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Assessing Tracheostomy as a Risk Factor for Hearing Loss in Neonates with Bronchopulmonary Dysplasia

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Abstract

Objectives: This study compares the risk of hearing loss in children diagnosed with bronchopulmonary dysplasia (BPD) based upon whether the child required tracheostomy.

Method: A retrospective chart review was conducted that included all children diagnosed with BPD from 2013–2020 at a single tertiary medical institution. Primary outcome was presence of hearing loss. Children without follow-up audiogram were excluded from analysis. Risk comparison was made using hazard analysis; Cox regression model controlled for exposure to ototoxic medications.

Results: There were 177 infants diagnosed with BPD who had sufficient follow-up for inclusion. Thirty-two children (18%) underwent tracheostomy placement. Children with tracheostomy were at significantly higher risk of developing hearing loss, with 13/32 (41%) demonstrating hearing loss during follow-up, compared with 16/145 (11%) of children without tracheostomy, p value < 0.001. Cox regression model found that children with tracheostomy were 5.9 times more likely to develop later onset hearing loss than children without tracheostomy, p value 0.011. Most patients diagnosed with hearing loss were shown to have mild conductive hearing loss.

Conclusion: Among children with BPD, those who required tracheostomy were at significantly higher risk of developing hearing loss, including later onset hearing loss. In this study, the hearing loss observed was typically mild and conductive in nature. Families of children with tracheostomy should be counseled regarding this risk and given recommendations for otologic and hearing surveillance.

Keywords: hearing loss, bronchopulmonary dysplasia, tracheostomy

Acronyms: BPD = bronchopulmonary dysplasia; JCIH = Joint Committee on Infant Hearing; NICU = neonatal intensive care unit

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Hearing loss affects thousands of infants annually, with 1.7 per 1000 newborns screened testing positive (CDC, 2021). Although screening programs have greatly improved our ability to detect infants who are born deaf or hard of hearing (Mehl et al., 1998; Wroblewska-Seniuk et al., 2017), delayed onset or progressive hearing loss poses a problem with detection once the infant leaves the hospital, with symptoms occurring weeks or months following birth. The implications of undetected hearing loss upon initial screenings can negatively impact critical developmental stages of the child (Yoshinaga-Itano, 1999).

The Joint Committee on Infant Hearing (2019) has aimed to address the issue of delayed onset hearing loss as described in the Guidelines for Early Hearing Detection. The JCIH recommends continued audiologic evaluation by nine months of age for a subset of infants with specific risk factors for delayed-onset or progressive hearing loss. They identified 12 risk factors, including a NICU stay of more than five days. Several studies have established that there is a higher incidence of delayed onset hearing loss in NICU graduates, despite an absence of known risk factors in some (Yoon et al., 2003; Wood et al., 2013). It has

been noted, however, that a NICU stay of over five days provides only small gains in detection of delayed onset hearing loss, and further exploration of this population is warranted (Kraft et al., 2014).

The population of infants remaining in the NICU for five days is often high. This close follow-up can prove difficult for families and audiology staff (Holte et al., 2012; Cheung et al., 2022). Furthermore, these babies have a variety of different etiologies, with different levels of risk for delayed onset hearing loss. For example, it is well-established that prolonged aminoglycoside therapy or in-utero Cytomagalovirus infection, frequently seen in the NICU, constitutes risks for hearing loss. For other infants with prolonged NICU stays, less is known about other entities that seem to contribute to a higher risk of hearing loss.

One of these entities is bronchopulmonary dysplasia (BPD). BPD is a chronic lung disease caused by premature arrest of pulmonary development, with neonates requiring oxygen supplementation for weeks after birth. Several studies have suggested a correlation between BPD specifically and hearing loss. Very low birth weight infants and premature infants with BPD have been observed to have more frequent recurrences of conductive hearing loss than those without BPD (Wang & Jiang, 2018; Zanchetta et al., 2010). One study suggests that these infants should be followed as the results of the hearing screening at discharge does not accurately predict future hearing loss (Gray et al., 2001).

Although papers have established a correlation between BPD and hearing loss, it is not clear what aspects of BPD heighten the risk. Since the updated recommendation of the JCIH, we have access to more data that include comprehensive audiologic evaluation for this subset of patients within nine months after hospital discharge. This study aims to evaluate factors contributing to hearing outcomes for infants with BPD.

The primary objective of this study was to determine the rate of referral on newborn hearing screening and diagnosis of hearing loss among children with a diagnosis of BPD. The secondary objective was to assess the risk of later onset hearing loss in those with BPD and tracheostomy placement.

Materials and Method

Institutional review board authorization for research on human subjects was obtained from the University of Michigan Medical Center Human Research Protection Program. A retrospective chart review was conducted using medical records from a single tertiary medical facility, Mott Children's Hospital. The population included all neonates diagnosed with BPD at Mott between January 2013 and December 2020. All audiometric data from these patients was reviewed, including newborn hearing screening results and all available audiograms. Patients were excluded from analysis if newborn hearing screening results were unavailable, or if they did not undergo diagnostic audiometric testing.

Other potential risk factors for hearing loss that were documented include the following: tracheostomy, course of ototoxic medications for greater than seven days (including aminoglycosides, macrolides, and loop diuretics), bacterial meningitis, persistent pulmonary hypertension of the newborn, craniofacial anomalies, syndromes associated with hearing loss, and family history of hearing loss.

Primary measures included results of newborn hearing screening and the rate of long-term hearing loss. Failure to pass newborn hearing screening was defined as a result of *refer* in one or both ears. Development of long-term hearing loss was assessed via auditory brainstem response or behavioral audiogram results, with hearing loss defined as a hearing level > 20dB. It is, however, important to note that other institutions may define hearing loss as a hearing level of > 25dB. Type and degree of hearing loss was determined from audiometry, with conductive hearing loss being defined as having an air-bone gap of > 10dB. Degree was defined as mild if hearing level was > 20 – ≤ 40dB, moderate if > 40 – ≤ 70dB, and severe if > 70dB. Requirement of tympanostomy tubes, hearing aids, cochlear implants, and speech language pathology intervention was noted.

Comparisons were made using hazard analysis with a Cox regression model due to varying lengths of follow-up among the patients. Models controlled for prolonged exposure to ototoxic medications.

Results

We identified 289 newborns with a diagnosis of BPD. Of those, 177 had sufficient follow up records to meet inclusion. Table 1 shows the clinical characteristics of children included in the study. Black patients represented 40% of all patients with hearing loss despite being only 23% of the study population, although there was no significant difference found in hearing loss outcomes between Black patients and patients of other races (HR 1.76, 95% CI 0.76–4.05, $p = 0.187$). No significant difference was found between genders (HR 0.94, 95% CI 0.42–2.09, $p = 0.872$).

Newborn hearing screening (NBHS) was not available for 53 of the newborns with BPD who were therefore excluded. Of the remaining 236 newborns, 177 (75%) passed their newborn hearing screening and 59 (25%) received a *refer* in at least one ear. Forty-three of the newborns who passed the NBHS and 16 of the newborns who did not pass received no further testing. Of the remaining 134 newborns who passed and received further testing, 11 (8%) were found to have hearing loss. Based on the audiogram data, nine of these newborns were found to have conductive hearing loss, one had sensorineural hearing loss, and one had an unknown type. Of the 43 remaining newborns who failed newborn hearing screening and received further testing, 18 (42%) were found to have hearing loss, which was determined to be conductive in 11 newborns, sensorineural in two, auditory neuropathy in three, and mixed in two. A flowchart of this grouping is seen in Figure 1.

Table 1

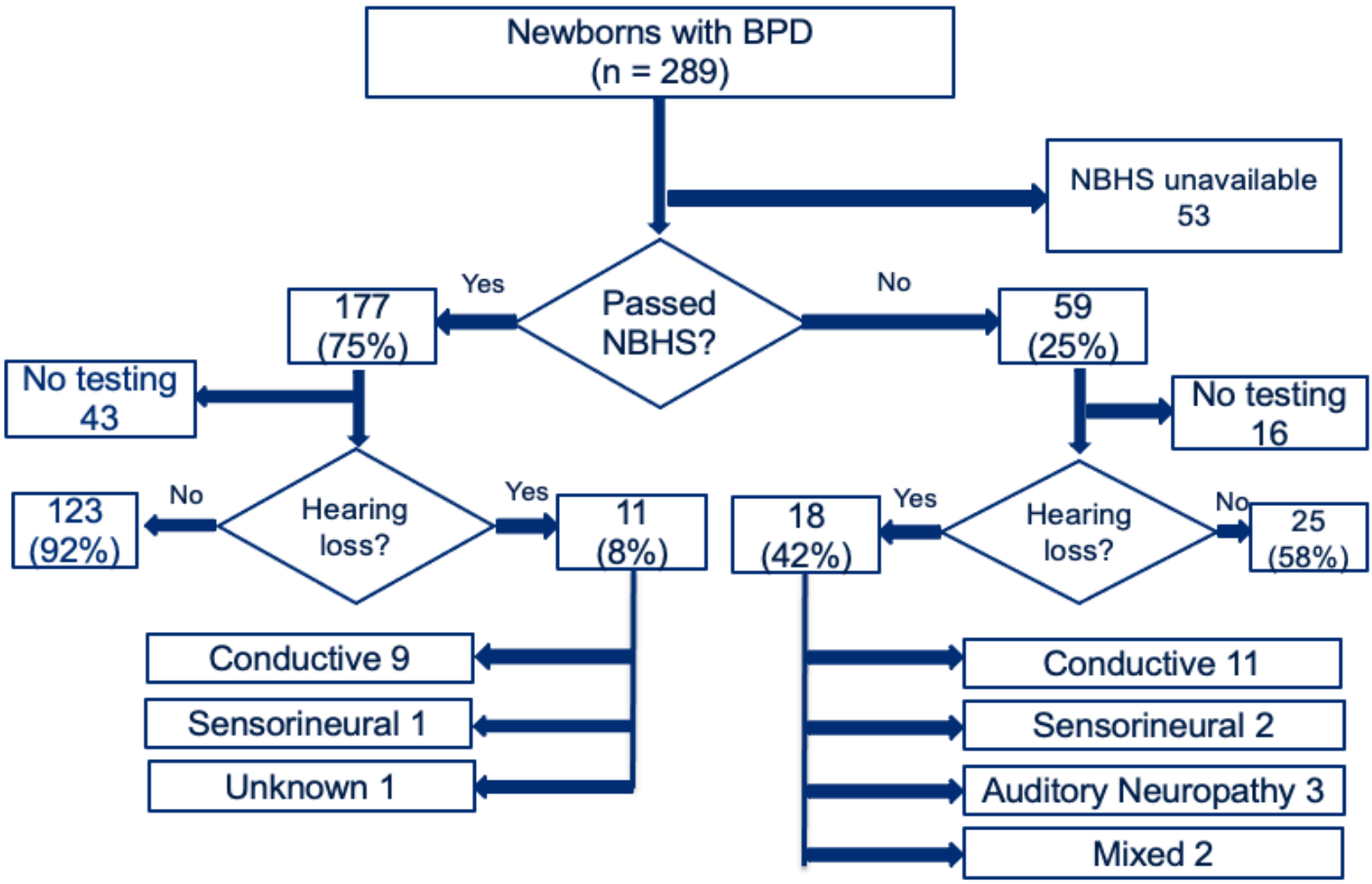
Baseline Characteristics of 177 Patients

	Total (%)	Hearing Loss (%)	Newborn Hearing Screening Refer (%)	Tracheostomy (%)
Gender				
Male	101 (57.1%)	17 (58.6%)	22 (51.2%)	16 (50.0%)
Female	76 (42.9%)	12 (41.4%)	21 (48.8%)	16 (50.0%)
Race				
White	120 (67.8%)	17 (58.6%)	25 (58.1%)	17 (53.1%)
Black	41 (23.2%)	12 (41.4%)	12 (27.9%)	13 (40.6%)
Asian	5 (2.8%)	0 (0.0%)	1 (2.3%)	1 (3.1%)
Other/Unknown	11 (6.2%)	0 (0.0%)	5 (11.6%)	1 (3.1%)

Thirty-two patients required tracheostomy, and 13 (41%) of those patients developed hearing loss, one of whom was also exposed to ototoxic medications. Of these 13 patients, 11 (85%) had conductive hearing loss, one had auditory neuropathy, and one had an undetermined type of hearing loss. Among those with conductive hearing loss, two were characterized as moderate (one of those patients had CHARGE syndrome), and the remaining nine had borderline or mild hearing loss. One patient with mild conductive hearing loss had Trisomy 21. Another patient

Figure 1

Flowchart of Hearing Loss by Newborn Hearing Screening Result



Note. BPD = Bronchopulmonary Dysplasia; NBHS = Newborn Hearing Screening.

had Trisomy 18 and ear canal stenosis. All except one of the patients with conductive hearing loss were noted to have eustachian tube dysfunction. Some of this population had fluctuating hearing loss, with occasional audiometry falling in the normal hearing range. Five patients had ear tubes placed, and two required hearing aids. Five of these patients required speech and language pathology follow up. No patients in this cohort have required cochlear implants. A Cox regression analysis controlling for ototoxic medication exposure was performed. There was an increased risk of hearing loss in patients requiring tracheostomy compared to those who did not require tracheostomy (HR 4.87, 95% CI 2.0–11.39, $p < 0.001$), as demonstrated in Figure 2 and Figure 3.

Patients receiving tracheostomies made up 6 of the 11 children who were later diagnosed with hearing loss after initially passing newborn hearing screening, despite being only 18% of the study population. Of the 20 patients with tracheostomies who passed their newborn hearing screening, six (30%) were diagnosed with later onset hearing loss, compared with five of 114 (4%) without tracheostomy. When looking at the subset of patients who passed their newborn hearing screening, there was a significant increase in risk for late-onset hearing loss in those requiring tracheostomies (HR 5.90, 95% CI 1.50–23.15, $p = 0.011$).

Figure 2

Hearing Loss by Tracheostomy Exposure

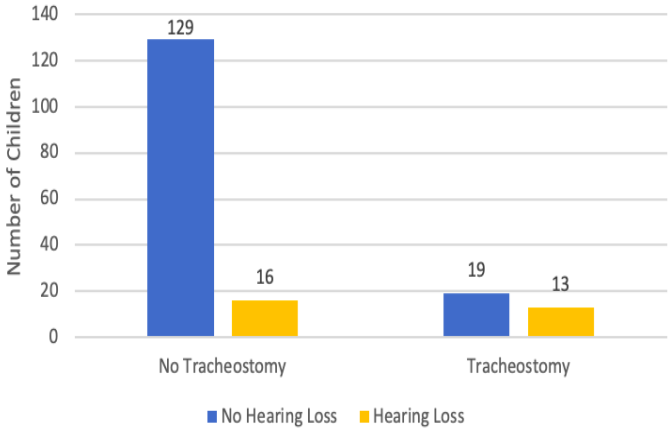
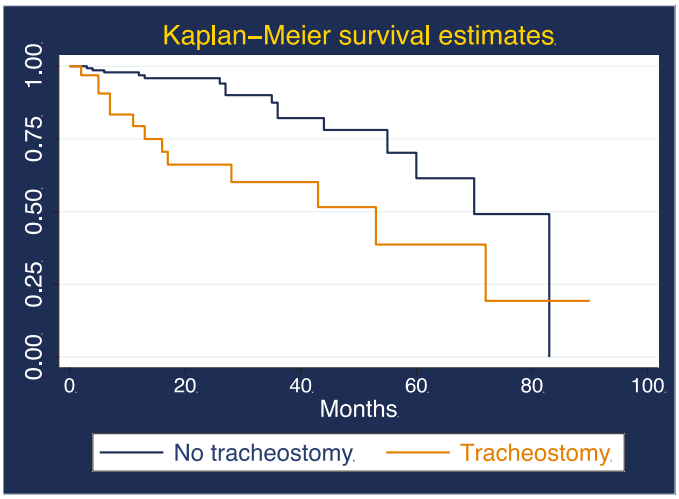


Figure 3

Hearing Loss Over Time by Tracheostomy Status



Thirty patients were exposed to prolonged ototoxic medications, five (17%) of whom developed hearing loss. Two patients who had no ototoxic medication or tracheostomy exposure required cochlear implants.

Characterization of hearing loss is shown in Table 2. Among the entire subset of patients with hearing loss, including those without tracheostomy, nine patients had hearing loss that was not found to be conductive in nature. One of these patients had an undetermined type of hearing loss. The remaining eight patients all had comorbid diagnoses that are known to be associated with hearing loss, including hyperbilirubinemia, pontocerebellar hypoplasia, myotonic dystrophy, cochleovestibular anomalies, absent cochlear nerve, sialic acid storage disorder, cerebral palsy, and epilepsy.

Discussion

Hearing loss is a known contributor to delays in childhood development (Yoshinaga, 1999). The current JCIH guidelines recommend follow up hearing screening for many groups of infants, including those with a NICU stay longer than five days, given their higher risk of hearing loss even in the absence of other known risk factors (JCIH, 2019; Yoon et al., 2003; Wood et al., 2013). However, little is known about exactly why these patients are at higher risk.

Table 2

Type of Hearing Loss by Tracheostomy Exposure

	Tracheostomy	No Tracheostomy
Conductive	11 (84.6%)	9 (56.3%)
Sensorineural	0 (0%)	3 (18.8%)
Auditory Neuropathy	1 (7.7%)	2 (12.5%)
Mixed	0 (0%)	2 (12.5%)
Unknown	1 (7.7%)	0 (0%)
Total	13	16

In the population of NICU patients with a diagnosis of BPD, some studies have suggested that certain subsets of patients experience a higher risk of hearing loss, including premature infants and those with very low birth weights (Wang & Jiang, 2018; Zanchetta et al., 2010). It has also been suggested that patients with a history of BPD are at higher risk of late onset hearing loss (Gray et al., 2001). In our analysis, we examined other possible factors that could contribute to hearing loss in these patients. We demonstrated an increased risk for hearing loss in patients with BPD who required tracheostomy.

In this study, the first aim was to assess the risk and onset of hearing loss in patients with BPD. The national rate of hearing loss is about 0.1–0.2% of children, and 0.17% of children nationwide are referred on newborn hearing screening (CDC, 2021). This study found a significantly higher rate of referral among patients with BPD, along with a high rate of hearing loss among those who had referred on newborn hearing screening. Additionally, among patients who had passed newborn hearing screening, a large proportion were found to have delayed-onset hearing loss.

The second aim of this study was to evaluate possible risk factors for hearing loss in the cohort of children with BPD. Tracheostomy was identified as a risk factor for developing hearing loss. This aligns with other research that has shown an association between tracheostomies and hearing loss, though there have not been reports previously on this specific subset of patients with BPD and the nature of said hearing loss has not been well characterized (DeMauro et al., 2014). Children with tracheostomy have been shown to be at higher risk for delayed onset hearing loss in particular (Narayanan et al., 2023). Two possible explanations that are common in patients with a tracheostomy include a delayed disease process causing their hearing loss or a delayed recognition of existing hearing loss as speech and language delays.

Qualitative analysis of the hearing loss characteristics showed that in the absence of other diagnoses contributing to sensorineural hearing loss or auditory neuropathy, patients with BPD are at high risk for conductive hearing loss secondary to eustachian tube dysfunction. No patients developed sensorineural hearing loss in the absence of other known comorbid diagnoses. The conductive hearing loss was characterized as mild in most cases, though it did fluctuate in severity in some patients. All children in this study who were profoundly deaf had other comorbid diagnoses.

Although almost all of the hearing loss that cannot be attributed to other diagnoses was mild in nature, even mild hearing loss can have effects on development of speech and learning in children (Tharpe, 2008). Children with even mild hearing loss can have speech and language delays and lower language levels compared to peers with normal hearing, and show improvement in their language abilities with interventions such as hearing aids (Mahomva et al., 2021). In a recent study of language outcomes for children with mild to severe hearing loss, children with mild hearing loss were found to benefit from hearing aids as much as those with severe hearing loss (Tomblin et al., 2015). A systematic review looking at outcomes for children with mild to moderate hearing loss noted that outcomes were improved when children with mild hearing loss had support from teachers, primary caregivers, and early intervention practitioners to work on speech, language, and literacy difficulties (Zussino et al., 2022). It is therefore important to support and monitor these children regardless of severity of hearing loss.

Our results suggest that we should continue to perform follow-up hearing screening for the subset of NICU patients with BPD. In particular, infants who had a tracheostomy need follow up given their increased risk of hearing loss (i.e., delayed onset, conductive hearing loss) in the setting of eustachian tube dysfunction. In the absence of other risk factors, tracheostomy alone does not appear to cause a severe, permanent hearing loss in patients with BPD. Despite the mild degree of hearing loss that is most commonly seen in children with BPD, caregivers should be counseled on the benefits of treating persistent mild hearing loss to improve speech and language outcomes. Treatments are variable and dependent on the child and may include placement of pressure equalization tubes, implementation of classroom accommodations and consideration of hearing aids or other devices when appropriate.

Although it was not statistically significant, the overrepresentation of Black patients requiring tracheostomy and developing hearing loss was concerning. It is not clear from our data whether the increased incidence of tracheostomy and hearing loss in this population was due to differences in the underlying disease or anatomy. It is possible that the increased incidence in hearing loss is, at least in part, related to the increased incidence of tracheostomy. There is evidence that Black children are 1.2 times more likely to undergo tracheostomy compared to children of other races within the United States, contributing to increased procedures, higher costs, and longer hospital stays (Brown et al., 2021). Another study looking specifically at tracheostomy in BPD found a statistically significant difference in rate of tracheostomy between Black neonates and other children (Karkoutli et al., 2022). Treatment of Black neonates appears to differ in other aspects of care for children with BPD, including duration of diuretic taper (Bhandari et al., 2010). Further exploration of these disparities is warranted to identify a cause and to help mitigate them.

Limitations of this study include the retrospective nature of collecting data, with respect to the audiometric findings, demographic characteristics, and other risk factors for hearing loss. Newborns with bronchopulmonary dysplasia often have other perinatal conditions that could contribute to hearing loss, and it is possible that not all of those conditions were accounted for in this study. Congenital cytomagalovirus, for example, was not routinely collected at this institution at the time when this data was gathered and therefore is not considered. Although type and severity of hearing loss was determined for most patients, there was one patient with an undetermined type and severity.

Conclusion

Compared to the general population, patients with BPD experience a higher rate of conductive hearing loss, including delayed-onset hearing loss. This hearing loss is typically mild, though it can fluctuate in severity. Patients with BPD in our study were only found to have severe, permanent hearing loss in the setting of other known comorbid risk factors. Additionally, patients with BPD requiring tracheostomy are at an even higher risk

compared to those with BPD who have not required tracheostomy. Further studies evaluating the broader NICU population as well as those with BPD specifically are necessary to help refine current guidelines.

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Preliminary Validation of the Conditioned Assessment of Speech Production in Spanish

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Abstract

Purpose: This study focused on the preliminary concurrent validity between the CASP and CASP-S to answer the following questions: (a) Do obtained scores on the CASP-S correlate to the CASP? (b) Do the two assessments categorize children with and without hearing loss similarly in terms of vocal development?

Method: Eighteen Spanish-English bilingual children (12–43 months) participated in this study; 12 had typical hearing; 6 had severe-to-profound hearing loss. During the study, the clinician modeled specified vocalizations over Zoom for the parent to repeat, then the parent imitated the vocalization for the child to repeat. Approximately a week later, the CASP-S was administered a second time.

Results: The results of this study yielded strong evidence for concurrent reliability, $r = .942$, $p < 0.001$. It revealed an Intraclass Correlation Coefficient of 0.986, (95% CI 0.962, 0.995), which indicates a high level of agreement between two raters. Lastly, for test-retest reliability, there was a statistically significant, positive relationship, $r = .803$, $p < 0.001$.

Conclusion: The CASP-S was found to be a reliable and valid measure to assess early vocal development in Spanish-speaking children with hearing loss.

Keywords: Spanish early speech production, speech assessment, telepractice

Acronyms: CASP = Conditioned Assessment of Speech Production; CASP-S = Conditioned Assessment of Speech Production-Spanish

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Standardized instruments to assess early vocal development in children who speak languages other than English are currently lacking (Tresoldi et al., 2018). The use of English norms for speakers of other languages is not possible, as each language has different rules for how consonants and vowels are produced, in what word positions they can be produced, and how complex words and syllables are, among other parameters (McLeod, 2007).

Early Speech Development in Children

Previous studies that have been conducted on early vocalizations have focused primarily on stages of vocal production and associated development of consonant repertoires; however, due to the scarcity of detailed information, studies are now transitioning their focus to early vocal development and its correlation with later speech and language development (Morgan & Wren, 2018). According to Sheppard and Lane (1968), prelinguistic, infant vocal behaviors serve as the basis for later language capabilities; therefore, it is an aspect that requires considerable analysis.

According to Kuhl & Meltzoff (1996), the process of acquiring speech is anatomically constrained yet auditorily led. Infants embody perceptual abilities that allow them to naturally learn from language exposure. A lack of exposure to language due to hearing loss, unenriched linguistic environments, or other similar scenarios may then lead to a decreased ability to develop speech (Kuhl, 2004). Numerous studies have shown that auditory input is critical for early speech development in infants especially, for the well-formation of syllables during their babbling period (Polka et al., 2007). Researchers have proposed that babbling is a highly canalized motor behavior. The process involves consistent feedback of self-produced sounds for infants to learn how to adequately coordinate their articulation, phonation, and rhythm of their vocalizations (Polka et al., 2007). Regarding treatment, recent evidence has suggested that early identification and aural habilitation provide a significant positive impact on speech development in very young children with hearing loss (Robbins et al., 2004). The subsequent section will thus discuss the effects of auditory-tech devices on speech development in children with hearing loss.

Hearing Loss Considerations

Two to three out of every 1,000 children in the United States are born with a detectable level of hearing loss in one or both ears (Centers for Disease Control and Prevention, 2010). Hearing loss during infancy restricts access to auditory input and feedback, which are necessary factors for children to adequately produce early speech sounds (Ambrose et al., 2016). According to Kent and colleagues (1987), infants with hearing loss demonstrate significant deficits in prelinguistic vocal development and differ in the phonetic properties of their babbling compared to infants with typical hearing. These deficits may possibly affect their emergence of purposeful expression and potentially lead to unintelligible, meaningless speech. It has been evidenced by Ching (2015) that earlier detection of hearing loss results in immediate, early intervention services and timely fittings of auditory devices, improving auditory access and resulting in better speech and language outcomes for the child.

Comparing Early Vocalizations of Infants with and without Hearing Loss

Although infants with hearing loss can also vocalize, the timetable of development and their productions are not as easily understood. Some studies demonstrate overlap in early vocal development between children with hearing loss and children with typical hearing (Lenneberg, 1966; Locke, 1983; Smith, 1984). In contrast, other studies show differences in early vocal development between children with hearing loss and children with typical hearing, demonstrating there is a difference in quality and quantity of prelinguistic vocalizations of children with hearing loss and children with typical hearing (Lyer & Oller, 2008; Maskarinec et al., 1981; Mavilya, 1972; Stoel-Gammon & Otomo, 1986).

Vocal development measures can be used not only to monitor progress of prelinguistic vocal development, but to also interpret auditory benefits from the use of auditory devices for children with hearing loss. Although there are various clinical tools available to assess early vocal development in English for children with typical hearing (Bayley, 1993; Coplan, 1986; Rossetti, 1990; Wetherby & Prizant, 1993; Zimmerman et al., 2002), instruments to assess speech and sound development in children who speak languages other than English are currently lacking (Tresoldi et al., 2018).

Assessing Early Vocalizations in Children with Hearing Loss

To measure the success of aural habilitation techniques on vocal development, early vocalization assessments would be deemed the most appropriate tool (Ambrose, et al., 2016). Assessments such as these are essential for two reasons: (1) to highlight the role of auditory abilities within the process of early vocal development and (2) to monitor the effectiveness of aural intervention strategies provided to infants with hearing loss (Ambrose et al., 2016). As previously mentioned, there are various clinical tools available to assess early vocal development in English for children with typical development. There are also a small number of tools available to assess early vocal development in children with hearing loss. These tools include the Vocal Development Landmarks Interview—Experimental Version (Ambrose et al., 2016), and the Conditioned Assessment of Speech Production (CASP; Ertmer & Stoel-Gammon, 2008). However, standardized instruments for the assessment of early vocal development in children who speak languages other than English are currently lacking (Tresoldi et al., 2018). The lack of instruments available in other languages leads to the use of norms based on studies of English-speaking participants. Unfortunately, this is not linguistically appropriate as it does not provide normative data specifically for each phoneme from another language and children will be assessed inaccurately (Tresoldi et al., 2018).

The CASP is an efficient, criterion-referenced tool that was constructed for early interventionists to analyze and monitor early vocal development in infants and toddlers with hearing loss who speak English (Ertmer & Stoel-Gammon, 2008). The CASP is not a linguistically appropriate tool to use with Spanish-speaking children with hearing loss; therefore, the CASP was adapted and constructed into the Conditioned Assessment of Speech Production-Spanish (CASP-S) as an evaluative tool to use with young children with hearing loss who speak Spanish (Alfano et al., 2022). Although an extensive review of the differences in the phonological systems of English and Spanish is not appropriate for this paper due to space constraints, the reader is referred to Alfano et al. (2022) for more detailed information. As Spanish is one of the most prominent spoken languages in the world, it was critical to determine if the Spanish-adapted version of the CASP would allow clinicians to effectively assess and monitor the early vocal development of Spanish-speaking and bilingual populations with hearing loss. Following the adaptation of the CASP-S, the aim of this current study was to provide preliminary reliability and validity data of the CASP-S.

Assessment Adaptation Process

Educational assessments are being translated and adapted at a higher rate now than they were before (Matsumoto & van de Vijver, 2011). Test adaptations involve deciding whether the assessment can measure the same constructs in a different language, selecting appropriate items to translate, deciding on appropriate changes to be made in preparing a test for a second language, adapting it, and ensuring both forms of the assessment are equivalent. Assessments need to be adapted to facilitate comparative studies of achievement across cultural and language groups, can be more cost-effective than developing new tests, and can achieve fairness in assessment methods through establishment of equivalence of scores (Hambleton et al., 2012). Adaptations involve much more than just simple translation to another language. It requires significantly more than the translation of literal words and is more highly involved with ensuring that they address the same concepts, words, and expressions that are culturally and linguistically equivalent in a second language and culture (Hambleton et al., 2012).

To adapt the CASP into Spanish, the authors reviewed and adapted each item of the CASP based on Spanish phonology. For vowels, it was noted that Spanish has five vowels and English has significantly more vowels. Due to a discrepancy in the number of vowels in each language, the assessment was adapted to include all five Spanish vowels in the CASP-S. In the warm-up section of the assessment, changes were not made because the salient high back vowel /u/ and the mid back vowel /o/ are both found in Spanish and English alike. For consonants, an adaptation from the CASP to the CASP-S was not necessary, as the specific English consonants in the CASP are also represented in Spanish. The mastery age of four years and six months in typically developing Spanish-speaking children was also equivalent to that development in English-speaking children.

There are three phases in formation of vocalizations with pre-canonical vocalizations, basic canonical syllables, and advanced forms of utterances. Pre-canonical vocalizations are sounds that children make that do not have true syllables. Basic canonical syllables are syllables that consist of consonant-vowel combinations that have adult-like timing and advanced forms of utterances consist of consonant-vowel-consonant syllables as well as a diphthongized vowel. Several adaptations were necessary when creating the CASP-S. In the pre-canonical vocalization section, changes were made because the vowels used in English did not exist in the Spanish phonetic repertoire. The mid-central /ʌ/ from the CASP was changed to the mid-low vowel /a/ because /ʌ/ is not found in the Spanish language. Similarly, the low-front /æ/ is not found in the Spanish repertoire and was changed to the mid-front /e/ in the CASP-S. In the basic canonical section, the only adaptation made was a change in the low-back vowel /ɑ/ to the mid-low vowel /a/. Finally, in the section of advanced forms the diversity of the consonant-vowel-consonant syllables is limited, and selecting a similar CVC sequence was challenging when adapting and validating the CASP-S. The cultural adaptation made in this section was to change /tʌk/ to /kon/, since syllables in Spanish do not typically end with /k/.

Purpose

This study focused on the preliminary concurrent validity between the CASP and CASP-S to answer the following questions: (a) Do obtained scores on the CASP-S correlate to the CASP? (b) Do the two assessments categorize children with and without hearing loss similarly in terms of vocal development? It is hypothesized that each assessment tool will demonstrate comparable developmental stages in the respective languages. The specific aims of this study were to provide preliminary reliability and validity data for the CASP-S. Since Spanish is one of the most prominent spoken languages in the world, it was critical to validate the Spanish version of the CASP for clinicians to effectively assess and monitor the early vocal development of Spanish-speaking and bilingual populations with hearing loss. This information can then be used to design future developmentally and linguistically appropriate aural habilitation for infants and toddlers with hearing loss who speak languages other than English.

Method

Participants

Following approval from Florida International University's Institutional Review Board, participants were recruited through a flyer that was shared across multiple settings and social media sources (e.g., e-mail, Facebook). Eighteen Spanish-English bilingual children between the ages of 12 and 43 months participated in this study as seen in Table 1. Each child was accompanied by a parent or caregiver throughout the entirety of the study. Prior to the study, parents confirmed that they had access to a phone, tablet/iPad, or a working computer to use Zoom, a video-conferencing platform that was used to conduct the evaluative sessions for this study. Twelve of these children (5 females, 7 males; ages 11–42 months) had typical hearing; the other six children (2 females, 4 males; ages 16–43 months) had been previously diagnosed by a certified audiologist with severe-to-profound hearing loss (pure-tone threshold average greater than 70 dB HL). Five of the six children with hearing loss were fitted with bilateral cochlear implants and one child used bilateral hearing aids. All 18 children met the following inclusion criteria: (a) typical development across all milestones, (b) came from a Spanish-English bilingual home, (c) presented with no intellectual, mental, or medical disabilities (other than hearing loss) and/or did not have any structural/anatomical abnormalities that could affect their ability to produce vocalizations.

Table 1

Participant Demographics

Participant	Gender	Age in Months	Participant	Gender	Age in Months
TH1	M	15	HL1	M	19
TH2	F	11	HL2	F	37
TH3	M	31	HL3	M	23
TH4	F	15	HL4	M	43
TH5	M	14	HL5	F	16
TH6	M	14	HL6	M	30
TH7	F	32			
TH8	M	42			
TH9	F	29			
TH10	F	11			
TH11	M	25			
TH12	M	14			

Note. TH = typically hearing, HL=hearing loss.

CASP and CASP-S

The Conditioned Assessment of Speech Production (CASP) is an efficient criterion-referenced tool that was constructed for early interventionists to analyze and monitor early vocal development in English-speaking infants and toddlers with hearing loss to test whether hearing aids or cochlear implants can lead to noticeable advancements in their vocalization abilities (Ertmer & Stoel-Gammon, 2008). This assessment presents three levels of speech stimuli: pre-canonical vocalizations, basic canonical syllables, and advanced forms. There are several different vowels displayed among these speech elements to test infants' vowel diversity since this is a factor that has been reported to increase after being fitted with hearing technology. This 10-item tool requires a parent/caregiver and clinician to serve as models of imitation paired with reinforcements (a ring stacker toy) to entice the child to produce the specified vocalizations. The CASP was developed to target early vocal development patterns in children with hearing loss who speak English. As English and Spanish do not contain the same phonological characteristics, the CASP is not an appropriate tool to use with Spanish-speaking children with hearing loss. The Conditioned Assessment of Speech Production-Spanish (CASP-S) was thus adapted and constructed as an adequate evaluative tool to use with young children with hearing loss who speak Spanish.

Procedure

Prior to the first visit and after the parent had signed and emailed the consent forms, the families were assigned a participant number which was used to label all data collected. The parent was provided toy materials, as needed, via non-contact home drop-off (due to the COVID-19 pandemic) to use with their child along with a background history form to be filled out based on the parent's observations of their child. The toy used in this study was a plastic ring stacker toy, as indicated in the original CASP. If the family already owned one, they were able to use their own. If they did not own one, a sanitized ring stacker was delivered.

For the first visit, the parent joined the individual Zoom link that was provided and the Zoom session was recorded using the Zoom record function. With the computer facing toward the parent and the child, they were asked to play for approximately 5–10 minutes with the child using toys in their home to habituate the child to the Zoom session and to allow the child to vocalize naturally with their parent. After the play session, the CASP and the CASP-S were both administered to the child, using their parents as a model. This was done by first having the researcher demonstrate the target vocalization for the parent, who then imitated the researcher. Following that demonstration, the parent would then demonstrate the target vocalization for the child, who was then encouraged to imitate the parent. Once completed, the Zoom session was terminated. During the second visit one week later, the parent joined the Zoom link that was provided. The session was recorded via Zoom record function, and the CASP-S was re-administered.

Tools

Concurrent Validity

Concurrent validity refers to a test's ability to be directly related with another measure of the same construct; this factor determines whether a test is valid by comparing it with an already-existing valid test. This study focused on the concurrent validity between the CASP and CASP-S to determine if obtained scores on the CASP-S correlate to the CASP and if the two assessments categorized children with and without hearing loss similarly in terms of vocal development. Both the CASP and CASP-S were administered to participants in ideal conditions on the same day within the same session; after both prelinguistic vocal development measures were scored, a Pearson's correlation was used to assess the magnitude and direction of the relationship between both test variables. Evidence of concurrent validity requires a validity coefficient of $r = 0.3$ or above. By obtaining this information, we will be able to determine whether these two evaluative sources truly measure prelinguistic vocal development across the range of age levels (12 to 43 months) in our study population.

Inter-rater Reliability

Inter-rater reliability refers to the consistency between the scores of two or more independent examiners. To determine this aspect, each protocol must be observed and scored by two or more examiners. In this study, a total of 18 CASP and 36 CASP-S assessment forms were independently scored by one of four research team members.

First Visit

Each researcher was assigned to four participants in which they collected and scored the CASP and CASP-S.

Second Visit

Each researcher collected and scored the CASP and CASP-S with the same participants a second time. This study specifically focused on the inter-rater reliability between the CASP-S protocols that were collected during the second visit for each participant. Using Excel (Microsoft Office 2016, Version 16.14.1), each researcher watched the recording and scored an additional CASP-S that they had not originally observed or scored; therefore, each CASP-S assessment conducted during the second visit was scored twice by two different raters. To measure whether there is a strong relationship between each set of CASP-S scores, the Intraclass Correlation Coefficient (ICC) was used. This coefficient is used to measure the level of agreement between two raters. If the results yield a one, this means that the two raters have the same ratings. ICC is deemed to be a highly powerful reliability statistic since the outcomes are measured at a continuous level to assess the agreement between two or more independent raters. Each observation did not involve an identical set of raters; therefore, a one-way random model of the ICC was used.

Test-retest Reliability

Test-retest reliability determines consistency of scores when administering an assessment. This is important to establish consistent and accurate scores. To determine evidence of consistency between scores from the first and second visits, the Pearson's correlation was used as the test-retest reliability coefficient. To obtain evidence of test-retest reliability, the *p*-value must be less than .05.

Results

For concurrent validity, a Pearson's correlation was run to assess the relationship between performance on the CASP and the CASP-S. There was a statistically significant, positive relationship between the results from the CASP and results from the CASP-S, $r = .942$, $p < 0.001$. These results are indicative of strong evidence for concurrent reliability.

The Intraclass Correlation Coefficient (ICC) was calculated to determine the level of agreement of performance between independent raters on the same CASP-S administration. The ICC was 0.986, (95% CI 0.962, 0.995), indicating a high level of agreement between the two raters.

For test-retest reliability, a Pearson's correlation was run to assess the relationship between the performance on the initial assessment and the score attained from the assessment administered one week later. There was a statistically significant, positive relationship, $r = .803$, $p < 0.001$. These results are indicative of strong evidence for test-retest reliability.

Discussion

The objective of this study was to identify preliminary validation the CASP-S, a criterion-referenced tool designed for early interventionists to analyze and monitor early vocal development in young children with hearing loss who use hearing technology. The original CASP was previously developed to assess early vocal development patterns in children who speak English; therefore, the CASP-S was adapted as an adequate evaluative tool to use with young children with hearing loss who speak Spanish. To successfully complete the aims of this study, it was essential to gather statistical evidence to ensure that this assessment tool produced reliable, sufficient, and valid results. To investigate these aspects, data was collected to determine the preliminary concurrent validity, interrater reliability, and test-retest reliability of the CASP-S. The role of validity in this circumstance was to identify how well the CASP-S measures early vocalizations in Spanish-speaking children with typical hearing and hearing loss; reliability factors were used to determine the assessment's internal structure.

Following data collection, two statistical analyses were used to interpret the values; the Pearson correlation was used to determine the concurrent validity and test-retest reliability, while the Intraclass Correlation Coefficient (ICC) was used to establish the inter-rater reliability. To ensure accuracy of test-retest reliability, researchers performed every possible effort to prevent external variables from influencing both administrations of the CASP-S to each participant. These efforts were deemed to be effective considering that the Pearson correlation analysis indicated strong evidence of consistency between each participant's performance from their first to their second administration of the assessment. This suggests that there were no significant differences found either across the children's performances or in the clinician's judgements across the two administrations of the CASP-S. Concurrent validity was then examined to gain information on the relationship between the original CASP and CASP-S. After further analysis, significant positive correlations were found between the CASP and CASP-S scores, demonstrating internal structure. This indicates that both assessment tools evaluated parallel early-vocalization abilities, specifically prelinguistic vocal development across the range of age levels (12 to 43 months). The ICC was then used to assess inter-rater reliability; findings showed there was a very high level of agreement between two examiners for each set of CASP-S scores. The high level of agreement indicated that raters of each second administration of the CASP-S gave similar ratings for each participant's vocalizations, therefore proving a linear relationship between each set of CASP-S scores. The overall results of this study support and confirm that the CASP-S is a statistically validated assessment that is clinically suitable to assess early vocalizations in young Spanish-speaking children with hearing loss.

Limitations

Several limitations of the current study warrant discussion. The first is a small sample size and varied age range for this study for this initial attempt to validate the CASP-S. Although this limits generalizability, positive results are encouraging. Secondly, due to the limitations presented by COVID-19, sessions were conducted via remote sessions on Zoom. It is possible that this may have impacted the effect on the participants and their performance on the assessments. There is always a possibility that the participants could have performed at a higher level, had the sessions been conducted in person. However, administration of the CASP-S over Zoom was found to be a feasible option, which may expand the number of possible children that can use the CASP-S. Another contributing factor that may present a limitation is excessive prompting on the parents' behalf throughout the assessment. The examiner should review the guidelines and emphasize the correct procedure to reduce the likelihood of incorrect assessment procedures.

Clinical Implications/Next Steps

These results provide useful, valid, and reliable initial evidence for clinicians considering the use of the CASP-S. The CASP-S is a tool that was adapted and constructed to use with young, Spanish-speaking children with hearing loss. Our recommendation is that additional validation be completed with a larger sample size to ensure this assessment provides valid information on a larger scale. However, initial results indicate that clinicians working with children with hearing loss who are from Spanish-speaking

homes may confidently use the CASP-S to document and monitor early vocal development.

Conclusion

The CASP-S, an assessment that documents early vocalizations in Spanish for children with hearing loss, was adapted to address the shortage of standardized instruments to assess early vocal development in young children with hearing loss who speak Spanish. This study set out to determine the preliminary reliability and validity of the Spanish adaptation of the CASP. The CASP-S was found to be an initially reliable and valid measure to assess early vocal development in Spanish speaking children with hearing loss. ICC was used and demonstrated high inter-rater reliability. Results were indicative of evidence suggesting test-retest reliability and concurrent validity.

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Factors Contributing to Late Identification of Deaf or Hard of Hearing Children in Louisiana

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Abstract
It is imperative to identify children who are Deaf or Hard of Hearing (DHH) as early as possible to ensure they receive supports needed to reach their full potential. Early Hearing Detection and Intervention benchmarks stipulate children who are DHH be enrolled in early intervention no later than 6 months of age. A major barrier to early enrollment is late identification. We reviewed records of children identified as DHH in Louisiana after 6 months of age for 2015–2020 birth cohorts to determine factors contributing to the late identification. Cases were examined in-depth after it was determined that a diagnosis was attainable by 6 months of age. For each case, factors contributing to the late identification were evaluated and assigned to three sources: (a) family, (b) provider, or (c) hospital. Results of the analysis revealed that 46% of late identifications were due to families not completing recommended testing, while provider factors accounted for 25% of late identifications. Hospital factors accounted for 5% of late identifications and 24% of late identifications were attributable to more than one source. The analysis indicated that the percentage of late identifications due to families increased from 2015 to 2020, while the percentages due to provider and hospital factors decreased.

Keywords: deafness, hearing loss, newborn hearing screening, late identification

Acronyms: DHH = deaf or hard of hearing; EHDI = Early Hearing Detection and Intervention; EI = Early Intervention; NHS = Newborn Hearing Screening

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The prevalence of congenital hearing loss is two to three per 1,000 infants (NIDCD, 2021). Prior to the implementation of state Early Hearing Detection and Intervention (EHDI) programs, children with congenital deafness were typically not identified until two to three years of age, or later for milder hearing levels (Coplan, 1987; Naarden et al., 1999; Wake et al., 2016). Universal Newborn Hearing Screening (NHS) has elicited an increase in the number of children who are deaf or hard of hearing (DHH) being identified in infancy, which allows for early intervention (EI) services to be initiated sooner than what was possible in the past. There is now a large body of research demonstrating the benefits of early identification and intervention for children who are DHH (Ching et al., 2017; Ching & Leigh, 2020; Meinzen-Derr et al., 2011, 2020, Moeller, 2000; Vohr et al., 2011; Yoshinaga-Itano et al., 1998, 2017, 2020). The Joint Committee on Infant Hearing (JCIH) made recommendations surrounding early hearing detection and intervention, which are now known as the 1-3-6 EHDI benchmarks: screen hearing by 1 month of age, diagnose hearing loss by 3 months of age, and enroll in early intervention services by 6 months of age (2019).

In 2020, 97% of infants born in the United States were reported to have received a hearing screening by one month of age (CDC, 2022a). Despite this progress, approximately 30% of infants who do not pass their NHS become lost to follow-up before receiving a final diagnosis, and many children who do receive follow-up testing do not obtain a diagnosis by three months of age (CDC, 2022c, 2022d). Poor achievement of the 3-month diagnosis benchmark has downstream effects on the 6-month early intervention benchmark. If children are identified as DHH near or beyond six months of age, it will be difficult or impossible for the child to be enrolled in early intervention services by six months of age.

For children who are born DHH, risk of language delay is high (Stika et al., 2015; Vohr et al., 2008; Yoshinaga-Itano, 2003). Enrollment in early intervention services as early as possible, but no later than six months of age, mitigates this developmental risk (Yoshinaga-Itano, 2003; Yoshinaga-Itano et al., 2017). Yet, despite the ubiquitous presence of universal newborn hearing screening, only 45% of children identified as DHH enroll in early intervention services by six months of age (CDC, 2022b). This leaves

a large number of children at an unacceptably high risk of language delay. The reasons for these poor EI enrollment rates are varied, including perceived barriers to accessing services, lack of perceived benefits, and disconnects between family and provider culture (Woodruff-Gautherin & Cienkowski, 2023). However, another major barrier to early enrollment is that many children are not identified by six months of age, therefore precluding enrollment by six months of age.

Since diagnosis before six months of age is imperative to achieving timely early intervention enrollment for children identified as DHH, improving the rates of early diagnosis would provide more families with the opportunity for timely early intervention. It is essential to determine why so few children born DHH are receiving an early diagnosis, even though many state EHDI programs have been in existence for more than 20 years now. Parents of children identified as DHH have reported a variety of experiences with the diagnostic journey. Common barriers to timely diagnosis reported by parents include poor communication of hearing screening results by providers, limited access to hearing healthcare services, and the need for multiple outpatient visits to secure a diagnosis (Elpers et al., 2016; Reynolds et al., 2023; Robinson et al., 2023). However, population-level research into late diagnosis of congenital hearing loss is lacking. Studies in the United States investigating demographic factors found associations between late diagnosis and low maternal education, low socioeconomic status, and maternal race (Deng et al., 2022; Meyer et al., 2020; Zeitlin et al., 2021). A population-based study in Canada reported common reasons for late identification were medical issues, middle ear dysfunction, and family follow-up concerns (Fitzpatrick et al., 2017). Further population-level studies are needed to supplement the qualitative parent perspective studies and uncover potential approaches for amelioration. Strategies to improve rates of identification by three months of age may, in turn, yield an improvement in rates of early intervention enrollment by six months of age. The objective of this study was to investigate reasons for identification after six months of age for children born DHH in Louisiana.

Method

Institutional Review Board Statement

This study was considered exempt by the Louisiana Department of Health Institutional Review Board.

Data Source and Study Population

A report was generated in the Louisiana Early Hearing Detection and Intervention Information System (LA EHDI-IS) for children born from 1/1/2015 to 12/31/2020 and identified as DHH. Children who were initially identified as DHH by six months of age (defined as at or before 180 days of age) were removed from the report. Of those cases remaining, further exclusions were conducted of children for whom obtaining a diagnosis by six months of age was not feasible. These late diagnoses occurred due to one of the following four reasons:

1) Child passed initial NHS.
2) Child passed outpatient follow-up testing (either rescreening or diagnostic evaluation) after not passing NHS.
3) Child was medically fragile or was still in the neonatal intensive care unit (NICU) at six months of age.
4) Child was born out of state and the birth record was not included in the LA EHDI-IS by six months of age.

Record Evaluation

The remainder of cases were included in the report for in-depth analysis to determine why a diagnosis was not obtained by six months of age for children who did not pass NHS. Each record was examined in the LA EHDI-IS, and all available information in the record was reviewed. This information included:

1) NHS reports
2) Follow-up testing reports
3) Notations of scheduled appointments
4) Communications from pediatricians, audiologists, and other providers
5) Communications between parents and EHDI team members.

Identified reasons contributing to each late diagnosis were assigned to one of three sources: family, provider, or hospital. Late diagnoses attributed to families occurred when appointments were missed or canceled, families declined to complete all recommended testing, or families were unresponsive or could not be contacted. Late diagnoses were attributed to providers if families were following recommendations made to them, but the outpatient providers they saw were not following evidence-based practices to ensure a timely diagnosis. Examples included delay in referring a child to a diagnostic evaluation, and failure of the primary care provider to order the recommended test. Late diagnoses attributed to hospitals included reporting incorrect newborn screening results (reporting a *did not pass* result as *pass*), and failure to schedule outpatient follow-up appointments for families of children needing additional testing.

In addition, records of children identified as DHH who passed NHS were reviewed in an effort to identify cases whose NHS results were reported incorrectly. Any information in the record which indicated results may have been inaccurate led to contact of the birth hospital for verification. Those verified as passing NHS remained excluded from the report, while those determined not to have passed NHS were included.

Results

Late Identification of Early-Onset Hearing Loss

A total of 771 children from 2015–2020 birth cohorts have been reported to the LA EHDI-IS as being identified as

DHH (Table 1). Of these, 315 (40.9%) were identified after six months of age. There were 174 cases removed from the analysis because a diagnosis was not deemed attainable by six months of age, for reasons described above. In-depth records analyses were performed on the remaining 141 cases. Of the 141 participants, 51.8% were female vs. 48.2% male; 42.6% non-Hispanic White, 37.6% non-Hispanic Black, 6.4% non-Hispanic other, 12.8% Hispanic, and 0.7% race/ethnicity unknown. One-quarter of mothers did not finish high school (25.5%), 43.3% completed high school as the highest level of education, and 31.2% attained some level of education beyond high school.

Table 1

Children Born in Louisiana in 2015–2020 and Identified as Deaf or Hard of Hearing (DHH)

Total DHH	771
Total late-identified DHH (> 6 months of age)	315
Cases removed from analysis	174
Passed Newborn Hearing Screening	90 (51.7%)
Passed outpatient follow-up testing	29 (16.7%)
Medically fragile	30 (17.2%)
Born out of state	25 (14.4%)
Cases included in analysis	141

Source of Late Identification

Evaluation of the reasons children were late-identified revealed the most common source to be the family, which was the sole source in 46% of cases (Figure 1). Providers were the sole source in 25% of cases, while hospitals were the sole source in 5% of cases. In 24% of cases, two or more sources contributing to late identification were observed.

Age at Identification

Over 70% of late-identified children were diagnosed before turning two years of age (Figure 2). Forty children (28%) received their initial diagnosis after turning two years of age, with the eldest reported at six years, 11 months of age.

Source of Late Identification by Birth Year

Sources of late identification were evaluated by birth cohort to visualize trends occurring over time (Figure 3). An analysis was performed to enable observation of the contributions of each source (family, provider, and hospital) to the total number of late identifications. For example, if a child was late-identified due to both the family and a provider, the source of the late identification was assigned as 50% family and 50% provider. Analysis by birth cohort over time demonstrated an increase in the percentage of late identifications attributable to families, and a decrease in the proportion of late identifications attributable to hospitals and providers.

Family Component to Late Identification

For children whose late identifications were attributed at least in part to the family, we reviewed what occurred after not passing NHS (Figure 4). In nearly half of these cases, the child was lost to follow-up after NHS. An additional 29% received one or more outpatient screenings, but did not complete a diagnostic evaluation by six months of age. In another 22% of cases (n = 20), diagnostic evaluations were completed prior to six months of age, but the findings indicated either transient conductive hearing loss (n = 7), or hearing loss of undetermined type (n = 13). These families did not complete all recommended follow-up testing, and subsequently became lost to follow-up.

Figure 1

Source of Late Identification Among Children Identified as DHH Receiving a Late Diagnosis (Greater Than 6 Months of Age; n *= 141)*

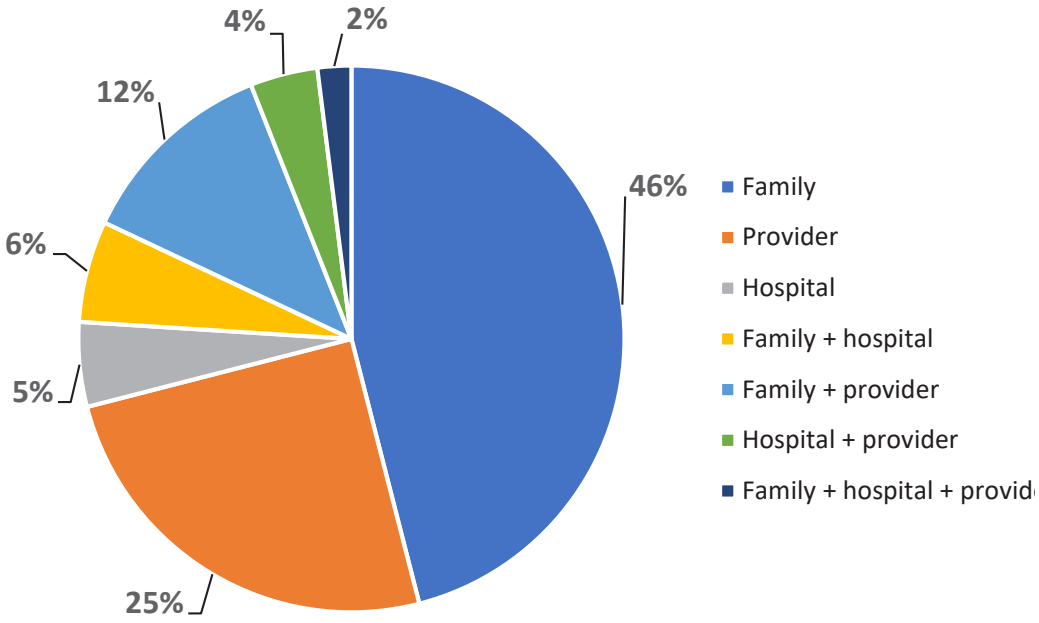


Figure 2

Number of Children Newly Identified as Deaf or Hard of Hearing (DHH) by Age Group

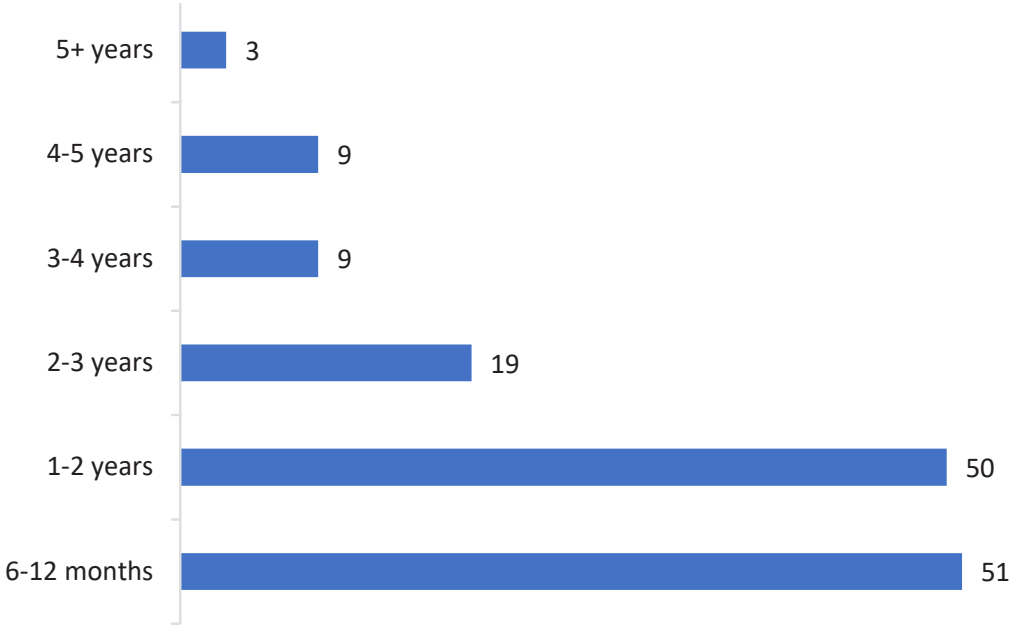
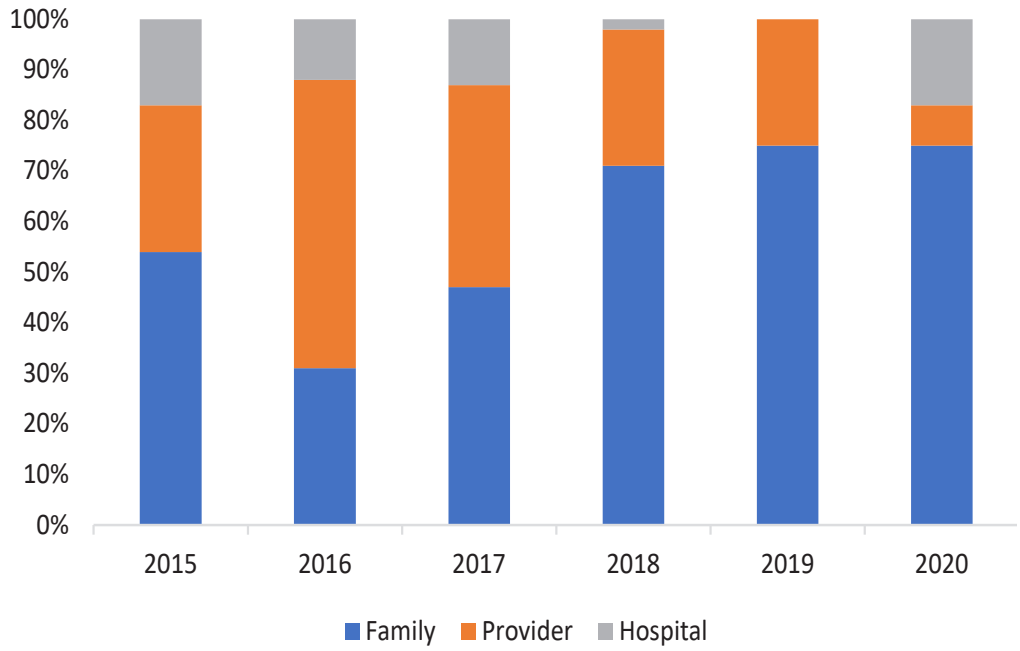


Figure 3

Sources of Late Identification by Birth Year



Note. The figure shows the contributions of each source to late identification (family, provider, and hospital) by each birth year cohort. Cases with more than one contributing factor were given equal weighting (i.e., two sources were each weighted as 50% contributors). A trend is seen over time with families being the most common contributing factor in the more recent birth cohorts.

Provider Component to Late Identification

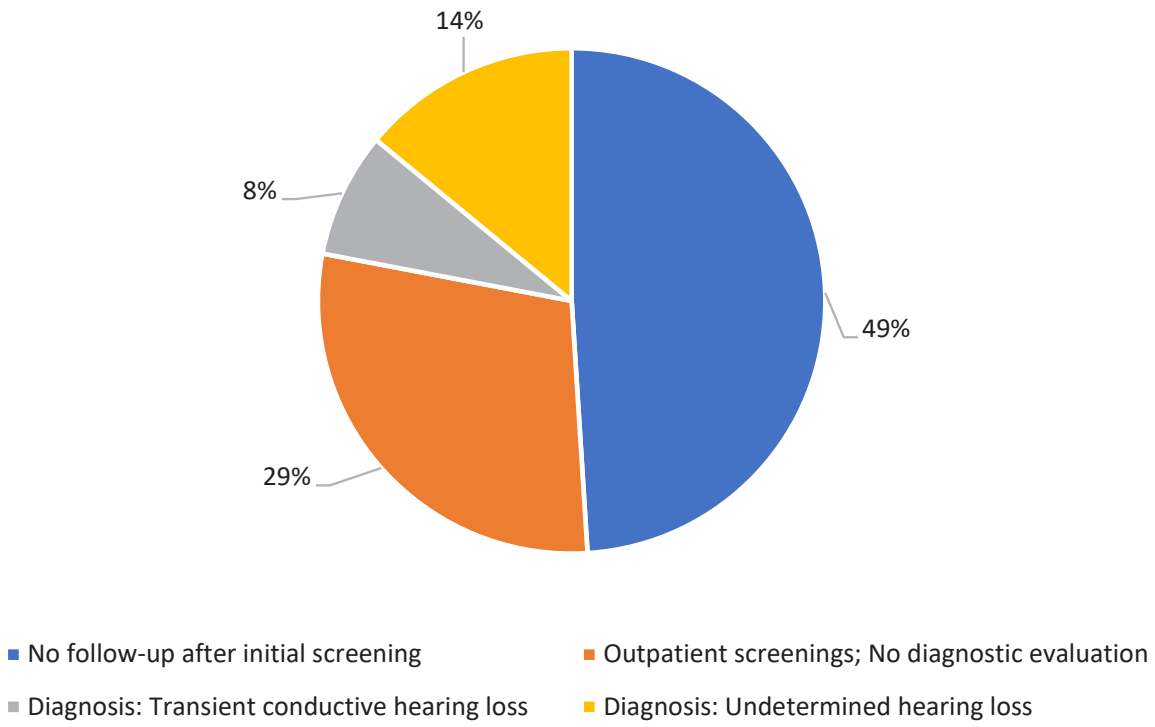
Provider contributions for late identifications were also evaluated (Figure 5). The most common provider issues were performing excessive rescreens prior to referring for diagnostic evaluation, and waiting too long to schedule a diagnostic evaluation (excessive scheduling gap between appointments). An excessive scheduling gap was defined as more than one month between appointments.

Hearing Levels of Children Late-Identified

Hearing laterality and levels of children late-identified were compared to the whole group of children identified as DHH from these cohorts (Figure 6). This indicated similar distributions in hearing levels between the two groups. The only hearing levels exhibiting between-group differences

Figure 4

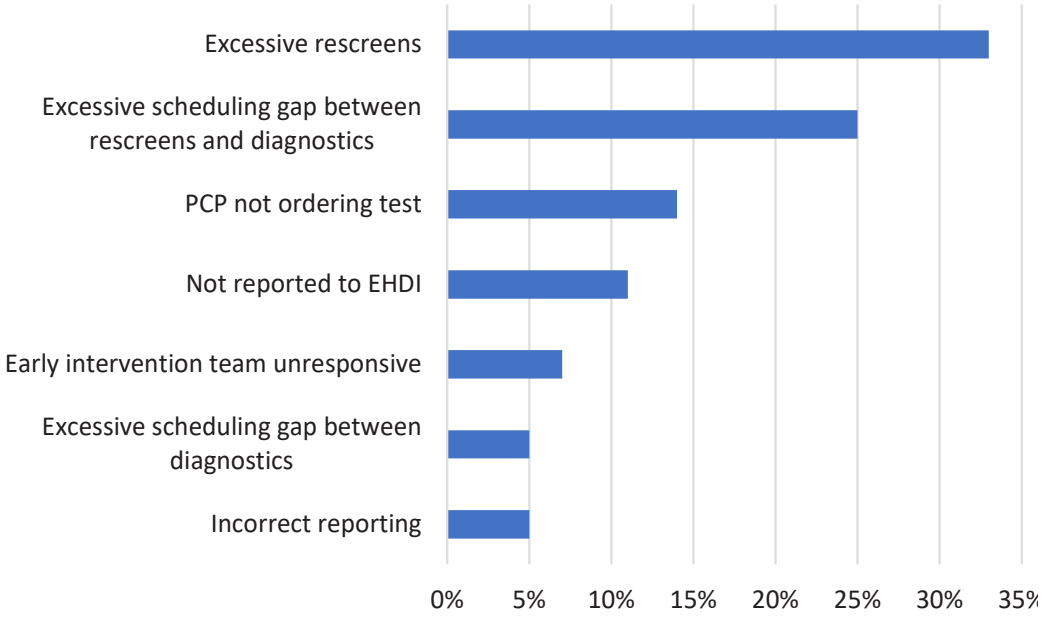
Follow-up Outcomes for Children Late-Identified as Deaf or Hard of Hearing due to Family Factors



Note. The graph displays outpatient follow-up outcomes for children who were late-identified as deaf or hard of hearing due to the family as a contributing source. Nearly half of these children received no outpatient testing in the newborn period after not passing newborn hearing screening.

Figure 5

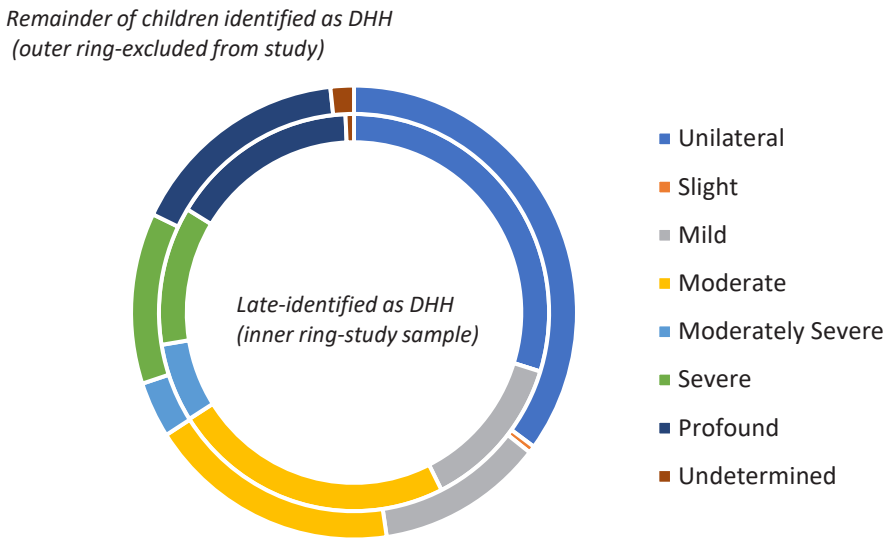
Follow-up Outcomes for Children Late-Identified as Deaf or Hard of Hearing due to Providers



Note. The graph displays outpatient follow-up outcomes for children who were late-identified as DHH due to the provider as a contributing source. The most common problems attributed to providers were performing excessive rescreens before scheduling a diagnostic evaluation and failing to schedule a diagnostic evaluation in a timely fashion (excessive scheduling gap between appointments). PCP = primary care provider; EHDI = Early Hearing Detection and Intervention.

Figure 6

Hearing Levels of Children Late-Identified as DHH vs. Remainder of DHH



Note. The graph shows the proportion of children late-identified as deaf or hard of hearing (DHH) with each hearing level (inner ring, n = 141), as compared to children identified as DHH who were excluded from the report (outer ring, n = 630). All hearing levels listed are bilateral hearing losses. The unilateral groups include unilateral hearing losses at all levels. The distribution of hearing levels was not substantially different between groups.

were unilateral hearing losses (greater proportion observed in the whole DHH group) and bilateral moderate hearing levels (greater proportion observed in the late-identified group). Percentages of other hearing levels were similar between groups.

Discussion

Of children born in Louisiana from 2015 to 2020 and identified as DHH, 41% received their initial diagnosis after six months of age (315/771). Of these 315 children identified after 6 months of age, 141 were included in our analysis because they did not pass newborn hearing screening and did not appear to have any medical barriers to obtaining a timely diagnosis. Factors attributed to families were observed more than any other source among children with late diagnoses, accounting for 46% of late diagnoses as the sole reason for late identification, and an additional 20% of cases as a contributing source. Thus, families were a contributing factor in a full 2/3 of cases in which children were late-identified. When evaluating late-identified children with only a family component, the most common scenario identified was no outpatient follow-up testing after not passing NHS (49%, Figure 4). In an additional 29% of cases with a family component to late identification, the child received one or more outpatient screenings in the first few months after hospital discharge, but the family did not follow through with a diagnostic evaluation. The remaining children with family components received a diagnostic evaluation by three months of age and were identified with hearing loss not deemed permanent (transient conductive or type undetermined). The families did not complete recommended follow-up to receive a timely final diagnosis. It is particularly concerning that half of the cases with a family component to the late diagnosis received outpatient testing indicating a hearing loss was present, or was likely present, even if this hearing loss was transient. In some of these cases, providers further contributed to the delayed diagnosis by not scheduling or recommending appropriate follow-up testing to rule out a permanent hearing loss.

There were a variety of ways outpatient providers were found to contribute to a late diagnosis (Figure 5). Providers were the sole source of late identification in 25% of cases, and were a contributing source in an additional 18% of cases. The most common provider issue was performing excessive rescreens before referring for a diagnostic evaluation, which was seen in a third of late identifications with a provider component. Per Joint Committee on Infant Hearing (JCIH) recommendations, no more than one outpatient rescreen following a referred initial hearing screening should be performed before sending an infant for a full diagnostic audiological evaluation (JCIH, 2019). When evaluating our birth cohorts, we found that the number of children who received excessive outpatient rescreens peaked in 2016, and has been declining since. Our program works to educate providers on the JCIH guidelines and to lend support in remediation. This has resulted in a reduction of children undergoing excessive rescreens since 2017, which is in large part responsible for the reduced contributions of providers to late identifications (Figure 3).

Another substantial issue discovered with providers was an excessive scheduling gap between visits, which was seen in a total of 30% of cases found to have a provider component. In the majority of these cases (18), the gap was seen between the rescreen and diagnostic evaluation appointments, but a smaller portion (four) exhibited gaps between two diagnostic appointments when the first evaluation did not yield a conclusive result. Time is of the essence in EHDI systems, so follow-up appointments

should be made as soon as possible if a firm diagnosis has not been obtained. If an immediate appointment is not available, the facility should provide a referral to another facility who can accommodate the family. We defined a scheduling gap as excessive if the facility scheduled the family's next appointment at greater than one month away from the previous appointment. However, in nearly all of these cases, the next appointment was far beyond this, with three to six months being common. Again, this issue was seen more frequently in birth cohorts prior to 2017, with one facility being responsible for most of the scheduling gaps. Remediation discussions with this facility successfully addressed this issue moving forward.

We found reporting errors by providers to account for a smaller, but significant, number (12; 16%) of provider component cases, including a lack of reporting into the EHDI-IS system and the reporting of incorrect results. We believe the numbers found in this study to be an underestimation, as we could only attribute a reporting error if documentation was available in our system to support that an error was made. In most of these cases, an outpatient follow-up report was not submitted to the EHDI program. Though this action may not be directly responsible for a late diagnosis, we consider it a contributing factor because our program contacts families and primary care providers to alert them of the need for further testing, and assist when they have challenges to obtaining an appointment. These actions may have resulted in timelier diagnoses for some of these cases. In addition, in four cases (5%), a diagnostic report was obtained indicating a diagnosis of hearing loss, yet the provider reported the child passed in the EHDI-IS system, which prevented our program from providing any further assistance. An additional 10 cases (14%) of provider late identifications were due to the primary care provider (PCP) failing or refusing to order a diagnostic evaluation, a problem that our program has virtually eliminated in recent years by communicating directly with the PCP office. We also found five children whose Part C early intervention family support coordinators were alerted by our program of the need to complete hearing follow-up testing, to no avail. Three of the children aged out of Part C before they were identified as DHH.

Issues arising from the hospitals who perform inpatient screenings represented a smaller portion of errors. Hospitals accounted for five percent of late-identified cases, and were contributing factors in an additional 11% of cases. Of the 23 cases with a hospital component to the late identification, 52% did not schedule a follow-up appointment for the family prior to hospital discharge, and 48% reported an incorrect NHS result (reported *pass* instead of *did not pass/further testing needed*; data not shown). Similar to provider issues, the bulk of hospital errors were found in the earlier birth cohorts, with only three errors found in birth cohorts 2018 to 2020 combined. This suggests that accurate entry of NHS results has improved at the hospital level, although it is more likely due to increased surveillance by our program. Louisiana EHDI has reviewed records of recent birth cohorts with greater vigilance, and we work with hospitals to make corrections when probable errors are found. This work has improved the accuracy of NHS data at the hospital level. We have also seen a greater number of outpatient appointments scheduled for families prior to hospital discharge, which has been demonstrated to improve compliance with follow-up (Tran et al., 2017).

Of the children who were late-identified, 36% were identified between six to 12 months of age, and 35% were identified between one to two years of age (Figure 2). A total of 15% of our study cohort were identified after turning three years of age, at which time they were no longer eligible for Part C early intervention services. We believe these numbers to be an underrepresentation, as reporting to EHDI programs tends to decline as children get older. This decline is exacerbated for children three years of age and older, who may see an audiologist who does not specialize in pediatrics, and therefore is not accustomed to reporting to their state EHDI program.

We reviewed the laterality and hearing levels to determine if there were differences in hearing levels between children who were late-identified as DHH, relative to the remainder of children identified as DHH (Figure 6). Since the latter group was composed largely of children identified by six months of age, we hypothesized that the group of late-identified children may consist more heavily of unilateral hearing losses and mild hearing levels, which may have caused less concern to parents and providers. However, the proportion of children with each hearing level was nearly identical between groups, and the only exceptions to this went in the opposite direction expected. Children with bilateral moderate hearing levels were more heavily represented in the late-identified group, while children with unilateral hearing loss were more common in the remainder of children, though the difference was only about five percent for each. All other hearing levels were nearly identical between groups.

Strengths and Limitations

The strengths of this study lie in the detailed analysis of late identification of congenital hearing loss. This analysis was possible due to the LA EHDI-IS, a robust statewide database with widespread adoption by providers. The profiles of many children in this study contained detailed records, including test reports, comments by providers, and actions by EHDI staff members. Date stamps for each entry allowed for historical tracking of actions taken. To our knowledge, there are no population-based studies in the United States investigating the diagnostic process from screening to identification for late diagnoses for children who are DHH. However, as approximately 30% of children who do not pass newborn hearing screening do not complete a diagnostic evaluation (CDC, 2022d), understanding the reasons testing is not completed can aid in targeting areas for improvement.

An additional strength of this study was that, for many children, the analysis enabled differentiation of late-onset hearing loss from congenital hearing loss that was late-identified. Of the 315 children identified after six months of age, 90 were reported as passing their newborn hearing screenings (28.6%) and 29 passed outpatient follow-up

testing (9.2%). The remaining 196 (62.2%) did not pass their newborn hearing screenings, and are therefore presumed to have congenital hearing loss. If all 119 children who passed their newborn hearing screenings or outpatient follow-up testing are presumed to have late-onset childhood hearing loss, the total percentage of DHH children from this birth cohort with late-onset hearing loss is 15.4% (119/771). However, records of many of these children included diagnoses of mild hearing loss, frequency-specific hearing loss, and auditory neuropathy spectrum disorder after a newborn screening via otoacoustic emissions. These hearing losses are all frequently missed by newborn hearing screening, suggesting the incidence of true late-onset childhood hearing loss in our cohort is relatively low, although we acknowledge that these cases may be underreported or underidentified.

There are several limitations of this study. First, we were limited to the information that was available in the child's initial and follow-up hearing screening records reported to the LA EHDI database. This means that relevant information was likely missing in some of our cases. This may be particularly pertinent to the birth hospitals, as we have no way of ascertaining what materials were provided to parents at the time of the screening in the hospital. Previous studies indicate that many parents do not follow up after NHS because the results were downplayed or were not clearly communicated to them (Elpers et al., 2016; Reynolds et al., 2023; Robinson et al., 2023). It is possible that poor communication from hospitals to parents contributed to some of the late identifications that were attributed to families. Although there were no parents of late-identified children who relayed this to us, we cannot rule this out as a factor. Second, reporting of children newly identified as DHH to EHDI programs is less consistent for older children than for children under three years of age. Though our program has worked hard to improve reporting, there may be children identified as DHH that are not known to us. Finally, data for the younger birth cohorts may be incomplete. At the time of data collection, the 2020 birth cohort was still relatively young, so there may be children who are DHH that were yet to be identified. The apparent improvement in provider and hospital errors over time may prove to be misleading as more children are identified as DHH, and previously unseen errors are revealed. It should be noted that this study defined a late diagnosis as being obtained after six months of age. This timeline was chosen because an identification by six months of age would still allow for enrollment into early intervention services by six months of age, per the JCIH goal (JCIH, 2019). However, if we had chosen the JCIH diagnostic goal of three months of age, 58% of our cohort of 771 children would be considered late-identified. If we additionally excluded the 119 children presumed to have late-onset hearing loss, 51% would be considered late-identified, which is only a slight improvement.

Conclusion

We reviewed records of 141 children born in Louisiana from 2015 to 2020 who were identified as DHH after six months of age, despite not passing their NHS. Our evaluation of reasons for each late diagnosis revealed that families not following up after NHS were a factor in 2/3 of cases, being the sole factor in nearly half of cases. Outpatient providers were responsible for another 1/4 of cases, with hospital errors accounting for a smaller proportion of cases. Our year-by-year analysis indicated that the percentage of cases late-identified due to families has been increasing over time, while the percentage due to hospitals and providers has been decreasing. This is likely due, in large part, to improvement strategies deployed by Louisiana EHDI to correct errors and mismanagement of cases by hospitals and outpatient providers. Change strategies targeting families have also been put into place, but the impact of these strategies has been smaller. Follow-up outcomes for children not passing NHS have steadily improved, but much progress is still needed. Future efforts must be geared toward connecting with families and communicating information clearly and effectively. This will be key to improving timely diagnosis and early intervention rates for children who are DHH.

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EHDInfo

World Health Organization

3 March is World Hearing Day. For more information, click here.

World Hearing Day is held on 3 March each year to raise awareness on how to prevent deafness and hearing loss and promote ear and hearing care across the world. Each year, WHO decides the theme and develops evidence-based advocacy materials such as brochures, flyers, posters, banners, infographics and presentations, among others. These materials are shared with partners in government and civil society around the world as well as WHO regional and country offices. At its headquarters in Geneva, WHO organizes an annual World Hearing Day event. In recent years, an increasing number of Member States and other partner agencies have joined World Hearing Day by hosting a range of activities and events in their countries. WHO invites all stakeholders to join this global initiative.

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Sociodemographic Factors Influencing Pandemic-Era EHDI Use and Access

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Abstract

Objective: The COVID-19 pandemic impact on Early Hearing Detection and Intervention (EHDI) programs is unknown. This research evaluated sociodemographic factors influencing adherence to EDHI diagnostic testing and the incidence of infant hearing loss during the pandemic.

Method: We evaluated EHDI adherence and incidence of hearing loss in Kentucky before and during the COVID-19 pandemic. Using univariate and multivariate analysis, we evaluated the association of these outcomes to sociodemographic variables.

Results: There were 71,206 births and 1,385 referred infant hearing screening tests during the study period. Infants during the pandemic had a 24% lower odds of hearing testing adherence (OR = 0.76, p = 0.05, 95%CI: 0.57–1). Hispanic infants have 45% lower odds of EHDI adherence (OR = 0.55, p = 0.03, 95%CI: 0.31–0.96) and infants of Swahili speaking families have 90% lower odds of EHDI adherence (OR = 0.10, p = 0.001, 95%CI: 0.02–0.42). Infants of mothers with a high school degree had a higher odds of adherence (OR = 1.50, p = 0.02, 95%CI: 1.06–2.17), presented earlier for testing (p = 0.003, 95%CI: -15.73– [-]3.32), and had a higher odds of normal hearing (OR = 1.63, p = 0.03, 95%CI: 1.06–2.51).

Conclusion: EHDI adherence is influenced by the COVID-19 pandemic and sociodemographic factors. EHDI programs are encouraged to use this data to promote timely and equitable access and use of diagnostic services.

Keywords: Infant hearing loss, Health disparities, COVID-19, EHDI, Access to care, Newborn hearing screening, Follow-up adherence

Acronyms: EHDI = Early Hearing Detection and Intervention; HA = hearing aid; UNHS = Universal Newborn Hearing Screening

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Hearing loss affects 2–3 of every 1,000 American newborns screened (Centers for Disease Control and Prevention [CDC], 2020a). Pediatric hearing loss can have a long-term impact on speech, language, and social development. For this reason, children with hearing loss require early intervention and multifaceted care. Children with congenital hearing loss who receive timely diagnosis and intervention have remarkably improved speech and language outcomes (CDC, 2019). Universal standard protocols of infant hearing screening and diagnostic testing have been developed by the CDC and are implemented by each state's Early Hearing Detection and Intervention (EHDI) program. These EHDI standards dictate that all newborns are to be screened at birth or no later than 1 month of age. An abnormal screening test should result in diagnostic testing at no later than 3 months of age. Babies with confirmed hearing loss should receive early intervention by 6 months of age (CDC, 2019).

Early detection of hearing loss initiates intervention earlier and improves overall outcomes. Universal Newborn Hearing Screening (UNHS) has been proven successful in identifying hearing loss at an earlier age leading to earlier intervention in several studies. A study disclosed that the median age at hearing aid (HA) fitting of the newborn hearing screened group was 3.9 months (IQR: 2.3-10.1) and the median age at HA fitting for the non-screened group was 17.3 months (IQR: 7.5-25.9; Ching & Leigh, 2020). Of those in the screening group, 72% received HA fitting no later than 6 months of age, comparatively, only 32% were fitted in the same time frame in the non-screened group (Ching & Leigh, 2020). The EHDI program has improved detection and early intervention; however, there has always been a long-standing problem with non-adherence to follow up for diagnostic testing after an abnormal hearing screening. This non-adherence leads to significant delays in diagnosis and may result in life-long negative consequences on language development. A number of factors may influence infants receiving timely hearing diagnostic testing. There is evidence that patients from low-income and rural backgrounds have lower rates of adherence. Parental education, insurance status, and proximity to hearing specialists may also influence access and use of infant hearing healthcare (Boss et al., 2011; Pynnonen et al., 2016). There is a paucity of research examining social and economic factors driving disparities that impact access to infant hearing healthcare. This gap is important to address as there is strong evidence that minority racial and ethnic groups have a higher incidence of infant hearing loss (Lantos et al., 2018). In addition to the existing factors of adherence, the COVID-19 pandemic resulted in additional factors influencing EHDI care and amplified healthcare disparities. However, the pandemic's impact on EHDI programs is unknown. Therefore, we hypothesized that: (a) Racial and ethnic minority infants have a higher rate of non-adherence to diagnostic testing after an abnormal infant hearing screening test, as compared to non-Hispanic Caucasian infants. (b) Infants undergoing a diagnostic hearing test during the COVID-19 pandemic (March 1, 2020–September 30, 2020) have a higher non-adherence rate as compared to those who underwent testing before the pandemic.

Method

Patients

This study was approved by the University of Kentucky Institutional Review Board before data analysis. A retrospective review of the EHDI program database of Kentucky newborns was performed. The database documents hearing screening within the birthing hospitals and reports follow-up documentation for infants born in the state of Kentucky. The database reported 75,132 infants born between April 1, 2019 and September 30, 2020. Inclusion criteria included documented hearing screening result and state of Kentucky residency. Out of state infants and infants with no hearing screening were removed. The demographic information for the 71,206 infants who met inclusion criteria can be found in Table 1.

Table 1

Demographic Data

Demographic Data	Number of Participants (%)
Gender	
Male	36,187 (51%)
Female	35,019 (49%)
Race	
White	57,733 (81%)
BIPOC*	13,475 (19%)
Ethnicity	
Non-Hispanic	66,137 (93%)
Hispanic	4,938 (7%)
Language	
English	68,464 (96%)
Non-English	2,744 (4%)
Maternal Education	
Less than High school	8,805 (12%)
High school or greater	62,036 (88%)
Location	
Urban Counties	84 (70%)
Rural Counties	36 (30%)
COVID-19	
Born prior to COVID-19 pandemic	43,843 (62%)
Born during the COVID-19 pandemic	27,365 (38%)
Insurance	
Private	12,121 (45%)
Medicaid	14,556 (55%)
Total Participants	**71,206**

Note. Numbers do not add up to 100% of total in each group due to incomplete reporting in some of the data fields. BIPOC = Black, Indigenous, and people of color.

Data Acquisition/Organization

The following variables were extracted from the database and coded to facilitate analyses: race/ethnicity, language preference, date of birth, ZIP code, maternal education, hearing screening outcomes and timing, diagnostic hearing testing (if attended) and timing, as well as insurance status. The categorical data were numerically coded to allow for statistical analyses. The coded data were analyzed both before **(**April 1, 2019–February 28, 2020) and during (March 1, 2020–September 30, 2020) the COVID-19 pandemic.

Data Analysis

Statistical analysis was done using SPSS 28.0. These analyses evaluated the impact of racial, ethnic, social, and linguistic factors on diagnostic testing adherence. Univariate and multivariate logistic regression were

calculated to generate odds ratios and 95% confidence intervals of diagnostic testing adherence. Receipt of infant diagnostic testing by 3 months and the timing of testing were analyzed as a dependent variable in the analyses and the independent variables are as follows: race, ethnicity, language, zip code, maternal education, and insurance status. A separate multivariate logistic regression analysis was conducted to assess the factors influencing the presence of hearing loss in infants. Multivariate linear regression was conducted to assess the relationship of timing (in days after birth) of diagnostic testing (dependent variable) with the above listed independent variables. A *p*-value equal to or less than 0.05 was considered statistically significant. Control variables were employed to enhance internal validity.

Results

Database Characteristics

The EHDI database included 75,132 infants born in the state of Kentucky between April 1, 2019 and September 30, 2020. Of those, 3,926 infants were excluded because they were not Kentucky residents or they never received an infant hearing screening test, leaving 71,206 infants included in the study. The database included 36,187 (51%) males and 35,019 females (49%). Regarding race and ethnicity, 57,733 (81%) newborns were white and 13,475 (19%) were black, indigenous or persons of color (BIPOC). Most newborns were non-Hispanic, 66,137 (93%) infants identified in this category and 4,938 (7%) newborns were Hispanic. There were 43,843 (62%) infants born prior to the COVID-19 pandemic **(**April 1, 2019–February 28, 2020) and 27,365 (38%) infants born during the COVID-19 pandemic (March 1, 2020–September 30, 2020). There was a multitude of different languages spoken by the mothers of infants included in this study. Twenty-eight different languages were identified. However, 68,464 (96%) mothers spoke English, and 2,744 (4%) spoke a non-English language. The three most common non-English languages were Spanish (*n* = 1,988), Swahili (*n* = 186), and Arabic (*n* = 125). Maternal education was also available in the database. Maternal education data was not available on 365 dyads; however, of those with education data, mothers with an education level less than high school accounted for 8,805 (12%) participants and those with a high school degree or greater included 62,036 (88%) mothers. Seventy percent of the newborns lived in urban counties and thirty percent lived in rural counties. The demographics of the sample are included in more detail in Table 1.

Study Results

Overall, 1,385 infants had abnormal hearing screening tests in Kentucky during the study period (Tables 2 and 3). Overall, 82% of those infants received diagnostic testing within 3 months of age as recommended by the CDC. Conversely, 18% of the infants in our study failed to adhere to these guidelines. Of those infants, 5% received delayed diagnostic testing at nine to sixteen months of age. Infants born before the COVID-19 pandemic had an adherence rate of 84%. Adherence dropped to 80% percent during the pandemic. The multivariate logistic regression showed that infants during the COVID-19 pandemic had a 24% lower odds of hearing testing adherence (OR = 0.76, *p* = 0.05, 95%CI: 0.57–1) compared to those infants during the pre-pandemic period. Additionally, the logistic regression model found that infants who had mothers with a high school degree or higher had 1.50 times higher odds of EHDI adherence (OR = 1.50, *p* = 0.02, 95%CI: 1.06–2.17). Additionally, a multivariate linear regression showed that infants of mothers with a high school degree or greater presented on average 9.5 days earlier for testing (*p* = 0.003, 95%CI: -15.73– -3.32). Further details are included in Table 4. Separate multivariate logistic regression analyses were conducted to evaluate the factors on the presence of infant hearing (variables included COVID-19 era, gender of infant, insurance status, rural residence, race/ethnicity, and maternal education). Only maternal education was a significant variable. We found that infants who had mothers with at least a high school degree have a 1.63 times higher odds of having normal hearing on EHDI testing (OR = 1.63, *p* = 0.03, 95%CI: 1.06–2.51). We found that about 1% of those infants who received diagnostic hearing testing were diagnosed with a hearing loss. We observed a trend that male infants have 14.8% lower odds of hearing testing adherence and infants born in rural locations have 15.7% lower odds of hearing testing adherence; however, these trends were not statistically significant. We did not identify any significant association between hearing loss incidence and sociodemographics such as sex, insurance status, or race/ethnicity.

There was no evidence in the primary logistic regression analysis of differences in adherence based on race/ethnicity between pre- and post-pandemic time frames. We conducted separate multivariate logistic regression analyses to assess the association of Hispanic ethnicity, Swahili language status, and the COVID-19 era with diagnostic adherence as these are much small populations in the state of Kentucky and the relationship of these factors on diagnostic adherence may be overlooked in the larger logistic regression model. In the separate models, we identified that infant ethnicity is associated with EHDI adherence. The logistic regression found that Hispanic infants have 45% lower odds of EHDI adherence when compared to non-Hispanic infants (OR = 0.55, *p* = 0.03, 95%CI: 0.31–0.96; Table 5**)**. Using multivariate logistic regression, we also evaluated language impact on diagnostic testing adherence. Analyses showed infants of Swahili-speaking families (*n* = 9) have 90% lower odds of EHDI adherence (OR = 0.10, *p* = 0.001, 95%CI: 0.02–0.42; Table 5). This finding is important as Swahili was the 3rd most common language spoken by the mothers in this study.

Discussion

Bilateral hearing loss is the most common congenital disorder seen in the United States, occurring in 2 to 3 per 1000 newborns (CDC, 2020a). Untreated hearing loss can lead to delayed language, psychosocial, and academic development. Early detection of hearing loss initiates intervention earlier and improves overall outcomes. Studies have shown that newborn hearing screening is more reliable than detection by clinician and caregiver. A

Table 2
EHDI Screening and Diagnostic Data 2019–2020

	Failed Hearing Screening*	Adherence YES	Adherence NO	Average Time to Diagnosis
Pre-COVID-19				
Infants born: (April 1, 2019–February 28, 2020)	*n* = 850 (1.93%)	*n* = 711 (83.65%)	*n* = 139 (16.35%)	61.4 days
COVID-19				
Infants born: (March 1, 2020–September 30, 2020)	*n* = 535 (1.96%)	*n* = 427 (79.81%)	*n* = 108 (20.19%)	58.4 days

Note. Includes inpatient hearing rescreen and outpatient hearing screening.

Table 3
Hearing Loss Incidence in Kentucky

	No Hearing Loss	Unilateral Hearing Loss	Bilateral Hearing Loss
Pre-COVID-19			
Infants born: (April 1, 2019–February 28, 2020)	*n* = 43,410 (99.01%)	*n* = 277 (0.63%)	*n* = 156 (0.36%)
COVID-19			
Infants born: (March 1, 2020–September 30, 2020)	*n* = 27,032 (98.78%)	*n* = 199 (0.73%)	*n* = 134 (0.49%)

Table 4
*Multivariate Linear Regression on the Impact of Maternal Education on Timing of Adherence (*n *= 1,134).*

	Co-efficient	*p* value	95% Confidence Interval
Maternal Education of at least a High School Degree	**-9.53**	**0.003**	**-15.73– -3.32**
Presence of infant hearing loss	-7.23	0.06	-14.78–0.31
COVID-19 era	-1.63	0.56	-7.14–3.87
Gender of infant (male)	0.65	0.81	-4.79–6.08
Rural residence	0.65	0.83	-5.11–6.43
Black, Indigenous, or persons of color	-12.37	0.72	-80.00–55.24
Hispanic ethnicity	-4.37	0.38	-14.06–5.31

Note. Bolded factor is statistically significant at $p \leq 0.05$.

Table 5

Multivariate Linear Regression on the Impact of Maternal Education on Timing of Adherence

COVID-19 and Sociodemographic Factors Impact on Diagnostic Adherence

Multivariate logistic regression *n* = 1,378	***p* value**	**Odds ratio of Diagnostic Testing Adherence**	**95% Confidence Interval**
COVID-19 era	0.05*	0.76	0.57–1
Gender of infant (male)	0.26	0.85	0.64–1.12
Rural residence	0.24	0.83	0.62–1.12
Maternal age	0.27	0.98	0.96–1.01
Maternal education of at least a high school degree	0.02*	1.5	1.06–2.17
Presence of infant hearing loss	0.42	1.20	0.76–1.88
Black, Indigenous, or persons of color	0.19	0.81	0.59–1.10

**Statistically significant $p \leq 0.05$*

Ethnicity Impact on Diagnostic Testing Adherence

Multivariate logistic regression *n* = 1,375	***p* value**	**Odds ratio of Diagnostic Testing Adherence**	**95% Confidence Interval**
Hispanic ethnicity	0.04*	0.55	0.31–0.96
COVID-19 era	0.07	0.77	0.58–1.02

Ethnicity Impact on Diagnostic Testing Adherence

Multivariate logistic regression *n* = 1,362	***p* value**	**Odds ratio of Diagnostic Testing Adherence**	**95% Confidence Interval**
Swahili language	0.001*	0.10	0.02–0.42
COVID-19 era	0.07	0.77	0.58–1.02

controlled trial of 53,781 infants born in different hospitals was done to evaluate the efficacy of newborn hearing screening. In the study, newborns with hearing loss who received newborn hearing screening were more likely to be detected (OR = 5, 95% CI: 1–23) and receive treatment (OR = 8, 95%CI: 1.5–41) at an earlier age than those who did not receive screening ("Controlled Trial of Universal Neonatal Screening," 1998).

The delivery of timely diagnostic testing after failed screenings is essential for EHDI programs. There are many factors that may influence adherence after a failed screening test. Although EHDI programs seek to improve their programs to promote timely access and use, there continues to be barriers to services that perpetuate disparities. To ensure that infants are meeting EHDI guidelines equitably, health disparities need to be addressed. Health disparities are preventable health differences associated with economic, social, or environmental factors (CDC, 2020b). Contributing factors to health disparities include race, ethnicity, religion, socioeconomic-status, gender, and geographic location (U.S. Department of Health and Human Services, n.d.b).

A study in rural Kentucky identified that 25% of Appalachian children that fail UNHS are lost to follow up (Bush, Bianchi, et al., 2014). Appalachian infants are 1.5 times more likely to be lost to follow-up when compared to non-Appalachian children given the unique challenges facing these rural communities (Bush, Bianchi et al., 2014). Distance to hearing healthcare providers and the socioeconomic status are pervasive issues facing pediatric hearing healthcare (Noblitt et al., 2018; Bush et al., 2013; Bush, Osetinsky, et al., 2014; Bush et al., 2015; Elpers et al., 2016). The socioeconomic depression within this rural region has been a pervasive problem. The per-capita income for residents in rural Kentucky is $39,917 and the poverty rate is 19.2% compared with $52,445 per-capita income and poverty rate of 12.0% in urban areas of the state (Economic Research Service, n.d.). Additionally, the

unemployment rate in rural Kentucky is 5.1% compared to 4.4% in urban Kentucky (Economic Research Service, n.d.). In our study, adherence to diagnostic testing did not differ between rural and urban Kentucky counties nor did the incidence of hearing loss.

Research also shows that racial and ethnic minority groups experience more challenges to receiving quality healthcare and overall have worse health outcomes. According to data reported by the United States Census Bureau, in 2020, 24.9% of Hispanics did not have health insurance. Comparatively, 7.7% of Caucasian Americans did not have insurance. Additionally, 75% of Caucasian Americans have private health insurance, compared to only 50% of Hispanics (Keisler-Starkey & Bunch, 2021). In our study, Hispanic infants were less likely to meet EHDI guidelines than non-Hispanic infants. Additionally, infants of Swahili-speaking families had lower odds of EHDI adherence when compared with English-speaking families. There is evidence that the prevalence of infant hearing loss is disproportionately higher in infants from minority races and ethnicities and infants from lower socioeconomic backgrounds (Lantos et al., 2018). Non-white infants had a 2.45 higher odds of hearing loss and infants born and living in urban low-income neighborhoods also had a higher prevalence of hearing loss (Lantos et al., 2018), however we did not see this in our study.

The mechanisms underlying these disparities are unclear. Additional research on the type, degree, and impact of health disparities related to EHDI services needs to be conducted to create equitable access and use of hearing healthcare for all infants regardless of race, ethnicity or socioeconomic status. Providing adequate follow-up instructions prior to discharge from the hospital may improve adherence to diagnostic testing (American Speech-Language-Hearing Association, n.d.). Moreover, instructions should be provided in a family's native language and at an appropriate reading level. Providing accurate and accessible information may also improve patient-provider communication and promote the notion of patient-centered care. A patient-centric environment has also been shown to improve patient adherence to follow-up and treatment (Roumie et al., 2011). Simple institutional changes are attainable and could potentially make immediate impact and ameliorate disparity. Additionally, increasing access to hearing healthcare services will increase equity of care. There is a longstanding challenge with access to hearing specialists in underserved and rural communities. Federal and state leaders need to work together to improve transportation, physician shortage, and affordability of care, especially within the specialty of hearing. Community and institutional interventions such as these have not been assessed as they relate to the pandemic, but would be worth further study.

Maternal education had a positive impact on adherence. In our study, infants' mothers with a higher level of education had increased odds of EHDI adherence and were more likely to have normal hearing. Additionally, these infants presented on average 9.5 days earlier to testing. Although we controlled for variables in our regression analysis, certain factors that are unknown to us can be a proxy for maternal education. The association between maternal education levels and infant health outcomes has been studied in the past and a causal link between the two has been established. It has been hypothesized that higher maternal education allows for more autonomy to navigate health institutions, act on health knowledge, manage fertility, and overall improve child health. Although this is most likely true, the effect is not nearly as strong as predicted (Mensch et al., 2019). Therefore, it is very likely that factors such as poverty play a more significant role in health outcomes, and this should be the focus of community interventions.

The COVID-19 pandemic also impacted access to and use of hearing healthcare services for infants and their families. At the onset of the pandemic, many audiology facilities and services were considered non-essential and suspended operations. This certainly played a role in the ability of patients to adhere to recommended diagnostic testing (Cunningham et al., 2021). Many hospitals and clinics put more precautions in place and created stringent protocols to limit the spread of the virus, thus creating barriers to receiving care. Although the incidence of hearing loss was similar before and during the COVID-19 pandemic, the adherence to diagnostic hearing testing was different between the time periods. Adherence to diagnostic testing before the COVID-19 pandemic was 83.6% compared to 79.8% during the pandemic. Infants born during the pandemic had a 24.3% lower odds of diagnostic hearing testing adherence. Our results were reflected in another study in which 100% of parents in the study felt that the COVID-19 pandemic impacted access to timely hearing healthcare services for their children (Ayas et al., 2020). Additional research would be required to evaluate the effects of COVID-19 on testing adherence beyond the peak of the pandemic. Many barriers that were present during the pandemic such as limited clinic hours, clinic closures, limited access to public transportation and overall fear of leaving home improved as the pandemic continued. Moving forward, it is important that audiology testing be established as an essential practice for newborns. If another shutdown were to take place in the future, newborns should not receive delayed diagnostic testing or treatment.

The purpose of this research was to evaluate sociodemographic factors influencing adherence to EDHI diagnostic testing and the incidence of infant hearing loss before and during the pandemic. Results from this study found evidence that COVID-19, maternal education, race, ethnicity, and language affected adherence to follow-up diagnostic testing. Similar research has shown there are other barriers to health equity in addition to those addressed in this study (U.S. Department of Health and Human Services, n.d.a). To address these disparities and promote equity, further research needs to be done to test and evaluate strategies or interventions related to EHDI services. Several immediate and attainable recommendations were also made in this paper and we encourage EHDI programs to use this research in programmatic planning and intervention work.

There were limitations to this research study. As with many retrospective studies, there is the possibility of missing data and inaccurate reporting. Secondly, patients were excluded if they were not residents of Kentucky, if they failed to receive a hearing screening test, or if the result was not reported. Therefore, results of the study may not be generalizable since only Kentucky residents were included. Moreover, the excluded infants who never received screening may represent an important group who were not captured in this particular study. It is possible that this group of infants may have missed screening due to the similar disparities analyzed in this study. Lastly, our study included infants born between April 1, 2019 and September 30, 2020. We chose that end date to facilitate analyses in a timely manner. March 1, 2020 was the start date for the COVID-19 pandemic group in our analyses due to cases rising in the United States at that time. As a result, the pre-COVID-19 group included infants born over an 11 month span, compared to 7 months for the COVID-19 group. This discrepancy in data collection periods exists in large part due to the natural history of the pandemic as it relates to the time of the study. Despite this discrepancy, both groups had adequate sample sizes to complete statistical analysis.

Conclusion

The COVID-19 pandemic negatively impacted adherence to EHDI diagnostic services in Kentucky. Race, ethnicity, and language impacted adherence to testing. Maternal education was also found to influence adherence and infant hearing outcomes. Additional research is needed to identify other differences in infant hearing healthcare among different patient populations to improve adherence to diagnostic testing. However, results from this research could be used in programmatic planning and intervention work to promote hearing healthcare among vulnerable populations. Furthermore, continued awareness of these health disparities is necessary to achieve equitable access and use of hearing healthcare and improve health outcomes for all.

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A Retrospective Analysis of Newborn Hearing Screening Outcomes in Infants Whose Mothers were COVID-19 Positive during Pregnancy

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Abstract

Background: This study examined newborn hearing screening referral rates over a three-year period, including testing performed pre-pandemic (2019) and during the first two years (2020 and 2021) of the onset of the spread of COVID-19.

Method: This study conducted a retrospective analysis of 8,130 newborn hearing screening referral records from 2019–2021, obtained from a large regional level II trauma center. These records were generated from the universal newborn hearing screening process that used automated auditory brainstem response devices.

Results: There was no significant change in the number of infants screened for hearing loss and hearing screening outcomes between the pre-pandemic and pandemic years. A significant portion of infants born in this hospital were premature. A significant portion of infants born to mothers who had COVID-19 during pregnancy were premature. There was no statistically significant finding for the referral rate of babies born to mothers who reported COVID-19 positivity during pregnancy when compared to babies whose mothers did not report such exposure.

Conclusion: COVID-19 during pregnancy may not have a significant negative effect on overall newborn hearing screening outcomes. However, infants born to mothers who had COVID-19 during pregnancy can be premature and those premature infants demonstrated higher referral rates.

Keywords: COVID-19, hearing loss, hearing screening, premature

Acronyms: ABR = auditory brainstem responses; TEOAE = transient evoked otoacoustic emissions

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High-risk infants are identified as a group of newborns who have a history of complications (viral or bacterial infections or reduced oxygen levels) during pre-natal or post-natal stages, are premature (< 37 weeks), or have extremely low birth weight. High-risk infants have a greater chance of developing hearing loss (Salamy et al.,1989). The Joint Committee on Infant Hearing (JCIH, 2019) has recommended neonatal hearing screening by 1 month of age, for early identification by 3 months of age, and intervention by 6 months of age for hearing loss in all infants, especially the high-risk infants.

Viral infections during pregnancy can significantly damage the still-developing auditory system, leading to permanent sensory neural hearing loss (Cohen et al., 2014; Grosse et al., 2008; Morton & Nance, 2006). The virus SARS-CoV-2 is an infectious virus that can cause Corona disease (COVID-19). The direct effect of COVID-19 during pregnancy and complications secondary to COVID-19 during pregnancy can have a significant negative effect on the developing fetus (Villar et al., 2021). Pregnant women with COVID-19 may be at risk of delivering a premature (< 37 weeks) infant (CDC, 2022). It is well established in the literature that premature infants are at higher risk of developing hearing loss or may show prolonged maturation of the auditory system (Borenstein-Levin et al., 2021; Jiang et al., 2007; Ping & Jiang, 2013; Wang et al., 2020). Currently, it is not clear how the COVID-19 infection during pregnancy affects the developing auditory system.

Globally, researchers have explored whether COVID-19 during pregnancy has any effect on the still developing auditory system. Alan and Alan (2021) reported that COVID-19 during pregnancy may have some temporary effect on newborn hearing screening outcomes. The authors retrospectively analyzed the newborn hearing

screening outcomes (between April 2020 to December 2020 in Konya, Turkey) of infants whose mothers were COVID-19 positive during pregnancy. The newborn hearing screening was carried out by recording the auditory brainstem responses (ABR). Researchers analyzed 4,663 files, out of which 141 (3.02%) mothers reported COVID-19 during pregnancy. The authors' findings support that when compared to control groups, infants whose mothers were COVID-19 positive during pregnancy were more likely to fail the newborn hearing screening.

Other studies have reported that COVID-19 during pregnancy is not a risk factor for hearing loss. Mostafa et al. (2021) conducted a multi-center survey on the effect of COVID-19 during pregnancy on newborn hearing screening outcomes (between November 2020–April 1, 2021 in Giza, Egypt). The newborn hearing screenings were carried out using automated ABR analysis or transient evoked otoacoustic emissions (TEOAE). A total of 908 infants were included in the study. Thirty-four (3.74%) of these infants' mothers had reported having COVID-19 during pregnancy. A significant portion of infants whose mothers were positive for COVID-19 during pregnancy failed the first hearing screening. However, upon re-testing, only one of these infants failed the hearing screening. The authors concluded that COVID-19 during pregnancy was not a significant factor for hearing loss.

Oskovi-Kaplan et al. (2022) retrospectively analyzed neonatal hearing screening outcomes (between March 2020–October 2020 in Ankara, Turkey) of infants whose mothers had COVID-19 during pregnancy (n = 458) compared with 339 infants who were born before the pandemic. The newborn hearing screenings were conducted using automated ABR or TEOAE. Analysis of the data revealed that the risk of congenital hearing loss in infants with a history of COVID-19 was only 1.3% and the authors concluded that COVID-19 during pregnancy is not a risk factor for hearing loss. Ghiselli et al. (2022) investigated the possible association between COVID-19 during pregnancy and its effect on an infant's auditory system (between February 15, 2020 and February 15, 2021 in Piacenza, Italy). In this study, the infant's hearing was screened using automated TEOAEs before discharge and a detailed audiological evaluation (tympanometry, Distortion Product Otoacoustic Emissions, ABR) was completed at the age of four months. Findings revealed a significant portion of infants whose mothers had COVID-19 during pregnancy showed test results within normal limits at the age of four months. The authors concluded that COVID-19 during pregnancy is not a risk factor for hearing loss. Goulioumis et al. (2023) investigated whether COVID-19 was responsible for congenital hearing loss. The authors reviewed 111 medical records of infants whose mothers had COVID-19 during pregnancy (between February 2020–June 2022 in Patras, Greece). The newborn hearing screening was conducted using TEOAEs and automated ABRs. Findings revealed that all infants passed the newborn hearing screening, indicating that COVID-19 during pregnancy was not a risk factor for hearing loss.

Kosmidou et al. (2022) retrospectively analyzed the newborn hearing screening outcomes of 32 neonates born to unvaccinated COVID-19-positive mothers (between March 2020–January 2021 in Patras, Greece). The newborn hearing screening was conducted by recording TEOAEs before three months of age and again at the age of nine months. The authors did not find any evidence of hearing loss in infants who were born to mothers who had COVID-19 during pregnancy. More recently, Tanyeri Toker et al. (2023) retrospectively analyzed the neonatal hearing screening outcomes of 60,233 newborns (between March 2020 and May 2021) using an automated ABR. Out of 60,233 newborns, 570 infants' mothers (0.94%) had COVID-19 during pregnancy. The ABR screening was conducted three times (within 30 days after birth). A significant portion of infants whose mothers had COVID-19 during pregnancy failed the first and second ABR screenings when compared to control groups, but passed the third hearing screening. The authors concluded that COVID-19 during pregnancy may not have any significant negative effect on an infant's hearing. Fancello et al. (2023) conducted a systematic review to examine the association between maternal COVID-19 infection and congenital hearing loss. The literature review suggested no correlation between COVID-19 infection and congenital hearing loss. In summary, a significant portion of the existing literature suggests that COVID-19 during pregnancy is not a risk factor for hearing loss. These studies analyzed newborn hearing screening outcome data from the pandemic period (2020 and 2021).

Further evidence from different regions and a large data set is required to confirm that COVID-19 during pregnancy does not affect newborn hearing screening outcomes. In this study, we examined the following questions using a large data set from a regional hospital in Mississippi. The purpose was to investigate if there was a change in the number of infants screened for hearing at the hospital during the pandemic years (2020 and 2021) when compared to the pre-pandemic period (2019). If COVID-19 during pregnancy is a risk factor for hearing loss, then a significant portion of infants whose mothers had COVID-19 during pregnancy should fail the newborn hearing screening when compared to infants whose mothers did not have COVID-19 during pregnancy during the pandemic years (2020 and 2021). The authors also examined whether the infants who were born to mothers who had COVID-19 during pregnancy were premature.

Method

This study conducted a retrospective analysis of 8,130 newborn hearing screening referral records between January 1, 2019 and December 31, 2021 from a large regional level II trauma center in southern Mississippi. The universal newborn hearing screening was conducted using automated ABR devices (Natus, Algo 5). Each infant was screened at an appropriate interval following birth according to Early Hearing Detection Intervention guidelines published by the Mississippi State Department of Health (MSDH, 2019). The MSDH guidelines mandate

all newborn hearing screenings should be conducted in a quiet environment and the first screening should be conducted 12 to 24 hours after birth. If either ear refers, a second screening should be conducted at least four hours after the first screening, and as close as possible to discharge. In addition, hospitals in Mississippi have the option of a third in-patient screening should referrals occur with the second screening. If any infant ultimately refers in either ear during the final screening, they are referred for an in-depth diagnostic evaluation of auditory function.

De-identified data consisting of hearing screening results, birth year, COVID-19 test results during pregnancy, and gestational age were extracted from the electronic medical records. This research was approved by the Institutional Review Board of The University of Southern Mississippi.

Results

In total 8,130 [2019 (n = 2,787), 2020 (n = 2,673), and 2021 (n = 2,670)] patients' records were retrospectively analyzed. Out of 8,130, 23 infants' data were excluded because no newborn hearing screening test results were reported either due to death or transfer for specialized care to other facilities. The average gestation age was 38.31 weeks (SD = 1.84 weeks). In total, there were 1,146 premature infants which is higher than the national average. In 2019, 13.9% of infants were born prematurely compared with 10.2% for the national percentage. In 2020, 13.3% of infants born were premature, compared to the national average of all babies born prematurely of 10.1%. In 2021, this sample had 15.6% of babies born prematurely, compared to 10.5% for the nation.[1] In 2019, there were no reports of COVID-19 during pregnancy. During the pandemic period (2020–2021), 266 (3.27%) mothers reported having COVID-19 during pregnancy (see Table 1).

Table 1

*Descriptive Statistics of the Study Sample (*N *= 8,130)*

Variable	Categories	*n* (%)	Mean *(SD)*
Baby Birth Year	2019 2020 2021	2,787 (34.3%) 2,672 (32.9%) 2,671 (32.9%)	
Gestation Age (Weeks)			38.31 (1.84)
Premature (born prior to 37 weeks)	Yes No	1,159 (14.3%) 6,971 (85.7%)	
Newborn Hearing Screening Test Status	Fail Pass Fail-COVID Pass-COVID No-Test	397 (4.9%) 7,445 (91.6%) 19 (0.2%) 246 (3.0%) 23 (0.3%)	
COVID onset	Yes No	265 (3.3%) 7,865 (96.7%)	

[1]CDC (2021). Premature Birth. https://www.cdc.gov/reproductivehealth/features/premature-birth/

Bivariate analysis was used for data analysis (refer to Table 2), which revealed no significant change in the newborn hearing screening outcomes during the pandemic period (2020 and 2021) when compared to the pre-pandemic year (2019; $\chi^2 = 5.512$, $p = 0.064$). During the pandemic years (2020 and 2021), there were no significant differences in newborn hearing screening outcomes between infants whose mothers had COVID-19 during pregnancy and infants whose mothers did not have COVID-19 during pregnancy ($\chi^2 = 2.34$, $p = 0.126$).

Table 2

Bivariate Analyses Between the Outcome Variable and other Variables

Variable 1	Variable 2	*n* (%)	Test Statistic *p*
Birth Year	Hearing Test		
2019	Pass	2,621 (94.4%)	
	Fail	155 (5.6%)	
2020	Pass	2,552 (95.7%)	$\chi2 = 5.512$
	Fail	115 (4.3%)	$p = 0.064$
2021	Pass	2,518 (94.5%)	
	Fail	146 (5.5%)	
Birth Year	Premature		
2019	Yes	386 (13.9%)	
	No	2,401 (86.1%)	
2020	Yes	355 (13.3%)	$\chi2 = 6.676$
	No	2,317 (86.7%)	$p = 0.035$*
2021	Yes	418 (15.6%)	
	No	2,253 (84.4%)	
Premature	Hearing Test		
Yes	Pass	1,078 (93.4%)	
	Fail	76 (6.6%)	$\chi2 = 5.847$
No	Pass	6,613 (95.1%)	$p = 0.016$*
	Fail	340 (4.9%)	
COVID-19 (2020, 2021)	Hearing Test		
Yes	Pass	246 (92.8%)	
	Fail	19 (7.2%)	$\chi2 = 2.338$
No	Pass	7,445 (94.9%)	$p = 0.126$
	Fail	397 (5.1%)	
Premature (2020, 2021)	COVID-19		
Yes	COVID	51 (6.6%)	
	No-COVID	721 (93.4%)	$\chi2 = 5.110$
No	COVID	214 (4.7%)	$p = 0.024$*
	No-COVID	4,345 (95.3%)	

As expected, the premature infants showed a statistically significant relationship with poorer newborn hearing screening outcomes ($\chi^2 = 5.847$, $p = 0.016$). If a baby was born before 37 weeks (premature), results demonstrated these infants were nearly 40% more likely to fail the hearing screening in one or both ears, (Odds = 1.37 [1.06 – 1.77], $p = 0.016$), which is consistent with published

literature. In addition, prematurity showed a statistically significant relationship with COVID-19 ($\chi^2 = 5.110$, $p = 0.024$). Data from infants who were born during the pandemic period (2020 and 2021) was used for this analysis. If a mother had COVID-19 during pregnancy in the pandemic period, they were more likely (21%) to have a premature baby (Odds = 1.21 [1.04 – 1.41], $p = 0.014$).

Discussion

In this research, the effects of COVID-19 during pregnancy on newborn hearing screening outcomes were examined. The current dataset (2019, 2020, and 2021) was extracted from a large hospital in southern Mississippi. Data revealed no significant changes in the number of infants screened for hearing loss between the pre-pandemic year (2019) and the pandemic years (2020 and 2021), indicating that the COVID-19 pandemic did not significantly affect newborn hearing screenings or the birth rate in this hospital. Similar findings have been reported by Roush et al. (2022).

During 2019, there were no reports of COVID-19 during pregnancy. During the pandemic years of 2020 and 2021, 266 mothers had COVID-19 during pregnancy. There were no significant changes in the referral rates on newborn hearing screens of infants whose mother had COVID-19 during pregnancy when compared to infants whose mothers did not report such exposure. The findings of this study are similar to existing literature suggesting that COVID-19 during pregnancy may not be a risk factor for a fail on a newborn hearing screening.

The present data set also showed that a significant portion of infants who were born in this regional hospital were premature. The percentage of premature infants is higher than the national average reported by the CDC (2022) in all three years (2019, 2020, and 2021). This is an incidental finding and mirrors published literature (Israel, 2020).

Additionally, the current data set demonstrated that a statistically significant portion of infants who were born to mothers who had COVID-19 during pregnancy were premature. Complications due to COVID-19 during pregnancy might have led to the premature birth of these infants. Similar suggestions are reported by the CDC concerning COVID-19 and its potential effects on the gestational cycle (2022).

A significant portion of studies have focused on the presence or absence of hearing loss in infants whose mother had COVID-19 during pregnancy. The present study also confirmed that COVID-19 is not a risk factor for a failed newborn hearing screening outcome in the sample included. However, this study did show significantly higher referral rates in infants that were born prematurely, which has not been reported in the studies we reviewed.

It is important to continue to monitor those infants identified as high risk for delayed or atypical auditory maturation, even with a passed newborn hearing screening, as the literature supports the fact that high risk infants can have an atypical ABR in the absence of hearing loss (Jiang et al. 2007; Ping & Jiang, 2013; Borenstein-Levin et al., 2021; Wang et al. 2020). A recent study (Ankmnal-Veeranna et al., 2022) demonstrated that in the absence of elevated electrophysiological thresholds, infants whose mothers had COVID-19 during pregnancy showed significantly prolonged wave V of the ABR, when compared to control groups, suggesting an atypical functioning of the auditory system. Infant subjects in that study were born in the same geographical region as those in the present study. It is not clear whether the abnormal ABRs observed in infants whose mothers had COVID-19 during pregnancy will change with maturation. Therefore, further monitoring of the auditory system maturation is warranted, and this should be a focus of research. Infant subjects who failed newborn hearing screenings in the current study should be monitored to make sure that the auditory function is intact.

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