumugam et al., 2021; Behl et al., 2017; Bunta et al., 2016; Chen & Liu, 2017; Costa et al., 2019; Davidson et al., 2021; Monshizadeh et al., 2019; Percy-Smith et al., 2018; Yanbay et al., 2014). Five studies reported a range of maternal education from less than high school to a graduate degree (Bunta et al., 2016; Davidson et al., 2021; Monshizadeh et al., 2019; Percy-Smith et al., 2018; Yanbay et al., 2014), three reported that all caregivers had at least some post-secondary education (Behl et al., 2017; Chen & Liu, 2017; Costa et al., 2019), and one reported that all participants were from a low socio-economic group (Arumugam et al., 2021).

Demographic information related to the study participants can be found in Table 2. On average, studies included 22 (SD = 24) experimental participants and 20 (SD = 21)

controls after removing one outlier with 702 experimental participants and 302 controls (Arumugam et al., 2021). Of the ten papers that reported participant sex, 51% (SD = 13%) of children in the experimental groups and 48% (SD = 11%) in the control groups were female. Children were between 10 and 72 months old when they participated in the studies.

Participant Hearing Status

Participants' hearing status appears in Table 3. Three studies did not report hearing level and 62% (n = 8) of those that did included children with a range of levels of hearing loss from mild to profound. The remaining studies (n = 5) included participants with only severe or profound hearing losses. Nine studies provided information on type of hearing loss. Of them, 56% (n = 5) reported that all participants had bilateral hearing loss while 11% (n = 1) reported a mix of bilateral and unilateral hearing loss, including atresia. Twenty-two percent (n = 2) of the studies explicitly stated that participants had sensorineural hearing loss. Another 11% (n = 1) of the studies included only participants with congenital hearing loss, 11% (n = 1)

Table 2
Participant Demographics

Study	N		Gender (Female)		Age (months)	
	Experimental	**Control**	**Experimental**	**Control**	**Experimental**	**Control**
Arumugam et al., (2021)	702	302	NA	NA	NA	NA
Behl et al. (2017)	23	25	NA	NA	20.2	19
Brooks (2017)	5	8	NA	NA	10–23	NA
Bunta et al. (2016)	10	10	5	5	55.3 (SD = 13.2)	55.6 (SD = 20.4)
Chen & Liu (2017)	5	5	4	2	60.6 (SD = 6.46)	58.2 (SD = 6.11)
Constantinescu et al. (2014)	7	7	3	4	29.4 (SD = 2.9)	29.16 (SD = 3.4)
Costa et al. (2019	15	12	9	9	51 (Mdn = 48)	49.5 (Mdn = 49)
Davidson et al. (2021)	32	16	11	10	42.8 (SD = 8.3)	66.8 (SD = 16.8)
Monshizadeh et al. (2019)	26	25	11	9	20–24	20–24
Moog & Geers (2010)	107	27	NA	NA	60–72	60–72
Nanjudaswamy et al. (2017)	10	10	5	3	45.6	44.4
Percy-Smith et al. (2018)	31	94	14	52	Mdn = 47	Mdn = 49
Talebi et al. (2015)	15	15	7	7	48–72	48–72
Yanbay et al. (2014)	14	14	8	7	50.52 (SD = 14.16)	56.76 (SD = 15.78)
Zamani et al. (2016)	33	33	NA	NA	29.06 (SD = 4.18)	28.78 (SD = 3.42)
Zhou et al. (2013)	19	15	NA	NA	14.8 (SD = 2.85)	13.95 (SD = 2.98)

Note. NA = Not Available; *SD* = Standard Deviation, *Mdn* = median.

Table 3

Participants' Hearing Status

Study	Hearing Level		Hearing Type		Hearing Technology	
	Experimental	**Control**	**Experimental**	**Control**	**Experimental**	**Control**
Arumugam et al. (2021)	Profound	Profound	NA	NA	702 CI	302 CI
Behl et al. (2017)	5 unilateral/atresia/bilateral mild; 3 mild-moderate; 2 moderate; 6 moderate-severe; 1 severe-profound; 6 profound	5 unilateral/atresia/bilateral mild; 4 mild-moderate; 3 moderate; 4 moderate-severe; 2 severe; 6 profound	Bilateral	Bilateral	2 unilateral CI; 12 bilateral CI	4 unilateral CI; 10 bilateral CI
Brooks (2017)	NA	NA	NA	NA	NA	NA
Bunta et al. (2016)	1 moderate; 2 moderate-severe; 1 severe; 1 severe-profound; 5 profound	1 mild; 2 severe; 7 profound	NA	NA	2 bilateral HA; 2 bilateral CI; 5 bimodal	3 bilateral HA; 2 bilateral CI; 5 bimodal
Chen & Liu (2017)	Mild to profound	Mild to profound	Bilateral	Bilateral	4 bilateral HA; 1 Bimodal	4 bilateral HA; 1 Bimodal
Constantinescu et al. (2014)	Mild-moderate to severe-profound	Mild-moderate to severe-profound	Bilatera	Bilateral	2 unilateral BAHA; 4 bilateral HA; 1 bilateral CI	6 bilateral HA; 1 bilateral CI
Costa et al. (2019)	Mild to profound	Mild to profound	5 congenital; 1 post-natal; 9 unknown	4 congenital, 1 post-natal, & 7 unknown	5 bilateral HA; 1 unilateral HA; 6 bilateral CI; 1 unilateral CI; 2 bimodal	6 HA & 5 CI
Davidson et al. (2021)	Mild to profound	Mild to profound	NA	NA	NA	NA
Monshizadeh et al. (2019)	NA	NA	NA	NA	NA	NA
Moog & Geers (2010)	Profound	Profound	NA	NA	4 bilateral CI; 104 unilateral CI	4 bilateral CI; 23 unilateral CI
Nanjudaswamy et al. (2017)	Moderately severe to profound	Severe to profound	Prelingual sensorineural bilateral	Prelingual sensorineural bilateral	10 bilateral HA	10 bilateral HA
Percy-Smith et al. (2018)	NA	NA	30 congenital; 1 other	85 congenital; 9 other	30 bilateral cochlear implants; 1 bimodal	78 bilateral CI; 16 unilateral CI
Talebi et al. (2015)	Moderate to severe	Moderate to severe	Bilateral sensorineural	Bilateral sensorineural	15 bilateral HA	15 bilateral HA
Yanbay et al. (2014)	Profound	Profound	Bilateral	Bilateral	2 unilateral CI; 12 bilateral CI	4 unilateral CI; 10 bilateral CI
Zamani et al. (2016)	Severe	Severe	NA	NA	33 HA	33 HA
Zhou et al. (2013)	Profound	Profound	Congenital	Congenital	19 CI	15 CI

Note. NA = not available; CI = cochlear implant; HA = hearing aid; BAHA = bone anchored hearing aid.

reported all participants had pre-lingual hearing loss, and 22% (*n* = 2) reported a mix of congenital, post-natal, and unknown etiologies.

Thirteen studies (81%) reported their participants' hearing technology. Of the experimental participants, 39% of the participants (*n* = 109) wore a unilateral cochlear implant, 31% (*n* = 86) wore bilateral cochlear implants, 26% (*n* = 73) wore bilateral hearing aids, and 3% (*n* = 9) wore bimodal hearing technology. The remaining 1% is comprised of two experimental participants who wore bone anchored hearing aids (BAHAs) and one who wore a unilateral hearing aid. Within the control groups, 49% (*n* = 125) wore bilateral cochlear implants, 30% (*n* = 77) wore bilateral hearing aids, 18% (*n* = 47) wore a unilateral cochlear implant, and the remainder were bimodal (*n* = 6).

Davidson et al. (2021) reported that 15 participants wore bilateral cochlear implants, 12 wore two hearing aids, 11 were bimodal, 3 wore BAHAs, 2 wore a hearing aid with an FM System, and 1 wore a bilateral contralateral routing of signal device. They did not, however, distinguish participants' device use by control or experimental group. In addition, all 1,004 of the participants in Arumugam et al. (2021) used cochlear implants, although the authors do not report if they were uni- or bilateral.

Interventions and Study Measures

Information related to the intervention programs investigated in each study is reported in Table 4. Each intervention is listed as described by the authors of the paper. Thirty-one percent of the studies reported these programs were provided by a combination of auditory-verbal therapists,

speech-language pathologists, audiologists, and teachers of the deaf. Nineteen percent were provided by auditory-verbal therapists alone, and 13% by speech-language pathologists alone. Psychologists implemented intervention in one study. One study investigated treatment provided by a software program monitored by an audiologist. Three studies in this group did not report who provided the service and one reported trained habilitationists implemented intervention. Seven of the studies confirmed that the professionals providing intervention were certified in their roles or specially trained to work with children who are DHH (Arumugam et al., 2021; Brooks, 2017; Bunta et al., 2016; Costa et al., 2019; Davidson et al., 2021; Percy-Smith et al., 2018; Yanbay et al., 2014). The remainder either did not describe clinician training or asked the professionals to self-

Table 4

Speech & Language Protocols

Study	Experimental	Control	Experimental	Control	Experimental	Control
Arumugam et al. (2021)	A standard structured set of exercises to build understanding and recognition of a sound signal conducted at a satellite center	A standard structured set of exercises to build understanding and recognition of a sound signal conducted at a cochlear implant clinic	12 months	Trained habilitationists	Speech perception; Speech intelligibility	NA
Behl et al. (2017)	Parent-focused intervention that incorporated daily routines via Telepractice	Parent-focused intervention that incorporated daily routines via In-person intervention	55 minutes sessions, 1x per week for 6 months	AVTs, ToDs, & SLPs	Receptive & Expressive Language; Vocabulary; Auditory Skills	Receptive: d = .3; Expressive: d = .17; Total: d = .26; Vocabulary: d = .01; Auditory Skills: d = .12
Brooks (2017)	Real-time Embedded coaching with the Application of Andragogical Principles	Auditory-oral	20–45 minute sessions 2x per month for 6 months	AVTs & ToDs	Vocabulary	NA
Bunta et al. (2016)	Bilingual AVT	Monolingual AVT	25 minute sessions, 2–3x per week for 29.8 (SD = 12.5) months	AVTs & ToDs	Receptive & Expressive Language	Receptive: d = .97; Expressive: d = 1.7; Total: d = 1.4
Chen & Liu (2017)	AVT via telepractice	AVT via in-person intervention	50.6 (SD = 2.64) months	NA	Receptive & Expressive Language	Receptive: d = .23[-1.46,1.03]; Expressive: d = .12[-1.98,.59]
Constantinescu et al. (2014)	AVT via telepractice	AVT via in-person intervention	1 hour sessions, 2x per month for 2 years	AVTs	Receptive & Expressive Language	Receptive: d = .5[-.57,1.56]; Expressive: d = 1.19[.02,2.32]; Total: d = .83[-.28,1.9]
Costa et al. (2019)	Parent-Child Interaction Therapy (PCIT)	Push-in & individual language services	1x per week for 16.2 (Mdn = 16) weeks	Psychologists	Vocabulary; mean length utterance (MLU); Negative Behaviors	Vocabulary: d = .74; MLU: d = 1.5; Negative Behaviors: d = 2.5
Davidson et al. (2021)	Confirmation of hearing loss, monitoring of hearing thresholds, provision of hearing devices, and instruction for families related to hearing loss and language acquisition before 3 years old.	Confirmation of hearing loss, monitoring of hearing thresholds, provision of hearing devices, and instruction for families related to hearing loss and language acquisition after 3 years old.	22 months (range = 3 to 34 months)	Audiologists, ToDs, & SLPs	Language; Receptive & Expressive Vocabulary	Language: d = 1.3[.71,2.0]; Receptive Vocabulary: NA; Expressive Vocabulary: d = 1.2[.54,1.83]

Table 4 (continued)

Speech & Language Protocols

Study	Experimental	Control	Experimental	Control	Experimental	Control
Monshizadeh et al. (2019)	An Education Package on Receptive Vocabulary Development for Persian Speaking Cochlear Implant Children	AVT	9–12 months	NA	Receptive & Expressive Language	Receptive: $d = 2.02[1.33,2.69]$; Expressive: $d = 1.26[.65,1.85]$; Total: $d = 1.78[1.12,2.42]$
Moog & Geers (2010)	Parent-infant program	Listening & Spoken Language (LSL) or mainstream classrooms	5 years	AVTs, ToDs, & SLPs	Receptive & Expressive Language; Vocabulary	NA
Nanjudaswamy et al. (2017)	Auditory training software	NA	45 minute sessions, 3x per week for 1 month	Computer software with audiologist support	Auditory Skills	NA
Percy-Smith et al. (2018)	AVT	Speech-language therapy not specialized for children who are DHH	1x per week/ month/quarter for 3 years	AVTs	Language; Vocabulary; Speech	Language: $d = 1.25[.64,1.85]$; Vocabulary: $d = 1.11[.55,1.68]$; Speech: $d = .59[.05,1.13]$
Talebi et al. (2015)	Traditional rehabilitation for children who are DHH & vowel training	Traditional rehabilitation for children who are DHH	2 hour sessions, 2x per week for 6 months	NA	Vowel identification; Reaction time	Identification!: /æ/: $d = 2.71[1.69,3.70]$, /u/: $d = 2.49[1.51,3.44]$; Reaction time!: /æ/: $d = 3.38[2.24,4.51]$, /e/: $d = 2.67[1.66,3.66]$, /u/: $d = 1.21[.42,1.99]$
Yanbay et al. (2014)	AVT	Auditory-Oral	Weekly or monthly for 4.05 ($SD = 1.18$) years	AVTs	Receptive & Expressive Language; Vocabulary	Receptive: $d = .05[-.05,.69]$; Expressive: $d = .12[-.62,.86]$; Vocabulary: $d = .15[-.89,.59]$
Zamani et al. (2016)	AVT with gestures	AVT	1 hour sessions, 1x per week for 15 weeks	SLPs	Receptive & Expressive Language	Receptive: $d = 1.64[1.08,2.19]$; Expressive: $d = 1.9[1.31,2.48]$
Zhou et al. (2013)	Speech-language pathology with a focus on developmentally appropriate auditory, speech, and language skills	No treatment	2–3x per week for 6–12 months	SLPs	Speech perception; Speech intelligibility	NA

Note. AVTs = auditory-verbal therapists; DHH = deaf or hard of hearing; ToDs = teachers of the deaf; SLPs = speech-language pathologists; NA = not available; AVT = auditory verbal therapy.

!Effect sizes reported for vowels the authors identified as significant.

identify their role. Treatment duration and frequency varied widely across studies. Interventions were provided from 15 weeks to 60 months and children attended treatment sessions once a quarter to three times a week for between 25 and 120 minutes.

The interventions reported by the reviewed studies included measures of language (10 studies), vocabulary (6 studies), and auditory skills (5 studies). Five studies reported more than one outcome measure (Behl et al., 2017; Davidson et al., 2021; Moog & Geers, 2010; Percy-Smith et al., 2018; Yanbay et al., 2014). One study

(Percy-Smith et al., 2018) also reported speech outcomes. Language results included receptive, expressive, and total language scores on standardized assessments. Vocabulary outcomes were also assessed using standardized assessments. Auditory skills were measured via speech perception testing, functional assessment tools, and auditory identification tasks.

Language Outcomes

Receptive and Expressive Language

Four of the studies reviewed here reported retrospective language outcomes for groups of children who received

different types of intervention specialized for children who are DHH. Davidson et al. (2021) reported that children who received listening and spoken language intervention services before three years of age had significantly higher language outcomes than those who received intervention later ($d = 1.3$ [.71,2.0]). Moog and Geers (2010) found that young children who received one-on-one intervention with a clinician and a caregiver had higher receptive and expressive language scores than peers in mainstream and specialized classrooms. As the children grew, however, more benefit was seen in the classroom environments. The paper did not report the necessary data to calculate effect size. Percy-Smith et al. (2018) and Yanbay et al. (2014) both investigated AVT. Percy-Smith et al. (2018) compared children in AVT to those who received an intervention that was "not specifically targeted" (p. 40) at children who were DHH. Participants in this non-AVT group were recruited from across Denmark and did not receive a consistent treatment protocol. Sixty-six percent of caregivers in the non-AVT group reported that they did not participate in therapy sessions, whereas 100% of the caregivers in the AVT group did. AVT had a very large effect on language ($d = 1.25$ [.64,1.85]), a large effect on vocabulary ($d = 1.11$ [.55,1.68]), and a moderate effect on speech outcomes ($d = .59$ [.05,1.13]) relative to the non-AVT intervention. Yanbay et al. (2014) compared the language outcomes of children in AVT to those receiving auditory-oral therapy. In this study, caregivers were included in both interventions. Yanbay et al. (2014) found no significant effect of intervention type on language outcomes (Receptive: $d = .05$ [-.05,.69]; Expressive: $d = .12$ [-.62,.86]) or vocabulary outcomes ($d = .15$ [-.89,.59]), and the size of the effects can be considered trivial because the confidence intervals include zero.

The principles of AVT state that intervention techniques should be integrated into daily activities through audition alone (AG Bell Academy for Listening and Spoken Language, 2007). These principles encourage listening and spoken language strategies be integrated into activities of daily living and that hearing be the primary sensory modality for language learning, rather than drill activities and visual cues. Two studies reviewed here, however, integrated these strategies in AVT. Zamani et al. (2016) added gestures when teaching verbs while Monshizadeh et al. (2019) added a vocabulary drilling activity to AVT. In both cases, they found very large and significant positive effects on receptive and expressive language compared to children receiving standard AVT (Monsizadeh et al., 2019: Receptive: $d = 2.02$ [1.33,2.69]; Expressive: $d = 1.26$ [.65,1.85]; Total: $d = 1.78$ [1.12,2.42]; Zamani et al., 2016: Receptive: $d = 1.64$ [1.08,2.19]; Expressive: $d = 1.9$ [1.31,2.48]). A third study by Bunta et al. (2016) found that providing bilingual AVT to bilingual families had a large effect on receptive language ($d = .97$) and a very large effect on expressive language ($d = 1.7$; Total Language: $d = 1.4$) relative to providing monolingual AVT to bilingual families.

Three studies investigated the use of telepractice to provide speech and language intervention to children who are DHH. Constantinescu et al. (2014) and Chen and

Liu (2017) found no significant differences in receptive language outcomes between AVT provided via telepractice relative to in-person AVT (Chen & Liu, 2017: $d = .23$ [-1.46,1.03]; Constantinescu et al., 2014: $d = .5$ [-.57,1.56]). Constantinescu et al. (2014) did find a large effect of telepractice compared to in-person AVT for expressive language ($d = 1.19$ [.02,2.32]) but Chen and Liu (2017) did not ($d = .12$ [-1.98,.59]). Behl et al. (2017) compared parent-focused intervention that incorporated daily routines and was provided via telepractice to a similar intervention provided in-person. They found a small effect in favor of telepractice over in-person intervention on receptive language ($d = .3$), but negligible effects for expressive language ($d = .17$) and vocabulary skills ($d = .01$).

Vocabulary

Three studies reported vocabulary measures as primary outcomes. Davidson et al. (2021) found that children who entered early intervention before three-years old had significantly higher receptive (effect size could not be calculated) and expressive ($d = 1.2$ [.54,1.83]) vocabulary scores than their peers who entered rehabilitation later. Brooks (2017) compared children whose caregivers were receiving real-time embedded coaching with the application of andragogical principles (i.e., principles of adult learning) to those receiving auditory-oral intervention. The amount and type of caregiver engagement in the auditory-oral intervention group was not clearly stated. Brooks reported over the course of 6 months of intervention, children in both groups showed increases in their receptive vocabulary age equivalents ranging from 2 to 11 months while the real-time coaching group improved their expressive vocabulary by 5 to 7 months and the auditory-oral group improved 2 to 6 months. However, data and analysis were not provided to calculate statistical significance or effect size, and the reporting of only age equivalent data limits interpretation. Costa et al. (2019) also implemented a caregiver coaching protocol, Parent-Child Interaction Therapy (PCIT). Designed as a method for reducing negative behaviors, rather than a language intervention, PCIT includes aspects of both play therapy and caregiver coaching focused on behavior management techniques. The children receiving PCIT were compared to children in a reverse inclusion classroom who also received individualized speech-language therapy. The authors found moderate and very large positive effects of PCIT on vocabulary outcomes ($d = .74$) and mean length utterance (MLU; $d = 1.5$), respectively, relative to the control intervention.

Auditory skills

Five studies measured auditory skills post-speech and language intervention. In one of the only studies reviewed here to compare an intervention group to a no-treatment group, Zhou et al. (2013) measured speech perception and speech intelligibility in children who received a cochlear implant and speech therapy, "with an emphasis on auditory training, speech orthodontic treatment, articulation training, and language training according to the child's performance" (p. 2), compared to those who had

only received a cochlear implant. No significant differences were found between the groups. Insufficient data was reported to calculate effect size. Arumugam et al. (2021) compared "a structured set of exercises designed to help the cochlear implant user to understand and recognize the sound signal" (p. 1) conducted in the primary cochlear implant clinic versus in satellite locations throughout the state. Like Zhou et al. (2013), Arumugam et al. (2021) found no significant differences in speech perception or speech intelligibility scores between groups and insufficient data was reported to calculate effect size.

Talebi et al. (2015) investigated a group of children receiving a "traditional rehabilitation program for their disability" (p. 15). Half of the participants also received vowel training in which six vowels were presented without visual cues in nonsense syllables with voiceless consonants. Participants were asked to verbally identify each syllable. They found that adding vowel training to "traditional rehabilitation" led to large improvements in speed and accuracy of vowel identification in half of the vowels. (Identification: /æ/: d = 2.71[1.69,3.70], /u/: d = 2.49[1.51,3.44]; Reaction time: /æ/: d = 3.38[2.24,4.51], /e/: d = 2.67[1.66,3.66], /u/: d = 1.21[.42,1.99]). There was no difference between the groups on the other vowels. Nanjundaswamy et al. (2017) designed an auditory training software program that caregivers used with their children. Their results on functional assessments were compared to a matched control group, but it was not clearly stated whether the control group received any form of language intervention. The children who received the computerized intervention made significantly greater improvements in parent report of listening skills in real word situations as measured by the Infant-Toddler Meaningful Auditory Integration Scale (IT-MAIS; Zimmerman-Phillips et al., 2001) but similar changes in hearing and communicating with others as measured by the Parents' Evaluation of Aural/Oral Performance of Children (PEACH; Ching & Hill, 2007) compared to peers who were not enrolled in the experimental intervention. Information to calculate effect size was not provided. Behl et al. (2017) measured auditory skills using a caregiver checklist with children receiving intervention (described previously) via telepractice versus in-person therapy and found no significant differences and negligible effects between the two modes of delivery on auditory skills (d = .12).

Discussion

This scoping review was conducted to summarize the extant literature on the effectiveness of speech and spoken language interventions for young children who are DHH. We not only wished to determine whether speech and language interventions have been shown to be effective, but which types might be most effective, for whom, and at which dosage. We identified 16 papers that investigated outcomes of speech and language interventions for children who are DHH. Two studies compared the presence versus absence of speech-language intervention. The remaining 14 studies compared two interventions to determine whether one led to better language and/or speech outcomes than the other. In

many cases, the papers described the control, and often the experimental, treatments in very broad terms. They referred to "traditional rehabilitation" (Talebi et al., 2015) or "speech-language therapy" (Percy-Smith et al., 2018) with little further explanation of what techniques and philosophies were employed. The studies also varied widely in the sample characteristics, including hearing status, of the children and in the duration and frequency of the interventions. As a result, determining essential ingredients for the most effective interventions for which children and at which dosage based on the scientific literature is, therefore, difficult. Nonetheless, a variety of themes did emerge that can inform future clinical research to support optimal spoken language outcomes for children who are DHH.

Caregiver-Centered Approaches May Positively Affect Outcomes

None of the studies reviewed here explicitly controlled for caregiver involvement in treatment. However, methodologies that specifically included caregiver-centered techniques positively affected language and vocabulary outcomes in children who are DHH compared to those in which caregiver participation was not overtly stated. AVT, which incorporates caregivers throughout treatment, had no differential effect on language outcomes compared to auditory-oral intervention involving a caregiver (Yanbay et al., 2014) and produced a very large effect compared to children receiving intervention with inconsistent caregiver attendance (Percy-Smith et al., 2018). Two studies reported interventions built on methods for coaching caregivers. Although Brooks (2017) did not provide statistical analysis or sufficient data to calculate the magnitude of effect for real-time parent coaching with the application of andragogical principles, Costa's team (2019) showed that PCIT can have a moderate effect on vocabulary outcomes. Neither of these studies, however, clearly excluded less formal or other methods of caregiver coaching. Moog and Geers (2010) also found that in young children, parent-infant therapy sessions yielded significantly higher language scores than classroom environments, although, again, effect sizes could not be calculated. Overall, this pattern of results provides converging evidence to suggest that caregiver-centered intervention approaches may be particularly effective for developing the spoken language skills of young children who are DHH and should be further investigated.

Caregiver-centered interventions have successfully improved outcomes for patients within a variety of allied health fields (Lawler et al., 2013), including pediatric speech and language disorders. By training caregivers, children with speech and language delays (like those associated with hearing loss) have the opportunity to receive the high quality language input they need to learn to listen and talk (Roberts & Kaiser, 2011). In addition, caregivers likely know their children better than any professional could and may, therefore, be more successful at integrating language goals into the child's daily life in a meaningful and motivating manner.

Taking a caregiver-guided approach when serving children who are DHH is especially fitting when considering that children typically learn language by engaging with adults (Romeo et al., 2018). By instructing caregivers to use strategies that allow young children who are DHH to engage with age appropriate language stimulation, the children can capitalize on their critical period for language learning. Optimizing this developmental window, in which most children are learning to listen and talk, can allow children who are DHH to achieve listening and spoken language skills similar to their peers with typical hearing. Focusing on parent-child interactions alone, however, may miss some important features of language learning. Although most language acquisition research investigates parent-child talk, the influences of peer-to-peer verbal interactions may also play an important role in language learning. Studies conducted in non-industrialized countries have found children receive a large proportion of their language exposure from other children (Shneidman & Goldin-Meadow, 2012). Additionally, studies have identified pragmatic difficulties in children who are DHH and suggest the need to expand intervention to include peer-to-peer communication (Most et al., 2010). These findings indicate the potential importance of peer-to-peer talk in many societies and highlight how these types of interactions may also influence language development in high-income countries. None of the studies reviewed here investigated intervention methods that included other children, nor did they measure pragmatic skills development.

Virtual Delivery May Produce Similar Outcomes to In-Person Interventions

Three studies reported on the use of teleintervention compared to in-person therapy. Two investigated AVT (Chen & Liu, 2017; Constantinescu et al., 2014) and one described a more general methodology that included a caregiver-centered approach (Behl et al., 2017). In all three studies, there were no differences (and any effects on language, vocabulary, and auditory skills were negligible in magnitude) between the two modes of delivery, with the exception of Constantinescu's team (2014) who found virtual AVT had a large effect on expressive language outcomes relative to in-person AVT. This large effect in the context of the small sample size (7 participants per group) suggest that the study may have been underpowered. Nonetheless, no evidence was found to suggest that virtual delivery is inferior.

Given social distancing mandates put in place as a result of the COVID-19 pandemic, evidence suggesting that telepractice may be as effective as in-person intervention for preschoolers who are DHH is encouraging. Families requiring specialized speech and language services being able to access effective care remotely can ensure better equity and accessibility of intervention to more families, both in the pandemic context and beyond. Telepractice protocols presented here were designed for children under five years old and, therefore, required a substantial amount of caregiver involvement. Caregiver-centered approaches, such as those reviewed here, reduce the need for

the child who is DHH to listen and process potentially degraded auditory signals from computer speakers during teleintervention. The clinician instead instructs the caregiver not just through the logistics of running the telepractice software but also toy manipulation and high-quality language stimulation provision, and reports the child's response back to the clinician in real-time. Out of necessity, teleintervention may thereby inherently increase caregiver participation in intervention. More research is needed to confirm the outcomes of children who receive speech and language intervention via telepractice.

Adding Other Speech-Language Techniques Improved AVT Outcomes

In two studies, the authors modified AVT with techniques that are relatively common in other speech-language treatment approaches and compared those outcomes to traditional AVT. Modifications included the addition of gestures (Zamani et al., 2016) and vocabulary drills (Monshizadeh et al., 2019). Both modifications yielded large or very large positive effects for the modified AVT programs relative to AVT alone. The addition of gesture, as described by Zamani et al. (2016), clearly violates the principles of auditory-verbal practice, which mandate that audition be the child's primary sensory mode for language learning (Estabrooks et al., 2020). However, in combination with formal AVT, the addition of pantomimed gestures for verbs did significantly and positively affect language outcomes. Similarly, AVT advocates for language learning through daily activities integrated into all aspects of the child's life (Estabrooks et al., 2020) rather than formal didactic drilling as proposed in Monshizadeh et al. (2019). Once again, however, in combination with other AVT methods, their protocol produced large positive effect sizes.

It should be noted that Monshizadeh et al.'s (2016) treatment program was specific to Persian. AVT was developed in North America (Estabrooks et al., 2020) and was, therefore, modeled after the language socialization practices followed there. Given that both culture and SES have been linked to language development (Hart & Risley, 1995; Ochs & Schieffelin, 2016), future study into the impact of cultural adaptation of AVT is needed, although the diversity of the countries from which the included studies originated, as well as the variety of languages in which services were provided, is encouraging. Bunta and colleagues' (2016) investigation of the effect of bilingual AVT (English/Spanish) compared to AVT provided in the culturally dominate language alone (English), found large positive effects on expressive language when bilingual families were treated in both the majority language and their home language. This protocol aligns well with the AVT commitment to having caregivers serve as primary language models (Estabrooks et al., 2020) while, at the same time, incorporating cultural differences into intervention in an effective manner.

Auditory Skills Outcomes of Speech and Language Interventions Remain Unclear

Five studies reported auditory outcomes using a variety of methods including functional assessments, auditory identification tasks, and speech perception testing. In a teleintervention study, Behl et al. (2017) found that virtual intervention was as effective as in person intervention for parent rated auditory skills. Talebi et al. (2015) added vowel recognition training to traditional intervention and found large effects on recognition skills for three of six vowels. Nanjudaswamy et al. (2017) reported differences between a group of participants who received auditory training via a software program and a control group of children (who may or may not have been receiving other intervention) on one of two functional assessments of auditory skills. Zhou et al. (2013) found no difference in speech perception or intelligibility between children with cochlear implants who did versus did not receive speech and language intervention and Arumugam et al. (2021) reported that the speech perception and intelligibility outcomes of children who received intervention at a cochlear implant clinic were the same as those who attend services at satellite centres. Neither Zhou et al. (2013), Arumugam et al. (2021), nor Nanjudaswamy et al. (2017) provided sufficient information to calculate effect size. The minimal and inconsistent effects of the intervention protocols reviewed here indicate that the impact of speech and language treatment for auditory skills development remains unclear. Further exploration of techniques and strategies to improve listening abilities for children who are DHH is needed. Future studies should include clear descriptions of both the experimental and control treatment protocols as well as effect sizes.

Effect of Hearing Status Could Not be Evaluated

Half of the papers reviewed reported participants had a range of hearing levels and five reported participants with exclusively severe or profound hearing losses. Due to the variability within studies and the lack of variability between studies, the effect of specialized interventions on different hearing levels could not be conducted nor compared across studies. Mild and moderate hearing losses have been associated with delays in both expressive and receptive vocabulary (Tomblin et al., 2015). Future research should explore differences in intervention outcomes for these children compared to those with more profound hearing losses.

Hearing type was inconsistently reported in the reviewed papers. Six studies specified that participants had bilateral hearing loss, although more study participants could be assumed to have bilateral hearing loss by the reported use of bilateral hearing technology. Like mild hearing loss, unilateral hearing loss can also negatively affect language outcomes (Lieu et al., 2010). Future studies should identify the intervention needs of children with both unilateral and bilateral hearing losses, as well as those with permanent conductive versus sensorineural hearing losses.

Limitations and Future Directions

This scoping review faced a number of limitations. Studies that potentially fit inclusion criteria were excluded due to being published in languages other than those the authors read fluently. Thirty studies that potentially fit the inclusion criteria could not be accessed. Of the studies that were reviewed, many had inadequate reporting of demographic information. Five did not include effect sizes or the data required to calculate effect size and six were manually calculated. Future studies should include effect size within the analysis. With only two exceptions (Davidson et al., 2021; Zhou et al., 2013), the studies reviewed compared two treatment groups but did not additionally examine whether clinically meaningful improvements attributable to the intervention were observed in either group. Additionally, Davidson and colleagues (2021) did not control for age at amplification, which is highly correlated with age at intervention. Without disentangling these two variables, the role of language therapy in a child's outcomes cannot be clearly identified, even though a *no-treatment* group was employed. Future studies should include designs and analyses to facilitate the evaluation of change due to intervention.

With two exceptions (Monshizadeh et al., 2019; Zamani et al., 2016), the studies examining AVT were retrospective, which creates opportunities for confounding variables, association rather than causation, and poor population representation in samples. Although retrospective studies allow researchers to capitalize on participants who have been receiving treatment for many years, results must be interpreted with caution. By contrast, the studies of speech-language approaches other than AVT were primarily prospective, which yield more accurate results but may, in this case, lack the same ecological validity as the retrospective AVT studies.

Although AVT and some of the other interventions explicitly stated the use of a caregiver-centered approach, the speech-language approaches other than AVT typical of the control groups in many of the reviewed studies did not overtly state the role of caregivers in intervention. It is possible that these other approaches reported here were encouraging significant caregiver involvement. Future studies should provide more detailed descriptions of their control interventions. In addition, length and dosage of treatment ranged significantly across studies. No conclusions could be made related to amount of intervention necessary to affect communication outcomes. Future studies should explore this question further.

Speech-language pathologists and teachers of the deaf provided the bulk of the interventions. In most high-income countries, these positions both require a graduate degree or certificate indicating extensive professional training. Within hearing loss intervention, it is not unusual for speech-language pathologists and teachers of the deaf to provide similar early intervention services. Most of the AVT protocols were provided by auditory-verbal therapists. Certification as Listening and Spoken Language Specialist Certified Auditory-Verbal Therapist requires a minimum of

three years of intensive training after receiving a degree in some form of clinical communication disorders (AG Bell Academy for Listening and Spoken Language, 2017). Although clinicians who are not certified can follow the 10 principles of AVT, regardless of their level of training, those with certification are more likely to report implementing listening and spoken language strategies consistently in intervention compared to those who were not (Rosenzweig & Smolen, 2021). None of the studies reported the certification status of the auditory-verbal therapists and it is, therefore, possible that AVT was administered in an inconsistent manner between and within studies, making comparisons and replications challenging.

Conclusion

This scoping review explored specialized speech and language interventions for children who are DHH. The results of these studies were often unclear due to poor reporting of intervention techniques and effects sizes. Future studies might seek to better define speech-language therapy as well as how clinicians and families choose one methodology over another.

Emerging themes, however, suggest that caregiver-centered approaches, teleintervention, adding other speech and language intervention techniques to AVT, and the effect of speech and language therapy on auditory skills should be further considered within the context of speech and spoken language therapy for children who are DHH. Additionally, the effect of intervention on children with different levels and types of hearing loss could not be calculated due to within sample variability. Continued investigations of the effects of specialized interventions are necessary for children from a wider set of demographics with different hearing statuses to ensure that all children who are DHH are receiving the most effective and efficient intervention.

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World Hearing Day 2022 (3 March)

On World Hearing Day 2022, WHO will focus on the importance of safe listening as a means of maintaining good hearing across the life course. In 2021, WHO launched the World report on hearing that highlighted the increasing number of people living with and at risk of hearing loss. It highlighted noise control as one of the seven key H.E.A.R.I.N.G. interventions and stressed the importance of mitigating exposure to loud sounds.

The World Hearing Day 2022 with the theme "To hear for life, listen with care" will focus on the importance and means of hearing loss prevention through safe listening.

Theory of Mind Acquisition in Children who are Deaf: The Importance of Early Identification and Communication Access

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Abstract

Objectives: The aim of this study was to compare Theory of Mind (ToM) acquisition in preschool-age children with typical hearing (TH), and children who are deaf and have hearing parents (DHP) who received a cochlear implant by 18 months of age, to determine if early access to spoken language via a cochlear implant affected ToM acquisition.

Methods: Participants included 25 children with cochlear implants ages 3.0 to 6.5 years and 25 age-matched children with TH all of whom were enrolled in preschools with typical peer models. The test battery included measures of expressive and receptive language and ToM.

Results: There were no differences between children who are DHP and their peers with TH on language or ToM performance. Hearing age was significantly different; children who are DHP had been exposed to spoken language for less time than their hearing counterparts by approximately 12 months. Language skills were correlated with ToM after controlling for chronological age.

Discussion: Early cochlear implantation may ameliorate some of the deleterious effects of congenital, profound deafness on oral language development; this could positively influence the development of social cognition.

Conclusions: Children who are deaf who receive a cochlear implant early and who have good oral language skills are more likely to acquire ToM in a typical time frame.

Key words: cochlear implants, theory of mind, social cognition, language

Acronyms: CI = cochlear implant; DDP = deaf with deaf parents; DHP = deaf with hearing parents; EHDI = Early Hearing Detection and Intervention; OWLS = Oral-Written Language Scales; TH = typical hearing; ToM = Theory of Mind

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Theory of mind (ToM) is one component of social cognition that reflects a child's developing understanding of the mind, and how mental and emotional states affect behavior (for reviews, see Wellman, 2011, 2014). In the early stages of ToM development, children understand that others can want different things (e.g., the child knows to give someone who likes vegetables a carrot for a snack rather than a cookie, even if the *child's* favorite snack is cookies) or believe different things (e.g., one person may believe a cat is hiding in the garage, and another may believe a cat is hiding in the attic). By 5 years of age, children with typical development have a relatively sophisticated understanding of the thinking and mental states of others. False belief understanding (the hallmark of ToM) is mastered by the end of preschool by most children and can be measured via several experimental tasks (Wellman and Liu, 2004). False belief understanding is signified by the child's realization that others can hold differing ideas or beliefs, that the beliefs of others can be false, and that these false cognitive representations can influence a person's actions (Apperly, 2010; Bretherton & Beeghly, 1982; Custer, 1996; Gopnik et al., 1994; Perner, 1991; Wellman, 2002). Having a mature ToM enables a child to predict, explain, and justify the actions of others;

it also supports their engagement in academic and social tasks, including inferring meaning from context, predicting and explaining the actions of people and characters, tricking others, lying, persuading, and understanding jokes (Moeller, 2002; Peterson, Slaughter, et al., 2016; Peterson et al., 2018; Watson et al., 1999). Preschoolers who possess better theory of mind skills are also more socially accepted and popular in their peer group (Slaughter et al., 2015), demonstrate more pro-social behaviors (Eggum et al., 2011), and tend to experience less friendlessness over time (Fink et al., 2014).

Although the sequence of ToM skill acquisition in preschoolers who are neuro-typical has been well established (Meltzoff et al., 1999; Wellman & Liu, 2004), the mechanisms underpinning acquisition and mastery of ToM are less well understood in children with risk factors for language delay. Language ability, in general, appears to influence ToM acquisition in children with typical development (Astington & Jenkins, 1999; Milligan et al., 2007). Specific language skills such as understanding advanced syntactic structures (de Villiers, 1995; de Villiers & de Villiers, 2000), use of mental state vocabulary (Grazzani & Ornaghi, 2012; Peterson & Slaughter, 2006; Ruffman et al., 2002), conversational exposure (Astington & Baird, 2005; de Rosnay & Hughes, 2006; Harris et al., 2005), and understanding of intentional behavior in infancy (Wellman et al., 2008) are also correlated with performance on ToM tasks in preschoolers with typical development.

In addition to language ability, language environment and conversational access to mental state terminology appear to play a role in the development of ToM and social competence in preschool age children that are typically developing. Mothers' conversational style and preference for mental state talk (talk about feelings, emotions, and thinking) is correlated with performance on false belief tasks (Peterson & Slaughter, 2003; Slaughter & Peterson, 2012) and children's mental state language usage can be predicted from their mothers' tendency to use mental state language (Taumoepeau & Ruffman, 2008). Children with more siblings tend to acquire false belief understanding earlier (Perner et al., 1994); and research shows a significant correlation between ToM and time in a preschool setting for children with typical development (Altun, 2019), and a positive correlation between social competence and peer play opportunities (Newton & Jenvey, 2010).

Research examining the development of ToM in children that are at high risk for late or atypical access to language supports the notion that language and conversational experiences are important for acquisition of ToM. Studies of children who are deaf indicate that ToM development is delayed in children who are deaf and whose parents have normal hearing (see Peterson, 2009 for a review), but is not delayed in children who are deaf whose parents are also deaf and who are immersed in sign language from birth (Courtin, 2000; Courtin & Melot, 2005), suggesting that early access to a natural language supports ToM development. The extant research on ToM in children

who are deaf indicates that ToM development is related to language ability, timing of access to a shared language, quality of language input, communication mode of the children in the sample, and hearing status of the parents (Moeller & Schick, 2006; Peterson, 2004; Peterson & Siegal, 1999, 2000; Remmel & Peters, 2009; Sundqvist et al., 2014) and is often delayed by many years, compared to children with typical hearing (TH; Peterson & Wellman, 2009; Peterson et al., 2012). Such delays can have important social consequences for school age children as well as for teenagers who are deaf (Peterson, O'Reilly, et al., 2016; Peterson, Slaughter, et al., 2016; Peterson et al., 2018; Slaughter et al., 2015).

ToM in Children who are Deaf

Numerous studies of ToM in children who are deaf and have hearing parents (DHP) have demonstrated that this population is characteristically delayed in ToM compared to peers with TH and to children who are deaf and have deaf parents (DDP), most of whom acquire a first language through care providers who are fluent users. Early research showed that children who were DHP were elementary school or even middle school age before they could pass a standard false belief task (Courtin, 2000; Courtin & Melot, 1998; de Villiers & de Villiers, 2000; Figueras-Costa & Harris, 2001; Jackson, 2001; Lundy, 2002; Peterson & Siegal, 1995, 1997, 1998, 1999; Steeds et al., 1997; Woolfe et al., 2002). Russell and colleagues (1998) showed that fewer than half of high school age students who were deaf demonstrated false belief understanding. Most children in these studies were classified as *late signers*—children who did not learn sign language until they entered formal schooling. Schick et al. (2007) measured ToM abilities in 176 children who were deaf or hard of hearing aged 3 years 11 months to 8 years 3 months who used either American Sign Language (ASL) or spoken English. Regardless of communication mode, all children who were DHP demonstrated significant ToM delays.

In one of the earliest studies to demonstrate the importance of early language access in ToM development, Courtin (2000) showed that 5 to 8-year-old children who were DDP outperformed hearing peers and children who were DHP (oral and signing) on several false belief tasks. The author concluded that referential shifting in sign language (changing body position or gesturing to indicate shifts among multiple referents) assists with specific aspects of perspective-taking and mental representation, and that early language access and exposure is critical to ToM development. In a follow-up study, Courtin and Melot (2005) found that 5 to 7-year-old children who were DDP outperformed children who were DHP (both those who acquired sign language later, and those who used spoken language) on an appearance-reality task (What does it look like? What is it really?), and a false belief task. Neither of these studies included measures of receptive and expressive language (other than a report that the participants could understand language and pass the control items). The authors wrote, "[T]hus the differences

in performances observed between deaf children groups may in part be due to some differences in their linguistic skills" (p. 23). Numerous studies since have supported the findings of Courtin and others, that children who are native sign language users do not demonstrate ToM delays (Edmonson, 2006; Hao et al., 2010; Jackson, 2001; Meristo & Hjelmquist, 2009; Meristo et al., 2007; Peterson and Siegal, 1999; Siegal & Peterson, 2008; Woolfe et al., 2002).

ToM in Children with Cochlear Implants

Statistically, more than 90% of children born deaf will have parents who have normal hearing (Mitchell & Karchmer, 2004). This can present significant communication and social challenges for families who do not use sign language naturally. Cochlear implants have altered the language-learning landscape for deaf children by providing an avenue by which some children who are DHP who receive a cochlear implant (CI) early and who have appropriate intervention and school supports can access spoken conversation and can develop intelligible spoken language (Geers & Sedey, 2011; Nicholas & Geers, 2017; Percy-Smith et al., 2017; Spencer et al., 2012).

Given the spoken language outcomes that some children achieve with cochlear implants, researchers have posited that the use of cochlear implants might mitigate some of the negative aspects of deafness and early auditory language deprivation on social cognition; however, ToM outcomes for this group are mixed. Meristo and colleagues (2012) compared the anticipatory looking behaviors of 10 infants who were deaf and 10 infants with normal hearing (age 24 months). All children who were deaf had been identified and amplified early (5 with CIs, 5 with hearing aids). The authors found significant differences between groups in false belief attribution, but not true belief attribution, suggesting that delayed language access affects the development of false belief reasoning. Remmel and Peters (2009) tested 30 children who were DHP with cochlear implants ages 3 to 12 years on a 5-item, developmentally ordered Theory of Mind scale developed by Wellman & Liu (2004). These children received cochlear implants on average at the age of 2.9 years and used spoken language as their only mode of communication. Findings indicated that false belief understanding was delayed, but not as significantly delayed as had been reported in previous studies, particularly for the younger participants. Peters and colleagues (2009) measured false belief use in a video description task to ascertain false belief task performance in 30 children with cochlear implants (the same cohort group as Remmel & Peters, 2009). The majority of children with cochlear implants used false belief reasoning when describing a character's anomalous actions, suggesting mature ToM despite poor performance on an experimental false belief task (unexpected contents). Similarly, Ziv and colleagues (2013), in their study of understanding of emotion and false belief among kindergarteners with normal hearing and those who were deaf, found that children who used oral language with cochlear implants outperformed children who used sign language on the false belief

measure. The authors reported delays in ToM performance relative to hearing children, however, and high variability on both the false belief measure and receptive vocabulary ability. Finally, Sundqvist and colleagues (2014) found that very early auditory access to spoken language through a cochlear implant (prior to about 2 years of age) correlated with better ToM development.

Although one might expect children who are DHP with cochlear implants who have caught up verbally to their peers to have typical ToM, age-appropriate language skill appears to be insufficient for ToM mastery. Ketelaar and colleagues (2012) found that desire and belief reasoning were significantly poorer for children who were DHP compared to hearing peers even in children with age-appropriate vocabulary skills. The authors found no differences in performance on desire, intention, or false belief tasks for children who used sign language compared with children who use speech; nor was age at implantation a significant predictor of ToM. The authors concluded that access to spoken language through a cochlear implant is insufficient for ToM development and that the focus of intervention and parent education must shift to the *quality* of early conversations.

The majority of research to date has shown that children who are DHP with cochlear implants significantly underperform on ToM tasks when compared to their peers with TH. Additionally, at least one study suggested that children with cochlear implants do no better than children who acquire sign language late (Peterson, 2009) and that "The use of spoken modality does not seem to benefit ToM development….Irrespective of whether they used cochlear implants or hearing aids, most of the oral deaf children were delayed in ToM development to the same extent as late-signers." (p. 476). Even children with moderate to severe hearing loss (who presumably have good acoustic access to spoken language using traditional amplification) demonstrated social cognitive deficits (Netten et al., 2017).

Several gaps in the ToM literature remain. Many ToM studies failed to measure expressive and receptive language ability at all, or only partially, in children who were DHP or DDP, making it difficult to determine the underlying mechanisms associated with ToM growth (or lack thereof). Ketelaar and colleagues (2012), for example, measured language abilities via a receptive vocabulary test (picture pointing). Such a vocabulary measure cannot accurately assess a child's understanding of non-observable concepts—the domain of language that is correlated with false belief performance (Grazzani & Ornaghi, 2012; Peterson & Slaughter, 2006; Ruffman et al., 2002). Also, receptive vocabulary knowledge might not be a reasonable proxy for the advanced morphology and syntax thought to correlate best with ToM understanding (Astington & Jenkins, 1999; Milligan, et al., 2007).

In studies in which language *was* measured, the majority of children who were DHP (either children who use oral communication or children who are late signers) were identified with hearing loss late, outside of the federal Early Hearing Detection and Intervention (EDHI) guidelines

(e.g., after the age of 6 months); received amplification or a cochlear implant after the age of 2 years; and as a result experienced significant delays in spoken language. Late identification and treatment of hearing loss results in long-term language learning delays regardless of language modality (Mayberry et al., 2002) or the form of first language input (Mayberry & Lock, 2003). Such language delays create subsequent delays in conversational access to a complete language model (including talk of the mind and other non-observable concepts) past the age at which many children with typical hearing are beginning to acquire early ToM skills (Wellman et al., 2005). This is true for children who are developing spoken language, sign language, or both. Peterson (2004) measured ToM in 52 children who were deaf, aged 4 to 12 years. There were 26 participants who used spoken language to some extent, half with cochlear implants and half with hearing aids, evenly divided between oral-only versus sign-plus-oral specialized schools for the deaf. Comparison groups of age-matched high-functioning children with autism and younger hearing children were also included.

> No significant ToM differences emerged between deaf children with implants and those using hearing aids, nor between those in oral-only versus sign-plus-oral schools….The finding that deaf children with cochlear implants are as delayed in ToM development as children with autism and their deaf peers with hearing aids or late sign language highlights the likely significance of peer interaction and early fluent communication with peers and family, whether in sign or in speech, in order to optimally facilitate the growth of social cognition and language. (Peterson, 2004, p. 1096)

However, the 13 children with cochlear implants in that study were all implanted after the age of 2 years; delayed ToM skills might be expected in these children, due to delays in conversational access. Early conversational access seems as important as closing language gaps in children who are deaf (which is often the primary goal in language intervention).

Finally, due to the relatively low incidence of childhood deafness, studies of ToM have relied on specialized, typically self-contained schools for the deaf to recruit participants. The downside of this approach is that these children are more likely to be conversing with other children that have language and ToM delays (Boyle, 1994), or concomitant disabilities affecting communicative competence (Shaver et al., 2013). This may reduce opportunities to converse about the mind and may affect ToM acquisition (De Rosnay & Hughes, 2006).

The above research suggests that the acquisition of a mature ToM in a typical timeframe depends on the ability to communicate early, easily, and proficiently about mental states with other skilled language users. Research shows that deaf children who are language delayed and/or late identified are likely to be delayed in ToM, and that children whose hearing and communication status match that of their parents are less likely to be delayed in language and

less likely to be delayed in ToM. Auditory access per se seems insufficient to ensure typical ToM development; rather conversational access to and understanding of language of the mind (mental, emotional, and cognitive terms) and the beliefs of others from an early age are key variables—regardless of communication mode. If children are identified late, receive technology late, and do not develop strong early language and conversational skills, a cochlear implant itself will confer little advantage in ToM acquisition. By contrast, children who are deaf and who are identified early, treated early, and acquire conversational language in a typical time frame should demonstrate ToM development that more closely approximates that of their hearing peers.

This paper measured language and ToM performance in a group of young children who are DHP and received cochlear implants prior to 18 months of age to determine if very early auditory access to spoken language facilitates social cognitive development. This study adds meaningful and unique information to the current research on ToM in children who are deaf in that it measured complex expressive and receptive language skills and ToM in very early implanted children who used spoken language at school and at home. It also included an age-matched control group with TH that completed identical ToM and language measures.

Method

Participants

Participants were 25 children who were DHP with cochlear implants and 25 children with typical hearing (TH); the groups were matched for chronological age. The children who were DHP (12 males and 13 females) ranged in age from 36 months to 76 months ($M = 57.32$, $SD = 10.67$) at the time of testing. Children in the DHP group received their first cochlear implant between 6 and 18 months of age ($M = 12.5$, $SD = 3.151$, median age of CI = 13 months) and had been using their implant(s) for an average of 44.84 months (range = 19 to 68 months, $SD = 10.92$) at the time of testing. For the purposes of data analyses, *hearing age* was operationalized as months of cochlear implant use. The children with typical hearing (13 males and 12 females) ranged in age from 42 to 71 months ($M = 56.36$, $SD = 8.276$) at the time of testing. Their hearing age and chronological age were equivalent. None of the children in either group had any known diagnosed developmental, cognitive, or neurological conditions, per school and parent report.

Children with cochlear implants were recruited through direct solicitation, word of mouth, newsletter advertisement, social media, and database retrieval from specialized cochlear implant clinics and schools for the deaf in the Midwest, Northeast, and Pacific Northwest. Subject recruitment and data collection occurred over approximately 3 years, primarily due to the wide geographical range from which participants were recruited and the time-intensive nature of data collection. Children with typical hearing were recruited by word of mouth from preschools and childcare centers in the Midwest and

Pacific Northwest. All children with cochlear implants used spoken English as their primary mode of communication. All children attended either mainstream preschool settings (children with TH), or specialized preschools for the deaf or hard of hearing in which peer models with TH were also enrolled (blended or co-enrolled preschools). Ninety percent of the mothers of children in both groups had either a college education or graduate degree; the remaining ten percent in each group were high school graduates or had at least one year of college. There was no significant between group difference with respect to socio-economic status.

Procedure

This study was approved by the Western Washington University Internal Review Board (IRB protocol #10-077) and the Indiana University-Purdue University Indiana Internal Review Board (IRB protocol #1007-63). All participants were individually tested in their home by a clinical professional familiar with speech and language development of children with cochlear implants. Children completed a measure of expressive and receptive language and a modified version of the ToM Scale (Wellman & Liu, 2004). Administration procedures were identical for children with CIs and those with typical hearing. All tests were administered in accordance with standard administration procedures provided in the testing manual or in published literature, unless otherwise specified.

Measures

Expressive and Receptive Language

Oral-Written Language Scales (OWLS; Carrow-Woolfolk, 1995). This standardized language test measures expressive and receptive language ability including lexical/semantic, syntactic, pragmatic, and supra-linguistic language structures in individuals ages three through twenty-one.

Theory of Mind. Theory of mind was assessed using the five-item scale developed by Wellman and Liu (2004) with one addition; a second false belief task was added (Change in Location task) to provide more robust data on this task. Items were presented exactly as described in the Wellman and Liu (2004) paper with minor modifications in props, but no deviation in script or scoring with exception of the Real-Apparent Emotion task where an alternate script was presented to eliminate the narrative of *teasing*.

1. Diverse Desires. This test measures a child's understanding that different people can have different *wants*. A child is presented with a picture of two different snacks, a carrot and a cookie and is asked which snack he/she would choose. The child is then introduced to a character Mr. Jones, and told that he likes the snack not chosen by the child. The child is asked which snack Mr. Jones will pick. The response is scored correct if the child picks the snack Mr. Jones likes.

2. Diverse Beliefs. This test measures a child's understanding that different people can *think* different things. A child is shown a picture of some bushes and a

garage and presented with a toy figure, Linda, who has lost her cat. The child is asked to guess where the cat is hiding and is provided two choices—in the garage or in the bushes (the actual location of the cat is unknown). The child is then told that Linda thinks her cat is in the location not chosen by the child (e.g., if the child chose garage, then Linda thinks the cat is in the bushes). The child is asked where Linda will look for the cat. The response is scored correct if the child chooses the location opposite to his/her own (i.e., responds to the question from Linda's perspective).

3. Knowledge Access. This test measures a child's understanding that perceptual information leads to knowledge. The child is asked to guess what is in a nondescript metal can. After the child responds, he/she is shown that a small toy dog is inside the can. The child is introduced to a character (Polly) and told that Polly has never seen inside the can. The child is asked if Polly knows what is inside the can. The response is scored correct if the child answers that Polly does not know what is in the can despite the child having seen inside the can (i.e., responds to the question from Polly's perspective).

4. Contents False Belief. This test measures a child's understanding that a person can believe something that the child knows to be untrue. The child is shown a Band-Aid box and is asked what is inside (most children say Band-Aids). The child is then shown that there is a pig inside the box. The child is introduced to a character (Peter) who has never seen inside the Band-Aid box. The child is then asked what Peter thinks is inside the box. The response is scored correct if the child answers Band-Aids.

5. Change in Location False Belief. Similar to the contents false belief task, this task measures a child's understanding that a person can believe that something is in a location that the child knows to be false. The child watches Ernie play with a marble and put the marble in a box before leaving the room. The child then moves the marble to a jar and Ernie returns to look for his marble. The child is asked where Ernie will look for his marble. The response is scored correct if the child answers "in the box."

6. Real-apparent Emotion. This test measures a child's understanding that a person's facial expression may not match the emotion they really feel inside. The child is shown illustrations of a happy, okay, and sad face and asked to identify the emotions. The child is then told the story of a boy (Matt) who loves toy trucks and gets a present from his grandmother which he hopes is a toy truck. When Matt opens the present, he finds a book. The child is told that Matt does not really like the book, but he does not want to hurt his grandmother's feelings. The child is asked to remember what toy Matt wanted to get and what toy Matt did get. The child is asked to label how Matt really feels inside (happy, sad, or okay) and then asked to label how Matt tried to look on his face (happy, sad, or okay). The response is scored correct if the child answers with a more negative response for how Matt felt inside than for the facial expression Matt displayed on his face (e.g., Matt really felt sad, but tried to look happy on his face).

Statistical Analyses

The main objective of this paper was to compare performance of children with TH and children who are DHP on measures of ToM, and expressive and receptive language. A second goal was to determine which variables were most strongly correlated with ToM for the group of children who are DHP. To that end, independent samples *t*-tests were conducted comparing the means on the ToM scale, and receptive and expressive language for the children who are DHP and and those with TH. Bivariate correlations were then conducted on the above variables for the group of children who are DHP with the ToM scale.

Results
Group Differences

Bonferroni corrections were applied to all between group comparisons to reduce the likelihood of a Type 1 error.

Children with cochlear implants were not significantly different from children with TH on chronological age ($p = .724$) and SES ($p = .885$; see Tables 1 and 2). There was a significant between group difference with respect to hearing age. The children with TH had been exposed to spoken language significantly longer than children who were DHP by about 12 months ($p = .000$). There were no significant differences on the total ToM Scale between the children who were DHP compared to the children with TH ($p = .716$); 16% percent of the children who were DHP passed all 6 ToM tasks compared to 20% of children with TH (see Table 3).

Correlation Analyses

To examine the relations between predictors and ToM scale performance for the children who were DHP, all predictor variables were correlated with ToM Scale

Table 1

Participant Demographics and Hearing History

| | Group | | | | |
| Variable | Children who are Deaf with Hearing Parents ($n = 25$) | | Children with Typical Hearing ($n = 25$) | | |
	M (SD)	Range	M (SD)	Range	t (df)
Age at implant (mos)	12.48 (3.15)	6.0–18.0		—	
Age at identification (mos)	3.71 (4.07)	1–14		—	
Chronological age (mos)	57.32 (10.67)	36–76	56.64	43–71	.249 (48)
Hearing Age[b]	44.84 (10.92)	19–68	56.65 (8.5)	43–71	-4.23(48)*
Maternal education[a]	6.32 (1.08)	4–7	6.48 (0.77)	4–7	-0.297 (48)

[a]Maternal education is coded on a scale from less than 7th grade (coded 1) to graduate degree (coded 7).
[b]Hearing age is defined as age at cochlear implantation subtracted from chronological age.
*p < .001

Table 2

Children with Typical Hearing (TH) compared to Children who are Deaf with Hearing Parents (DHP): Language Measures and Theory of Mind (ToM)

| | Group | | | | | | |
| | Children who are DHP | | | Children with TH | | | |
Variable	n	M	SD	n	M	SD	t(df)
Expressive language age[a]	25	62.68	20.211	25	63.60	17.428	-.172 (48)
Receptive language age[a]	25	65.84	19.356	25	64.24	14.652	.330 (48)
Expressive language SS[b]	25	104.84	19.334	25	108.88	14.578	-.834 (48)
Receptive language SS[b]	25	107.56	17.628	25	108.00	11.680	-.104(48)
ToM 6-item scale	25	3.80	1.443	25	3.96	1.645	-.223 (48)

[a]Oral-Written Language Scales (OWLS) age equivalent
[b]OWLS standard score (SS)

Table 3

Percentage of Correct Responses on a 6-item Theory of Mind (ToM) Scale

Task	Children who are Deaf with Hearing Parents		Children with Typical Hearing	
	n	Percent Passed	*n*	Percent Passed
Diverse Desires	25	80	25	84
Diverse Beliefs	25	92	25	76
Knowledge Access	25	64	25	76
Contents False Belief	25	36	25	48
Location False Belief	25	60	25	68
Hidden Emotion	25	48	25	44
All 6 ToM tasks	25	20	25	16
Mean total score (0–6)	25	3.80	25	3.96
SD Total Score		1.443		1.645
Mean Age (months)		57.32		56.64
Mean Hearing Age (months)		44.84		56.64
SD Age		10.668		8.495

Scores. Additionally, partial correlations were conducted controlling for chronological age to attempt to exclude effects of maturation. These correlations are presented in Tables 4 and 5. Expressive and receptive language skills were significantly positively correlated with scores on the ToM Scale for the children who were DHP group, even after controlling for age. Maternal education level was significantly correlated with expressive and receptive language scores, but not ToM performance.

Discussion

In this study of 25 young early implanted children who were deaf and used cochlear implants and spoken language, and 25 children with TH, there were no differences between children with cochlear implants and their age-matched peers with TH on expressive language, receptive language, or ToM performance. The only significant difference between these two groups of children was their hearing age; children who were DHP had been exposed to spoken language for significantly less time than their TH counterparts by 12 months on average. Expressive and receptive language skills were correlated with ToM performance in the group of children who were DHP, even after controlling for the effects of chronological age. These results provide evidence that early cochlear implantation can ameliorate some of the deleterious effects of congenital, profound deafness on language development, which in turn may positively influence social cognition; and that children who are DHP who receive cochlear implants relatively early *and* who have age-appropriate language skills are more likely to acquire ToM in a typical time frame. The present findings contrast with earlier literature showing that children who are DHP

Table 4

Bivariate Correlations for Children who are Deaf with Hearing Parents

Variable	1	2	3	4	5	6	7	8	9
1. Theory of Mind score	-	.348	.363	-.079	.422*	.471*	.509**	.542**	.381
2. Chronological age		-	.958**	.068	-.149	.011	.447*	.501*	-.023
3. Hearing age			-	-.222	-.144	.027	.410*	.489*	.046
4. Age at implant				-	-.007	-.057	.093	.004	-.237
5. Receptive language SS+					-	.897**	.790**	.666**	.586**
6. Expressive language SS+						-	.795**	.845**	.633**
7. Receptive language age							-	.915**	.486*
8. Expressive language age								-	.524**
9. Maternal Education									-

Note. N = 25
+Standard Score (SS; where 85–115 represents average range)
*$p < .05$
**$p < .01$

Table 5

Chronological Age Controlled Partial Correlation for Children who are Deaf with Hearing Parents

1. Theory of Mind score	-	.421*	.453*
2. Receptive language age		-	.893**
3. Expressive language age			-

Note. n = 25 for all variables.
p < .05. **p* < .001

who used cochlear implants performed no differently than children who used hearing aids on a ToM test battery (Peterson, 2009). However, in contrast with previous studies, this study was the first to include only children who received cochlear implants early, and who used spoken English as their primary language at home and at school. In this regard, the present sample of children was more similar to children with typical hearing and children who are DDP in that they shared a natural language with their parents from an early age. In addition, this study was unique in that all participants who were deaf attended mainstream, or co-enrolled/blended preschool programs. This educational environment provided them with opportunities to interact frequently with typical language and social peer models, and to observe and participate in typical conversational exchanges among other children.

A novel finding of this study is that children who are DHP performed no differently than children with TH on measures of expressive and receptive language and social cognition. This result was observed despite the fact that the children who were DHP had fewer months of language access than the hearing control group. Linguistic deprivation has been raised as a troubling phenomenon in children who are deaf and whose parents have normal hearing (the majority of congenitally deaf children; Hall, 2017; Hall et al., 2019). Children who are born deaf are not eligible for cochlear implants until at least 9 months of age (per FDA guidelines), although some children receive a cochlear implant as early as 6 months of age. This lag in auditory language access is concerning as it may lead to short and long-term language, social, cognitive, and academic delays. However, this study suggests that some children who receive cochlear implants by 18 months of age can function similarly to children with typical hearing, not only in their spoken language ability, but also in their social cognitive skills indexed by tests of ToM. Social cognitive abilities correlate with pro-social behaviors, social skills, and social well-being in preschoolers with normal hearing (Eggum et al., 2011; Fink et al., 2014) and children who are deaf (Peterson, O-Reilly et al., 2016; Peterson, Slaughter, et al., 2016). In this group of children who had CIs implanted early, 20% passed all ToM tasks, compared to 16% of the participants with TH (this difference was not

statistically significant). In the group of children who were DHP, only three out of 25 performed greater than one standard deviation below the mean on receptive language and only four of the 25 fell greater than one standard deviation below the mean on expressive language; one child out of 25 exhibited expressive language scores greater than two standard deviations below the mean. Nine children in the group of children who were DHP demonstrated receptive language skills that were greater than one standard deviation above the mean on the OWLS, and 11 children who were DHP demonstrated expressive language skills above the average range.

One caution about these language findings is that all participants in this study were young (kindergarten age at the oldest), and therefore did not possess mature linguistic skills. Language delays can emerge in middle and high school despite advanced early language function (Marschark & Knoors, 2019). Language plateau in this population may also affect the acquisition of more advanced ToM skills such as understanding of deceit, irony, and sarcasm. Research on college students that are deaf shows that they are vulnerable to delays in these advanced ToM skills (Marschark et al., 2019), reinforcing the need for diligence in supporting language and social skill development as children who are deaf progress through elementary and secondary school.

This study also found that expressive and receptive language skills were strongly correlated with ToM in children who are DHP, even after controlling for the effects of maturation. This finding is supported by most of the literature on children with TH (Milligan et al., 2007; Astington & Jenkins, 1999), children who are DHP (Peterson, 2004; Peterson & Siegal, 1999; Peterson & Siegal, 2000; Remmel & Peters, 2009; Sundqvist et al., 2014), and children who are DDP (Courtin, 2000). One exception is research by Ketelaar et al. (2012) who found that children who are DHP with age-appropriate receptive vocabulary still did not pass the desire and belief reasoning tasks. It is possible that receptive vocabulary is not a good proxy for the domains of language that might support ToM mastery. The current study included more comprehensive measures of expressive and receptive language, including vocabulary, figurative language, morphology, and syntax. This study also compared the children who were DHP with the control group that was TH on all measures, which provided for a direct comparison of language and ToM skills, as well as the relationship between measured language (versus inferred language based on chronological age) and ToM for both groups. It is possible that language skill alone is insufficient to ensure typical ToM acquisition. The participants in Ketelaar and colleagues' study were older at the time of receiving their CI and as such, experienced a shorter period of access to auditory language and, by extension, spoken conversation. It may be that language competence combined with opportunity for practice are important for the acquisition of ToM. In this study, children who were DHP not only had good language skills, but likely more exposure to social exchanges and more opportunities for conversational

practice than deaf children of the same age who received auditory language access later.

Results of this study when considered in light of previous research on ToM in children that are deaf suggests that technology alone is insufficient for addressing social cognitive deficits. Cochlear implants are a sensory aid and neural prosthesis that can improve auditory access to sound and speech and, with appropriate early intervention, can facilitate language development and conversational access for many deaf children. This, in turn, might provide an avenue for ToM development. Children who are profoundly deaf and who have hearing parents are still at risk for language delays (Nittrouer et al., 2018). These language deficits are likely to put them at higher risk for ToM delays as well. Children who learn sign language from adults who are not proficient sign language users are also at risk for ToM delays (Moeller & Schick, 2006). Very early access to conversation (whether signed or spoken) appears to facilitate ToM acquisition. Professionals should focus on strategies that build linguistic fluency and social engagement to promote strong social cognitive skills. For children who are deaf and who have typically hearing parents, cochlear implants may provide auditory access to natural, complex conversations about more abstract concepts such as cognitive, emotional, and mental states. On the other hand, if care providers and family members acquire conversational competence in ASL relatively quickly, including the vocabulary and syntax required to convey cognitive (unobservable) concepts, this could also be a reasonable means by which a child who is deaf can be exposed to theory of mind language and concepts at an early age.

Study Limitations

This is a relatively small sample of mostly middle-class children. In this group of participants, language ability was predicted by maternal education level, a finding observed in previous research on children with cochlear implants (Szagun & Stumper, 2012). Such children may be advantaged in other ways as well; they may have more access to attentive care providers and more intensive, specialized therapy services—both of which might positively influence ToM acquisition. In fact, all of the children who participated in this study were receiving speech-language and listening therapy at specialized clinics for children who are deaf or hard of hearing in addition to school-based speech pathology services. This may have influenced both language and ToM development; Percy-Smith and colleagues (2017) suggested that children who are deaf and who receive intervention from providers with expertise in developing listening and spoken language skills of preschoolers who are deaf or hard of hearing have better outcomes than children who receive speech language therapy alone.

Another limitation was that the ToM tasks used for this research were binary (children either passed or failed each task) and not standardized—although widely used in research with this population. They are not necessarily a robust measure of all ToM behaviors exhibited by neurotypical 3 to 6-year-olds. Standardized measures of ToM such as the ToMI-2 (Hutchins et al., 2017), could further elucidate ToM gaps in children who are deaf across a wider age range, and describe the impact of early identification and treatment of hearing loss on a multitude of ToM skills.

Several gaps in the research remain. Studies that include children implanted prior to 12 months of age are necessary. Dettman and colleagues (2021) found that children implanted by 9 months of age demonstrated significantly better long-term language outcomes than children implanted later; this could positively influence social cognitive acquisition and development. Additionally, studies that include preschoolers who are classified as hard of hearing might provide further insights into the contribution of acoustic hearing (and overhearing) to ToM development. Studies of early implanted children who are bilingual-bimodal (use both spoken language and sign language fluently) would also be useful in ascertaining if use of a visual language enhances access to social cues and abstract, mental state talk in children who also use speech. Children who have used signed supported speech may also demonstrate a different trajectory of ToM development, assuming that supplemental visual language cues enhance vocabulary and/or language development (van Berkel-van Hoof et al., 2019).

Studies of teenagers who are deaf and received a cochlear implant at a very early age could provide information about the longitudinal trajectory of ToM (second order ToM, advanced ToM, future thinking). Language and learning gaps tend to show up later for children who are deaf, regardless of their abilities in elementary school (Marschark & Knoors, 2019); language delays in middle and high school might affect acquisition of these more advanced ToM skills.

Finally, the development of ToM in children who are deaf with additional developmental and cognitive disabilities has not been described at all in the literature. The clinical implications of such research would be valuable to both parents and educators.

Conclusion

Theory of mind acquisition for children who are deaf and who have hearing parents (DHP) is a complex process and probably the result of several intersecting variables: expressive and receptive language ability, high-quality and frequent linguistic and social input by care providers, early exposure to conversations about the mind, opportunities to engage regularly in conversation about the mind with adults and peers, and typical sensorimotor and neurocognitive abilities.

The findings of this study suggest that children who receive cochlear implants by 18 months of age and who acquire age-appropriate spoken language skills may acquire ToM in a timeframe comparable to their peers with typical hearing; ToM acquisition can be supported through optimizing communication access and function from a very early age.

Future research should include more children from a variety of home environments and educational settings, children who are bilingual-bimodal, and children who receive cochlear implants by 6 to12 months of age. Longitudinal studies of very early implanted children would provide further insights into the developmental trajectory of ToM and the possible influence of language plateau on ToM development. The influence of language input and environment on ToM acquisition should be studied systematically, using standardized measures; and the effectiveness of therapy approaches to enhance ToM in young children who are deaf should be reviewed, as this remains a significant gap in the literature.

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