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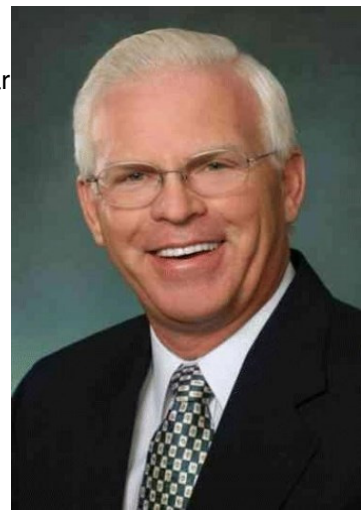
IN MEMORIAM

Leslie Robert Schmeltz Jr.
05/18/1943 - 07/23/2024

On July 23, 2024 the Early Hearing Detection and Intervention (EHDI) community lost one of its earliest and most ardent supporters. I first met Les in the summer of 1994. We were both attending a conference in Colorado where I had been invited to speak about implementing hospital-based universal newborn hearing screening programs and the recent recommendation by the ***National Institutes of Health Consensus Statement on Early Identification of Hearing Impairment in Infants and Young Children*** that "universal [hearing] screening be implemented for all infants within the first 3 months of life." At a reception that evening, Les sought me out to learn everything he could about implementing a universal newborn hearing screening program in Bettendorf, Iowa. I had never heard of Bettendorf, but it was clear to me that Les would ensure that Bettendorf had a universal newborn hearing screening program – with or without our help. He was polite and respectful, but he had a goal, and it was obvious after just a few minutes that he would not rest until that goal was achieved. Over the next 29 years I came to understand that Les lived his life that way.

Les was working as a school audiologist in Bettendorf, Iowa, but recognized how much more effective we could be if we identified children who were deaf or hard of hearing and began providing services in the first few months of life. As the Director of the National Center for Hearing Assessment and Management (NCHAM) at Utah State University, I was looking for people with a desire to help infants and young children who were deaf or hard of hearing and it was easy to see that Les was a tireless advocate and worker. No task was too big or too small for him as he traveled throughout the region helping hospitals implement newborn hearing screening programs. He always coordinated his work with NCHAM and in 2000 he became a member of NCHAM's first National Network of EHDI Experts. He continued working as a key member of NCHAM's team until his death.

After working for the Area Education Agency in Bettendorf, Iowa for over 30 years, Les relocated to Mesa, AZ to earn his doctorate of Audiology degree from A. T. Still University of Health Sciences in 2001. In 2005 he accepted a position as an Associate Professor of Audiology at A. T. Still University of Health Sciences. His specialties included educational audiology, early hearing detection and intervention (EHDI), pediatric audiology, and assistive hearing devices. He was also a clinical coordinator for student externships outside the Phoenix metro area. He served as the President of the Educational Audiology Association and wrote extensively and presented on numerous topics related to EHDI and educational audiology. He had a particular interest in developing information management systems for tracking infants who are deaf or hard of hearing and providing resources for researchers and clinicians providing services to these children.



In 2020 Les *retired* from A. T. Still University, but he never really retired. He was always looking for ways to improve and expand EHDI programs. Two of the best examples of his contributions that continued beyond his retirement as largely volunteer efforts are the *EHDI eBook* and the *Journal of Early Hearing Detection and Intervention (JEHDI)*. In both cases, he recognized a need, proposed a solution, and served as the Editor-in-Chief from the first publication until he passed away. The *EHDI eBook* was designed to provide people unfamiliar with EHDI programs with an overview of the basic components of the EHDI system. It has been used in hundreds of graduate classes and has become a trusted reference for professionals working in EHDI and family members who have a child who is deaf or hard of hearing. The *Journal of Early Hearing Detection and Intervention (JEHDI)* was established in 2016 as a semi-annual scholarly peer-reviewed online publication dedicated to advancing EHDI programs by publishing articles that describe current research, evidence-based practice, and standards of care. His careful and dedicated work has guided *JEHDI* from its inception to the point that it is now one of the most important sources of peer-reviewed information about EHDI throughout the world. As of today, articles published by *JEHDI* have been downloaded more than 160,000 times by people in 194 countries.

Everyone who knew Les was struck by his playful sense of humor and kind demeanor. He left this world a better place and we miss him deeply. But, his accomplishments and the lessons we learned from him will carry on.

With appreciation and admiration,

Karl R. White
Director, National Center for Hearing Assessment and Management
Utah State University

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A Review of Social Media Platforms of State Early Hearing Detection and Intervention Programs in the United States

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Abstract

Purpose: Social media tools are increasingly used to support good health practices. Almost every Early Hearing Detection and Intervention (EHDI) program has a website, and it would be useful to know how they are using social media. This article analyzes how social media is being used by Early Hearing Detection and Intervention programs in the United States, as well as how frequently.

Method: The present study analyzed existing social media handles of the Early Hearing Detection and Intervention programs. The search for social media accounts and data extraction were carried out between January 1, 2022 and July 31, 2023.

Results: Eight (14.8%) EHDI program websites had Facebook accounts/pages, three (5.6%) had Twitter accounts, two (3%) used YouTube, and 1 (1.9%) had Instagram. Overall, for most states with a social media account, the accounts were inactive, had limited content, or had limited followers/subscribers. Generally, the use of social media accounts was very limited by state EHDI programs.

Conclusion: The findings of the present study highlight the need for EHDI programs to have more active social media accounts to captivate and cater to the needs of the present digital generation.

Keywords: Social media, EHDI, Internet, Health

Acronyms: EHDI = early hearing detection and intervention

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Website use among adults in the United States has grown dramatically from 52% in 2000 to 93% in 2021 (Pew Research Center, 2021). The use of social media has also grown even more rapidly in recent years across all age groups from 27% in 2009 to 86% in 2019 (Chou et al., 2021). A recent national poll found that one in every five Americans used social media to search health related information (Gordon, 2021). The health sector has adapted to this change and accepted it as one of the means to reach their target audiences. Griffis and colleagues (2014) reported the use of social media varied across different U.S. hospitals with most of them using Facebook (94.41%) and Twitter (50.82%). There is a plethora of information available on different websites and social media handles. These can serve as a useful tool to spread awareness among the masses as well as promote the importance of early hearing detection and intervention. Social media can also help foster better engagement with the audience and can also improve networking opportunities with the professionals involved in the Early Hearing Detection and Intervention (EHDI) programs.

Use of social media has been widely studied by researchers in various fields including hearing sciences. Social media applications such as YouTube, Facebook, Instagram, and Twitter are among the tools that have been frequently studied for their contents related to hearing health (Crosier et al., 2016; A. K. Deshpande et al., 2019; Gunjawate et al., 2021; Manchaiah et al., 2018; Manchaiah, Bellon-Harn, Godina, et al., 2020; Ni et al., 2020; Rotondi et al., 2019). The areas studied include tinnitus (Choudhury & Renken, 2018; A. K. Deshpande et al., 2018; Manchaiah et al., 2018; Ni et al., 2020; Ulep et al., 2022), hearing aids (Chundu et al., 2021; Manchaiah, Bellon-Harn, Michaels, et al., 2020), infant hearing loss (Gunjawate et al., 2021), central auditory processing disorder (S. B. Deshpande et al., 2019), Hyperacusis (A. K. Deshpande et al., 2019), and auditory hallucinations (Crosier et al., 2016).

In a study exploring media use by older adults with hearing loss found that 54% of the participants used the internet for their hearing health care needs (Manchaiah, Bellon-Harn, Kelly-Campbell, et al., 2020). In two studies investigating Twitter usage for hearing loss & tinnitus, it was noted that out of the 100 most active accounts the majority were accounts of commercial companies instead of individuals (Crowson et al., 2018; Ni et al., 2020). Twitter was mainly used as a medium for advocacy and sharing of personal experiences and information. Studies about hearing aids and tinnitus have reported YouTube and Twitter platforms to have the highest level of activity (Choudhury et al., 2017; Choudhury & Renken, 2018). Deshpande and colleagues (2018) explored tinnitus related information on social media platforms and reported that the greatest activity was seen on Facebook pages, followed by YouTube. Further, these platforms were also used to learn more about tinnitus, seek support, advocate, and connect with other professionals. A varied degree of misinformation is reported across the different social media. Gunjawate and colleagues (2021) studied the YouTube videos about infant hearing loss with more than 1,000 views. The most common theme of the videos was diagnosis of hearing loss followed by EHDI and universal newborn hearing screening.

Since 1993, state and federal governments in the United States have invested substantially in improving children's hearing health. The internet and social media are frequently used to find hearing health, but most information about hearing health currently on the internet is focused on adults. Every state has an Early Hearing Detection and Intervention (EHDI) program that aims to detect childhood hearing loss early and provide accurate information to parents. Federal grants have been awarded to EHDI programs in every state (NCHAM, n.d.). One of the conditions of those grants is for every state to have a website that is updated and has relevant information.

Although the use of social media to support good health has expanded dramatically in recent years, it is important that the information conveyed by social media is evidence-based, reliable, valid, and follows ethical principles. Almost every EHDI program has a website, and it would be useful to know how they are using their social media. The purpose of this article is to analyze how EHDI programs are using social media, as well as how frequently.

Method

The present study aimed to explore how state EHDI websites in the United States are incorporating the use of social media. A list of EHDI/Universal Newborn Hearing Screening program websites by state for all the 59 states and territories (hereafter referred to as "states") were obtained from The National Center for Hearing Assessment and Management (NCHAM) website (NCHAM, n.d.). These websites were inspected to identify the social media account/page for the respective state. The states that did not have a dedicated social media account/page for EHDI on their health department website were excluded. Only those social media account/pages were considered that were linked to the health department website. Social media account/pages on Facebook, YouTube, Twitter, or Instagram were included for analysis.

This search was carried out between January 2022 and July 2023. The names and lists at all the stages were populated on Microsoft Excel by the first author (RR) and cross-checked for accuracy by another author (KW).

The following data were extracted from each social media account:

- information on name of the handle
- date of creation of account
- number of followers/subscribers and following
- number of posts/tweets and views
- contents on the social media handle between January1, 2022 and July 31, 2023.

The data was extracted by the first author (RR). To reduce the bias and ensure accuracy, 30% of the extracted data was verified by another author (KW).

Results

In this study, the websites of EHDI programs and social media accounts were analyzed. A total of 54 EHDI websites across different states and territories were included in the present study. The 5 states and territories (American Samoa, Mariana Islands, Marshall Islands, Palau, and Virgin Islands) not included did not have their own EHDI website. Websites that included links to social media with their details are in Table 1.

Eight (14.8%) EHDI program websites had Facebook account/pages, three (5.6%) had Twitter accounts, two (3%) had YouTube accounts, and one (1.9%) had an Instagram account. Overall, for most states with a social media account, the accounts were inactive or had limited followers/subscribers. Facebook was among the most active social media site linked to EHDI websites. Most of the Facebook accounts contained contents that were reposted from other accounts. The other content shared included information about upcoming events, webinars, newsletters, conferences, and holiday announcements. Two out of three (Arizona, Guam) Twitter accounts were inactive. Of the two states with links to YouTube (Illinois and South Dakota), both had limited content and subscribers and were inactive (most recent videos being posted in 2017 and 2021, respectively). South Dakota and Utah had an Instagram account which was created in May 2022 and October 2020, respectively. South Dakota had 15 posts, 88 followers, and was followed by 61 people. Whereas, Utah Instagram had 163 posts, 370 followers, and 120 following.

Discussion

Websites and social media accounts have become a primary source of information for medical needs. In the United States, 93% of adults use the internet (Pew Research Center, 2021) and 72% of those adult internet users browsed the internet for health-related issues related to specific diseases and their management (Fox, 2014).

Table 1

Social Media Accounts	States	Name of Handle	Date of Account Creation	Number of Followers/ Following Members/ Subscribers	Numbers of Posts/ Tweets/Views
Facebook	Arizona	AZEHDI	10/2009	289 followers	15 posts
	California	LEAD-K Family services-parent group	03/2021	58 members, private closed group	N/A
	Colorado	(@coehdi), Colorado EHDI Alliance	04/2020	166 followers	49 posts
	Guam	Guam Early Hearing Detection & Intervention	12/2011	305 followers & 8 following	52 posts
	Massachusetts	MassNewbornHearingScreening	12/2009	1K followers	307 posts
	North Dakota	North Dakota Early Hearing Detection and Intervention Program (ND EHDI)	04/2010	240 followers	398 posts
	Rhode Island	Rhode Island Early Hearing Detection Intervention Program	05/2014	361 followers, 205 following	152 posts
	South Dakota	SD EHDI Collaborative	04/2020	188 followers, 48 following,	111 posts
Twitter	Arizona	ArizonaEHDI	11/2014	37 following, 19 followers	18 tweets, mostly retweet of other organization; last tweet, 2015
	Guam	GuamEHDI	12/2011	14 followers, 4 following	348 tweets, last tweet, June 30, 2019
	North Dakota	NDEHDI_NDCPD	03/2017	1072 following, 325 followers	421 tweets
YouTube	Illinois	@illinoisehdi4414	11/2013	68 subscribers	120 videos; last post, August 30, 2017
	South Dakota	sdehdicollaborative2249	01/2021	3 subscribers	6 videos
Instagram	South Dakota	sd_ehdicollaborative	05/2022	102 followers, 63 following	18 posts
	Utah	utahehdi	10/2020	370 followers, 120 following	163 posts

The use of social media has emerged as an important source of information and guidance for different audiological conditions such as hearing loss, tinnitus, central auditory processing disorder, and hearing aids. The internet is increasingly being used for gaining knowledge, reaching larger audiences, and improving interactions and engagements (Choudhury et al., 2017; Choudhury & Renken, 2018; Chundu et al., 2021; Crowson et al., 2018; Deshpande et al., 2018; S. B. Deshpande et al., 2019; Gunjawate et al., 2021; Manchaiah, Bellon-Harn, Kelly-Campbell, et al., 2020; Ni et al., 2020; Ulep et al., 2022).

Only two of the EHDI websites (Illinois & South Dakota) linked to YouTube accounts and both of those had been inactive for two or more years. In a study of YouTube videos that were relevant to hearing health for infants and young children, Gunjawate et al. (2021) found that most of the videos (58%) were created by organizations while 5.48% of the videos were created by consumers and had the most views. The remaining videos were blogposts or television sources such as news. These videos were rated as poor for actionability and understandability. It is possible that the overall actionability and understandability of the YouTube videos for children's hearing health would improve if organizations are involved with creating more content.

Three state EHDI programs had links to Twitter on their websites (Arizona, Guam, North Dakota) but only North Dakota was active. For all three states, most of the twitter

content involved retweeting posts from other accounts which included news and events. In analyzing the use of social media in the context of hearing health, Crowson et al. (2018) concluded that accounts belonging to commercial organizations outnumbered those belonging to individuals by a 2:1 ratio. Thus, it is puzzling why more EHDI programs don't have social media accounts.

Facebook accounts were linked to websites for eight of the EHDI programs (California, Colorado, Arizona, Massachusetts, Rhode Island, South Dakota, North Dakota, and Guam). In a recent review (Ulep et al., 2022) examining the use of social media (Facebook, Twitter, and YouTube) in the context of hearing health, the authors concluded that misinformation was a matter of concern and an area that needed to be addressed. According to Deshpande and colleagues (2018) the various platforms had misinformation as follows: Facebook group (44.4%); Facebook pages (42.7%); Twitter (34.6%), and YouTube (21.5%).

Most of the content across all social media was found to be somewhat informative, but many of the authors have reported lack of educational value and misinformation (Deshpande et al., 2018; Kahn et al., 2021; Ulep et al., 2022). However, in comparison from the findings of the present study, it can be noted that the information on the EHDI websites and their social media handles was relevant and backed with evidence-based practices and recommendations. This could be the case as these websites and their social media handles are maintained by professionals trained and involved in EHDI. Overall, there is a need for the majority of EHDI programs to keep up-to-date information on their websites and come up with a greater number of social media accounts along with regular updates, posts, and active users to reach more people. Further studies are needed to determine and find means to mitigate the causes of the lack of social media tools available through EHDI websites.

Conclusion

Websites and social media are the backbone of the digital world and disseminate information, including health-related information. Although professional websites are a primary source of information, recent trends show that social media platforms such as Facebook, YouTube, Instagram, and Twitter have also been widely used to convey health-related information. In the present study, most of the states had their own websites which had information related to Early Hearing Detection and Intervention. However, very few of their websites were linked to social media accounts which were active and regularly updated. The findings of the present study highlight the need for these programs to have more active social media accounts to captivate and cater to the needs of the present digital generation.

References

Chou, W. Y. S., Gaysynsky, A., Trivedi, N., & Vanderpool, R. C. (2021). Using social media for health: National data from HINTS 2019. *Journal of Health Communication*, *26*(3), 184–193. https://doi.org/10.1080/10810730.2021.1903627

Choudhury, M., Dinger, Z., & Fichera, E. (2017). The utilization of social media in the hearing aid community. *American Journal of Audiology*, *26*(1), 1–9. https://doi.org/10.1044/2016_AJA-16-0044

Choudhury, M., & Renken, L. (2018). The "buzz" about tinnitus: A social media study. *The Hearing Journal*, *71*(5), 20. https://doi.org/10.1097/01.HJ.0000533803.94719.58

Chundu, S., Allen, P. M., Han, W., Ratinaud, P., Krishna, R., & Manchaiah, V. (2021). Social representation of hearing aids among people with hearing loss: An exploratory study. *International Journal of Audiology*, *60*(12), 964–978. https://doi.org/10.1080/14992027.2021.1886349

Crosier, B. S., Brian, R. M., & Ben-Zeev, D. (2016). Using Facebook to reach people who experience auditory hallucinations. *Journal of Medical Internet Research*, *18*(6), e160. https://doi.org/10.2196/jmir.5420

Crowson, M. G., Tucci, D. L., & Kaylie, D. (2018). Hearing loss on social media: Who is winning hearts and minds? *The Laryngoscope*, *128*(6), 1453–1461. https://doi.org/10.1002/lary.26902

Deshpande, A. K., Deshpande, S. B., & O'Brien, C. (2018). A study of social media utilization by individuals with tinnitus. *American Journal of Audiology*, *27*(4), 559–569. https://doi.org/10.1044/2018_AJA-18-0033

Deshpande, A. K., Deshpande, S. B., & O'Brien, C. A. (2019). Hyperacusis and social media trends. *Hearing, Balance and Communication*, *17*(1), 1–11. https://doi.org/10.1080/21695717.2018.1539321

Deshpande, S. B., Deshpande, A. K., O'Brien, C. A., & McMonagle, K. L. (2019). A study of the portrayal of information related to (central) auditory processing disorder on social media. *Hearing, Balance and Communication*, *17*(2), 134–144. https://doi.org/10.1080/21695717.2019.1591004

Fox, S. (2014). *The social life of health information*. Pew Research Center. https://www.pewresearch.org/short-reads/2014/01/15/the-social-life-of-health-information/

Gordon, D. (2021, October 6). *1 in 10 Americans turn to social media for health information, new survey shows*. Forbes. https://www.forbes.com/sites/debgordon/2021/10/06/1-in-10-americans-turn-to-social-media-for-health-information-new-survey-shows/

Griffis, H. M., Kilaru, A. S., Werner, R. M., Asch, D. A., Hershey, J. C., Hill, S., Ha, Y. P., Sellers, A., Mahoney, K., & Merchant, R. M. (2014). Use of social media across US hospitals: Descriptive

analysis of adoption and utilization. *Journal of Medical Internet Research,16*(11), e264. https://doi.org/10.2196/jmir.3758

Gunjawate, D. R., Ravi, R., Bellon-Harn, M. L., & Manchaiah, V. (2021). Content analysis of YouTube videos addressing infant hearing loss: A cross-sectional study. *Journal of Consumer Health on the Internet*, *25*(1), 20–34. https://doi.org/10.1080/15398285.2020.1852387

Kahn, C. I., Wang, R., Shetty, K., Huestis, M. J., Cohen, M. B., & Levi, J. R. (2021). Assessing the educational quality of Facebook videos as an informative resource on otitis media. *Otolaryngology–Head and Neck Surgery*, *164*(1), 110–116. https://doi.org/10.1177/0194599820933887

Manchaiah, V., Bellon-Harn, M. L., Godina, I. M., Beukes, E. W., & Vinay. (2020). Portrayal of hearing loss in YouTube videos: An exploratory cross-sectional analysis. *American Journal of Audiology*, *29*(3), 450–459. https://doi.org/10.1044/2020_AJA-19-00121

Manchaiah, V., Bellon-Harn, M. L., Kelly-Campbell, R. J., Beukes, E. W., Bailey, A., & Pyykkő, I. (2020). Media use by older adults with hearing loss: An exploratory survey. *American Journal of Audiology*, *29*(2), 218–225. https://doi.org/10.1044/2020_AJA-19-00039

Manchaiah, V., Bellon-Harn, M. L., Michaels, M., Swarnalatha Nagaraj, V., & Beukes, E. W. (2020). A Content analysis of YouTube videos related to hearing aids. *Journal of the American Academy of Audiology*, *31*(09), 636–645. https://doi.org/10.1055/s-0040-1717123

Manchaiah, V., Ratinaud, P., & Andersson, G. (2018). Representation of tinnitus in the US newspaper media and in Facebook pages: Cross-sectional analysis of secondary data. *Interactive Journal of Medical Research*, *7*(1), e9. https://doi.org/10.2196/ijmr.9065

National Center for Hearing Assessment and Management. (n.d.) *Home.* https://www.infanthearing.org/index.html

Ni, J., Bellon-Harn, M. L., Zhang, J., Li, Y., & Manchaiah, V. (2020). Twitter usage using common reference to tinnitus. *American Journal of Audiology*, *29*(2), 206–217. https://doi.org/10.1044/2020_AJA-19-00055

Pew Research Center. (2021, April 7). *Internet/Broadband Fact Sheet*. https://www.pewresearch.org/internet/fact-sheet/internet-broadband/

Rotondi, L., Zuddas, M., Marsella, P., & Rosati, P. (2019). A Facebook page created soon after the amatrice earthquake for deaf adults and children, families, and caregivers provides an easy communication tool and social satisfaction in maxi-emergencies. *Prehospital and Disaster Medicine*, *34*(02), 137–141. https://doi.org/10.1017/S1049023X19000086

Ulep, A. J., Deshpande, A. K., Beukes, E. W., Placette, A., & Manchaiah, V. (2022). Social media use in hearing loss, tinnitus, and vestibular disorders: A systematic review. *American Journal of Audiology*, *31*(3S), 1019–1042. https://doi.org/10.1044/2022_AJA-21-00211

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Understanding Barriers to Timely Enrollment of Early Intervention Services for Children who are Deaf and Hard of Hearing

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Abstract
The Joint Committee on Infant Hearing guidelines recommend that children who are deaf and hard of hearing should begin early intervention by six months of age. However, prior work has revealed a substantial percentage of children who receive a diagnosis of hearing loss by three months of age, but do not enroll in early intervention by six months of age (Grey et al., 2022). To further understand barriers to enrollment in early intervention for these families, we completed qualitative semi-structured interviews with 10 caregivers whose children were diagnosed with hearing loss by three months of age but did not begin early intervention by six months. We recruited from participants in Grey et al. (2022). Interviews were coded using the Bioecological Model of Human Development (Bronfenbrenner & Morris, 2006) as a guiding framework. The interviews revealed widespread barriers encountered by families of children who are deaf and hard of hearing across ecological systems, ranging from child characteristics to macro-level issues like insurance coverage. To ensure that all children who are diagnosed with hearing loss have timely access to early intervention, changes to current policy and practice are needed across multiple ecological systems.

Keywords: early intervention, barriers, family perspectives, qualitative design, children who are deaf and hard of hearing

Acronyms: DHH = deaf or hard of hearing; EHDI = early hearing detection and intervention; FCEI = family-centered early intervention; UNHS = universal newborn hearing screening

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The average age at which children who are deaf and hard of hearing (DHH) receive an audiological diagnosis and enroll in early intervention services has steadily decreased since the advent of universal newborn hearing screening (UNHS) programs. The Early Hearing Detection and Intervention (EHDI) system was created to accelerate the screening, diagnosis, and intervention process for children who are DHH. The current EHDI guidelines recommend that infants should receive a hearing screening by one month of age, be diagnosed with a hearing loss by three months of age, and enroll in early intervention by six months of age (1-3-6 guidelines; Joint Committee on Infant Hearing [JCIH], 2019).

For the past decade, various agencies and research teams have examined the percentage of children who are DHH who have met the screening, diagnosis, and intervention timeline outlined in EHDI's 1-3-6 guidelines. Based on data collected in 2020 from 50 states and seven territories of the United States, the Center for Disease Control and Prevention (CDC) reported 98.2% of newborn babies received a UNHS before one month of age (CDC, 2020). Of the infants who failed their UNHS, only 60.0% received a diagnosis of hearing loss or no hearing loss by three months of age, and 61.4% of those diagnosed with hearing loss enrolled in early intervention services before six months of age.

Research samples are consistent with this population-based data. Despite UNHS being routine practice, it appears that timely follow up for diagnosis and intervention is not as common. Across two research samples,

Yoshinaga-Itano et al. (2017) and Grey et al. (2022) reported that only 58% and 57%, respectively, of children who are DHH met all three EHDI benchmarks. McLean et al. (2019) reported that a little more than 50% of those children diagnosed with a hearing loss began early intervention services before their six-month birthday, and Holte et al. (2012) found that 75% of children who are DHH were enrolled in early intervention by six months of age. Out of 76 children who are DHH across 31 states who were enrolled in a longitudinal study of language and literacy acquisition, Grey et al. (2022) reported that 95% received a hearing screening by one month of age, 70% were diagnosed with a hearing loss by three months of age, and only 62% of those who received a hearing loss diagnosis began intervention by six months of age.

Therefore, it appears that a substantial percentage of children who are DHH, 25–50%, do not begin early intervention services by the recommended 6 months of age. It is crucial to understand barriers encountered by families of children who are DHH after a failed newborn hearing screening. We have previously reported on barriers during the diagnosis process in Reynolds et al. (2023). The purpose of this study, therefore, was to understand families' barriers to timely enrollment in early intervention services. We approached our research questions using Bronfenbrenner's Bioecological Model of Human Development (Bronfenbrenner & Morris, 2006) as our theoretical lens.

The Bioecological Model of Human Development

The Bioecological Model of Human Development (Bronfenbrenner & Morris, 2006) is the culminating model of the work of Urie Bronfenbrenner, who proposed the Ecological Model of Human Development originally in 1979. In this model, an individual's development is affected by a series of nested systems beginning with the individual and branching outwards into the extended community and broader culture (Bronfenbrenner, 1979). The core of the Bioecological Model is the individual and includes specific personal traits that influence the course of their development. These traits can include birth complications, diagnoses, and temperament. Working outwards, the microsystem is characterized by the relationships, environments, and communities that the developing individual has direct interactions with on a regular basis. Interactions between components in the microsystem make up the mesosystem. The mesosystem categorizes the complex interrelations that exist between the immediate communities influencing the individual. An example interaction in the mesosystem might encompass how the caregiver's work schedule relates to a child's school; for example, the work schedule might make it difficult to schedule an appointment to talk with the child's teacher or a child's ability to participate in extracurricular activities. Beyond the mesosystem, the exosystem consists of the organizations and environment outside of the immediate community that hold influence on the individual, but do not directly include the individual. Within the exosystem exists a broad range of formal and informal social structures. A caregiver's workplace, the home and neighborhood of the child, the media a child may be exposed to, and ruling government bodies (local, state, and federal) are all considered influencing factors that fall into the exosystem. The final sphere is the macrosystem, which is characterized by overarching societal systems, including but not limited to the political, economic, educational, and healthcare systems used by the culture in which the developing individual lives.

The Bioecological Model of Human Development has been used to guide qualitative research since it was first proposed and is especially prevalent in research concerning early education and intervention (Swick & Williams, 2006; Tudge et al., 2021). The model's value lies in its holistic approach, which allows investigators to examine the impact and relationship of multiple factors on a child's development (Eriksson et al., 2018; Swick & Williams, 2006). Barriers of interest in the present study, for example, may exist across any of the systems in the Bioecological Model, some of which can be addressed by the caregiver and some of which lie out of their control (Awad et al., 2019; Holte et al., 2012; Shulman et al., 2010).

Prior Work on Barriers to Hearing Healthcare Services

Recently, there has been increased attention to barriers to meeting EHDI's three-month diagnosis benchmark (Bush et al., 2015; Holte et al., 2012; Kingsbury et al., 2022; McLean et al., 2019; Richlin et al., 2023; Shulman et al., 2010). From this work, it is clear that barriers faced by families are systemic, complex, and multidimensional. These barriers can occur at multiple levels, beginning at the patient and fanning out to society at large, making the Bioecological Model of Human Development an ideal framework through which to comprehensively examine the issue. At the level of the patient, barriers to diagnosis by three months consist of birth complications, speech and language delays (perceived or documented), severity and configuration of hearing loss, and comorbidities (Awad et al., 2019; Holte et al., 2012; McLean et al., 2019). Family-level barriers include conflicting work responsibilities, transportation challenges, rural location, minimal to no insurance coverage, and lack of understanding the importance of follow up (Awad et al., 2019; Bush et al., 2015; Holte et al., 2012; McLean et al., 2019; Shulman et al., 2010). Provider barriers include a lack of knowledgeable professionals (e.g., pediatric audiologists, speech-language pathologists, early interventionists), limited early intervention and family support programs, and complex diagnostic appointments requiring multiple evaluations and lengthy wait lists (Awad et al., 2019; Holte et al., 2012; Shulman et al., 2010). Similar barriers were identified by Reynolds et al. (2023), who examined caregivers' impressions of their path to obtaining a diagnosis of hearing loss for their child. Provider barriers were the most frequently reported, with caregivers describing limited access to providers in their area and inadequate informational counseling when they were put into contact with providers. In the present study, we focus specifically on barriers to timely start of early intervention services.

The Need to Focus on Access to Early Intervention for Children who are DHH

The EHDI benchmark of 6 months for early intervention enrollment is based on decades of research showing its benefit to spoken language outcomes (e.g., Yoshinaga-Itano et al., 2010; Yoshinaga-Itano et al., 2017). Grey et al. (2022) reported that the only unique predictor of preschool omnibus spoken language outcomes between children who met EHDI benchmarks and those who did not is enrollment in early intervention by six months of age, highlighting the importance of this final benchmark for developmental outcomes. Ideally, early screening and early diagnosis of hearing loss leads seamlessly to immediate enrollment in appropriate early intervention services. However, there is a subset of families who meet the one month hearing screening and three month diagnosis EHDI benchmarks, but do not meet the six month early intervention benchmark (Findlen et al., 2023). To be able to reduce or eliminate barriers and/or develop methods for intervening, it is necessary to understand why a large number of children who are diagnosed with hearing loss by three months of age do not begin early intervention by six months of age. The purpose of the present study was to examine the barriers that prevent individuals from meeting the final goal of the 1-3-6 EHDI recommended timeline after meeting the one-month screening and three-month diagnosis goals, and to categorize reported barriers using Bronfenbrenner's Bioecological Model of Human Development as our theoretical lens. We addressed the following research questions in this study:

1. What barriers prevent children who are DHH who met previous EHDI benchmarks from meeting the final EHDI goal of enrollment in early intervention by six months of age?
2. Which systems of the Bioecological Model of Human Development represent barriers for children who are DHH trying to enroll in early intervention?

Method

All study procedures were approved by the University of South Carolina Institutional Review Board. All participants provided informed consent prior to participating in the interviews.

Participants

In this study, we specifically recruited caregivers of children in Grey et al. (2022) whose children met the EHDI guideline for diagnosis of hearing loss by three months of age but did not meet the guideline for enrollment in early intervention by six months of age. We invited via email or phone call all participants whose children met these criteria, and all agreed to participate in this follow-up study. Participants included 10 caregivers whose children are deaf or hard of hearing (DHH) and use amplification and spoken English as their primary language. Table 1 presents demographic information for each caregiver and their child who is DHH.

Procedures

Interview process. Each caregiver of a child who is DHH participated in individual semi-structured interviews conducted by the third author. The interviews were conducted virtually via Zoom for Telehealth and were recorded using Zoom's internal capabilities. Eight of the interviews were with the child's mother only and two of the interviews were with the child's mother and father. The average length of interviews was 21 minutes (range: 11 to 36 minutes). The interview questions focused on barriers to enrollment in early intervention. The interview protocol was developed based on Bronfenbrenner's Bioecological Model (Bronfenbrenner & Morris, 2006) and specifically addressed barriers related to each system of the model. The interview protocol is displayed in the Appendix.

Transcription process. Each interview was transcribed verbatim by either the third author or a graduate student research assistant. Accuracy of interview transcription was verified by the other. Thus, final transcriptions represented consensus of the two transcribers.

Coding process. Analysis was completed by the first, second, and fourth authors. We used a combined deductive and inductive coding approach in this study. Deductive coding is a top-down approach to qualitative analysis in which the research team develops the initial codebook based on an established framework prior to coding interviews (Saldaña, 2021). Our framework in this study was Bronfenbrenner's Bioecological Model of Human Development. Therefore, our initial codes matched to each system in the model: Child, Microsystem, Mesosystem, Exosystem, and Macrosystem. Inductive coding is a bottom-up approach to qualitative analysis in which the research team develops codes as the dataset is analyzed (Saldaña, 2021). As the coding progressed, new codes were added to the codebook based on the data. Codes that emerged during this process were related to specific barriers experienced by families within each of the bioecological systems.

Prior to reading or listening to any interview, the first author created an a priori codebook in collaboration with the second and fourth authors, which was based on the Bioecological Model of Human Development. Then, the second and fourth authors separately coded each interview using the initial codebook and adding additional codes as they were identified from the data. They met with the first author regularly during coding to discuss disagreements and refine the codebook. The final analysis represents consensus of the coders and first author on the final codebook and themes/subthemes that were identified in the interviews. Finally, the entire research team reviewed and agreed on the final codebook and themes identified.

Table 1

Demographic Information for Participants

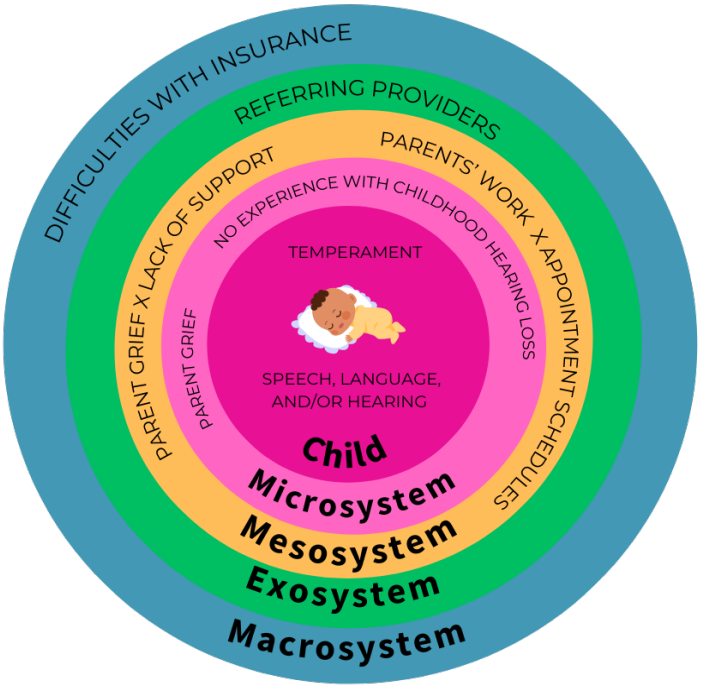
Participant Number	Caregiver(s) Interviewed	Child Amplification	Child Degree of Hearing Loss	Child Sex Assigned at Birth	Geographic Region in U.S.	Languages at Home	Race	Ethnicity	Age at Early Intervention Enrollment	Maternal Education
1	Mother + Father	Bimodal	Severe to Profound	Girl	West South Central	English 100%	White	Not Hispanic or Latino	14 months	Graduate degree
2	Mother	Bilateral Cochlear Implant	Profound	Boy	East North Central	English 75% Albanian 25%	White	Not Hispanic or Latino	9 months	Some college
3	Mother	Bilateral Cochlear Implant	Severe to Profound	Boy	Mountain West	English 100%	White	Not Hispanic or Latino	10 months	Bachelor's degree
4	Mother	Bilateral Hearing Aids	Mild to Moderate	Girl	South Atlantic	English 100%	White	Not Hispanic or Latino	7 months	Bachelor's degree
5	Mother	Bilateral Hearing Aids	Severe	Girl	South Atlantic	English 100%	Black or African American	Not Hispanic or Latino	24 months	Graduate degree
6	Mother	Bilateral Hearing Aids	Mild to Moderate	Girl	West North Central	English 100%	White	Not Hispanic or Latino	9 months	Some college
7	Mother + Father	Bilateral Hearing Aids	Moderately Severe	Boy	Pacific West	English 100%	White	Not Hispanic or Latino	50 months	Some college
8	Mother	Unilateral Hearing Aid	Moderate	Girl	West South Central	English 100%	White	Not Hispanic or Latino	17 months	Bachelor's degree
9	Mother	Bilateral Hearing Aids	Mild to Moderate	Boy	East South Central	English 100%	Native Hawaiian or Other Pacific Islander	Not Hispanic or Latino	17 months	Bachelor's degree
10	Mother	Bilateral Hearing Aids	Moderate	Boy	Pacific West	English 60% Spanish 40%	Not Reported	Hispanic or Latino	9 months	Some college

Results

Themes identified from caregiver interviews are presented below. Overall, barriers to timely access of early intervention reported by caregivers spanned the systems of the Bioecological model, including child, microsystem, mesosystem, exosystem, and macrosystem factors (see Figure 1). Subthemes in each system are described below.

Figure 1

Barriers to On-Time Enrollment in Early Intervention by System of the Bioecological Model of Human Development.



Note. This figure is based on the description of nested systems in Bronfenbrenner and Morris (2006).

Barriers Related to Child

We identified two barriers to early intervention related to child factors: child temperament and child hearing or speech/language skills. First, caregivers reported that child temperament influenced their ability to access early intervention; however, the specific temperament characteristics varied child-to-child. For example, some caregivers reported that a difficult temperament caused a delay in accessing early intervention. This temperament was seen primarily in opposition to wearing amplification devices, which influenced the speed at which caregivers sought early intervention.

Some caregivers reported that they believed that early intervention wouldn't be useful until they felt their child was well adjusted to their amplification devices. Other caregivers reported that a slow-to-warm-up temperament influenced the speed at which they enrolled in early intervention. One caregiver, for instance, reported that their child's shyness resulted in professionals being hesitant to use test results to make eligibility decisions, which delayed their enrollment in early intervention services: "A lot of the testing that was done in that timeframe wasn't the most accurate because she … didn't want to participate."

Additionally, several caregivers reported that their enrollment in early intervention was delayed because they were told that their child's hearing and/or speech and language did not qualify for the state program. Caregivers reported that child factors that led to being told they would not qualify included mild to moderate degrees of hearing loss, as well as speech and language development that was on target at three to six months of age. It is important to note that decisions about early intervention eligibility for children who are DHH were being made on the basis of no delay in speech or language at six months of age or younger.

Barriers Related to Microsystem

The two main barrier categories within the microsystem were lack of caregiver experience with childhood hearing loss and caregiver grief. First, caregivers reported a lack of experience related to childhood hearing loss, expressing the idea that they did not feel equipped to make decisions about early intervention. For example, one caregiver reported that they felt pressure to "make a fairly quick decision" about their child's communication modalities after diagnosis. They reported that they thought they would just need hearing aids: Probably just get hearing aids and he'll be fine. No one in the family really knew. I'm a physical therapist. I know about therapy in general but speech is a lot different." Many caregivers reported that they did not feel a sense of urgency in getting their child enrolled in early intervention because they had no experience with childhood hearing loss—they didn't recognize the need for intervention prior to their child beginning to talk.

Second, caregivers reported grief and a feeling of loss after their child's hearing loss diagnosis. A caregiver recalled being "kind of sad at first realizing that he wouldn't be able to have normal hearing like I've had my whole life." Another said, "I remember feeling very overwhelmed and very scared. I'm very worried about my child. What was the future going to look like for her? Cause this was something that I had not experienced before and no one in our family had hearing loss. I think as parents we all envision our children being perfect. And not that she's not perfect but I knew that with this we were going to have some obstacles and some challenges that we were gonna have to learn how to overcome in order to help her succeed." This grief encompassed feelings that their child would miss out on a normal life, as well as feelings of guilt for any potential role they had in causing the hearing loss, and fear for their child's future. Some caregivers reported being in denial, whereas others reported feeling overwhelmed without a clear picture of where to go next. Both led to delays in early intervention.

Barriers Related to Mesosystem

Relevant to the mesosystem, two primary interactions were identified: (a) caregivers' grief and lack of support from extended family members or community members, as well as (b) caregivers' work and appointment scheduling. For some families, caregiver grief was amplified by a lack of support from extended family or

communities. Caregivers reported that without buy-in from their support systems, they questioned the need for intervention for their child. This interaction of caregiver grief and lack of support from family or community resulted in delays to enrollment in early intervention. Caregivers reported switching focus from following recommendations for early intervention to convincing others to support and participate in their decisions regarding their child's development (e.g., wearing hearing aids at grandma's house). In other cases, caregivers reported getting mixed messages from community members regarding the use of amplification and choice of communication mode and waited to enroll in early intervention while they attempted to learn more about their child's options.

Additionally, caregivers' schedules impacted the timely enrollment in early intervention for some children. Several caregivers reported that it was difficult to schedule appointments with hearing healthcare professionals and/or early intervention systems while also maintaining their work schedules and productivity. This difficulty was sometimes, but not always, related to distance from the family to a specialized service provider. Caregivers discussed their difficulty in navigating all the appointments needed for their child in light of the time away from work required; often this included substantial travel time as well as the time in appointments.

Barriers Related to Exosystem

The primary barrier within the exosystem was related to referring providers. Caregivers reported a lack of communication from medical professionals, difficulty obtaining referrals for early intervention, and professionals who did not follow the EHDI 1-3-6 guidelines. In many cases, caregivers reported that providers, primarily pediatricians, did not know that they should refer their child to early intervention, and in some cases explicitly declining caregivers' requests for a referral. Provider lack of knowledge of the EHDI guidelines was widespread. Less common, but reported by some caregivers, were providers who do not believe in referring any child for early intervention prior to certain ages (in this study, some caregivers reported they were told their pediatrician never refers before 14–18 months). In hindsight, caregivers reported that had early intervention been recommended by their provider, they would have enrolled earlier. Caregivers whose providers declined to provide a referral for early intervention report they wished they had pushed the issue more or with a different professional. A caregiver said**, "**originally a lot of the doctors told us don't do anything cause it's single sided and she'll develop fine." Although pediatricians were the primary medical professional related to this issue, hearing healthcare professionals (encompassing multiple professions including otolaryngologists, audiologists, and speech-language pathologists) were also mentioned by a minority of caregivers for downplaying the need for early intervention for their child.

Barriers related to Macrosystem

Finally, the primary barrier within the macrosystem was difficulties with insurance. Seventy percent of caregivers reported difficulties with insurance. These difficulties included their insurance declining coverage of early intervention, as well as overly complicated protocols to obtain coverage. Some caregivers reported having to change providers because of insurance changes, which led to additional delays. A caregiver details their difficulty with insurance coverage: "We fought like hell. We got out of the NICU and we had severe insurance drama go on because I was very naïve… My work's benefits advisor advised that we get the HMO... I go to call and make the doctor's appointments and they're like oh no no no. You have an HMO. You can't do anything. You have to go through all these loops and hoops and scoops to get that. So we're trying to get the insurance fixed. I was on the phone everyday fighting for two or three hours."

Discussion

The purpose of this study was to understand families' barriers to timely enrollment in early intervention services for their children who are DHH. We approached our inquiry through a lens of Bioecological Model of Human Development (Bronfenbrenner & Morris, 2006). By situating our findings within this model, we were able to gain a comprehensive understanding of the multi-layered and multi-leveled barriers to early intervention for this population. Our findings suggest that barriers to timely access of early intervention for children who are DHH are numerous and span bioecological systems for the child. Child factors such as temperament that made testing difficult ranging to macrosystem factors such as difficulties with insurance coverage delayed enrollment in early intervention for the children who are DHH in this study. Increasing the percentage of children who are DHH who meet the six month EHDI early intervention guideline will require comprehensive, widespread improvements to the current hearing healthcare system.

Child

First, professionals must be aware of potential child-level factors that may influence the speed at which early intervention is accessed. Many infants who are DHH initially resist wearing amplification devices (Visram et al., 2021), and caregivers have previously reported that child behavior impacts their time of hearing aid use (Muñoz et al., 2015). For families who choose a spoken language communication modality, hearing healthcare professionals should spend time explaining expectations and provide strategies to increase usage when introducing new devices. Additionally, professionals should explicitly tell families that this resistance does not prevent the child and family from accessing and benefiting from early intervention. Likewise, hearing healthcare professionals should not delay early intervention based on child temperament. Recall that our participants all received a diagnosis of hearing loss by three months of age, so temperament played a role only in their access to early intervention, not diagnosis of hearing loss. Importantly,

Sanson (1996) reported that ratings of child shyness did not stabilize until after one year of age and even then only showed moderate stabilization. Children who are diagnosed with mild degrees of hearing loss should also not be the basis of declining access to early intervention. Children with mild, unliateral, or even minimal hearing loss are likely to experience later academic difficulties, and early intervention may prevent these difficulties (e.g., Bess et al., 1998; Fitzpatrick et al., 2019; Walker et al., 2020).

Microsystem

Next, professionals should be aware that most caregivers have no prior experience with childhood hearing loss. Hearing loss health literacy is extremely low even in caregivers who have more than a year of experience with their child's hearing loss (Cooper & Werfel, 2024). Therefore, it is vital that professionals provide multimodal supports, be mindful of how much information is conveyed at one time, and check caregiver understanding (Richlin et al., 2023). Richlin and colleagues' 2023 article, "Living in the Void Between Hearing Health Care Encounters: Evaluation of the Barriers Families Face" is an excellent resource for providers to read more about informational counseling with families of children diagnosed with hearing loss.

Caregivers also reported experiencing many facets of grief during the 1-3-6 timeline, including feeling shame, wondering what they could have done differently, mourning the loss of an idealized view of their child's future, and feeling paralyzed in the decision-making process. Hearing healthcare providers can help caregivers navigate their grief by providing appropriate counseling and referring families to appropriate mental health professionals. Providers should practice a client-centered model of counseling, in which the provider practices selfless listening, the parent participates in testing so the family can take ownership of their child's diagnosis from the beginning, and the provider shares information while acknowledging the family's painful feelings (for more details, see Luterman, 2021). Luterman recommends that, often, information sharing should be done in a subsequent appointment, particularly if the family exhibits behaviors consistent with grief and/or shock. Additionally, there is a need to explicitly consider how to convey information to families in multiple formats and multiple times to scaffold families in learning about their child's hearing loss and the full range of their treatment options.

Mesosystem

Professionals also must be aware of key interactions that occur within family systems. Specifically, in this study we identified two interactions that served as barriers to early intervention enrollment. First, caregivers reported feelings of grief that were compounded by lack of support. This lack of support sometimes came from extended family members, such as grandparents, which highlights the need for family-centered early intervention (FCEI) approaches. FCEI approaches emphasize the need for considering multiple family members and how their needs may differ when planning early intervention services for children (Dirks & Szarkowski, 2022). Involving extended family members and encouraging caregivers to involve them in interactions with hearing healthcare providers, including audiologists and speech-language pathologists, may provide an avenue for informational counseling with family beyond the primary caregivers to relieve some burden from the family. Other caregivers reported lack of support from their communities. Caregivers often sought support from the local DHH community but reported that they were sometimes met with hostility regarding their communication and/or amplification choices. Importantly, this reported hostility goes both ways: some caregivers reported being told their child would never learn spoken language if they used sign language, whereas others reported being told that cochlear implantation was abusive to their child. Both of these perspectives are highly emotionally charged, and there is no evidence to suggest that either is true. As providers, we must be aware of these potential support system difficulties and prepare families to navigate them. As a field, it's time to step past these extremes and be respectful of family choices.

The second interaction we identified was the interaction of caregiver work schedules and appointment scheduling. The field has long recognized the hearing healthcare disparities faced by families resulting from distance from a hearing healthcare facility (e.g., Bush et al., 2013). The caregivers in this study also highlighted the burden to families that comes from time off work, particularly in some cases immediately following parental leave. The time required for follow-up appointments and the lack of time off work to complete them is an issue the field must be aware of and help families to address; we believe this burden should fall on providers, not families. This finding also indicates the need for provider flexibility in scheduling visits as well as institutional commitments to investing in mobile service delivery. Telepractice has the potential to alleviate some of this burden, and families in rural areas are enthusiastic about its use (Bush et al., 2015).

Exosystem

At the level of the exosystem, caregivers reported tremendous difficulty getting referred to early intervention by their providers. Primarily, these difficulties centered on pediatricians; however, a minority of families reported that hearing healthcare specialists downplayed the need for early intervention for their child. The majority of caregivers reported that their pediatricians lacked knowledge about the EHDI guidelines for early intervention enrollment, and some caregivers reported that their pediatricians never refer any child to early intervention prior to specific ages (e.g., 18 months), with the potential loss of a full year of early intervention progress. Our field must provide better support to front-line professionals in knowing who, when, and how to refer. Hearing healthcare providers should explore ways in which information about the need for early intervention for children who are DHH and basic knowledge of the EHDI guidelines can be better conveyed to pediatricians to enact systemic change in this area. It is also important to consider how individuals with lived experiences related to childhood hearing loss, including

deaf mentors and parents who have previously navigated the hearing healthcare system, may collaborate with hearing healthcare providers to ensure that families have appropriate support.

Macrosystem

The prevailing theme from the macrosystem was difficulties with insurance. Consistent with prior research (Bush et al., 2015; Kingsbury et al., 2022), caregivers whose children are DHH reported dealing with overly complicated insurance protocols, minimal to no coverage for early intervention services, and forced changes in providers (e.g., in the case of enrolling their child in Medicaid) as significant barriers to enrolling in early intervention. Because insurance and Medicaid rules and regulations are largely beyond the control of families and hearing healthcare professionals, the need for change in hearing healthcare requires work to change systems via top-down, as well as bottom-up, approaches. Local advocacy efforts have been quite successful at a state level for initiatives like insurance coverage for pediatric hearing aids, and similar approaches may be appropriate for early intervention services. It is vital that hearing healthcare providers know the specific IDEA Part C eligibility rules in their states and provide this information to families via informational counseling, along with information about how families can self-refer via ChildFind.

Conclusion

Families of children who are DHH face widespread, systemic, multi-layer barriers to enrolling in early intervention services for their child. The barriers identified herein spanned Bronfenbrenner's Bioecological Model of Human Development, from child-level factors such as temperament to macrosystem-level factors such as insurance. Hearing healthcare professionals must be aware of these barriers and take steps to ensure that all families are able to access early intervention services in a timely manner for their children who are DHH.

References

Awad, R., Oropeza, J., & Uhler, J. (2019). Meeting the Joint Committee on Infant Hearing standards in a large metropolitan children's hospital: Barriers and next steps. *American Journal of Audiology*, *28*, 251–259. https://doi.org/10.1044/2019_AJA-18-0001

Bess, F., Dodd-Murphy, J., & Parker, R. (1998). Children with minimal sensorineural hearing loss: Prevalence, educational performance, and functional status. *Ear & Hearing*, *19*, 339–354. https://doi.org/10.1097/00003446-199810000-00001

Bronfenbrenner, U. (1979). *The ecology of human development: Experiments by nature and design.* Harvard University Press.

Bronfenbrenner, U., & Morris, P. (2006). The bioecological model of human development. In R. Lerner & W. Damon (Eds.), *Handbook of Child Psychology, Vol.1: Theoretical Models of Human Development* (6th ed., pp. 793–828). Wiley. http://doi.org/10.1002/9780470147658.chpsy0114

Bush, M., Burton, M., Loan, A., & Shinn, J. (2013). Timing discrepancies of early intervention hearing services in urban and rural cochlear implant recipients. *Otology & Neurotology*, *34*, 1630–1635. https://doi.org/10.1097/MAO.0b013e31829e83ad

Bush, M., Hardin, B., Rayle, C., Lester, C., Studts, C., & Shinn, J. (2015). Rural barriers to early diagnosis and treatment of infant hearing loss in Appalachia. *Otology & Neurotology*, *36*, 93–98. https://doi.org/10.1097/MAO.0000000000000636

Centers for Disease Control and Prevention. (2020). 2020 CDC Early Hearing Detection and Intervention (EHDI) hearing screening & follow-up survey. https://www.cdc.gov/ncbddd/hearingloss/2020-data/documents/2020-hsfs-explanations-508.pdf

Cooper, E., & Werfel, K. (2024). High general health literacy does not ensure hearing loss health literacy in mothers of children who are deaf or hard of hearing. *American Journal of Speech-Language Pathology*, *33*, 468–475. https://doi.org/10.1044/2023_AJSLP-23-00110

Dirks, E., & Szarkowski, A. (2022). Family-centered early intervention (FCEI) involving fathers and mothers of children who are deaf or hard of hearing: Parental involvement and self-efficacy. *Journal of Clinical Medicine*, *11*(3), 492. https://www.mdpi.com/2077-0383/11/3/492

Eriksson, M., Ghazinour, M., & Hammarström, A. (2018). Different uses of Bronfenbrenner's ecological theory in public mental health research: What is their value for guiding public mental health policy and practice? So*cial Theory & Health*, *16*, 414–433. https://doi.org/10.1057/s41285-018-0065-6

Findlen, U. M., Davenport, C. A., Cadieux, J., Gehred, A., Frush Holt, R., Vaughn, L. M., Houston, D., & Hunter, L. L. (2023). Barriers to and facilitators of early hearing detection and intervention in the United States: A systematic review. *Ear and Hearing*, *44*(3), 448–459. https://journals.lww.com/ear-hearing/fulltext/2023/05000/barriers_to_and_facilitators_of_early_hearing.3.aspx

Fitzpatrick, E., Gaboury, I., Durieux-Smith, A., Coyle, D., Whittingham, J., & Nassrallah, F. (2019). Auditory and language outcomes in children with unilateral hearing loss. *Hearing Research*, *372*, 42–51. https://doi.org/10.1016/j.heares.2018.03.015

Grey, B., Deutchki, E., Lund, E., & Werfel, K. (2022). Impact of meeting early hearing detection and intervention benchmarks on spoken language. *Journal of Early Intervention*, *44*, 235-251. https://doi.org/10.1177/10538151211025210

Holte, L., Walker, E., Oleson, J., Spratford, M., Moeller, M. P., Rousch, P., Tomblin, J. B., & Ou, H. (2012). Factors influencing follow-up to newborn hearing screening for infants who are hard-of-hearing. *American Journal of Audiology*, *21*, 163–174. https://doi.org/10.1044/1059-0889(2012/12-0016)

Joint Committee on Infant Hearing. (2019). Year 2019 position statement: Principles and guidelines for early hearing detection and intervention programs. *Journal of Early Hearing Detection and Intervention*, *4*(2), 1–44. https://doi.org/10.15142/fptk-b748

Kingsbury, S., Khvalabov, N., Stirn, J., Held, C., Fleckenstein, S., Hendrickson, K., & Walker, E. (2022). Barriers to equity in pediatric hearing health care: A review of the evidence. *Perspectives of the ASHA Special Interest Groups*, *7*, 1060–1071. https://doi.org/10.1044/2021_persp-21-00188

Luterman, D. (2021). Counseling parents at the time of diagnosis: Moving toward client-centered practice. *American Journal of Audiology*, *30*(1), 226–230. https://doi.org/doi:10.1044/2020_AJA-20-00122

McLean, T., Ware, R., Heussler, H., Harris, S., & Beswick, R. (2019). Barriers to engagement in early intervention services by children with permanent hearing loss. *Deafness & Education International*, *21*, 25–39. https://doi.org/10.1080/14643154.2018.1528745

Muñoz, K., Olson, W., Twohig, M., Preston, E., Blaiser, K., & White, K. (2015). Pediatric hearing aid use: Parent-reported challenges. *Ear & Hearing*, *36*, 279–287. https://doi.org/10.1097/AUD.0000000000000111

Reynolds, G., Werfel, K., Vachio, M., & Lund, E. (2023). Early experiences of parents of children who are deaf or hard of hearing: Navigating through identification, intervention, and beyond. *Journal of Early Hearing Detection and Intervention*, *8*, 56–68. https://doi.org/10.26077/6d9d-06f3

Richlin, B., Blaiser, K., & Bush, M. (2023). Living in the void between hearing health care encounters: Evaluation of the barriers families face. *Perspectives of the ASHA Special Interest Groups*. https://doi.org/10.1044/2023_PERSP-23-00038

Saldaña, J. (2021). *The Coding Manual for Qualitative Researchers* (4th ed.) SAGE Publications.

Sanson, A. (1996). Shyness ratings: Stability and correlates in early childhood. *International Journal of Behavioral Development*, *19*, 705–724. https://doi.org/10.1080/016502596385532

Shulman, S., Besculides, M., Saltzman, A., Ireys, H., White, K., & Forsman, I. (2010). Evaluation of the universal newborn hearing screening and intervention program. *Pediatrics*, *126*, S19–S27. https://doi.org/10.1542/peds.2010-0354F

Swick, K., & Williams, R. (2006). An analysis of Bronfenbrenner's bioecological perspective for early childhood educators: Implications for working with families experiencing stress. Ea*rly Childhood Education Journal*, *33*, 371–378. https://doi.org/10.1007/s10643-006-0078-y

Tudge, J., Navarro, J., Merçon-Vargas, E., & Payir, A. (2021). The promise and the practice of early childhood educare in the writings of Urie Bronfenbrenner. *Early Child Development and Care*, *191*, 1079–1088. https://doi.org/10.1080/03004430.2020.1844193

Visram, A., Roughley, A., Hudson, C., Purdy, S., & Munro, K. (2021). Longitudinal changes in hearing aid use and hearing aid management challenges in infants. *Ear & Hearing*, *42*, 961–972. https://doi.org/10.1097/AUD.0000000000000986

Walker, E., Sapp, C., Dallapiazza, M., Spratford, M., McCreery, R., & Oleson, J. (2020). Language and reading outcomes in fourth-grade children with mild hearing loss compared to age-matched peers. *Language, Speech, & Hearing Services in Schools, 51*, 17–28. https://doi.org/10.1044/2019_LSHSS-OCHL-19-0015

Yoshinaga-Itano, C., Baca, R. L., & Sedey, A. L. (2010). Describing the trajectory of language development in the presence of severe-to-profound hearing loss: A closer look at children with cochlear implants versus hearing aids. *Otology & Neurotology*, *31*(8), 1268–1274. https://doi.org/10.1097/MAO.0b013e3181f1ce07

Yoshinaga-Itano, C., Sedey, A., Wiggin, M., & Chung, W. (2017). Early hearing detection and vocabulary of children with hearing loss. *Pediatrics*, *140*, e20162964. https://doi.org/10.1542/peds.2016-2964

Appendix

Interview Protocol

1. How did you feel or what was your initial reaction to the hearing loss diagnosis?

2. Had you had any experience with hearing loss prior to your child's diagnosis?

3. Did you encounter any difficulties with your child receiving hearing aids after diagnosis? If so, what were they?

4. What were you told was the next step after being diagnosed and fitted with hearing aids?

5. How did you find your early intervention (EI) provider? Who helped you?

6. What went well about the process of finding an EI provider and starting services?

7. What was the main difficulty that you encountered when trying to find an EI provider and begin services?

8. First, can you talk about experiences in beginning EI that might have been related to your child?

9. Next, can you talk about experiences in beginning EI that might have been related to your family?

10. Next, can you talk about experiences in beginning EI that might have been related to your community?

11. Next, can you talk about experiences in beginning EI that might have been related to your healthcare providers?

12. Last, can you talk about experiences in beginning EI that might have been related to society and culture?

Note. EI = early intervention.

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Diagnostic Overshadowing: A Delayed Diagnosis of Autism Spectrum Disorder in a Child who is Deaf

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Abstract

Diagnostic overshadowing occurs when a patient with a pre-existing diagnosis presents with symptoms that are attributed to the diagnosis, but actually reflect a separate issue. This diagnostic overshadowing can lead to delays in diagnosis, as well as disparities in health care and outcomes. The following case will provide an example in which diagnostic overshadowing contributed to the delay of an autism diagnosis in a child who is deaf. With 40 to 50% of children who are deaf or hard of hearing having at least one co-existing medical/developmental condition, and the current prevalence of autism at 1 in 36 children in the United States, the intersection of these conditions is substantial. Pediatricians, otolaryngologists, audiologists, and speech therapists are all likely to encounter young children who are deaf or hard of hearing and in the process of acquiring language. Using interdisciplinary collaboration can support identification of children with coexisting conditions. Clinicians can benefit from increased familiarity with the presentation of children who are both deaf and neurotypical, and how it tends to differ from children with autism, reducing the chance of delayed or incorrect diagnosis and strengthening the formation of care plans.

Keywords: autism, deaf, diagnostic overshadowing, hearing loss, language development

Acronyms: ASD = Autism Spectrum Disorder; EHDI = early hearing detection and intervention; DHH = deaf or hard of hearing

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Diagnostic overshadowing occurs when a patient with a pre-existing diagnosis presents with symptoms that are attributed to the diagnosis, but truly reflect a separate issue. When considering a diagnosis of autism in an individual who also has a sensory issue (deaf or hard of hearing, low vision, etc.), diagnostic overshadowing can occur in two distinct manners: *misdiagnosis* of autism (symptoms that are attributed to autism but are actually due to the sensory diagnosis), and *missed diagnosis* of autism (symptoms that are attributed to the sensory diagnosis when they are actually due to autism; Ludwig et al., 2022). Discerning whether a child is deaf or hard of hearing and/or has autism provides some diagnostic challenges. Most states in the United States currently have a formal early hearing detection and intervention (EHDI) program that oversees newborn hearing screening and follow-up policies (National Center for Hearing Assessment and Management, 2024). EHDI programs have contributed to children who are deaf or hard of hearing being identified earlier in life, and often as infants if they are congenitally deaf or hard of hearing. A reliable diagnosis of autism can be made by an experienced professional starting around 2 years of age, although children with a milder presentation may not raise clinical suspicion until they are older (Centers for Disease Control and Prevention, 2022). Thus, for the majority of children who are congenitally deaf or hard of hearing, identification of hearing status will be well established prior to the time suspicion for possible autism may arise. This has the potential to confound the identification of autism due to some shared presenting (but etiologically differing) symptoms, such as delayed or lack of language development, reduced responsiveness, and diminished social engagement, all of which may be inappropriately attributed to lack of hearing and access to spoken language. The following case will provide an example of diagnostic overshadowing that contributed to the delay of an autism diagnosis in a child who is deaf.

Case Report

History

A 2-year, 8-month-old female who had been diagnosed as deaf presented to our specialty clinic for deaf and hard of hearing children. She had been identified with late onset profound bilateral sensorineural hearing loss by an outside

pediatric otolaryngologist at 14 months of age after concerns for lack of receptive and expressive language development in both spoken English and sign language. Review of available records from her otolaryngologist (who was part of a pediatric cochlear implant program) included her diagnosis of bilateral profound sensorineural hearing loss, a Magnetic Resonance Imaging (MRI) scan of her internal auditory canals which was normal and specifically made reference to normal cochlear development and normal caliber cochlear nerves, a statement of her having been determined to be a good candidate for cochlear implant, and her intra-operative record. Copies of her aided and unaided audiogram assessment prior to cochlear implantation were unfortunately not provided for direct review. Parents reported that she had not benefited substantially from traditional hearing aid amplification. At 22 months old, she had bilateral cochlear implants placed at the outside pediatric cochlear implant program that housed the pediatric otolaryngologist and audiology team that had been seeing her.

She was enrolled in regular occupational therapy for sensory defensiveness, as well as twice weekly auditory verbal speech therapy, which had been initiated prior to cochlear implantation. An initial speech therapy evaluation done when she was two years old included the Receptive Expressive Emergent Language Scale, Third edition (REEL-3) assessment, which demonstrated receptive language, expressive language, and language ability scores in the *extremely low* range (standard scores [SS] all < 55). Follow-up re-evaluation included re-administration of the REEL-3 ten months later, and standard scores had changed marginally (receptive language SS = 55, expressive language SS = 57, language ability SS = 55). Diagnosis at both assessments was noted to be consistent with a mixed expressive/receptive language disorder. Additionally, her family had routine exposure to sign language learning through a deaf education parent advisor via Early Childhood Intervention who worked with them weekly. Observation of what the family was using with her during our visit was most consistent with a CASE model (Conceptually Accurate Signed English) using signs based on conceptual meaning consistent with American Sign Language but using an English-based syntax and grammar system.

Parents reported that for several months, she had been increasingly refusing to wear her cochlear implant processors, and they were concerned with lack of progress with language development (both spoken and signed), as she had extremely limited expressive language. Her mother reported occasional spontaneous non-word vocalizations and an occasionally recognizable spoken “mama”, as well as occasional spontaneous signs for “want” and “more”. Her occupational therapist suggested the possibility of a co-existing developmental disorder in addition to hearing differences. However, parents reported that when they had brought up concerns with other members of the treatment team, they had been reassured that she had “lots of strengths,” and had been informed by her audiologist that she was “hearing at 35 decibels with her cochlear implants” (unaided/aided audiograms were requested but ultimately not able to be provided for independent review).

Exam

Head, ears, eyes, nose, and throat examination were all unremarkable. Limited eye contact and social engagement was noted for the duration of the visit. She demonstrated persistent repetitive motor and verbal behaviors including constantly circling the room, spinning in a circle, beating her arms against her sides, and flapping her hands. She was repeatedly screeching loudly and saying, “ah ah ah!” In addition, she appeared to be fixated on objects (e.g., pen, ID badge), and enjoyed scribbling repeatedly with a pen. She did not demonstrate any formal expressive language, in American Sign Language or spoken language, throughout the visit.

Assessment and Plan

Due to her clinical presentation, a co-occurring developmental disorder was suspected in addition to hearing status. The patient was referred to our hospital’s autism and developmental disorders clinic for further evaluation by a team including a licensed psychologist and a speech language pathologist with a Certificate of Clinical Competence, who also reviewed her available previous testing. Assessment included, but was not limited to, Developmental Profile 3rd edition (DP-3), Brief Observation of Symptoms of Autism—Minimally Verbal (BOSA-MV, which was administered in lieu of an Autism Diagnostic Observation Scale due to COVID restrictions), Childhood Autism Rating Scale 2nd edition standard version (CARS2-ST), Adaptive Behavioral Assessment System 3rd edition (ABAS-3), and Early Classroom Assessment Scoring System (Early CLASS) Functional Communication Assessment. She was ultimately identified with autism spectrum disorder as well as language disorder. Multiple resources were provided, and recommendations were made, including social skills intervention, continuation of speech and occupational therapy, consideration of Augmentative and Assistive Communications technology, consideration of neurodevelopmental evaluation, recommendations for educational programming, and recommendation for Applied Behavioral Analysis (ABA) therapy.

At subsequent follow-up visits, the patient’s mother reported that ABA had been a helpful intervention for her daughter, to the extent that they chose to enroll her in a full-time ABA program rather than in a more traditional self-enclosed or accommodated education classroom. The family continued therapy services, but also elected to find a different Auditory Verbal speech therapist who had more experience in working with children with autism and was able to collaborate with her ABA program. Her parents reported the new therapist was a better fit for the family as well as the patient. The patient’s mother shared with us at subsequent follow-up visits that although the initial discussion around the possibility of autism had been very hard, it was also a relief to get confirmation that something else was contributing to their daughter’s developmental delay. Additionally, she stated that the diagnosis had changed how her family functioned around her daughter, both in how they attempted to connect with her and the educational decisions they made for her in terms of therapies, school enrollment, and use of amplification.

Discussion

In their 2022 Sentinel Event Alert, the Joint Commission describes diagnostic overshadowing as "the attribution of symptoms to an existing diagnosis rather than a potential co-morbid condition." Cognitive bias can lead a clinician away from considering other alternatives to presenting symptoms, and toward viewing a patient solely through the lens of their established diagnosis. Speed, stress, and lack of training all contribute to an increased risk of inappropriately assigning presenting symptoms to a pre-existing condition (Ospina et al., 2019, p.1).

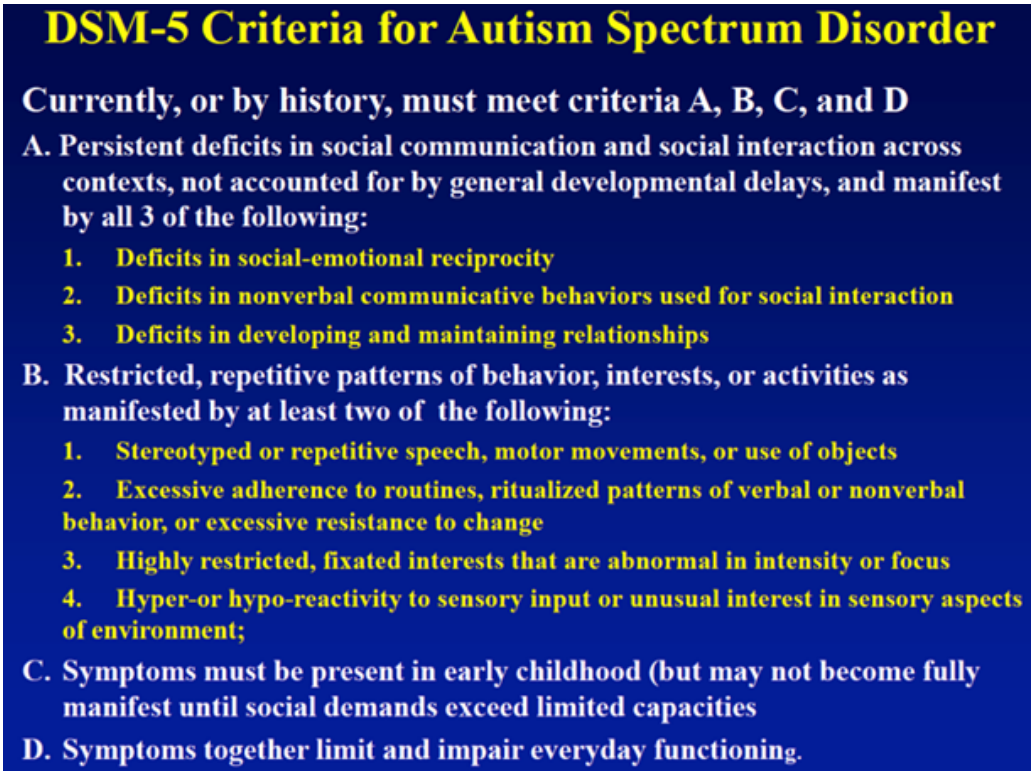
In this case, the patient's deafness appeared to steer most of her clinicians and therapists away from appreciating her additional underlying developmental disorder, potentially due, in part, to viewing her through a *specialty lens* and underappreciating her overarching presentation. This issue can be exacerbated further by the fact that professional efforts to take the time to collaborate and communicate across specialties and disciplines are not traditionally rewarded in the current U.S. medical system, which tends to prioritize productivity, efficiency, and high volume workflow. Professionals may not feel they have the *bandwidth*, depending on their practice environment, to consider involving interdisciplinary specialists when they are not seeing the outcomes they expect as there is often little protected time for this. Consequently, they may continue down the same clinical track with the hope that more time and continuation of therapy may improve outcomes. It is very difficult for one specialist to be all things to all patients and increasing patient complexity warrants increasing diversity of both evaluation and therapy, as well as making system changes that allow for more multidisciplinary collaboration.

Atypical sensory responsiveness is one of the salient diagnostic features of autism, and children with autism who are deaf or hard of hearing can have difficulty tolerating hearing technologies such as hearing aids or cochlear implants (Ludwig et al., 2022; Beers et al., 2014). This may be due to an intolerance for the physical feel of the appliance on or around the ear, the actual sound input, or both. This patient's language development delay may have been mistakenly attributed to implant non-use before further interdisciplinary evaluation revealed an underlying diagnosis of autism that resulted in diminishing sensory tolerance to implants.

In some ways, it is understandable that a clinician unfamiliar with how children who are deaf and not neurotypical present may attribute certain behaviors to "being deaf," particularly if the provider is not familiar with sign language and which movements constitute true language as opposed to repetitive behaviors. Yet, with the current prevalence of autism in the United States estimated at 1:36 children (Maenner et al., 2023), many clinicians have a baseline familiarity with characteristics of autism in children who are hearing (see Figure 1). At least 40–50% of children who are deaf or hard of hearing have one or more coexisting medical/developmental conditions (Bowen & Probst, 2023), which makes clinical evaluation more complex. More specifically, studies indicate an overall increased prevalence of autism in children who are deaf or hard of hearing at 7–9% (Do et al., 2017; Kancherla et al., 2013; Van Naarden Braun, et al., 2015). If this child had been hearing and had presented with lack of eye contact, social avoidance, repetitive behaviors, preoccupation with objects, and lack of expressive language development, the suspicion for a developmental disorder may have been raised sooner due to relatively increased diagnostic familiarity.

Figure 1

DSM-5 Criteria for Autism Spectrum Disorder



Note. Reprinted from Open-Access Journal, *Inquiries*. Singh, A. N. (2014). Increases in the prevalence of autism disorder: Exploring biological and socio-environmental factors. *Inquiries Journal/Student Pulse, 6*(09). http://www.inquiriesjournal.com/a?id=913

This patient's limited access to auditory input stemming from deafness prior to implantation, limited cochlear implant adherence, and delays in spoken language development contributed to diagnostic overshadowing between deafness and autism. To add to the complexity, delays in spoken language development related to being deaf can present with features that resemble those seen in autism: not responding to name, atypical social engagement, limited communication, and so forth. However, children who are deaf and neurotypical, even with delayed linguistic development, demonstrate some marked differences from children who are deaf with autism (Ludwig et al., 2022). Children who are deaf and neurotypical are rarely avoidant of eye contact as they are visual learners, and will make significant use of joint attention to ensure understanding in a conversation (whether using spoken or sign language). They also tend to respond well when provided with adequate access to formal language (sign, spoken, or both), while a child with severe autism may not progress with language development despite supported

Table 1

ASD Diagnosis Considerations in Children who are Neurotypical and Children who are DHH

Symptoms of Autism Spectrum Disorder

- Deficits in social-emotional reciprocity
- Deficits in non-verbal communication in social interactions
- Deficits in relationship building
- Stereotyped or repetitive movements, speech, use of objects
- Inflexible adherence in routines or ritualized behavior
- Fixated interests
- Hyper or hypo-reactivity to sensory input

Child who is Neurotypical and DHH	Child who has ASD and is DHH
• Appropriate social smile • Appropriate eye contact • Engages with others verbally and non-verbally • Can imitate behaviors • Appropriate joint attention • Builds relationships when provided with communication means • May use more gestures/classifiers if exposed to ASL • Shows varied play and interests • Is flexible and can transition without major difficulty	• Social emotional reciprocity and language delays are lower than what is expected for child with hearing loss • Poor eye contact • Reciprocal conversation difficult • Atypical social approach • Poor joint attention, lack of pointing • Difficulty building relationships • Does not respond to name or attention-getting movements • Difficulty understanding others' needs and social cues, including signed emotional cues • Pronoun reversal • Failure to initiate or respond to peers in communication or make/sustain friendships • Language acquisition delays • Difficulty recognizing Deaf culture norms • Shows reduced shared enjoyment • Delayed acquisition of symbolic play skills inconsistent with non-verbal IQ • May engage in echolalia through sign and palm rotation errors (Shield et al., 2017; Shield & Meier, 2012) • Idiosyncratic and made up gestures despite formal sign being taught • Rocking, twirling, flapping, spinning • Highly repetitive play • Resistance to change and difficulty in shifting from preferred interest • Highly specific interests that are atypical in topic • Sensitive to sounds or resistant to wearing hearing aids or cochlear implants

Considerations to Avoid Diagnosis Overshadowing

- Could the social reciprocity and conversation engagement be impacted by language skills of child who is DHH?
- Does the language modality of gestures and vocalization match child's communication partner?
- Does child use eye contact and facial expressions as nonverbal communication (typically a strength for children who are DHH)?
- Does child make eye contact with their interpreter if one is being used?
- Is repetitive speech a symptom of ASD or reflective of receptive language deficits because language development was impacted?
- Is difficulty with change a result of not understanding what is happening (DHH) or inflexible behavior (ASD)?
- Are hearing aids and cochlear implants being monitored by audiologist to ensure proper fit and function, thereby ruling out sensitivity that is not indicative of ASD?

Note. Lugwig et al. (2022) provides a detailed comparison of children who are neurotypical and DHH versus children who are DHH and present with ASD. ASD = Autism Spectrum Disorder; DHH = deaf or hard of hearing.

access to it, regardless of language mode(s) provided. (A significant red flag in this particular case was that this child was consistently being presented with two communication modalities, spoken and visual, and was not developing expressive language in either.) Facial expression will often be reduced and restricted compared with children who are deaf and neurotypical. Language development may also have atypical features such as sign copying (sign echolalia) or abnormal hand position when making signs. Repetitive and restricted patterns of behavior, often a hallmark of autism, are not usually a presenting feature in children who are deaf and neurotypical. Table 1 outlines key presenting differences between children who are neurotypical and deaf or hard of hearing (DHH) and those who have Austism Spectrum Disorder (ASD) and are DHH. The child's hearing change in this case study was diagnosed at age 14 months, so this case seems more consistent with diagnostic overshadowing rather than unidentified hearing status presenting similarly to autism.

Conclusion

Diagnostic overshadowing is a challenge for many clinicians, which can be addressed in part by gaining understanding of how the intersectionality of multiple characteristics and conditions in a single individual can collectively impact their functioning. The need for interdisciplinary input and evaluation is critical in identifying and viewing these children in the context of their whole person, and not just according to a particular specialty or discipline. Children who are deaf with autism spectrum disorder can present a unique diagnostic challenge due to lack of familiarity with how children who are deaf and neurotypical function in comparison, as well as overlapping symptomatology. Providers who recognize clinical features of both children with autism as well as those of neurotypical children who are deaf will be better poised to recognize and support a child who is deaf with autism.

References

Beers, A., McBoyle, M., Kakande, E., Dar Santos, R., & Kozak, F. (2014). Autism and peripheral hearing loss: a systematic review. *International Journal of Pediatric Otorhinolaryngology, 78*, 96–101.

Bowen, S. K., & Probst, K. M. (2023). Deaf and hard of hearing students with disabilities: An Evolving Landscape. *Education Sciences*, *13*(7), 752. https://doi.org/10.3390/educsci13070752`

Centers for Disease Control and Prevention. (2022). *Autism Spectrum Disorder (ASD).* https://www.cdc.gov/autism/

Do, B., Lynch, P., Macris, E. M., Smyth, B., Stavrinakis, S., Quinn, S., & Constable, P. A. (2017). Systematic review and meta-analysis of the association of autism spectrum disorder in visually or hearing impaired children. *Ophthalmic and Physiological Optics*, *37*(2), 212–224. https://doi.org/10.1111/opo.12350

Kancherla, V., Van Naarden Braun, K., & Yeargin-Allsopp, M. (2013). Childhood vision impairment, hearing loss, and co-occurring autism spectrum disorder. *Disability Health*, *6*, 333–342. https://doi.org/10.1016/j.dhjo.2013.05.003

Ludwig, N., Jashar, D. T., Sheperd, K., Pineda, J. L., Previ, D., Reesman, J., Holingue, C., & Gerner, G. (2022). Considerations for the identification of autism spectrum disorder in children with vision or hearing impairment: A critical review of the literature and recommendations for practice. *The Clinical Neuropsychologist, 36*(5), 1049–1068. https://doi.org/10.1080/13854046.2021.2002933

Maenner, M. J., Warren, Z., Williams, A. R., Amoakohene, E., Bakian, A. V., Bilder, D. A., Durkin, M. S., Fitzgerald, R. T., Furnier, S. M., Hughes, M. M., Ladd-Acosta, C. M., McArther, D., Pas, E. T., Salinas, A., Vehorn, A., Williams, S., Esler, A., Grzybowski, A., Hall-Lande, J., . . . Nguyen, R. H. N. (2023). Prevalence and characteristics of autism spectrum disorder among children aged 8 years—Autism and developmental disabilities monitoring network, 11 sites, United States, 2020. *MMWR Surveillance Summaries, 72*, 1–14. https://www.cdc.gov/mmwr/volumes/72/ss/ss7202a1.htm

National Center for Hearing Assessment and Management. (2024). *The "State" of Early Hearing Detection and Intervention in the United States.* www.infanthearing.org/states/index.html#states_list

Ospina, N. S., Phillips, K. A., Rodriguez-Gutierrez, R., Castaneda-Guarderas, A., Gionfriddo, M. R., Branda, M. E., & Montori, V. M. (2019). Eliciting the patient's agenda—Secondary analysis of recorded clinical encounters. *Journal of General Internal Medicine*, 34, 36–40. https://doi.org/10.1007/s11606-018-4540-5

Shield, A., Cooley, F., & Meier, R. P. (2017). Sign language echolalia in deaf children with autism spectrum disorder. *Journal of Speech, Language, and Hearing Research*, *60*(6), 1622–1634. https://doi.org/10.1044/2016_JSLHR-L-16-0292

Shield, A., & Meier, R. P. (2012). Palm reversal errors in native-signing children with autism. *Journal of Communication Disorders*, *45*(6), 439–454. https://doi.org/10.1016/j.jcomdis.2012.08.004

Singh, A. N. (2014). Increases in the prevalence of autism disorder: Exploring biological and socio-environmental factors. Inquiries Journal/Student Pulse, 6(09). http://www.inquiriesjournal.com/a?id=913

The Joint Commission. (2022) Diagnostic overshadowing among groups experiencing health disparities. *Sentinel Event Alert,* (*65*), 1–7. https://www.jointcommission.org/-/media/tjc/documents/resources/patient-safety-topics/sentinel-event/sea-65-diagnostic-overshadowing-6-16-22-final.pdf

Van Naarden Braun, K., Christensen, D., Doernberg, N., Schieve, L., Rice, C., Wiggins, L., Schendel, D., & Yeargen-Allsopp, M. (2015). Trends in the prevalence of autism spectrum disorder, cerebral palsy, hearing loss, intellectual disability, and vision impairment, metropolitan Atlanta, 1991-2010. *PLOS ONE*, *10*(4), Article e0124120. https://doi.org/10.1371/journal.pone.0124120

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Cost-Effectiveness Analysis: Automated Auditory Brainstem Response Diagnostic Test Compared to Transient Evoked Otoacoustic Emission Screening for Universal Newborn Hearing Screenings in High-Risk Neonates

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Abstract

Purpose: Comorbidities and ototoxic medications increase risk of hearing loss (HL) in infants admitted to neonatal intensive care units (NICU; 6 per 1000 births) compared to well-babies (1.7 per 1000 births). For newborn hearing screening (NBHS), transient evoked otoacoustic emissions (TEOAE) testing is more efficient and less costly. Automated auditory brainstem response (aABR) testing yields higher sensitivity and specificity. This study will identify if aABR is cost-effective compared with TEOAE for NBHS in high-risk neonates.

Methodology: Cost-effectiveness analysis was conducted from a healthcare system perspective. Prevalence and outcomes data for aABR, TEOAE, and auditory brainstem response (ABR) were obtained from a published study with 144 neonates admitted to the same Thailand NICU. Sensitivity and specificity were used to evaluate effectiveness. Cost was calculated from published Medicaid rates across 34 states in the United States of America. A decision tree developed in TreeAge modeled diagnostic pathways of congenital HL. Consolidated Health Economic Evaluation Reporting Standards (CHEERS) guidelines were followed.

Conclusions: aABR was more costly (mean $34.09) with higher sensitivity (.917) and specificity (.921) than TEOAE (mean $29.03; sensitivity .787; specificity .888). The incremental cost-effectiveness ratio (2.80) indicates the aABR costs an extra $2.80 per each additional true positive screening. Equity considerations are vital for ensuring cost-effective NBHS and appropriate audiology referrals.

Keywords: newborn hearing screening, economic evaluation, cost-effectiveness, automated auditory brainstem response, transient evoked otoacoustic emissions

Acronyms: a ABR = Automated auditory brainstem response; CEA = cost effective analysis; CHEERS = Consolidated Health Economic Evaluation Reporting Standards; HL = hearing loss; ICER = Incremental Cost-Effectiveness Ratio; NBHS = newborn hearing screening; nHL = normal hearing level; NICU = Newborn Intensive Care Unit; TEOAE = transient evoked otoacoustic emissions; UNHS = Universal Newborn Hearing Screening

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Universal newborn hearing screening (UNHS) is federally mandated and designed for early detection of congenital hearing loss (HL) in infants (Joint Committee on Infant Hearing [JCIH], 2019). Early detection and intervention are crucial for childhood speech, language, cognitive development and future academic achievement (American Speech-Language-Hearing Association, n.d.-b). Infants in the neonatal intensive care unit (NICU) are at higher risk for HL (6 per 1000 births) compared to well babies (1.7 per 1000 births; Butcher et al., 2019; National Center on Birth Defects and Developmental Disabilities, 2019; White et al., 1994). A highly effective screening approach is needed to identify HL within this at-risk population.

The two most used tools in UNHS are otoacoustic emissions (OAE) and automated auditory brainstem response (aABR; Wroblewska-Seniuk et al., 2017). In OAE, a small probe is placed in the ear canal to deliver sound stimuli through the middle ear to the inner ear (Wroblewska-Seniuk et al., 2017). In response to sound stimuli, the outer hair cells of the cochlea produce sounds and the OAE represents the sum of those active responses (Wroblewska-Seniuk et al., 2017). In aABR, disposable surface electrodes are placed near the ear, on the mastoid, or earlobe to record auditory brainstem response to sound. Although aABR has higher sensitivity and specificity, it is more costly and complicated to

administer (Heidari et al., 2017; Khaimook et al., 2019; Sheng, 2021). OAE detects middle ear and cochlear outer hair cell functioning, but it does not detect auditory nerve and brainstem functioning and yields high false-positive rates which lead to high referral rates (Akinpelu et al., 2014; Heidari et al., 2017; Khaimook et al., 2019; Sheng, 2021; Wroblewska-Seniuk et al., 2017). Compared to babies in the well-baby nursery, there is a higher incidence of auditory neuropathy among neonates who stay in the NICU; thus, aABR is indicated in the NICU (Colella-Santos et al., 2014; Heidari et al., 2017; Sheng, 2021; Wroblewska-Seniuk et al., 2017).

When an infant does not pass a newborn hearing screening (NBHS), rescreening should occur in the outpatient setting as close to hospital discharge as possible and always before one month of age (JCIH, 2019). If the newborn does not pass the outpatient rescreen, they should be referred to a pediatric audiologist for diagnostic ABR testing (JCIH, 2019). The JCIH recommends maintaining a referral rate of 4% or less (Akinpelu et al., 2014; JCIH, 2019; Wroblewska-Seniuk et al., 2017). Referral rates are lower with two-stage screening, such as conducting a second screening in the hospital with the same tool (OAE or aABR twice) or with both tools (OAE then aABR; Akinpelu et al., 2014; Clemens & Davis, 2001; Colella-Santos et al., 2014; Levit et al., 2015; Wroblewska-Seniuk et al., 2017). It is recommended that aABR is used for babies admitted to the NICU for more than 5 days because aABR is highly effective for identifying infants who are later diagnosed with auditory neuropathy. However, aABR may not detect infants later diagnosed with mild HL and HL at isolated frequencies; thus two-stage screening with aABR and OAE is ideal for NBHS in the NICU population (JCIH, 2019; Levit et al., 2015; Wroblewska-Seniuk et al., 2017). Not all programs have the resources to fund the cost of a two-tool approach and the JCIH recommends NBHS programs design feasible protocols that best serve their community (JCIH, 2019). Conducting a cost-effectiveness analysis (CEA) on these tools may assist programs in identifying the optimal tool for purchase.

Study Question and Hypothesis

The scope of this article is to conduct a CEA on the NBHS tools aABR and transient-evoked otoacoustic emissions (TEOAE). Among newborns admitted to a NICU, in the context of CEA, what is the optimal choice between aABR and TEOAE for NBHS prior to hospital discharge? Based on existing literature, this study was designed to assess the hypothesis that aABR is the optimal choice for NBHS in the NICU population.

Method

A health economic analysis plan was not developed for this study.

Study Population and Inclusion Criteria

This analysis was conducted with data from a published study which provided hearing screening and diagnostic data for 144 neonates admitted to the NICU in Songklanagarind Hospital in Thailand (Khaimook et al., 2019). The original study, approved by the ethics committee of the Songklanagarind Hospital, was designed to compare the screening reliabilities (i.e., sensitivity and specificity) of TEOAE and aABR based on ABR diagnostic results (Khaimook et al., 2019). The analysis for this article was conducted entirely based on published, deidentified data which does not meet the University of South Florida's Department of Research Integrity and Compliance's Institutional Review Board's (IRB) definition of human subject review and thus IRB review and approval was not required. All neonates were screened and tested in the hospital with the following protocol. First, nursing staff conducted TEOAE screening prior to NICU discharge. Second, with infants under sedation, a trained technician in the hospital audiology department conducted aABR screening. Third, while the infant was still under sedation, the technician conducted ABR diagnostic testing (Khaimook et al., 2019). The study was conducted over a 40-month time frame because of the need for anesthesiologist expertise and time to monitor infant sedation (Khaimook et al., 2019). Data from other studies were considered for this analysis and excluded (Akinpelu et al., 2014; Burke et al., 2012; Chesnaye et al., 2018; Heidari et al., 2017; Levit et al., 2015). Much of the literature does not provide data necessary to address the research question, such as lacking NICU-specific data and sufficient screening and/or diagnostic results necessary to calculate sensitivity and specificity (Akinpelu et al., 2014; Burke et al., 2012; Chesnaye et al., 2018; Heidari et al., 2017; Levit et al., 2015). For the purposes of this analysis, screening and diagnostic measures from Khaimook and colleagues (2019) provided valid and replicable HL prevalence and screening outcomes data for a sample of neonates.

Study Design

Cost-effectiveness analysis examines costs and outcomes of alternative approaches (Bang & Zhao, 2012; Office of Policy, 2021). The status-quo approach is compared to a new approach by estimating the cost to gain a unit of a health outcome with each approach (Office of Policy, 2021; Snowsill, 2023). In previous studies, aABR presents superior sensitivity and specificity compared to TEOAE (Sheng, 2021; Song et al., 2015). TEOAE was chosen as a comparator, or the alternative method of testing, because it is a common tool used in UNHS and it is relatively less costly and easier to operate than the aABR (Heidari et al., 2017). CEA was conducted to identify the optimal approach from a healthcare system perspective with a focus on health-related benefits. Findings can inform screening decisions for NICU populations.

Operational Definitions

For modeling cost-effectiveness of diagnostic tests, a decision tree approach reflects the diagnostic pathway and allows for predicting the volume of neonates who follow a particular diagnostics pathway (Burke et al., 2012; Snowsill, 2023). Diagnostic pathways each generate a unique cost-effectiveness outcome (Burke et al., 2012).

CEA produces an Incremental Cost-Effectiveness Ratio (ICER), calculated by dividing the difference in total costs (incremental cost) by the difference in the outcome of the screening tool (incremental effect). The ICER is a ratio of additional cost per additional unit of effectiveness for aABR compared to TEOAE (Bang & Zhao, 2012).

Cost Parameters

As of June 2023, 85,614,581 individuals were enrolled in Medicaid (Centers for Medicare & Medicaid Services, 2023). Medicaid is a federally mandated program managed by each state (Centers for Medicare and Medicaid Services, n.d.). Thus, applying Medicaid reimbursement rates as a cost of care reflects the cost incurred by U.S. society. Indirect patient costs were not included and the cost of providers, materials, and facilities were not separately accounted for in this analysis. Medicaid reimbursement rates are determined by the state, with federal guidelines that rates are set high enough to attract provider participation and do not exceed Medicare rates (Burney et al., 1979). Thus, published state Medicaid rates were collected from 34 state Medicaid websites (effective 2021–2023). Descriptive statistics were conducted to describe Medicaid reimbursement rates and results revealed positively skewed distributions. Cost for TEOAE and aABR was calculated as the mean Medicaid reimbursement rate. Per CHEERS guidelines, costs were not converted from the mean Medicaid rate. United States currency was not converted.

Effectiveness Parameters

As in similar studies (Heidari et al., 2017), effectiveness is defined as the subset of identified neonates who were confirmed as having HL with diagnostic testing compared to the total number of neonates indicated as being at risk for HL by a positive screener. The screening tools' clinical validity, or accuracy in detecting infants later diagnosed with HL, was identified via the published observations of the TEOAE and aABR screening true and false positive and negative results. Khaimook and colleagues (2019) aimed to identify permanent bilateral HL of at least moderate severity thus imposing a 40dB threshold. True positives cases were those exceeding 40dB nHL (normal hearing level; Khaimook et al., 2019), including conductive, reversible HL, which led to accurate detection of risk for HL and referral for necessary diagnostic testing. This is reflected in the calculation of the prevalence rate for the sample (Snowsill, 2023). True negative cases were those below 40dB nHL (Khaimook et al., 2019) and led to accurate identification of those not at risk for HL and prevented over-referral to audiology. True positive and negative cases were selected because of the benefits to the healthcare system. Sensitivity and specificity were calculated relative to the published follow-up ABR diagnostic results that confirmed HL status in all participants (Khaimook et al., 2019; Snowsill, 2023). Sensitivity and specificity were the outcomes used to reflect benefits and harms to the healthcare system. Formulas for effectiveness are reflected in Table 1.

Table 1

Formulas for Calculating Screening Effectiveness (Khaimook et al., 2019)

Effectiveness	Effectiveness formula
Eff. of identifying T+	*N**prev. of HL*sens. of screening tool
Eff. of identifying F-	*N**prev. of HL*(1-sens. of screening tool)
Eff. of identifying T-	*N**prev. of HL*spec. of screening tool
Eff. of identifying F+	*N**prev. of HL*(1-spec. of screening tool)

Note. *N* = total sample of newborns, Eff. = Effectiveness, prev. = prevalence, T+ = true positive, F- = false negative, T- = true negative, F+ = false positive, sens. = sensitivity, spec. = specificity

Effectiveness

OAE screening yielded a sensitivity of 78.7% and a specificity of 88.8%; whereas, aABR screening yielded a sensitivity of 91.7% and a specificity of 92.1% (Khaimook et al., 2019). Costs in Table 2 reflect median reimbursement rates for OAE (current procedural terminology CPT code 92587 without a modifier) and aABR (current procedural terminology CPT code 92650). The 92587 CPT code is a limited evaluation code for distortion product evoked otoacoustic emissions or transient evoked otoacoustic emissions and assumes an audiologist provided a report and interpretation describing the screening results (American Speech-Language-Hearing Association, n.d.-a). Whereas the CPT code 92558 describes a pass/fail automated evoked otoacoustic emissions screening often used with NBHS, CPT 92587 is a better representation of the TEOAE screening services conducted for the 144 newborns in the Khaimook study (American Speech-Language-Hearing Association, n.d.-a; Khaimook et al., 2019). UNHS are conducted within the first month of life and a discount rate was not used for this analysis. An audiologist was consulted to confirm CPT code selection for cost calculations and in the overall design of the study.

Economic Analysis Procedures

Using TreeAge, a decision tree software program used in CEA, a decision tree was designed to model the diagnostic pathway of congenital HL to branch by true disease and test results (TreeAge Pro, 2021). We used the decision tree tool as a methodological approach to provide the results of the cost-effectiveness analysis. The decision tree begins with the number of neonates (n = 144) included in the CEA of hearing screening tools and then branches out into the two screening tools: aABR and TEOAE. Following a validated modeling approach (Snowsill, 2023), each screening tool first branches into true disease as reflected by chance nodes with the prevalence of HL and normal hearing then branches further into chance nodes reflecting clinical validity of the screening tool: true positive (sensitivity), false negative, true negative (specificity), and false positive (Snowsill, 2023). The final nodes end with the cost-effectiveness formula.

Each device has four branches and end nodes (Figure 1). Expected cost is determined as:

- Branch aABR (or TEOAE) screen + (true +): Cost of screening newborns with a definite diagnosis of HL and identifying risk for HL with screening.
- Branch aABR (or TEOAE) screen – (false -): Cost of screening newborns with a definite diagnosis of HL and not identifying risk for HL with screening.
- Branch aABR (or TEOAE) screen – (true -): Cost of screening newborns without a diagnosis of HL and correctly identifying those not at risk for HL in screening.
- Branch aABR (or TEOAE) screen + (false +): Cost of screening newborns without a diagnosis of HL and incorrectly identifying as at risk for HL in screening.

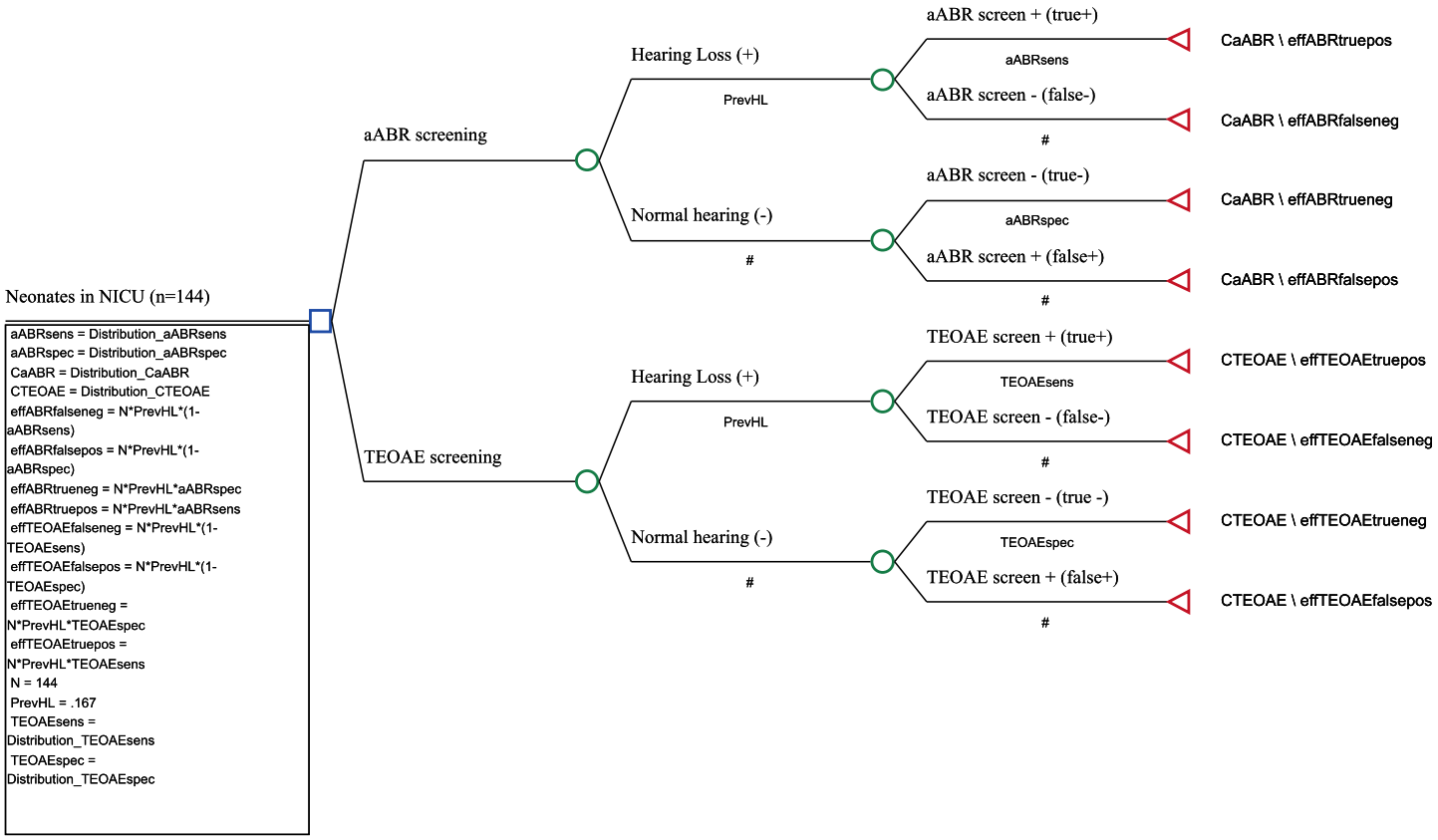
Cost-effectiveness values are multiplied by the number of neonates associated with each branch to calculate expected costs (Burke et al., 2012). The ICER will provide a ratio of additional cost per additional unit of effectiveness for aABR compared to TEOAE (Bang & Zhao, 2012). Applying screening results for a cohort of neonates from Thailand to U.S. currency, this analysis will identify the optimal strategy for NBHS in a Thailand NICU (Bang & Zhao, 2012).

Additional Economic Evaluation Practices

Consistent with standard practice for published economic evaluations, the CHEERS guidelines were followed for this analysis (Husereau et al., 2022). There were no anticipated ethical issues expected in this study. There were no methods used to statistically transform the effectiveness data, nor were any extrapolation methods used. Standard

Figure 1

Decision Tree, aABR compared to TEOAE for UNHS in NICU



Note. aABR = Automated Auditory Brainstem Response, HL = hearing loss; NICU = Newborn Intensive Care Unit, TEOAE = Transient Evoked Otoacoustic Emissions, UNHS = Universal Newborn Hearing Screening.

deviations for effectiveness data were not provided in the literature. Reimbursement rates varied by state. Screening equipment effectiveness can vary in terms of clinical accuracy, malfunctioning, maintenance, administration, and clinical populations presenting different prevalence rates of HL. Thus, there was uncertainty regarding costs, sensitivity, and specificity. Gamma and beta distributions were applied for cost and clinical validity uncertainties, respectively, because gamma distribution is suggested for non-negative parameters whereas beta distribution is suggested for binomial outcomes (Briggs, 2006).

Additional CEA

The study sample was very specific to babies at risk for HL who were born in Thailand (Khaimook et al., 2019). Two additional analyses were conducted to assess results for different levels of prevalence and assess for heterogeneity across HL severities. In general, a higher prevalence rate of disease is associated with higher sensitivity and lower specificity (Murad et al., 2023). Thus, the first additional CEA was conducted to identify the optimal approach given a U.S. NICU prevalence rate (2.3 per 100), sensitivity and specificity from the Thailand study, and Medicaid median rates.

Khaimook and colleagues (2019) reported the severity of HL diagnosed among neonates identified with HL. Additional CEA was conducted, and results were compared across (a) babies without HL (n = 122) and those with bilateral moderate HL (n = 8) and (b) babies without HL (n = 122) and those with bilateral severe to profound HL (n = 6). To account for the wide range of cost data, further analyses of the cost data were conducted with interquartile Medicaid reimbursement rates across the 34 states.

Sensitivity Analyses

Sensitivity analysis with tornado diagrams developed in TreeAge reflected the variables in the model that had the greatest impact on the ICER. Sensitivity and specificity are not fully independent of prevalence; thus, sensitivity analysis was considered to evaluate differences in HL prevalence and screening tool sensitivities and specificities for U.S. and Thailand NICU cohorts. Prevalence, sensitivity, and specificity for this study were based on a cohort of infants in a Thailand NICU. Risk factors for HL may differ in Thailand and the United States. Higher prevalence is associated with higher sensitivity and lower specificity (Murad et al., 2023). A comparison of prevalence rates of HL in a U.S. NICU (2.3 per 100) and a Thailand NICU (16.7 per 100) is warranted (Khaimook et al., 2019; White et al., 1994). However, the U.S. NICU cohort analysis did not provide follow-up on infants who passed the screening to confirm hearing status. Thus, ROC analysis could not be conducted to compare the prevalence of HL in Thailand and U.S. NICUs (Khaimook et al., 2019; White et al., 1994).

Results

Study Parameter Values

Study parameters included effectiveness (sensitivity, specificity, prevalence), cost, and benefit. Values are reflected in Table 2. In this analysis, the prevalence of HL among the 144 newborns was .167 because 24 newborns were diagnosed with HL based on diagnostic ABR (Khaimook et al., 2019). The sensitivity of the TEOAE was .787 and the sensitivity of the aABR was .917 (Khaimook et al., 2019). The specificity of the TEOAE was .888 and the specificity of the aABR was .921 (Khaimook et al., 2019). For this analysis, costs were the mean values of published Medicaid reimbursement rates per screen across 34 states ($29.03 for TEOAE and $34.09 for aABR).

Table 2
Cost-Effectiveness Study Parameters

Parameter	TEOAE	aABR	ABR	Reference
Prevalence	--	--	24/144 (0.167)	(Khaimook et al., 2019)
Sensitivity	0.787	0.917	--	(Khaimook et al., 2019)
Specificity	0.888	0.921	--	(Khaimook et al., 2019)
Cost	$29.03	$34.09	--	Mean state Medicaid reimbursement rate

Adjustments

Given the uncertainty of costs and screening equipment accuracy, adjustments were made to the parameters in the model (e.g., costs and outcomes). Standard deviations were not provided in the literature and thus assumptions were made for distribution purposes. Due to the range in cost per TEOAE and aABR, the gamma distribution was applied to costs for TEOAE (M = $29.03, SD = $15.42) and aABR ($M$ = $34.09, SD = $20.08). Further, as the sensitivity and specificity of the equipment may vary across populations with different prevalence rates of HL, beta distributions were applied to these values for the aABR (sensitivity M = .917, SD = $\pm$ .02; specificity .921, SD = $\pm$ .02) and the TEOAE (sensitivity M = .787, SD = $\pm$ .03; specificity .888, SD = $\pm$ .03). See Table 3.

Histograms shown in Figures 2a–2f reflect the appropriateness of the distributions for the analysis using 10,000 samples and established mean and standard deviation values (see also Table 4). The Medicaid reimbursement rate of aABR (M = $34.09; SD = $20.08; Mdn = $26.06) and the cost of TEOAE (M = $29.03; SD = $15.42; Mdn = $25.75) reflect slightly right-skewed distributions which indicate the means of these parameters are greater than the medians and thus overestimate common values in the reimbursement rates across states. The sensitivity of aABR (M = .917, SD = $\pm$ .02), the specificity of aABR (M = .921, SD = $\pm$ .02), the sensitivity of TEOAE (M = .787, SD = $\pm$ .03), and the specificity of TEOAE (M = .888, SD = $\pm$ .03) reflect slightly left-skewed distributions which indicate the means of these parameters are less than the medians and thus underestimate common values in the distribution or the true rate of identifying true positives and true negatives. This could lead to the cost-effectiveness analysis results reflecting a slightly lower value than the true value of the effectiveness of the screening tools.

Table 3
Distributions, Means, Standard Deviations for Parameters

Parameter	Distribution	*Mean* ($\pm$ *SD*)
Cost per screening	Gamma	
aABR		$34.09 ($\pm$20.08)
TEOAE		$29.03($\pm$15.42)
Sensitivity	Beta	
aABR		0.917($\pm$.02)
TEOAE		0.787($\pm$.03)
Specificity	Beta	
aABR		0.921($\pm$.02)
TEOAE		0.888($\pm$.03)

Note. Costs were identified from published Medicaid reimbursement rates for 34 states. aABR = Automated Auditory Brainstem Response, TEOAE = Transient Evoked Otoacoustic Emissions

Figures 2a-f

Histograms for Each of the Parameters from 10,000 Samples

Figure 2a. Distribution of Costs for aABR

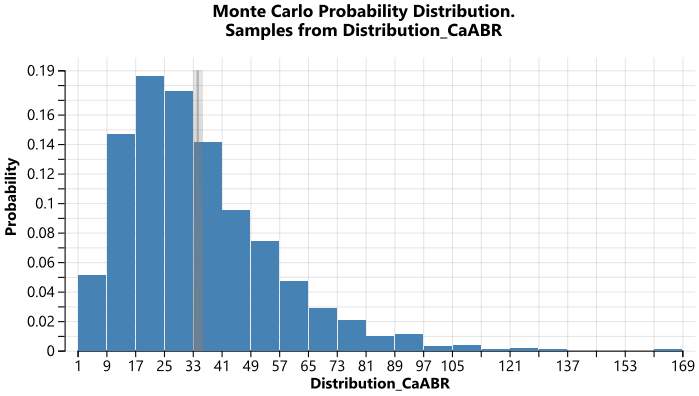


Figure 2b. Distribution of Costs for TEOAE

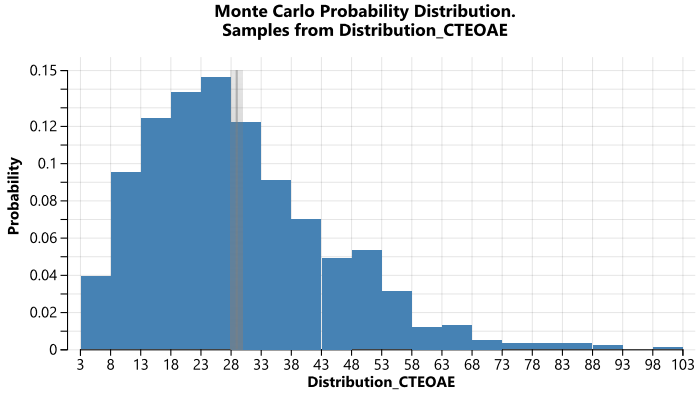


Figure 2c. Distribution of Sensitivity for aABR

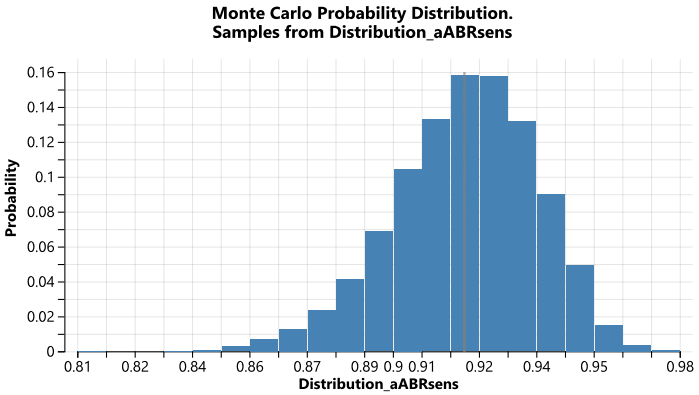


Figure 2d. Distribution of Sensitivity for TEOAE

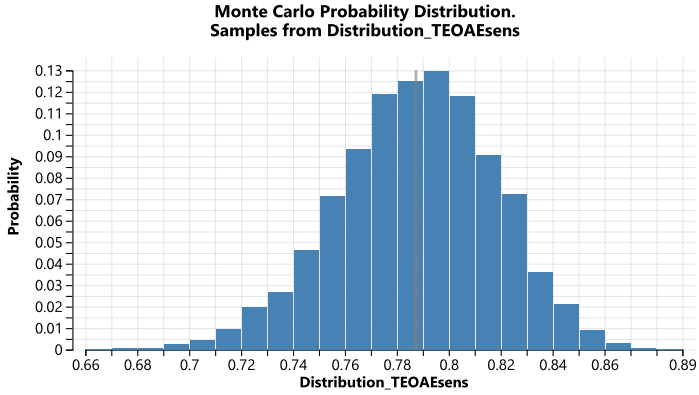


Figure 2e. Distribution of Specificity for aABR

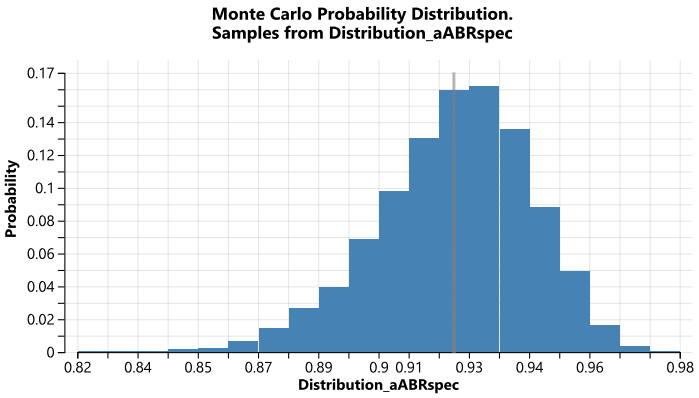
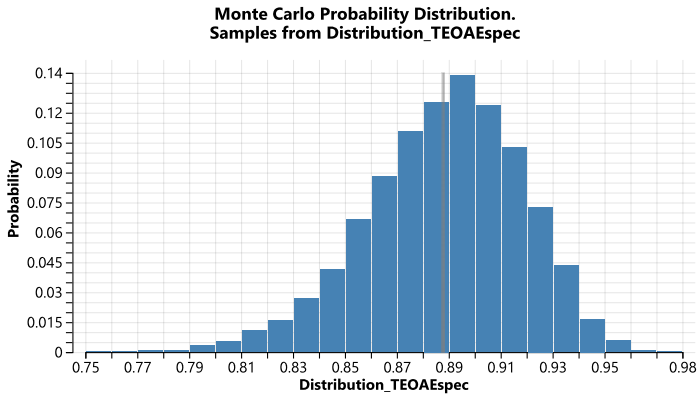


Figure 2f. Distribution of Specificity for TEOAE



Note. aABR = Automated Auditory Brainstem Response, TEOAE = Transient Evoked Otoacoustic Emissions

Table 4

Mean Values for Costs and Outcomes

Parameter	TEOAE	aABR
Cost per screening	$29.03	$34.09
Incremental Cost	--	$5.06
Outcomes (effectiveness)	18.72	20.52
Incremental effectiveness	--	1.80
Incremental Cost-effectiveness ratio (ICER)	--	2.80
Net monetary benefit	813.23	889.38

Note. aABR = Automated Auditory Brainstem Response, TEOAE = Transient Evoked Otoacoustic Emissions

Primary CEA Results

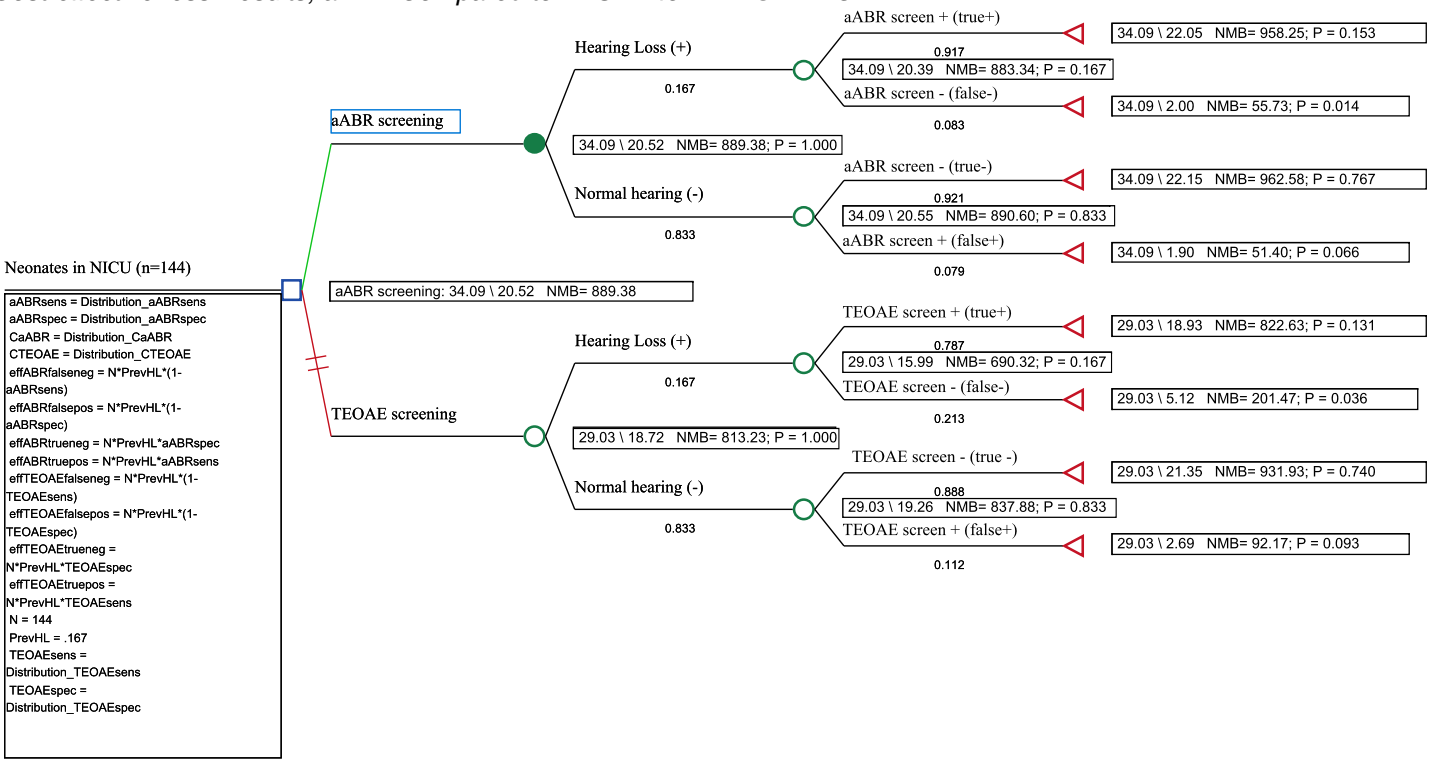
Among the 144 newborns participating in the study, 24 were ultimately diagnosed with HL via a diagnostic ABR. Of the 24 diagnosed with HL, 20 failed the aABR and 18 failed the TEOAE. This suggests a higher false negative rate associated with the TEOAE. Overall results indicate that in a sample of 144 newborns at higher risk for HL than well-babies, the aABR will accurately identify 20 of 24 babies in a Thailand NICU with HL at a cost of $34.09 per screening while the TEOAE will accurately identify 18 of 24 babies in a Thailand NICU with HL at a cost of $29.03 per screening (U.S. currency; see Figure 3). CEA criteria suggests that a dominant treatment is one that is both more effective and less costly (Snowsill, 2023). Although the aABR is not dominant due to a higher cost than the TEOAE, results (ICER = 2.80) indicate the aABR costs an additional $2.80 per each additional true positive screening (see Figure 4).

Additional CEA Results with U.S. Prevalence Rate

Additional cost-effectiveness analyses were conducted to identify if the cost-effectiveness results differ given a U.S. NICU prevalence rate (6 per 1000; Butcher et al., 2019), using sensitivity and specificity from the Thailand study and U.S. Medicaid mean rates. In a sample of 144 newborns at high risk for HL, both tools will accurately identify 2 of 3 babies with HL at a cost of $29.03 per TEOAE screening and $34.09 per aABR screening (U.S. currency). An ICER of 27.10 reveals the aABR costs

Figure 3

Cost-effectiveness Results, aABR Compared to TEOAE for UNHS in NICU



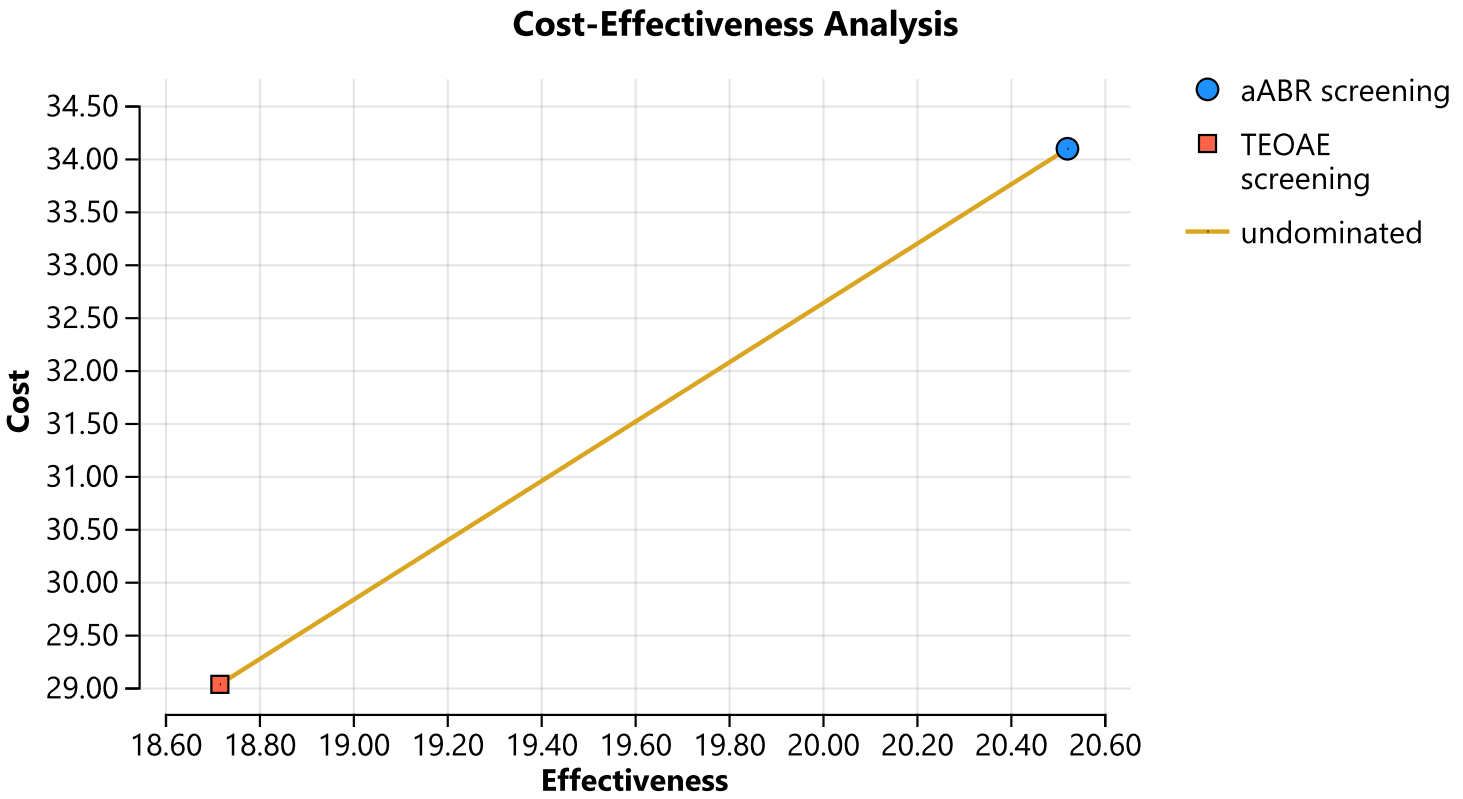
Note. aABR = Automated Auditory Brainstem Response, NICU = Newborn Intensive Care Unit, TEOAE = Transient Evoked Otoacoustic Emissions, UNHS = Universal Newborn Hearing Screening.

an additional $27.10 per each additional true positive screening. Thus, the lower prevalence yields a higher ICER. Results should be viewed with caution because the higher prevalence rate in the Thailand NICU may yield higher sensitivity and lower specificity than the lower prevalence associated with a U.S. NICU cohort. This is a general analysis using published prevalence rates for the entire United States. It is important to note that prevalence rates differ by populations. Audiologists would be wise to consider both these CEA results and the recommendations from the Joint Committee on Infant Hearing regarding the application of aABR in the NICU population.

Additional CEA Results by HL Severity

Further cost-effectiveness analyses were conducted to determine if results differ by HL severity. Using published thresholds for HL severity, Khaimook and colleagues (2019) identified eight infants who were ultimately diagnosed with mild HL (21–40 dB HL), eight infants who were ultimately diagnosed with moderate HL (41–70 dB HL), and six infants who were ultimately diagnosed with severe (71–90 dB HL) to profound (more than 90 dB HL) HL. Both screening tools were used to accurately identify 6 of 8 babies, who were ultimately diagnosed with moderate HL, from a sample of 130 newborns (those babies with typical hearing and those with moderate HL) in the Thailand NICU. The diagnosis cost $29.03 per TEOAE screening and $34.09 per aABR screening (U.S. currency). An ICER of 10.60 reveals the aABR will cost an additional $10.60 per each additional true positive screening for newborns in the NICU who will later be

Figure 4
Cost Effectiveness Analysis



Note. aABR = Automated Auditory Brainstem Response, TEOAE = Transient Evoked Otoacoustic Emissions

diagnosed with moderate HL. In a sample of 128 newborns in the Thailand NICU (those babies with typical hearing and those with severe to profound HL), it was found that 5 of 6 babies were accurately identified by both screening tools and later diagnosed with severe to profound HL. The cost was $29.03 per TEOAE screening and $34.09 per aABR screening (U.S. currency). An ICER of 13.20 reveals the aABR will cost an additional $13.20 per each additional true positive screening for newborns in the NICU who will later be diagnosed with severe to profound HL. Thus, among this cohort, the ICER is higher for the sample of newborns in the NICU who will later be diagnosed with severe to profound HL compared to the ICER for the babies later diagnosed with moderate HL.

Sensitivity Analyses for Primary CEA

Uncertainty regarding the inputs' exact values may affect CEA findings; thus, sensitivity analysis was warranted. Reflected in the tornado diagram (Figure 5) "ICER for aABR Screening Versus TEOAE Screening," the effectiveness of a TEOAE true negative had the greatest impact on the ICER, followed by the effectiveness of an aABR true negative and the specificity of aABR. Higher values for effectiveness post TEOAE true negative, cost of aABR screening per newborn, sensitivity and specificity of TEOAE, and effectiveness post TEOAE true positive were associated with higher ICER values. Whereas with effectiveness post aABR true negative, specificity of aABR, cost of TEOAE screening per newborn, sensitivity of aABR, effectiveness post aABR true positive, and prevalence of HL in high-risk neonates, there was an inverse relationship reflected between these variables' values and the ICER values such that higher values for these variables were associated with lower ICER values. The effectiveness post aABR true negative having the greatest impact on the ICER as an inverse relationship indicated that the aABR screening tool is the optimal choice.

Sensitivity Analysis of Cost Data

Published Medicaid reimbursement rates for TEOAE (CPT 92587) and aABR (CPT 92650) varied across 34 states. The interquartile range for TEOAE reimbursement rate was $25.75 to $41.31; whereas the interquartile range for aABR reimbursement rate was $26.06 to $44.48. Further cost-effectiveness analyses were conducted to identify if results differed by cost data. Applying the second quartile cost rates (TEOAE = $25.75; aABR = $26.06) in conjunction with the prevalence, sensitivity, and specificity associated with the original sample of 144 newborns' results (ICER 0.17) indicates the aABR will cost an additional $0.17 per each additional true positive screening for newborns in the NICU who will be later diagnosed with HL. The third quartile cost rates (TEOAE = $41.31; aABR = $44.48) applied in conjunction with the prevalence, sensitivity, and specificity associated with the original sample of 144 newborns' results (ICER 1.76) indicate the aABR will cost an additional $1.76 per each additional true positive screening for newborns in the NICU who will be later diagnosed with HL.

Discussion

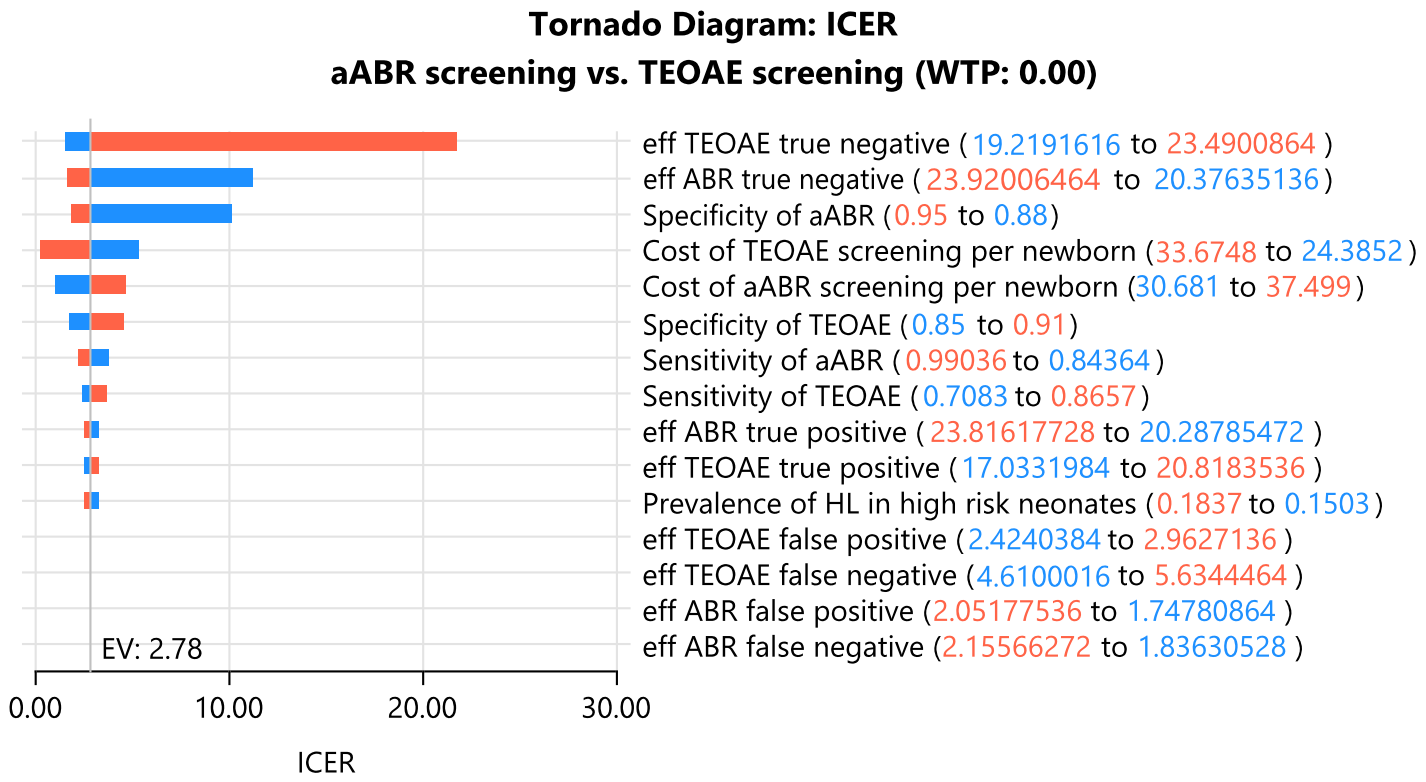
Primary CEA results

Higher sensitivity and specificity values indicate the aABR screening tool will accurately identify more NICU infants who will ultimately be diagnosed with HL than the alternative, TEOAE. The cost per screening with the aABR is higher which indicates the aABR is not a dominant strategy. The increased cost to administer the aABR per additional unit of effectiveness, as defined by the number of neonates whose risk of HL was correctly detected (Snowsill, 2023), was only $2.80 per additional true positive screening. The effectiveness of a true negative with both tools and the specificity of aABR had a strong influence on the outcome of this analysis.

Current U.S. policy mandates UNHS for all babies born in the United States. Although reputable guidelines reflect recommendations, specific equipment or protocols are not a required component of the federal mandate (JCIH, 2019; Wroblewska-Seniuk et al., 2017). Although two-stage screenings with both technologies are advantageous for detecting auditory neuropathy and identifying higher rates of minimal to mild hearing thresholds, not all hearing

Figure 5

Tornado Diagram: Incremental Cost-Effectiveness Ratio



Note. aABR = Automated Auditory Brainstem Response, eff. = effectiveness, TEOAE = Transient Evoked Otoacoustic Emissions, WTP = Willingness to Pay Threshold, set at $0.00.

screening programs have the funding for both screening tools and leaders may need to select one tool for all NBHS (JCIH, 2019). Results of this analysis indicate that although the aABR equipment is more expensive, it is a more effective screening tool than the TEOAE. These results can aid program leaders in decision-making when they must choose between purchasing TEOAE or aABR equipment for the NICU setting.

Subgroup and Cost Differences

Additional cost per positive screening varies depending on prevalence rates, HL severity, and cost per screening. The lower prevalence in the U.S. NICU resulted in a more expensive increased cost to administer aABR per increased unit of effectiveness ($27.10) compared to the Thailand NICU aABR cost per increased unit of effectiveness ($2.80). Prevalence varies by region (Khaimook et al., 2019; Rein et al., 2024; White et al., 1994) which may yield different needs in terms of surveillance, routine screenings, and provider network volume. Some populations, such as Mexican Americans and those with lower household incomes, are at greater risk for pediatric HL (Mehra et al., 2009). Thus, hospitals that serve a NICU population at higher risk for HL might consider investing in aABR screening equipment to ensure more accurate screening results.

Different costs per increased unit of effectiveness were associated with those identified with moderate HL and those with severe HL. There was a higher increased cost to administer aABR per increased unit of effectiveness ($13.20) among those at risk for severe HL compared to the aABR cost per increased unit of effectiveness ($10.60) to accurately identify those at risk for moderate HL. A NICU

population is at greater risk for more severe types of HL due in large part to comorbidities and ototoxic, life-saving interventions and these severe types of HL are often better detected by the aABR as opposed to the TEOAE (Wroblewska-Seniuk et al., 2017). It is recommended that infants who stay in the NICU for more than 5 days should be screened using aABR (JCIH, 2019; Wroblewska-Seniuk et al., 2017). Audiologists advocating for hospital and program investment in aABR equipment should communicate these CEA results because a relatively small increase in cost for more effective screening outcomes will appeal to fiscally prudent administrators.

Accounting for the potential range in Medicaid reimbursement rates by using the second and third quartile values revealed the second quartile cost rates (TEOAE = $25.75; aABR = $26.06) were associated with a $0.17 increased cost to administer aABR per each additional newborn accurately identified at risk for HL. Whereas, the third quartile cost rates (TEOAE = $41.31; aABR = $44.48) were associated with a $1.76 increased cost to administer aABR per each additional newborn accurately identified at risk for HL. Costs and reimbursement rates differ by region and these differences can affect a program's decision to invest in new equipment. Programs with lower costs will see a lower cost increase per additional accurate detection associated with the aABR.

Limitations

Limitations of this cost-effectiveness analysis include study sample size, potential selection bias associated with a single NICU, and uncertainty of the outcomes and cost data. Further, this study involved a small cohort of newborns in the NICU in a hospital in Thailand and applied U.S. currency. Newborns admitted to the NICU present with a higher risk for HL and, as indicated by future diagnostic results, a higher prevalence of HL compared to well babies. Thus, results cannot be generalized to another population with potentially different outcomes, such as babies who did not require a NICU stay or babies born in countries with a much lower prevalence rate, such as the United States. Per the recommendations of JCIH (2019), babies whose neonatal intensive care exceeded five days and who passed the NBHS should participate in diagnostic follow-up by nine months of age. This practice should offer additional study opportunities among babies born in the United States with a history of a NICU stay. Further, the limitation of a single NICU cohort may introduce selection bias as NICU admissions criteria vary by hospital and region. Future research should evaluate the cost-effectiveness of aABR compared to TEOAE in populations of newborns in other countries and who are not at high risk for HL. In the United States, researchers should prioritize follow-up measures for all infants, even those who pass the initial screening, so that screening effectiveness data includes true negatives and false negatives.

Prevalence and effectiveness data were acquired from published literature. Cost data was acquired from published reimbursement rates. There are ambiguities regarding costs, clinical outcomes, and the true impact on the ICER. Due to a wide range of Medicaid reimbursement rates, the mean values for TEOAE and aABR may not reflect the actual cost per screening. States have discretion to set fee-for-service Medicaid rates and although rate changes have lagged behind increases in cost during economic downturns, states tend to consider increases to Medicaid rates to ensure provider participation (Cunningham et al., 2016). Thus, reimbursement rates are probably close to the true cost of screening. Costs vary, depending on regional equipment costs, staffing costs, and applied procedural codes. Screening equipment effectiveness can vary in terms of clinical accuracy, malfunctioning, maintenance, administration, and clinical populations presenting different prevalence rates of HL. Some hospitals may use screeners rather than audiologists to administer screening and thus may use a CPT code that yields a different reimbursement rate.

The focus of this CEA was on the immediate health-related benefit of accurate identification of risk for HL and not broader benefits (e.g., long-term health-, developmental-, and educational-related benefits). Societal-level, modifiable risk factors, such as birth weight, nutritional deficiencies, and socioeconomic status, and certain health conditions (e.g., genetic syndromes) are associated with pediatric HL (Mehra et al., 2009; Vasconcellos et al., 2014). This data was not available for the cohort in this study. Future research on the cost-effectiveness of UNHS tools should incorporate long-term outcomes for newborns, such as medical and academic costs and outcomes. Cost-effectiveness analysis of NBHS tools should also account for infants presenting with known risk factors for HL other than NICU admission alone.

Access to Hearing Healthcare

In many countries, the mandated UNHS yields a high volume of screenings annually (JCIH, 2019; Wroblewska-Seniuk et al., 2017). This is in large part due to the implications of undetected, untreated HL (JCIH, 2019). The current policy in the United States mandates UNHS, but the use of specific equipment is not reflected in the mandate (JCIH, 2019; Wroblewska-Seniuk et al., 2017). Infants with significant medical needs, like newborns who stay in the NICU post-delivery (National Center on Birth Defects and Developmental Disabilities, 2019; White et al., 1994), should participate in screenings with tools that provide the greatest effectiveness (JCIH, 2019; Wroblewska-Seniuk et al., 2017). Ethical or equity considerations should be made for providing the most cost-effective equipment and methods for identifying risk for HL in infants. Certain populations, like those in the NICU, benefit from more tailored access to screening care than the well-baby populations. Using the most cost-effective tool for screening the highest-risk infants could reduce unnecessary medical care for infants, lead to more accurate identification of referrals to audiology for diagnostic testing and thus reduce the burden of unnecessary testing on parents and providers, preventing overuse of hearing healthcare.

References

Akinpelu, O. V., Peleva, E., Funnell, W. R., & Daniel, S. J. (2014). Otoacoustic emissions in newborn hearing screening: A systematic review of the effects of different protocols on test outcomes. *International Journal of Pediatric Otorhinolaryngology*, *78*(5), 711–717. https://doi.org/10.1016/j.ijporl.2014.01.021

American Speech-Language-Hearing Association. (n.d.-a). *Billing and Coding for Pediatric Audiology Services*. https://www.asha.org/practice/reimbursement/coding/billing-and-coding-for-pediatric-audiology-services/

American Speech-Language-Hearing Association. (n.d.-b). *Effects of Hearing Loss on Development*. https://www.asha.org/public/hearing/effects-of-hearing-loss-on-development

Bang, H., & Zhao, H. (2012). Median-based incremental cost-effectiveness ratio (ICER). *Journal of Statistical Theory and Practice*, *6*(3), 428–442. https://doi.org/10.1080/15598608.2012.695571

Briggs, A., Sculpher, M., & Claxton, K. (2006). *Decision modelling for health economic evaluation*. Oxford University Press.

Burke, M. J., Shenton, R. C., & Taylor, M. J. (2012). The economics of screening infants at risk of hearing impairment: An international analysis. *International Journal of Pediatric Otorhinolaryngology*, *76*(2), 212–218. https://doi.org/10.1016/j.ijporl.2011.11.004

Burney, I. L., Schieber, G. J., Blaxall, M. O., & Gabel, J. R. (1979). Medicare and Medicaid physician payment incentives. *Health Care Financing Review*, *1*(1), 62–78.

Butcher, E., Dezateux, C., Cortina-Borja, M., & Knowles, R. L. (2019). Prevalence of permanent childhood hearing loss detected at the universal newborn hearing screen: Systematic review and meta-analysis. *PloS One*, *14*(7), e0219600. https://doi.org/10.1371/journal.pone.0219600

Centers for Medicare & Medicaid Services. (2023). *June 2023 Medicaid and CHIP enrollment data highlights*. Retrieved October 20, 2023 from https://www.medicaid.gov/medicaid/program-information/medicaid-and-chip-enrollment-data/report-highlights/index.html

Centers for Medicare and Medicaid Services. (n.d.). *Medicaid*. Retrieved October 1, 2023 from https://www.medicaid.gov/medicaid/index.html

Chesnaye, M. A., Bell, S. L., Harte, J. M., & Simpson, D. M. (2018). Objective measures for detecting the auditory brainstem response: Comparisons of specificity, sensitivity and detection time. *International Journal of Audiology*, *57*(6), 468–478. https://doi.org/10.1080/14992027.2018.1447697

Clemens, C. J., & Davis, S. A. (2001). Minimizing false-positives in universal newborn hearing screening: A simple solution. *Pediatrics*, *107*(3), E29. https://doi.org/10.1542/peds.107.3.e29

Colella-Santos, M. F., Hein, T. A., de Souza, G. L., do Amaral, M. I., & Casali, R. L. (2014). Newborn hearing screening and early diagnostic in the NICU. *Biomed Research International*, *2014*, 845308. https://doi.org/10.1155/2014/845308

Cunningham, P., Rudowitz, R., Young, K., Garfield, R., & Foutz, J. (2016). *Understanding Medicaid hospital payments and the impact of recent policy changes*. https://www.kff.org/report-section/understanding-medicaid-hospital-payments-and-the-impact-of-recent-policy-changes-issue-brief/

Heidari, S., Manesh, A. O., Rajabi, F., & Moradi-Joo, M. (2017). Cost-effectiveness analysis of automated auditory brainstem response and otoacoustic emission in universal neonatal hearing screening. *Iranian Journal of Pediatrics*, *27*, e5229. https://doi.org/10.5812/ijp.5229

Husereau, D., Drummond, M., Augustovski, F., de Bekker-Grob, E., Briggs, A. H., Carswell, C., Caulley, L., Chaiyakunapruk, N., Greenberg, D., Loder, E., Mauskopf, J., Mullins, C. D., Petrou, S., Pwu, R. F., & Staniszewska, S. (2022). Consolidated health economic evaluation reporting standards 2022 (CHEERS 2022) statement: Updated reporting guidance for health economic evaluations. *Value in Health*, *25*(1), 3–9. https://doi.org/10.1016/j.jval.2021.11.1351

Joint Committee on Infant Hearing. (2019). Year 2019 position statement: Principles and guidelines for early hearing detection and intervention programs. *Journal of Early Hearing Detection and Intervention, 4*(2), 1–44. https://doi.org/10.15142/fptk-b748

Khaimook, W., Pantuyosyanyong, D., & Pitathawatchai, P. (2019). Accuracy of otoacoustic emissions, and automated and diagnostic auditory brainstem responses, in high-risk infants. *Journal of Laryngology and Otology*, *133*, 363–367. https://doi.org/10.1017/S0022215119000872

Levit, Y., Himmelfarb, M., & Dollberg, S. (2015). Sensitivity of the automated auditory brainstem response in neonatal hearing screening. *Pediatrics*, *136*(3), e641–647. https://doi.org/10.1542/peds.2014-3784

Mehra, S., Eavey, R. D., & Keamy, D. G., Jr. (2009). The epidemiology of hearing impairment in the United States: Newborns, children, and adolescents. *Otolaryngology and Head and Neck Surgery*, *140*(4), 461–472. https://doi.org/10.1016/j.otohns.2008.12.022

Murad, M. H., Lifeng, L., Haitao, C., Bashar, H., Reem, A. A., Alzhraa, S. A., Reem, A. M., & Zhen, W. (2023). The association of sensitivity and specificity with disease prevalence: Analysis of 6909 studies of diagnostic test accuracy. *Canadian Medical Association Journal*, *195*(27), E925. https://doi.org/10.1503/cmaj.221802

National Center on Birth Defects and Developmental Disabilities, C. f. D. C. a. P. (2019). *2019 Summary of diagnostics among infants not passing hearing screening*. https://www.cdc.gov/ncbddd/hearingloss/2019-data/06-diagnostics.html

Office of Policy, Performance, and Evaluation. (2021). *Cost-effectiveness analysis*. https://www.cdc.gov/policy/polaris/economics/cost-effectiveness/index.html

Rein, D. B., Franco, C., Reed, N. S., Herring-Nathan, E. R., Lamuda, P. A., Alfaro Hudak, K. M., Hu, W., Hartzman, A. J., White, K. R., & Wittenborn, J. S. (2024). The prevalence of bilateral hearing loss in the United States in 2019: A small area estimation modelling approach for obtaining national, state, and county level estimates by demographic subgroup. *The Lancet Regional Health Americas*, *30*, 100670. https://doi.org/10.1016/j.lana.2023.100670

Sheng, H., Zhou, Q., Wang, Q., Yu, Y., Lui, L., Liang, M., Zhou, X., Wu, H., Tang, X., & Huang, Z. (2021). Comparison of two-step transient evoked otoacoustic emissions and one-step automated auditory brainstem response for universal newborn hearing screening programs in remote areas of China. *Frontiers in Pediatrics*, *9*. https://doi.org/10.3389/fped.2021.655625

Snowsill, T. (2023). Modelling the Cost-Effectiveness of Diagnostic Tests. *Pharmacoeconomics*, *41*, 339–351. https://doi.org/10.1007/s40273-023-01241-2

Song, C., Kang, H. S., & Ahn, J. H. (2015). Analysis of audiological results of patients referred from newborn hearing screening program. *Acta Oto-Laryngologica*, *135*, 1113–1118. https://doi.org/10.3109/00016489.2015.1063785

TreeAge Pro. (2021). *TreeAge Software*. http://www.treeage.com

Vasconcellos, A. P., Colello, S., Kyle, M. E., & Shin, J. J. (2014). Societal-level risk factors associated with pediatric hearing loss: A systematic review. *Otolaryngology and Head and Neck Surgery*, *151*(1), 29–41. https://doi.org/10.1177/0194599814526561

White, K. R., Vohr, B. R., Maxon, A. B., Behrens, T. R., McPherson, M. G., & Mauk, G. W. (1994). Screening all newborns for hearing loss using transient evoked otoacoustic emissions. *International Journal of Pediatric Otorhinolaryngology*, *29*(3), 203–217. https://doi.org/10.1016/0165-5876(94)90167-8

Wroblewska-Seniuk, K. E., Dabrowski, P., Szyfter, W., & Mazela, J. (2017). Universal newborn hearing screening: Methods and results, obstacles, and benefits. *Pediatric Research*, *81*(3), 415–422. https://doi.org/10.1038/pr.2016.250

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Documented Newborn Hearing Screenings in Florida Administrative Hospital Data: State Policy Compliance by Hospital Types

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Abstract

Purpose: Florida policy mandates newborn hearing screenings (NBHS) in hospitals. United States inpatient administrative hospital data reflects low rates of documented screenings. This analysis investigates inconsistencies between Florida policy and administrative records.

Method: Analysis of Florida statutory language was completed. Florida hospital administrative data was retrospectively analyzed using various statistical methods to explore differences in proportions of documented NBHS among distinct hospital types based on profit and teaching statuses.

Results: Florida mandate requires NBHS completion in the hospital prior to discharge from the birth facility or within 21 days after birth and allows for billing a third-party payer. The median proportions of screenings in Florida hospitals were as follows: not-for-profit teaching hospitals: 0.35 (σ: 0.00–0.83), for-profit teaching hospitals: 0.00 (σ: 0.00–0.07), not-for-profit non-teaching hospitals: 0.08 (σ: 0.00–0.36), and for-profit non-teaching hospitals: 0.05 (σ: 0.00–0.27). Hospital types exhibit significantly different proportions of documented NBHS ($\chi 2 = 194{,}321.85$, $p < .0001$).

Conclusion: Improving administrative documentation practices to align with policy will enhance adherence to statutory regulations. Boosting volume of documented screenings could lead to increased hospital revenue and present opportunities to invest in infrastructure for the NBHS program.

Keywords: newborn hearing screening, hearing test, claims analyses, policy analysis

Acronyms: EHDI = early hearing detection and intervention; HL = hearing loss; NBHS = newborn hearing screening

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The federal Early Hearing Detection and Intervention (EHDI) program aims to identify risk of congenital hearing loss (HL) within the first month of life using mandated Newborn Hearing Screenings (NBHS; American Speech-Language-Hearing Association, n.d.; Joint Committee on Infant Hearing [JCIH], 2019). Florida statute requires NBHS for all newborns prior to discharge from a hospital or birth facility and within 21 days of birth (Florida Department of Health Newborn Screening Program, n.d.; "Newborn and Infant Hearing Screening," 2023).

NBHS Identify At-Risk Newborns

Universal NBHS is a crucial first step in early identification of congenital HL (American Speech-Language-Hearing Association, n.d.). Children born with HL experience decreased language input secondary to HL (Tomblin et al., 2015). Undetected, untreated congenital HL leads to risk for significant delays in early childhood and poor academic outcomes (American Speech-Language-Hearing Association, n.d.; Khaimook et al., 2019; National Center on Birth Defects and Developmental Disabilities, 2022).

Identification of HL is crucial before children with HL can gain access to language input through use of hearing technology (e.g., hearing aids, cochlear implants). NBHS in the hospital meets the first EHDI aim and the Florida mandate. Despite mandates, analysis of U.S. claims data indicates lack of alignment between policy and record of documented NBHS for many states (Do et al., 2020).

Low Rates of Documented NBHS

Claims data reflect that low rates of documented NBHS are a problem in the United States. Findings from a study by Do and colleagues (2020) revealed 84.3% of infants born in the United States between 2013 and 2014 did not have a filed claim for NBHS (n = 384,587 among 456,407 private insurance reimbursement records). Lack of documented NBHS was likely due to a commonly used bundled claims approach (Do et al., 2020). There is a gap in understanding reasons for undocumented NBHS in administrative data. Mandates may provide clarity in understanding documentation practices. Clearly written, understandable mandates are key to provider and healthcare system compliance. A thorough review of Florida policy is necessary to understand policy-driven NBHS requirements, such as NBHS timing and location. This research will address gaps in understanding Florida state policy.

The Problem with Undocumented NBHS in Administrative Data

Florida EHDI staff report annual data to the Centers for Disease Control and Prevention (CDC; Florida Department of Health, 2021). Annual Florida CDC data reflects the percentage screened before the first month of age ranges from 95.1% to 98.2% which indicates most newborns in Florida receive a NBHS (*Annual Data: Early Hearing Detection and Intervention [EHDI] Program*, 2011-2020). Hospital factors account for 5% of late identifications of HL (Mercer et al., 2023). Screening documentation within administrative hospital records is important for patients, healthcare disciplines, and hospital systems. Lack of documented NBHS in hospital administrative records could interfere with surveillance efforts and recommended EHDI program timelines. NBHS in the hospital leads to earlier identification, diagnosis, and intervention, compared to timelines for children without a NBHS (Neumann et al., 2020; Sequi-Canet & Brines-Solanes, 2021). The average age of HL diagnosis is 4.6 months among children who received a NBHS and 34.9 months for children who do not receive a NBHS (Neumann et al., 2020). Missing documentation could delay necessary healthcare.

Policy Influences Practice

Identification of root causes for low rates of documented NBHS procedures can inform recommended approaches for improved documentation. State policy and procedures are a good source for initial understanding of expected practices. Florida policy and *Florida Department of Health (DOH) guidelines for Newborn Hearing Screening* show that Florida maintains multiple reporting systems for NBHS: the newborn's medical record, the newborn screening specimen card, the electronic state portal, and the Newborn Screening Web Order Application (Florida Department of Health, 2021; Florida Department of Health Newborn Screening Program, n.d.; "Newborn and Infant Hearing Screening," 2023). Final NBHS results must be reported within seven to 10 days following birth of a well-baby using the specimen card, the electronic portal, or the Web Order Application (Florida Department of Health, 2021). This suggests potential for provider burden of duplicative record keeping across multiple systems. Accurate record keeping of NBHS may be a challenge in Florida due to multiple documentation systems. Multiple reporting systems could explain discrepancies across providers and hospitals and may lead to providers prioritizing one reporting system over the other. Statutory language in state policy could provide clarity that informs more efficient documentation practices.

Reimbursement Policy Allowances Influence Practice

State mandates might reflect specific reimbursement requirements which in turn might affect NBHS documentation practices. NBHS are frequently bundled into claims for delivery and newborn care in the United States (Do et al., 2020). In such cases, the NBHS may not be submitted as a claim separate from comprehensive newborn care and thus, documentation of NBHS may be omitted from administrative hospital records. Florida statutes reflect NBHS is billable to Medicaid and commercial insurance companies; however, statutes do not suggest providers or hospital systems are required to submit a claim for NBHS (Florida Department of Health Newborn Screening Program, n.d.; "Newborn and Infant Hearing Screening," 2023). However, third-party payers will not reimburse for a service without administrative documentation of a procedure. Reimbursement potential of documented NBHS should motivate providers and hospital systems to document NBHS in administrative hospital records.

Hospital Factors Influences Practice

Hospital teaching and profit statuses may be associated with practice and outcomes differences (Herrera et al., 2014; Shahian et al., 2012). Comparing the proportion of documented NBHS in administrative data across hospital types could reveal which hospitals document NBHS in administrative hospital records and which hospitals can improve administrative hospital documentation. This may also suggest that facilities that lack documentation of NBHS in their administrative hospital records may bundle NBHS in their comprehensive newborn care. This research will evaluate administrative data records to identify hospital types associated with a higher proportion of documented NBHS.

Although policies inform practice, patterns in documentation of service provision may reflect reimbursement and local facility policies. This study was designed to evaluate administrative hospital data in the context of Florida statutory language. This research will answer the following research questions: (a) What requirements are reflected in current Florida policy on NBHS? and (b) What hospital factors are associated with newborn encounters reflecting a documented NBHS prior to discharge from a Florida hospital? First, a statutory language text analysis will be applied to identify requirements reflected in Florida policy

(Clinton, 2017). Second, a retrospective administrative data analysis will be conducted with multiple group comparisons across hospital types to identify differences in proportion of documented NBHS (Elliott & Hynan, 2011). Four hospital types will be defined by profit and teaching statuses: not-for-profit teaching hospitals, for-profit teaching hospitals, not-for-profit non-teaching hospitals, and for-profit non-teaching hospitals.

Method

Statutory language text analysis of Florida policy was conducted to address three questions:

1) Does policy reflect required timing of NBHS completion?
2) Does policy require completion of NBHS in hospital of birth prior to discharge?
3) Does policy provide for payer reimbursement for NBHS?

This statutory language text analysis was conducted to identify Florida mandate specifics (Clinton, 2017). The Florida policy was reviewed and answers to the above questions were recorded to develop a more nuanced understanding of the policy.

Administrative Data Analysis

Data Sources

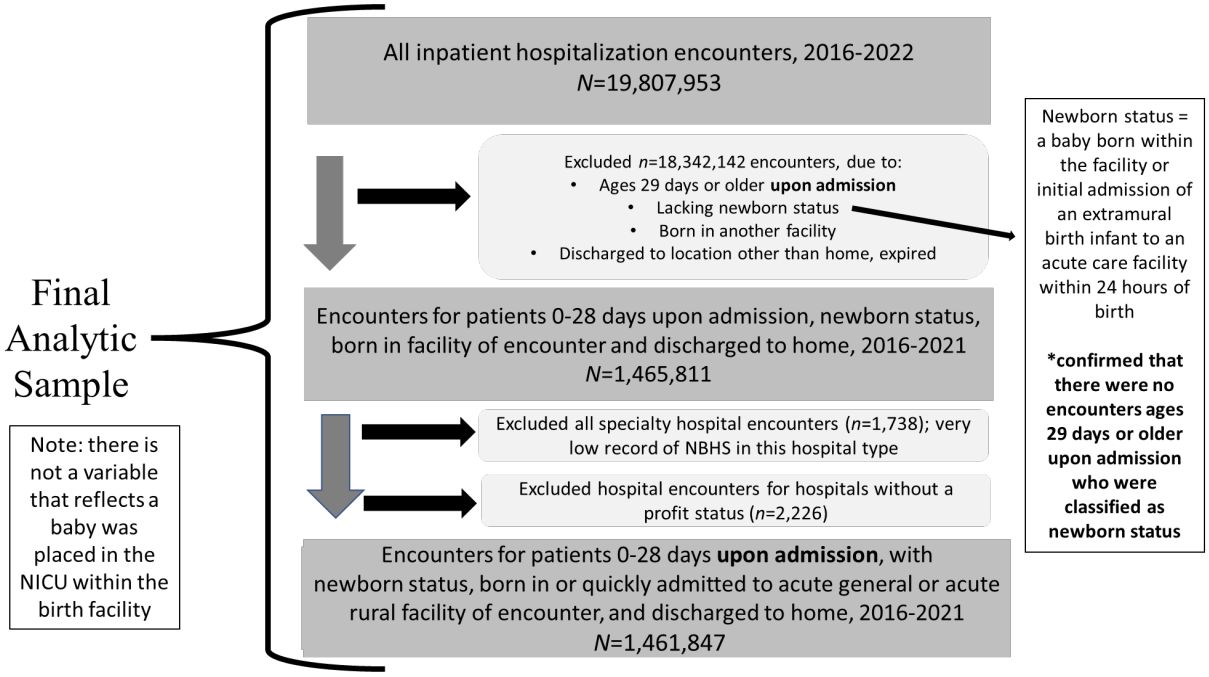
Three data sources were merged for this analysis: (a) patient care episode level, administrative hospital data from the Florida Agency for Healthcare Administration (AHCA) for inpatient hospitalizations from 2016–2022 (Agency for Health Care Administration [Florida], 2022); (b) 2023 teaching hospital records from the Centers for Medicare & Medicaid Services [CMS], 2023); and (c) 2017–2019 hospital profit status data from Florida Health Finder (*Florida Health Finder Hospital Profit Status*, 2023). AHCA inpatient data includes Florida hospital inpatient, encounter-level, administrative data with variables including patient demographics, hospital data, patient healthcare data (e.g., diagnostic codes), and encounter data (e.g., length of stay). Similar to electronic health record data, administrative hospital data is associated with an encounter and provides details about the patient and the services rendered; however, administrative hospital data is different from medical records in that it does not reveal everything recorded in the medical record. Additionally, this data does not account for Florida babies with an extramural birth (i.e., born in birthing facilities or in the home) who are not admitted for an inpatient hospitalization within 24 hours of birth. CMS hospital records reflect hospital teaching or non-teaching statuses. Florida Health Finder hospital profit status data reflects hospitals as for-profit or not-for-profit.

Study Design and Hypothesis

This retrospective administrative data analysis examined the proportion of documented NBHS for newborns in administrative hospital records associated with hospital teaching or non-teaching status and profit or not-for-profit status. This analysis of Florida inpatient hospitalization encounters for 2016–2022 included administrative hospital records of encounters for patients with a newborn status who were zero to 28 days upon admission, born in the hospital of encounter, and discharged to home (Figure 1). AHCA data encounters reflecting newborn status include Florida babies born in birthing facilities or homes who were admitted for inpatient hospitalization within 24 hours of birth. The authors hypothesized that there would be differences in the proportion and odds of documented NBHS between teaching and non-teaching hospitals, as well as between for-profit and not-for-profit hospitals.

Figure 1

Exclusion Process to Reach Final Analytic Sample



Dependent Variable

Documented NBHS was identified in an encounter if any hearing screening code was present at least once in AHCA variables for principal procedure and/or other procedure codes 1–30. Eight hearing screening codes from the CMS valid ICD-10-PCS list were considered NBHS for this analysis (F13Z0ZZ, F13Z00Z, F13Z01Z, F13Z02Z, F13Z03Z, F13Z08Z, F13ZM6Z, F13ZMZZ). This comprehensive list for hearing screening codes includes some procedures codes not intended for NBHS. However, each hearing screening code was coded at least once in the total sample of encounters across 2016–2022 and indicated a documented hearing screening for the encounter (F13Z0ZZ: *n* = 63,781; F13Z00Z: *n* = 7; F13Z01Z: *n* = 4,909; F13Z02Z: *n* = 6; F13Z03Z: *n* = 1; F13Z08Z: *n* = 33; F13ZM6Z: *n* = 141,189; F13ZMZZ: *n* = 7,190). Thus, encounters reflecting any of the eight hearing screening codes was identified as a completed and documented NBHS for the analyses. Encounters reflecting more than one hearing screening code (i.e., for rescreening) were counted once as an encounter with a documented screening.

Independent Variables

Explanatory variables included hospital teaching and profit statuses. Hospital teaching status was described as teaching or non-teaching, as reflected in CMS 2023 teaching hospital records. Hospital profit type indicated a status of for-profit or not-for-profit for each hospital, reflected in Florida Health Finder records. Multi-factor hospital types were created by combining teaching and profit status to yield not-for-profit teaching, for-profit teaching, not-for-profit non-teaching, and for-profit non-teaching hospitals.

Exclusionary Criteria

As shown in Figure 1, this analysis of Florida inpatient hospitalization encounters from 2016 to 2022 excluded encounters for patients 29 days and older upon admission, those without newborn status, individuals born in another facility, those who expired, and babies who discharged to a location other than home (*n* = 18,342,142). Newborn status was not present on any encounters with an admission age of 29 days or older. Preliminary analysis revealed newborn encounters reflected three of eight hospital types: acute general, acute rural, and specialty. Encounters for newborns in specialty hospitals (*n* = 1,738) revealed zero documented NBHS and thus were excluded. The study sample was merged with Florida Health Finder hospital profit status data for 2017–2019. Encounters lacking hospital profit status were excluded (*n* = 2,226, 0.15%). The final analytic sample included 1,461,847 encounters for newborns ages zero to 28 days upon admission, born at the acute general or rural facility of encounter, and discharged to home.

Statistical Analysis Methods

Explanatory variables were selected based on the notion that policy and hospital type influence practice. Analysis was conducted to identify proportions of NBHS among multiple hospital types (teaching, non-teaching, for-profit, not-for-profit). Methods involved calculating annual rates of documented NBHS across all newborn encounters in Florida hospitals 2016–2022 and descriptive statistics of the study sample. Bivariate statistics were calculated using chi-square analyses to examine proportions of documented NBHS in the hospital types. The odds were calculated as the probability of documented screening divided by (1- the probability of documented screening). The proportion of documented NBHS across the hospital types (not-for-profit teaching, for-profit teaching, not-for-profit non-teaching, and for-profit teaching) was tested with the Shapiro-Wilk test to examine assumptions of normality. Across all hospital types, Shapiro-Wilk test results rejected assumptions of normality (p-values < .05) indicating a non-normal distribution and need for non-parametric alternatives. The Kruskal-Willis test, a non-parametric alternative to test for significant differences, was conducted to examine if mutually exclusive hospital groups (for-profit teaching, not-for-profit teaching, for-profit non-teaching, not-for-profit non-teaching) differed significantly by proportion of documented NBHS (Elliott & Hynan, 2011). SAS version 9.4 and Microsoft Excel version 2311 were used for these analyses.

Results

Florida Policy

Preliminary statutory language text analysis of Florida NBHS policy revealed three requirements that could influence documentation of NBHS (Clinton, 2017; "Newborn and Infant Hearing Screening," 2023): (a) screening required within a few days of birth, (b) screening required in the birth facility, and (c) screening reimbursable by third party payer sources ("Newborn and Infant Hearing Screening," 2023). Review of Florida policy revealed a requirement for completion of NBHS for all newborns prior to discharge from their birth facility or within 21 days after birth (Florida Department of Health Newborn Screening Program, n.d.; "Newborn and Infant Hearing Screening," 2023). The mandate does not indicate Florida providers are required to charge for a NBHS; however, policy reflects the NBHS can be billed to Medicaid and commercial insurance ("Newborn and Infant Hearing Screening," 2023). This indicates the procedure does not need to be bundled into newborn care.

Administrative Data Analysis

Descriptives

Florida hospital administrative data for newborn encounters reflect a low rate of documented NBHS procedures across years 2016–2022 (Table 1). Annual rates of documented NBHS ranged from 11.76% to 16.08% among 1,461,847 newborn encounters. The proportion of documented NBHS (Table 2) in teaching hospitals (29.20%) and in not-for-profit hospitals (18.52%) far exceeded that of non-teaching hospitals (7.44%) and for-profit hospitals (3.98%) across the study sample.

Odds

The odds of documented NBHS varied by hospital type (Table 3). The odds of a documented NBHS were higher

Table 1

NBHS Performed Prior to Discharge Among Babies Born in Florida Acute Rural and Acute General Hospitals by Year, 2016–2022

Year	Encounters for all newborns *N* = 1,461,847 (100%)	Encounters with hearing screenings *n* = 216,486 (14.81%)	Encounters without hearing screenings *n* = 1,245,361 (85.19%)
	n	*n* (%)	*n* (%)
2016	214,558	30,700 (14.31)	183,858 (85.69)
2017	212,490	32,246 (15.18)	180,244 (84.82)
2018	210,795	33,887 (16.08)	176,908 (83.92)
2019	209,566	32, 427 (15.47)	177,139 (84.53)
2020	199,861	29,918 (14.97)	169,943 (85.03)
2021	204,254	32,596 (15.95)	171,758 (84.05)
2022	210,223	24,712 (11.76)	185,511 (88.24)

Note. NBHS = Newborn Hearing Screening.

Table 2

Bivariate Descriptive Statistics for Documented Newborn Hearing Screenings in Newborn Encounters in Florida Hospitals, 2016-2022

Hospital Types	Encounters for all newborns, total sample *N* = 1,461,847 (100%)	Encounters screened *n* = 216,486 (14.81%)		Encounters not screened *N* = 1,245,361 (85.19%)	Significance
	n	*n (%)*		*n (%)*	χ2 *(DF, N), p-value*
Hospital Teaching Type					
Teaching	494,810	144,501 (29.20)		350,309 (70.80)	122,844.00 (1, 1,461,847)*
Non-teaching	967,037	71,985 (7.44)		895,052 (92.56)	
Hospital Profit Type					
For-profit	373,140	14,848 (3.98)		358,292 (96.02)	46,578.76 (1, 1,461,847)*
Not-for-profit	1,088,707	201,638 (18.52)		887,069 (81.48)	
	n	*n* (%)	*Median* (95% *CI*)	*n* (%)	χ2 *(DF, N)*
Hospital Profit and Teaching Type					194,321.85 (3, 1,461,847)*
Not-for-profit teaching hospital	407,164	144,078 (35.39)	0.35 (0.00–0.83)	263,086 (64.61)	
For-profit teaching hospital	87,646	423 (0.48)	00.00 (0.00-0.07)	87,223 (99.52)	
Not-for-profit non-teaching hospital	681,543	57,560 (8.45)	0.08 (0.00-0.36)	632,983 (91.55)	
For-profit non-teaching hospital	285,494	14,425 (5.05)	0.05 (0.00-0.27)	271,069 (94.95)	

*p-value < .001.

in teaching (.41) versus non-teaching hospitals (0.08) and in not-for-profit (.22) versus for-profit hospitals (.04). The odds of documented NBHS were highest in not-for-profit teaching hospitals (0.54) and lowest in for-profit teaching hospitals (0.00). All non-teaching hospitals presented with odds of less than 10% for documented NBHS. Not-for-profit non-teaching hospitals presented with higher odds of documented NBHS (0.09) than for-profit non-teaching hospitals (0.05). Odds ratios revealed teaching (*OR* 5.12; 5.07–5.17) and not-for-profit (*OR* 5.48; 5.39–5.57) hospitals were more than 5 times more likely to document a NBHS than non-teaching and for-profit hospitals. Not-for-profit teaching (*OR* 112.92; 102.61–124.27) hospitals were 112 times more likely to document a NBHS than for-profit teaching hospitals.

Table 3

Odds of Documents Newborn Hearing Screening (NBHS) by Hospital Type, 2016–2022

Factors	Encounters for all newborns, total sample N = 1,461,847 (100%)	Encounters screened n = 216,486 (14.81%)	*Odds*	*Odds Ratio (95% CI)*
Hospital Teaching Status				
Teaching	494,810	144,501 (29.20)	0.4125	Teaching v. Non-Teaching 5.1289 (5.0791, 5.1792)*
Non-teaching	967,037	71,985 (7.44)	0.0804	
Hospital Profit Status				
For-profit	373,140	14,848 (3.98)	0.0414	Not-for-profit v. For-profit 5.4851 (5.3920, 5.5797)*
Not-for-profit	1,088,707	201,638 (18.52)	0.2273	
Hospital Profit and Teaching Type				
Not-for-profit teaching hospital	407,164	144,078 (35.39)	0.5476	Not-for-profit teaching v. For-profit teaching 112.9251 (102.6145–124.2716)*
For-profit teaching hospital	87,646	423 (0.48)	0.0048	
Not-for-profit non-teaching hospital	681,543	57,560 (8.45)	0.0922	Not-for-profit non-teaching v. For-profit non-teaching 1.7335 (0.5501, 1.7663)
For-profit non-teaching hospital	285,494	14,425 (5.05)	0.0532	

Note. Wilcoxon two-sample test statistic, p value < .0001; *p value <.05.

Bivariate Statistics

Chi-square tests of independence were performed to examine relationships between hospital teaching and profit statuses and proportion of documented NBHS. The relation between hospital teaching status and proportion of documented NBHS (teaching hospital: n = 144,501, 29%; non-teaching hospital: n = 71,985, 7%) was significant, χ^2 (1, N = 1,461,847) = 122,844.01, p < .001 (Table 2).

Teaching hospitals were more likely than non-teaching hospitals to document NBHS. The relation between hospital profit status and proportion of documented NBHS (for-profit hospital: n = 14,848, 3%; not-for-profit hospital: n = 201,638, 18%) was also significant, χ^2 (1, N = 1,461,847) = 46,578.76, p < .001 (Table 4). Not-for-profit hospitals were more likely than for-profit hospitals to document NBHS. Teaching hospitals presented with the highest rate of documented NBHS in the analysis.

Table 4

Post-hoc Analysis, Difference in Mean Documented Newborn Hearing Screenings by Hospital Type

Hospital Types	For-profit teaching hospital (x~0.00, σ: 0.00-0.07)	Not-for-profit non-teaching hospital (x~0.08, σ: 0.00-0.36)	For-profit non-teaching hospital (x~0.05, σ: 0.00-0.27)
Not-for-profit teaching hospital (x~0.35, σ: 0.00-0.83)	0.35*	0.27*	0.30*
For-profit teaching hospital (x~0.00, σ: 0.00-0.07)	0.00	0.08*	0.05*
Not-for-profit non-teaching hospital (x~0.08, σ: 0.00-0.36)	0.08*	0.00	0.03*

*p-value < .0001.

Kruskal-Willis test

There was a significant difference in the proportion of NBHS across all four hospital types (see Table 2). Not-for-profit teaching hospitals (x~ 0.35, σ: 0.00-0.83) presented with the highest median proportion of documented NBHS (see Table 2 and Figure 2b).

Post-hoc analysis (Table 4) revealed the greatest difference in proportion screened was between not-for-profit teaching (x~0.35, σ: 0.00-0.83) and for-profit teaching (x~0.00, σ: 0.00-0.07) hospitals.

Teaching Hospital Analysis

There were 20 teaching hospitals among 123 Florida hospitals included in this analysis. Of the 20 teaching hospitals, most were not-for-profit (*n* = 14, 70%) and some were for-profit (*n* = 6, 30%; Table 5, Figure 2a). Hospitals (*n* = 6, 30%) with the highest rates of documented NBHS were not-for-profit teaching hospitals, many associated with the same hospital system. Proportions of encounters with documented NBHS among not-for-profit teaching hospitals ranged from 0% to 93.76%. Some not-for-profit teaching hospitals (*n* = 6, 42.85%) reflected more than 75% of their encounters with documented NBHS. All for-profit teaching hospitals were in the same hospital system and presented with 0.11%–1.44% encounters with documented NBHS.

Table 5

Bivariate Descriptive Statistics of Newborn Hearing Screening in Encounters Among Florida Teaching Hospitals, 2016-2022

Hospital	Encounters for all newborns *n* = 494,810 (100%)	Encounters Screened *n* = 144,501 (29.20%)	Encounters not screened *n* = 350,309 (70.80%)
For-profit hospitals (FP)	*n*	*n* (%)	*n* (%)
FP A*	11,085	12 (0.11)	11,097 (99.89)
FP B*	23,061	331 (1.44)	22,730 (98.56)
FP C*	13,207	20 (0.15)	13,187 (99.85)
FP D*	13,130	15 (0.11)	13,115 (99.89)
FP E*	19,488	31 (0.16)	19,457 (99.84)
FP F*	7,663	14 (0.18)	7,649 (99.82)
Not-for-profit hospitals (NFP)			
NFP G	19,192	40 (0.21)	19,152 (99.79)
NFP H	20,964	0 (0.00)	20,964 (100.00)
NFP I	4,316	0 (0.00)	4,316 (100.00)
NFP J	44,162	33,900 (76.76)	10,262 (23.24)
NFP K	23,155	21,710 (93.76)	1,445 (6.24)
NFP L	31,974	29,612 (92.61)	2,362 (7.39)
NFP M	15,839	14,851 (93.76)	988 (6.24)
NFP N	21,009	19,660 (93.58)	1,349 (6.42)
NFP O	97,580	1,034 (1.06)	96,546 (98.94)
NFP P	26,460	23,271 (87.95)	3,189 (12.05)
NFP Q	17,415	0 (0.00)	17,415 (100.00)
NFP R	34,877	0 (0.00)	34,877 (100.00)
NFP S	24,203	0 (0.00)	24,203 (100.00)
NFP T	26,018	0 (0.00)	226,018 (100.00)

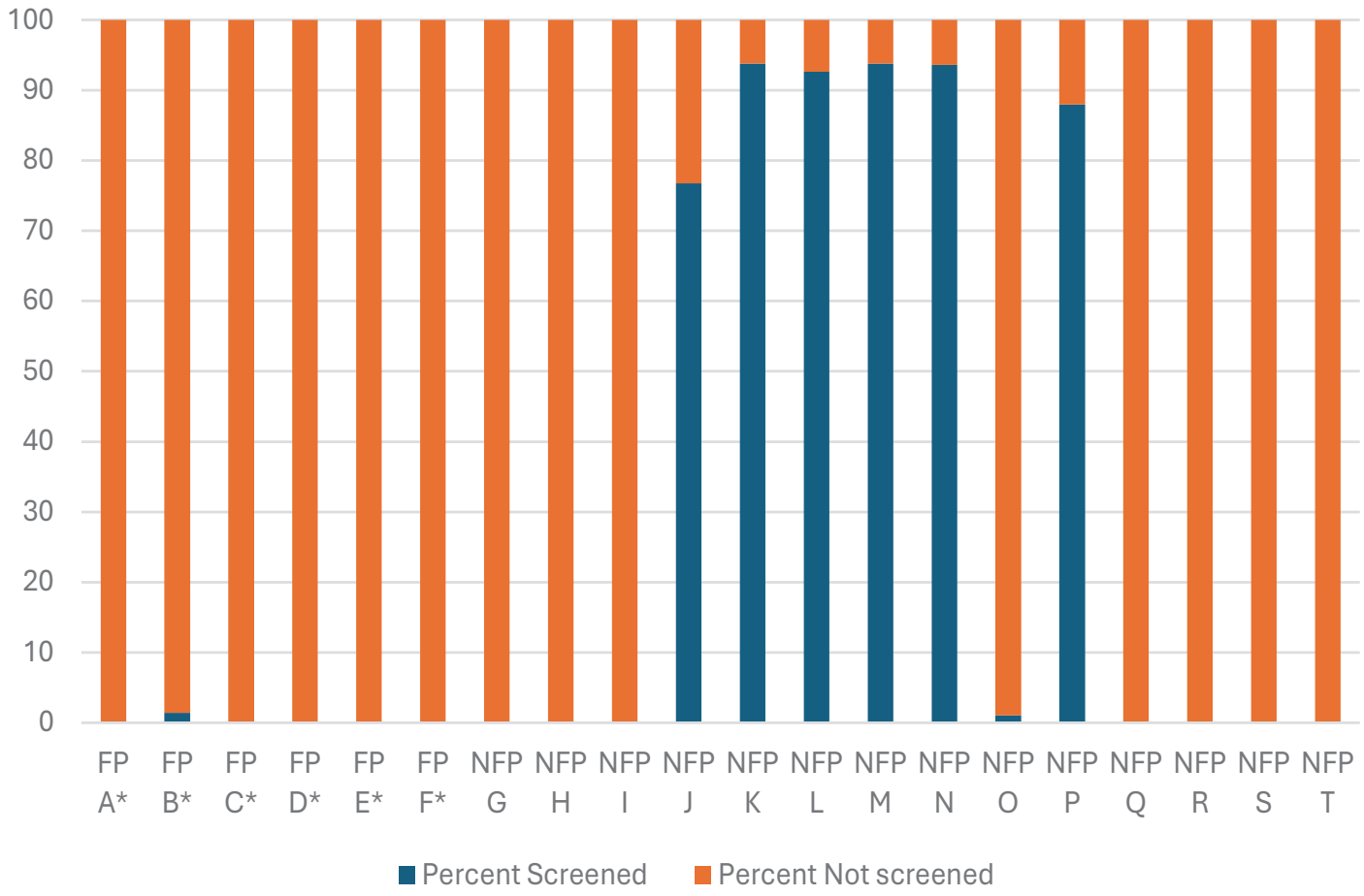
Note. Significance of facility number by screened: 408,260 $\chi 2$, $p < .0001$.

Discussion

This study addressed gaps in understanding the Florida NBHS state policy and identified hospital types associated with higher proportions of documented NBHS in Florida administrative data. Florida mandate requires that NBHS

Figure 2a

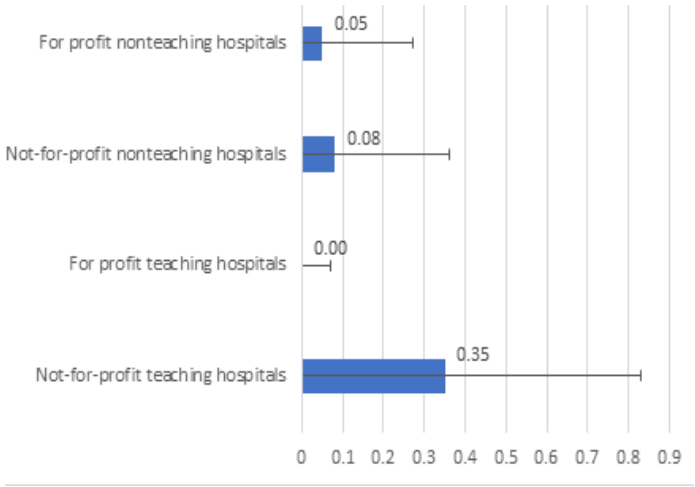
Proportion of Documented Newborn Hearing Screening Among Teaching Hospitals, For-Profit and Not-For-Profit



Note. Total newborns n = 494,810; total screened n =144,501 (29.20); total not screened n = 350,309 (70.80); FP = For-profit; NFP = Not-for-profit; *same hospital system

Figure 2b

Median Proportion of Documented Newborn Hearing Screening by Hospital Group



are completed in the birth facility prior to discharge or within 21 days of birth and permits billing a third-party for the NBHS service. Florida administrative hospital records analysis revealed a low rate of documented NBHS. Proportions of documented NBHS were different across hospital types with the highest proportion in not-for-profit teaching hospitals and the lowest proportion in for-profit teaching hospitals.

Policy Informs Practice

NBHS policy differs across states relative to location of birth, timing of NBHS relative to newborn age, and/or Medicaid and commercial insurance reimbursement allowances (Early Hearing Detection and Intervention National Technical Resource Center, 2023). Not all mandates provide clear NBHS guidelines for location or timing of screening and if billing a third-party payer is permissible. When statutory language does not clearly describe the expectations or criteria, providers may not understand requirements and allowances which could lead to discrepancies in documentation (Clinton, 2017). To ensure provider and hospital system compliance, policy should be written clearly and understandably.

Need for Clearly Articulated Policy

Florida hospitals with documented NBHS prior to newborn discharge comply with mandates (Joint Committee on Infant Hearing, 2019; "Newborn and Infant Hearing Screening," 2023). Florida mandate reflects hospitals can bill Medicaid and commercial insurance for the NBHS ("Newborn and Infant Hearing Screening," 2023). Specific statutory language informs providers what is permissible in the context of documenting and billing for NBHS. Florida mandate does not require bundling NBHS into newborn care ("Newborn and Infant Hearing Screening," 2023). This differs from other state mandates that reflect requirements to bundle NBHS into newborn care (Do et al., 2020; Early Hearing Detection and Intervention National Technical Resource Center, 2023). Clearly written policy should easily translate to practice.

Hospital Type Influences Practice

Typically, policy informs practice; however, there is a discrepancy between Florida policy and some Florida administrative hospital data. Although Florida policy reflects requirements of NBHS in the birth hospital, Florida AHCA administrative hospital data revealed that not-for-profit teaching hospitals are most likely to document NBHS. This is consistent with research that reflects differences in teaching and non-teaching hospitals and for-profit and not-for-profit hospitals (Herrera et al., 2014; Shahian et al., 2012). Teaching hospitals are known for advanced clinical capabilities and often serve as industry leaders in medical research and innovation (Shahian et al., 2012). Some research has indicated that for-profit hospitals have higher costs and mortality rates than not-for-profit hospitals (Herrera et al., 2014). Differences might extend to policy compliance, clinical documentation, and billing practices. National hospital networks with presence in multiple states may implement and enforce system-wide policies based on the strictest state mandates, to ensure compliance. Florida for-profit hospitals associated with a national network all demonstrated low rates of documented NBHS. In contrast, not-for-profit Florida hospitals associated with a different network all demonstrated higher rates of documented NBHS. Differences across state NBHS policies could explain the rate differences of documented NBHS in Florida hospital administrative data for hospitals in national networks.

Consequences for Documented NBHS

There is an opportunity to improve the proportion of documented NBHS in administrative hospital records across Florida hospitals. Improvement in the proportion of documented NBHS in administrative hospital records could have positive implications for patients, populations, clinicians, and healthcare systems. Accurate, documented NBHS are crucial for future diagnosis and treatment. Babies who do not pass the NBHS in the hospital could lack follow-up for recommended diagnostic appointments (Sequi-Canet & Brines-Solanes, 2021). Documented NBHS in administrative hospital records can contribute to surveillance efforts designed to prevent loss-to-follow-up. Procedure records inform data-driven advocacy for clinical procedural terminology (CPT) code changes with the American Medical Association (AMA, n.d.). The AMA maintains a CPT advisory committee of providers nominated by national medical professional associations (AMA, n.d.). The committee advises the CPT Editorial Panel regarding procedure coding relevant to the associated discipline and provides documentation for codes under consideration (AMA, n.d.). Accurate administrative data informs this process. Further, reimbursement requires documentation of procedures. Improved documentation could yield increased revenue for Florida hospitals that currently do not document NBHS in their administrative hospital records. Increased revenue could fund new NBHS equipment and surveillance infrastructure.

Limitations and Future Research

This study is limited by reliance on accurate documentation in the administrative hospital records, lack of accounting for third-party screening vendors or outpatient screening, and the use of retrospective data. AHCA relies on accurate clinical documentation. The low rate of documented NBHS reflected in the AHCA administrative hospital data probably reflects lack of documentation as opposed to lack of service provision. This distinction is important to determine the best course of action. Additionally, this study did not account for documentation of other attempts at NBHS, such as outpatient re-screening, or outside screening vendor record keeping maintained outside of hospital records. Analysis of outpatient NBHS and outside NBHS vendor records may reveal more consistent documentation as these services may not be bundled in the care of a newborn. This study involved retrospective secondary data analysis and no causal inference can be concluded.

Many research opportunities could address gaps in understanding the reasons for discrepancy between Florida NBHS policy and administrative hospital records. First, mandates differ across states and there is opportunity to conduct a similar analysis with other states' administrative hospital data to determine if the difference in mandates and practice are common across states (Clinton, 2017). Second, future research could evaluate the effect of state mandate changes on the proportion of documented NBHS in administrative data. Third, researchers could evaluate single versus multiple documentation methods and processes to identify outcome differences. Fourth, additional research could identify provider and hospital administrator understanding of policy. Finally, research could also assess reimbursement outcomes following implementation of improved documentation practices.

Recommendations

State policies with clear statutory language could yield more consistent provider compliance in clinical documentation. The commonly used clinical training adage "if it was not documented, it did not happen" should be a consideration in the context of NBHS administrative hospital data. Policy should explicitly describe documentation and reimbursement requirements to ensure practice aligns with policy.

Providers, families, and healthcare systems would benefit from a universal policy with documentation requirements in one system that links to state and federal agencies. Single data entry linked to other systems reduces documentation burden, simplifies record access, increases likelihood of statutory compliance, and leads to potential increase in revenue. There is a complex intersection between policy, technology, and healthcare delivery, particularly with clinical documentation (Johnson et al., 2021). Electronic health records (EHR) provided solutions for communication and safety; however, clinicians are frustrated with EHRs (Johnson et al., 2021). Multiple reporting systems will only expound frustration. Multiple system data entry perpetuates the problem of fragmented medical information systems that disrupt workflows (Janett & Yeracaris, 2020). Complex records and access points cause concern among families about accessibility. Patients are concerned about poor usability of complex medical record systems (Zarcadoolas et al., 2013). Multiple systems could contribute to complexity. Simplifying and linking NBHS documentation could aid in accurate records and quality surveillance.

References

Agency for Health Care Administration (Florida). (2022). *Hospital Discharge Data/Inpatient Data.* https://ahca.myflorida.com/agency-administration/florida-center-for-health-information-and-transparency/office-of-data-collection-quality-assurance

American Medical Association. (n.d.). *The CPT code process*. https://www.ama-assn.org/about/cpt-editorial-panel/cpt-code-process

American Speech-Language-Hearing Association. (n.d.). *Effects of Hearing Loss on Development*. https://www.asha.org/public/hearing/effects-of-hearing-loss-on-development

Centers for Disease Control and Prevention. (2024). *Annual Data: Early Hearing Detection and Intervention (EHDI) Program*. (2011-2020). https://www.cdc.gov/ncbddd/hearingloss/ehdi-data.html

Centers for Medicare & Medicaid Services. (2023). *Open Payments List of Teaching Hospitals*. https://www.cms.gov/files/document/open-payments-2023-report-cycle-teaching-hospital-listp.pdf

Clinton, J. D. (2017). Coding the ideological direction and content of policies. *Annual Review of Political Science*, *20*(1), 433–450. https://doi.org/10.1146/annurev-polisci-062615-031524

Do, T. Q. N., Chung, W., & Grosse, S. D. (2020). Private insurance reimbursements for newborn hearing screening in the United States, 2013-2014 birth cohort. *Journal of Early Hearing Detection and Intervention*, *5*(1), 13–19. https://doi.org/10.26077/q64a-ce52

Early Hearing Detection and Intervention National Technical Resource Center. (2023, June 29). *State EHDI/UNHS mandates: Summaries*. https://www.infanthearing.org/legislative/summary.html

Elliott, A. C., & Hynan, L. S. (2011). A SAS® macro implementation of a multiple comparison post hoc test for a Kruskal–Wallis analysis. *Computer Methods and Programs in Biomedicine*, *102*(1), 75–80. https://doi.org/10.1016/j.cmpb.2010.11.002

Florida Department of Health. (2021). *Florida guidelines for newborn hearing screening*. https://www.floridanewbornscreening.com/wp-content/uploads/Florida-Guidelines-for-Newborn-Hearing-Screening-4-2.pdf

Florida Department of Health Newborn Screening Program. (n.d.). *Your newborn's screening is comprised of 3 components: for parents.* https://floridanewbornscreening.com/

Florida Health Finder Hospital profit status. (2023). https://quality.healthfinder.fl.gov/

Herrera, C. A., Rada, G., Kuhn-Barrientos, L., & Barrios, X. (2014). Does ownership matter? An overview of systematic reviews of the performance of private for-profit, private not-for-profit and public healthcare providers. *PloS One*, *9*(12), e93456-e93456. https://doi.org/10.1371/journal.pone.0093456

Janett, R. S., & Yeracaris, P. P. (2020). Electronic medical records in the american health system: Challenges and lessons learned. *Cien Saude Colet*, *25*(4), 1293–1304. https://doi.org/10.1590/1413-81232020254.28922019

Johnson, K. B., Neuss, M. J., & Detmer, D. E. (2021). Electronic health records and clinician burnout: A story of three eras. *Journal of the American Medical Informatics Association*, *28*(5), 967–973. https://doi.org/10.1093/jamia/ocaa274

Joint Committee on Infant Hearing. (2019). Year 2019 position statement: Principles and guidelines for early hearing detection and intervention programs. *Journal of Early Hearing Detection and Intervention, 4*(2), 1–44. https://doi.org/10.15142/fptk-b748

Khaimook, W., Pantuyosyanyong, D., & Pitathawatchai, P. (2019). Accuracy of otoacoustic emissions, and automated and diagnostic auditory brainstem responses, in high-risk infants. *Journal of Laryngology and Otology*, *133*, 363–367. https://doi.org/10.1017/S0022215119000872

Mercer, D., Tran, T., McCabe, D., Ibieta, T., & Hubbard, D. (2023). Factors contributing to late identification of deaf or hard of hearing children in Louisiana. *The Journal of Early Hearing Detection and Intervention*, *8*(2), 16–25. https://digitalcommons.usu.edu/cgi/viewcontent.cgi?article=1211&context=jehdi

National Center on Birth Defects and Developmental Disabilities, C. f. D. C. a. P. (2022, July 18). *About Hearing Loss in Children*

https://www.cdc.gov/hearing-loss-children/about/?CDC_AAref_Val

Neumann, K., Euler, H. A., Chadha, S., & White, K. R. (2020). A survey on the global status of newborn and infant hearing screening. *Journal of Early Hearing Detection and Intervention*, *5*(2), 63–84. https://doi.org/10.26077/a221-cc28

Newborn and Infant Hearing Screening, § 1 (2023). http://www.leg.state.fl.us/statutes/index.cfm?App_mode=Display_Statute&URL=0300-0399/0383/Sections/0383.145.html

Sequi-Canet, J. M., & Brines-Solanes, J. (2021). Keypoints to successful newborn hearing screening. Thirty years of experience and innovations. *Healthcare*, *9*(11), 1436. https://doi.org/10.3390/healthcare9111436

Shahian, D. M., Nordberg, P., Meyer, G. S., Blanchfield, B. B., Mort, E. A., Torchiana, D. F., & Normand, S. L. (2012). Contemporary performance of U.S. teaching and nonteaching hospitals. *Academic Medicine*, *87*(6), 701–708. https://doi.org/10.1097/ACM.0b013e318253676a

Tomblin, J. B., Harrison, M., Ambrose, S. E., Walker, E. A., Oleson, J. J., & Moeller, M. P. (2015). Language outcomes in young children with mild to severe hearing loss. *Ear and Hearing*, *36*(Suppl 1), 76s–91s. https://doi.org/10.1097/aud.0000000000000219

Zarcadoolas, C., Vaughon, W. L., Czaja, S. J., Levy, J., & Rockoff, M. L. (2013). Consumers' perceptions of patient-accessible electronic medical records. *Journal of Medical Internet Research*, *15*(8), e168. https://doi.org/10.2196/jmir.2507

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Caregiver Perceptions on Telehealth-Based Audiological Services Offered Within a Mobile-Health Clinic for Infants

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Abstract

Purpose: Telehealth audiological services offered within a mobile clinic can expand the range of hearing healthcare services to rural and remote areas where many patients, in particular infants with hearing loss, go undetected due to limited access to specialist care. This study describes caregiver perceptions on the use of synchronous telehealth-based auditory brainstem response (ABR) services.

Method: Forty caregivers rated their perceptions of the mobile telehealth-based service through a self-administered questionnaire comprised of 15 questions.

Results: Caregivers were satisfied (76.8%) with mobile telehealth-based services on aspects of access, satisfaction with the mobile health clinic, privacy, comfort, technical and non-technical experiences, distance, quality of care, travel costs and time; and noted they would use telehealth in the future. Interestingly, 17.5% of the participants agreed that using computer technology to receive health services is not culturally appropriate and 15% agreed that it felt *unnatural* to them. There was a strong association ($p = 0.04$) between the participants that earned below the minimum wage and the choice to use telehealth in the future.

Conclusion: The findings of the study are a positive indicator for the use of synchronous telehealth-based ABR services through a mobile clinic as a service delivery model for infants. This service is particularly beneficial to those residing within rural and remote areas with limited access to specialized services. However, consideration of how telehealth services may influence cultural practices and beliefs is important.

Keywords: mobile-health clinic, telehealth-based ABR, caregiver perceptions, audiological care, infant ABR

Acronyms: ABR = auditory brainstem response; PHC = primary healthcare

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Telehealth-based audiological services are defined as the use of technology-based virtual platforms to offer a variety of assessments stretching across hearing screening, follow-up auditory brainstem response (ABR) examinations, behavioral audiometry, cochlear implants programming and intervention (Hatton et al., 2019; Molini-Avejonas et al., 2015). In this article, mobile health services is defined as delivery of health services through a mobile clinic. The use of telehealth-based audiological services offered within a mobile clinic could potentially improve early detection and intervention services for infants with hearing loss. Mobile audiological services ensure that infants receive hearing healthcare services within close proximity to their homes. Caregivers are often faced with cost challenges and long travel times, as well as limited access to audiologists, which can deter caregivers from taking their infants for audiology care visits at hospitals. Furthermore, the COVID-19 pandemic created delays and disruption in the auditory healthcare setting with many caregivers being discharged with their infants before infants received hearing screening (Ben-David et al., 2021; D'Onofrio & Zeng, 2022; Jenks et al., 2022;

Panahi et al., 2023). In the South African context, due to the socio-economic challenges, caregivers are unlikely to come back for a routine hearing test. It is also likely that the COVID-19 outbreak had psychological impacts on the population (Naidu, 2020) and caregivers may fear taking their infants back to the hospital for hearing assessments due to the risk of exposing their infant's vulnerable immune system to sick patients at the hospital. Telehealth-based audiological services may help alleviate some of these concerns especially if such services are brought into close proximity of the patient.

There has been a rapid growth in technological advances and telehealth technology, coupled with an increase in the use of telehealth-based services, particularly since the COVID-19 pandemic (Bhamjee et al., 2022; D'Onofrio & Zeng, 2022; Hoi et al., 2021; Manchaiah et al., 2022; Saunders & Roughley, 2021; Talbott et al., 2022). Bhamjee and colleagues (2022) found that telehealth-based services for hearing loss in South Africa's public healthcare system increased during the COVID-19 pandemic. Prior to the pandemic, only 7.2% reported using hearing healthcare via telehealth, but nearly 19.6% used it during the COVID-19 pandemic. It is therefore plausible to assume that the use of telehealth technology provides an opportunity for increasing the provision of audiology services (Fernandes et al., 2020; Kim et al., 2021). However, it is important that end-users of a service are consulted in terms of their perceptions and experience with the services to ensure compliance and sustainability of health programs. Caregivers are key stakeholders in the provision of newborn audiological services. Ongoing consultation of their experiences, opinions, suggestions, and perceptions must occur so that the derived information can be assimilated into the development and refinement of health service delivery programs.

The overall feedback from caregivers regarding telehealth-based services reflected in the literature has been positive (Hatton et al., 2019; Talbott et al., 2022). The main benefits pointed out by caregivers are related to the overall increased accessibility to health services offered by synchronous telehealth services and the associated reduction in travel time (Hatton et al., 2019; Talbott et al., 2022). Additionally, they felt that their privacy was respected, and they felt comfortable in the area where the sessions were being conducted (Hatton et al., 2019). Furthermore, they indicated that they would use telehealth-based services in the future (Hatton et al., 2019). A study conducted by Dharmar and colleagues (2016) on reducing loss to follow-up with tele-audiology diagnostic evaluations reported that all caregivers rated the importance of tele-based hearing healthcare services *extremely important*. It is therefore plausible that continued development and validation of the telehealth-based programs will help improve audiological services and expand the reach to patients located in remote areas (Talbott et al., 2022). A scoping review of telehealth services to facilitate audiological management for children concluded that further telehealth research must be focused on technology assessment, protocol design, cost-effectiveness, and stakeholder perception to bridge the gap between knowledge and action (Govender & Mars, 2017).

Although there are known advantages to offering auditory care services via telehealth, there are currently few studies describing caregiver perceptions of the use of mobile telehealth-based ABR services, especially for caregivers accessing care at primary healthcare (PHC) clinics. Caregiver perceptions are useful when designing and developing ABR service delivery programs. For instance, ABR services can be used for both screening and diagnostic objective hearing testing in the pediatric population who are unable to be tested by conventional hearing testing. Thus, understanding caregiver perceptions is valuable to the sustainability of the program. Caregivers as key stakeholders can provide valuable insight into the service delivery model.

The current study aimed to describe caregiver perceptions of the use of mobile telehealth-based ABR services offered to their infants when accessing care at primary healthcare clinics (PHC) within the Winterveldt district/region of Pretoria North, South Africa.

Method

Study Design

A questionnaire on perceptions of telehealth was used to measure the effectiveness of the ABR telehealth service that was offered within a mobile clinic. Questions were adapted from Weaver and colleagues (2020) and worded to suit the current research study that is based on caregiver perceptions of telehealth-based audiology services. A self-administered questionnaire comprising 15 questions in the form of a Likert scale (*strongly disagree* to *strongly agree*) was used. The tool was translated into Setswana by a professional Setswana translator to ensure its validity. The questionnaire was back-translated to ensure that it was accurate.

Setting

The research study was conducted in three PHC facilities situated in the rural areas of Winterveldt, situated in the northwest of Pretoria, and falling under the City of Tshwane Metropolitan Municipality of the Gauteng Province, South Africa. Selection of the PHC facilities was based on permission obtained from the various sites. These sites were also selected as they do not provide audiology services.

Study Population and Sampling Strategy

The sample included caregivers visiting Winterveldt PHC clinics during the data collection period who volunteered to have their infants' hearing evaluated through an ABR assessment. The three participating PHC clinics in this study have a total number of 51,100 infants receiving post-natal care services on an annual basis. Of these, an average of 358 infants require follow-up audiologic

testing. Approximately 10% of caregivers seeking service (40 caregivers taking care of 40 infants) were asked to complete the questionnaire regarding their experience with audiological care in a mobile-based telehealth setting. Participants were included in the study if they were seeking postnatal care for their infants who ranged from 3 days to 6 months old. Caregivers were invited to participate in the study, regardless of whether their infants had risk factors for hearing loss, such as being born prematurely or being born with low or very low birth weight. All participants were residents of Winterveldt. The sample consisted of caregivers of infants (n = 40) who were residing in rural areas and who were accessing health services at the PHC clinics. Purposive and convenient sampling strategies were employed to recruit caregivers for the study. The caregivers could converse in either English or Setswana. The researchers explained the aim of the study to the caregivers and invited them to participate. Only caregivers who met the inclusion criteria and who provided consent were included in the study.

Data Collection

Data Collection Tools

Caregivers were asked to complete a questionnaire adapted from a survey by Weaver and colleagues on perceptions of telehealth (Weaver et al. 2020). The questionnaire collected information about the effectiveness of telehealth-based hearing healthcare services. The questionnaire was designed in the form of a Likert scale (*strongly disagree* to *strongly agree*) and was available in English and was translated to Setswana. The questionnaire had fifteen questions about the perceptions of the caregivers regarding tele-diagnostic ABR testing. The first section of the questionnaire elicited demographic information (educational levels, minimum wage range, and linguistic profile). The second section explored participants' experience with the ABR telehealth service that was offered in the mobile clinic (quality of the care received, videoconferencing experience, comfort, privacy, and cultural considerations). A copy of the questionnaire is included as Appendix A.

Data Collection Procedure

Caregivers observed their infants undergo a mobile telehealth-based ABR assessment using a synchronous telehealth model with the assistance of a community healthcare worker. Caregivers were then asked to complete a questionnaire to evaluate their perceptions about telehealth-based services.

Each of the 40 infants underwent a face-to-face and synchronous telehealth ABR assessment. All infants were aged six months or less with 90% of the infants aged between 1 and 2 months old. The face-to-face ABR assessment was conducted in a sound controlled environment within the nearest hospital and the telehealth assessment was conducted in a mobile clinic that was stationed just outside PHC facilities. The researchers conducted testing in a counter-balanced manner in that the researcher started with in-person testing for the first patient, followed by tele-diagnostic testing. The reverse of this process occurred for the next patient. This pattern continued for the entire sample. Mobile tele-diagnostic ABR testing was conducted by the audiologist (researcher) and required the assistance of a community healthcare worker in the mobile clinic to prepare the infants for testing. Two laptops were used: the laptop in the mobile clinic (used by a community healthcare worker and a caregiver) formed part of the PATH Medical Sentiero Advanced (ABR or ASSR) system and used videoconferencing (TeamViewer installed) for the audiologist to test the infants remotely, monitoring and providing guidance to the community healthcare worker. This laptop was charged with a portable power supply. The second laptop was with the audiologist who was situated in the nearest hospital to test the infants as it mirrored (duplicated) the laptop in the mobile clinic van. Caregivers were seated next to the bed where their infant underwent the ABR assessment. After both assessments (face-to-face and telehealth-based ABR), caregivers were given a questionnaire to complete.

Data Analysis

Descriptive and inferential statistical analysis was used to summarize and analyze results of caregivers' responses obtained through self-administered questionnaires. Caregivers' responses were collected and captured in a Microsoft Excel spreadsheet. Participant numbers were used to ensure anonymity. Microsoft Excel was also used to organize and format the data. Both researchers checked the accuracy of the recorded data to ensure reliability of the data capturing process. Data were analyzed using the SPSS software v28.0.18. This software was used for both descriptive and inferential statistics. The results of the study have been presented as frequency, and percentages for categorical variables, and mean ±*SD* for continuous variables. To compare groups, Fisher's exact test was used for categorical variables, while an independent student *t*-test was performed for continuous variables. A p value of less than 0.05 was considered statistically significant. A statistician assisted with data interpretation and verification, result analysis, and confirmation of the reliability of the analysis.

Results

Demographics

A total of 40 participants completed the questionnaire. All participants were black females residing in the Winterveldt region of Pretoria who volunteered to bring their infants for a face-to-face and synchronous telehealth ABR assessment in a mobile clinic. The majority of participants (67.5%, n = 27) had matriculated while 7.5% (n = 3) attended College and 7.5% (n = 3) attended University. One of the 40 participants left school after grade 11. Six (15%) participants' did not report education level. All of the participants reported they could read and write, and regarded their literacy as good. The majority of participants

(80%) earned less than the minimum wage (under ZAR25.42 per hour) while 20% (*n* = 8) earned above the minimum wage. The majority of the participants (92.5%, *n* = 37) were Setswana speaking and 7.5% (*n* = 3) were Shona speaking. Of the Setswana speaking participants, only 8.1% (*n* = 3) requested to complete the questionnaire translated to Setswana with the rest preferring the English questionnaire. The average age of the participants was 26.25 years, ranging between 17 and 44 years. The demographic information is summarized in Table 1.

Table 1
Demographical Information of Participants

Variable	Description	Frequency	Percentage
Education level	College	3	7.5
	Grade 11	1	2.5
	Matric	27	67.5
	University	3	7.5
	Missing	6	15
Minimum wage	Less	32	80
	More	8	20
Linguistic profile	Setswana	37	92.5
	Shona	3	7.5

Questionnaire Results

Participants were requested to rate their perceptions of the telehealth ABR services that they received from a mobile clinic using a five-point Likert scale questionnaire. The options included two extremes (*strongly disagree* and *strongly agree*), two intermediate (*disagree* and *agree*) and one *neutral* opinion. Participants rated their perceptions in terms of access to the mobile services, satisfaction with the tele-diagnostic ABR services, privacy, comfort, and comparison of services to in-hospital care, as well as various other aspects detailed in the Method section.

Participants were asked whether the telehealth service they received improved their access to hearing healthcare services. Thirty-nine participants (98%) either *strongly agreed* or *agreed* and one participant was *neutral*. Thirty-eight (95%) participants *agreed* that they were satisfied by the telehealth service being delivered in a mobile health clinic and 5% (*n* = 2) *disagreed* as they did not find the service satisfactory.

All participants *agreed* that their privacy and the privacy of their infant was respected. A majority of participants (87.5%, *n* = 5) *agreed* or *strongly agreed* that receiving services in the mobile clinic was comfortable, however 7.5% (*n* = 3) experienced some discomfort whilst receiving services within the mobile clinic which largely related to space. Participants were asked if they thought that using computer technology to receive health services was culturally inappropriate. Interestingly, 15% (*n* = 6) *strongly agreed* with this statement whilst 52.5% (*n* = 21) *strongly disagreed*.

All participants (100%, *n* = 40) agreed that they would access telehealth services again. Participants were asked whether it was easier for them to attend the mobile health service than to attend a face-to-face service at the local hospital and 85% (*n* = 34) either *agreed* or *strongly agreed* whilst 10% (*n* = 4) *disagreed*. Regarding the distance, 80% (*n* = 32) of participants *agreed* that the mobile service reduced their overall travel distance whilst 20% (*n* = 8) did not feel that the mobile health service impacted their travel time to the local hospital.

Regarding the technical experience, a total of 32 (80%) participants *strongly agreed* that they had observed the audiologist communicate clearly through videoconferencing during testing, 12.5% (*n* = 5) *agreed* with the statement, 5% (*n* = 2) either *disagreed* or *strongly disagreed* with the statement. Caregivers were given feedback through videoconferencing, and 90% (*n* = 36) indicated that they could hear the audiologist clearly during the feedback session after testing their infants, and 5% (*n* = 2) did not agree with the statement.

Regarding the quality of care, 77.5% (*n* = 31) *strongly agreed* or *agreed* that the quality of care over the telehealth model is the same as in-person visits, however, 20% (*n* = 8) *disagreed* with this statement. A total of 39 participants (97.5%) indicated that telehealth is an acceptable way to receive hearing healthcare services, and 2.5% (*n* = 1) *strongly disagreed* with the statement. The majority of the participants (97.5%, *n* = 39) indicated that they would use telehealth again for their infants' hearing healthcare services in the future, and 2.5% (*n* = 1) neither *agreed* nor *disagreed*. Lastly, 97.5% (*n* = 39) of the caregivers indicated that telehealth saved them and their family time and/or money, and 2.5% disagreed (*n* = 1).

Table 2 provides detailed findings of participants' responses from the questionnaire. As shown in Table 3, there was a statistically significant difference (*p*-value < 0.0001) between participants that agreed (76.8%) and those that disagreed (18.4%) on aspects of access, satisfaction with the tele-diagnostic service, privacy, comfort, using telehealth again, experience, distance, videoconferencing experience, and feedback from the audiologist.

Table 4 shows a strong association (*p* = 0.04) between the participants earning below minimum wage and the choice to use telehealth in the future suggesting that participants earning below minimum wage would access telehealth services in the future due to saving costs and less traveling time.

Discussion

This study investigated caregiver perceptions on the use of mobile telehealth-based ABR services and found that a majority of caregivers are positive about this model of hearing healthcare service delivery. The findings of the study indicated that the majority of participants strongly agreed that the telehealth mobile service was easy to access, was comfortable, and that their privacy was respected. Participants indicated that they would use the

Table 2

Participants' Responses on the Questionnaire

	Strongly disagree	%	Disagree	%	Neutral	%	Agree	%	Strongly agree	%	Total	%
Q 1: Access	0	0.0%	0	0.0%	1	2.5%	12	30.0%	27	67.5%	40	100%
Q2: Satisfaction	0	0.0%	2	5.0%	0	0.0%	10	25.0%	28	70.0%	40	100%
Q 3: Privacy	0	0.0%	0	0.0%	0	0.0%	7	17.5%	33	82.5%	40	100%
Q 4: Comfort	0	0.0%	3	7.5%	2	5.0%	7	17.5%	28	70.0%	40	100%
Q 5: Computer tech for culture	21	52.5%	11	27.5%	1	2.5%	1	2.5%	6	15.0%	40	100%
Q 6: Natural or not	19	47.5%	14	35.0%	1	2.5%	1	2.5%	5	12.5%	40	100%
Q 7: Use telehealth again	0	0.0%	0	0.0%	0	0.0%	7	17.5%	33	82.5%	40	100%
Q 8: Experience	1	2.5%	3	7.5%	2	5.0%	9	22.5%	25	62.5%	40	100%
Q 9: Distance	1	2.5%	2	5.0%	5	12.5%	9	22.5%	23	57.5%	40	100%
Q 10: Videoconfencing experience	1	2.5%	1	2.5%	1	2.5%	5	12.5%	32	80.0%	40	100%
Q 11: Feedback	1	2.5%	1	2.5%	2	5.0%	8	20.0%	28	70.0%	40	100%
Q 12: Quality of care	0	0.0%	8	20.0%	1	2.5%	9	22.5%	22	55.0%	40	100%
Q 13: Telehealth	1	2.5%	0	0.0%	0	0.0%	12	30.0%	27	67.5%	40	100%
Q 14: Use of telehealth in future	0	0.0%	0	0.0%	1	2.5%	8	20.0%	31	77.5%	40	100%
Q 15: Travel costs and time	0	0.0%	1	2.5%	0	0.0%	0	0.0%	39	97.5%	40	100%
Average	3	7.5%	3	7.7%	1	2.8%	7	17.5%	26	64.5%	40	100%

Table 3

Analysis of Variance Between Caregivers that Agreed and Disagreed

Summary

Groups	*Count*	*Sum*	*Average*	*Variance*
Disagree	16	2.95	0.184375	0.086823
Neutral	16	0.75	0.046875	0.00649
Agree	16	12.3	0.76875	0.113125

ANOVA

Source of Variation	***SS***	***df***	***MS***	***F***	***P-value***	***F crit***
Between Groups	4.701354	2	2.350677	34.16061	<0.0001	3.204317
Within Groups	3.096563	45	0.068813			
Total	7.797917	47				

Table 4

Minimum Wage

Variable	***Description***	***Less***	***%***	***More***	***%***	***Total***	***%***	***Chi square***	***df***	***p-value***
Q 15: Will use telehealth in future	Neutral	0	0.0%	1	12.5%	1	2.5%	6.129a	2	0.0467
	Agree	8	25.0%	0	0.0%	8	20.0%			
	Strongly agree	24	75.0%	7	87.5%	31	77.5%			
	Total	32	100.0%	8	100.0%	40	100.0%			

telehealth mobile service again because they had a good experience, and did not have to travel a long distance for these services. They reported that the videoconferencing experience was good, they could hear the audiologist well during testing, and the feedback session and overall quality of care via telehealth was comparable to the in-person visit. Participants' answers indicated that mobile telehealth services is an acceptable and easy way to receive hearing healthcare services.

Our study indicated that the telehealth model has the potential to expand access to audiological care. Studies conducted by both Hatton et al. (2019) and Dharmar et al. (2016) found many of the same benefits of telehealth for participants including: spacing in the mobile clinic was comfortable, their privacy was respected, and they were satisfied with the technical experience for videoconferencing throughout the session. Liu and colleagues (2021) found that telehealth visits had a higher frequency of visits than other modes of consultation and care due to easy accessibility. Our study builds on these findings by showing the potential to expand access to audiological care through the telehealth model.

However, in this study, a minority of participants did not agree that the mobile telehealth-based service was culturally appropriate, and noted that it did not feel *natural* compared to the traditional in-hospital care approach. None of the existing studies included cultural appropriation matters in their survey questionnaires which adds to the relevance of the present study findings. The findings of the study are similar to that of Ncube and colleagues (2023) who found cultural and traditional beliefs were identified as inhibitors to telehealth services. Therefore it is important that cultural factors and beliefs be considered in the development of telehealth programs as culturally-appropriate services will ensure sustainable uptake (Caffery et al., 2018).

Caregivers indicated that the quality of care during telehealth-based care services is the same as in-person visits. These findings are similar to that of Bilimoria et al. (2021), Slightam et al. (2020), and Street et al. (2022) where patient experiences using telehealth resulted in similar rates of patient experiences between face-to-face and telehealth-based services. Participants in the present study indicated that they would access mobile telehealth services in the future and the findings were in agreement with those of Atreya et al. (2020) and Hatton et al. (2019).

The study findings revealed a statistically significant difference between those working below and those working at or above minimum wage and the choice to access telehealth services in the future. There are socio-economic inequalities in South Africa where a significant percentage of the population earn below minimum wage (Anwar and Brukwe, 2023). This suggests that those working below minimum wage may not have the necessary finances to access healthcare. It is well known that the majority of the population earning below the minimum wage reside in rural and remote geographical areas (Anwar & Brukwe, 2023). Because of this, they may face barriers to accessing specialized healthcare. This includes long travel distances to hospitals and high transportation costs (Harris et al., 2011; Rural Health Information Hub, 2023). Mobile telehealth-based ABR services through the use of a synchronous modality could bridge the service gap, allowing infants to receive services remotely at an early age, and, thereby, mitigating the negative impact of unidentified or late identified hearing loss.

The study findings show that a mobile telehealth ABR service for infants can be offered within rural community contexts as it appears that caregivers are accepting of this service. This means that it has the potential to reduce loss to follow-up as patients do not always visit hospitals due to various circumstances. This also contributes positively to the initiative of early detection and intervention of hearing loss among infants, thus, reducing the high prevalence and effects of infant and childhood hearing loss which according to the literature have been suggested to have a negative impact on development and on quality of life (Butcher et al., 2019; Mostafa et al., 2022; Neumann et al., 2022). A telehealth screening and/or diagnostic ABR service offered within a mobile clinic could improve access to audiological services and reduce the impact of hearing loss on the pediatric population. A mobile health service with screening programs that offer immediate access to diagnostic services could reduce loss to follow-up.

Strengths and Limitations

This research project was carried out in a mobile clinic located at the clinic, parked in an area that is accessible for patients who were visiting the facilities for postnatal care services. This implies that early detection and intervention programs using synchronous, telehealth services delivered from a mobile clinic can be used to make services more accessible to communities.

Although this study used several PHC facilities, the sample consisted of 40 participants. The small number of participants limits the extent to which the description of caregiver perceptions can be considered fully representative and the extent to which it can be generalized to a larger region. Additionally, the sites were relatively homogenous (all within the Winterveldt), the data collection period was only 8 weeks (which is relatively short), and the infants ranged in age from 3 days to 6 months. These variables affect generalization of results in terms of demographics.

Recommendations

Future studies should repeat the study with a larger sample size to increase the applicability of the results to a larger population. It would also be valuable to replicate the study across different populations to explore possible effects of age, populations, and geographical locations. Furthermore, future studies should evaluate cultural dynamics using the mobile telehealth-based model to deliver hearing healthcare services in South African rural

communities. Lastly, a cost-benefit analysis for infant telehealth-based ABR through a mobile clinic in a South African public health setting would be beneficial.

Conclusion

The majority of caregivers responded positively and were satisfied with the mode of hearing healthcare service delivery. Although offering telehealth-based services comes with some challenges such as the need to ensure culturally-appropriate services, the model of care provides an opportunity to improve audiological services to rural and remote communities. This research project was carried out in a mobile clinic located close to PHC facilities, parked in an area that is accessible for caregivers and their infants who were visiting the facilities for postnatal care services. The findings suggest there may be multiple benefits for *at home* (direct-to-patient) telehealth services that could save costs and improve access to audiological services.

References

Anwar, M. A., & Brukwe, K. (2023). 'We endure because we need money': Everyday violence, COVID-19 and domestic workers in South Africa. *Canadian Journal of Development Studies/Revue canadienne d'études du développement*, 1–19. https://doi.org/10.1080/02255189.2023.2166024

Atreya, S., Kumar, G., Samal, J., Bhattacharya, M., Banerjee, S., Mallick, P., Chakraborty, D., Gupta, S. & Sarkar, S. (2020). Patients'/Caregivers' perspectives on telemedicine service for advanced cancer patients during the COVID-19 pandemic: An exploratory survey. *Indian Journal of Palliative Care*, *26*(Supp. 1), S40. https://doi.org/10.4103%2FIJPC.IJPC_145_20

Ben-David, B. M., Mentzel, M., Icht, M., Gilad, M., Dor, Y. I., Ben-David, S., Carl, M., & Shakuf, V. (2021). Challenges and opportunities for telehealth assessment during COVID-19: iT-RES, adapting a remote version of the test for rating emotions in speech. *International Journal of Audiology*, *60*(5), 319–321. https://doi.org/10.1080/14992027.2020.1833255

Bhamjee, A., Le Roux, T., Swanepoel, D. W., Graham, M. A., Schlemmer, K., & Mahomed-Asmail, F. (2022). Perceptions of telehealth services for hearing loss in South Africa's public healthcare system. *International Journal of Environmental Research and Public Health*, *19*(13), 1–14. https://doi.org/10.3390/ijerph19137780

Bilimoria, K. Y., Zhan, T., Durst, D. A., Merkow, R. P., Sama, P. R., Bahaveolos, S. A., & Chrisman, H. B. (2021). Comparison of patient experience with telehealth vs. in-person visits before and during the COVID-19 pandemic. *Joint Commission Journal on Quality and Patient Safety*, *47*(8), 533–536. https://doi.org/10.1016%2Fj.jcjq.2021.01.009

Butcher, E., Dezateux, C., Cortina-Borja, M., & Knowles, R. L. (2019). Prevalence of permanent childhood hearing loss detected at the universal newborn hearing screen: Systematic review and meta-analysis. *PLoS One*, *14*(7), 1–21. https://doi.org/10.1371/journal.pone.0219600

Caffery, L. J., Bradford, N. K., Smith, A. C., & Langbecker, D. (2018). How telehealth facilitates the provision of culturally appropriate healthcare for Indigenous Australians. *Journal of Telemedicine and Telecare*, *24*(10), 676–682. https://doi.org/10.1177/1357633X18795764

Dharmar, M., Simon, A., Sadorra, C., Friedland, G., Sherwood, J., Morrow, H., Deine, D., Nickell, D., Lucatorta, D., & Marcin, J. P. (2016). Reducing loss of follow-up with tele-audiology diagnostic evaluations. *Telemedicine and e-Health, 22*(2), 159–164. https://doi.org/10.1089/tmj.2015.0001

D'Onofrio, K. L., & Zeng, F. G. (2022). Tele-audiology: Current state and future directions. *Frontiers in Digital Health*, *3*(207), 1–11. https://doi.org/10.3389/fdgth.2021.788103

Fernandes, F. D. M., Lopes-Herrera, S. A., Perissinoto, J., Molini-Avejonas, D. R., Amato, C. A. de la H., Tamanaha, A. C., de Souza, A. P. R., Montenegro, A. C., de A., Machado, F. P., Segeren, L., & de Goulart, B. N. G. (2020). Use of telehealth by undergraduate students in speech therapy: Possibilities and perspectives during COVID-19 pandemic. *Codas, 32*(4), 1–3. https://doi.org/10.1590/2317-1782/20192020190

Govender, S. M., & Mars, M. (2017). The use of telehealth services to facilitate audiological management for children: A scoping review and content analysis. *Journal of Telemedicine and Telecare*, *23*(3), 392–401. https://doi.org/10.1177/1357633X16645728

Harris, B., Goudge, J., Ataguba, J. E., McIntyre, D., Nxumalo, N., Jikwana, S., & Chersich, M. (2011). Inequities in access to health care in South Africa. *Journal of Public Health Policy*, *32*, S102–S123. https://doi.org/10.1057/jphp.2011.35

Hatton, J. L., Rowlandson, J., Beers, A., & Small, S. (2019). Telehealth-enabled auditory brainstem response testing for infants living in rural communities: The British Columbia Early Hearing Program experience. *International Journal of Audiology, 58*(7), 381–392. https://doi.org/10.1080/14992027.2019.1584681

Hoi, K. K., Curtis, S. H., Driver, L., Wisnosky, E., Zopf, D. A., & Bohm, L. A. (2021). Adoption of telemedicine for multidisciplinary care in pediatric otolaryngology. *Annals of Otology, Rhinology & Laryngology*, *130*(10), 1105–1111. https://doi.org/10.1177/0003489421997651

IBM SPSS Statistics for Windows, Version 28.0. Armonk, NY: IBM Corp. https://www.ibm.com/support/pages/downloading-ibm-spss-statistics-28010

Jenks, C. M., DeSell, M., & Walsh, J. (2022). Delays in infant hearing detection and intervention during the COVID-19 pandemic: Commentary. *Otolaryngology–Head and Neck Surgery*, *166*(4), 603–604. https://doi.org/10.1177/01945998211067728

Kim, J., Jeon, S., Kim, D., & Shin, Y. (2021). A review of contemporary teleaudiology: Literature review, technology, and considerations for practicing. *Journal of audiology & otology*, *25*(1), 1–7. https://doi.org/10.7874%2Fjao.2020.00500

Liu, X., Goldenthal, S., Li, M., Nassiri, S., Steppe, E., & Ellimoottil, C. (2021). Comparison of telemedicine versus in-person visits on impact of downstream utilization of care. *Telemedicine and e-Health*, *27*(10), 1099–1104. https://doi.org/10.1089/tmj.2020.0286

Manchaiah, V., Eikelboom, R. H., Bennett, R. J., & Swanepoel, D. W. (2022). International survey of audiologists during the COVID-19 pandemic: Effects on the workplace. *International Journal of Audiology*, *61*(4), 265–272. https://doi.org/10.1080/14992027.2021.1937348

Molini-Avejonas, R. D., Rondon-Melo, S., de La Higuera A. C. A., & Samelli, A. G. (2015). A systematic review of the use of telehealth in speech, language and hearing sciences. *Journal of Telemedicine and Telecare*, *21*(7), 367–376. https://doi.org/10.1177/1357633X15583215

Mostafa, B. E., Mostafa, A., Fiky, L. M. E., Omara, A., & Teaima, A. (2022). Maternal COVID-19 and neonatal hearing loss: A multicentric survey. *European Archives of Oto-Rhino-Laryngology*, *279,* 3435–3438. https://doi.org/10.1007/s00405-021-07098-5

Naidu, T. (2020). The COVID-19 pandemic in South Africa. *Psychological Trauma: Theory, Research, Practice, and Policy*, *12*(5), 559–561. https://psycnet.apa.org/doi/10.1037/tra0000812

Ncube, B., Mars, M., & Scott, R. E. (2023). Perceptions and attitudes of patients and healthcare workers towards the use of telemedicine in Botswana: An exploratory study. *PloS one*, *18*(2), e0281754. https://doi.org/10.1371/journal.pone.0281754

Neumann, K., Mathmann, P., Chadha, S., Euler, H. A., & White, K. R. (2022). Newborn hearing screening benefits children, but global disparities persist. *Journal of Clinical Medicine*, *11*(1), 1–12. https://doi.org/10.3390/jcm11010271

Panahi, R., Nemati, S., Akbarpour, M., Nasirmohtaram, S., Maroufizadeh, S., & Hosseinian, A. (2023). Neonatal hearing screening and prevalence of hearing impairment in the time period of COVID-19 epidemic in the north of Iran. *Auditory and Vestibular Research*, *32*(1), 32–38. https://doi.org/10.18502/avr.v32i1.11318

Rural Health Information Hub. (2023). Healthcare access in rural communities introduction. https://www.ruralhealthinfo.org/topics/healthcare-access#health-insurance

Saunders, G. H., & Roughley, A. (2021). Audiology in the time of COVID-19: Practices and opinions of audiologists in the UK. *International Journal of Audiology*, *60*(4), 255–262. https://doi.org/10.1080/14992027.2020.1814432

Slightam, C., Gregory, A. J., Hu, J., Jacobs, J., Gurmessa, T., Kimerling, R., Blonigen, D., & Zulman, D. M. (2020). Patient perceptions of video visits using veterans affairs telehealth tablets: Survey study. *Journal of Medical Internet Research*, *22*(4), e15682. https://doi.org/10.2196/15682

Street, R. L., Jr., Treiman, K., Kranzler, E. C., Moultrie, R., Arena, L., Mack, N., & Garcia, R. (2022). Oncology patients' communication experiences during COVID-19: Comparing telehealth consultations to in-person visits. *Supportive Care in Cancer*, *30*(6), 4769–4780. https://doi.org/10.1007/s00520-022-06897-8

Talbott, M. R., Lang, E., Avila, F., Dufek, S., & Young, G. (2022). Short report: Experiences of caregivers participating in a telehealth evaluation of development for infants (TEDI). *Journal of Autism and Developmental Disorders*, *52*, 5266–5273. https://doi.org/10.1007/s10803-022-05607-x

Weaver, M. S., Robinson, J. E., Shostrom, V. K., & Hinds, P. S. (2020). Telehealth acceptability for children, family and adult hospice nurses when integrating the pediatric palliative provider during sequential rural home hospice visits. *Journal of Palliative Medicine, 23*(5), 641–649. https://doi.org/10.1089/jpm.2019.0450

Appendix

Questionnaire Used in Study

Participant No. _____

Dear Caregiver:

In order for us to gather good information about our study, we require your assistance in identifying your experiences with the use of mobile auditory brainstem response (ABR) service using telehealth that was conducted on your child.

Please answer the following questions and please be assured that your information will remain confidential.

Please tick (√) in the most appropriate box

	Strongly Disagree 1	Disagree 2	Neutral 3	Agree 4	Strongly Agree 5
1. Telehealth improves my access to hearing health services.					
2. I am satisfied with the telehealth visit.					
3. I felt my privacy was respected.					
4. I felt comfortable in the mobile clinic van.					
5. I do not think that computer technology to receive health services is culturally appropriate.					
6. I think that receiving health services through telehealth feels unnatural to me.					
7. I would use telehealth again.					
8. It was easier for me to attend using telehealth rather than in person.					
9. If telehealth was not available, I would have travelled a long distance for my visit.					
10. I have observed the Audiologist communicate clearly through videoconferencing during testing.					
11. I could hear the Audiologist clearly during the feedback session after testing my child.					
12. I think quality of care over telehealth system is same as in-person visits.					
13. Telehealth is an acceptable way to receive hearing health services.					
14. I would use telehealth again for my child's hearing health services again in the future.					
15. Telehealth saved me and my family time/or money.					