JEHDI
The Journal of Early Hearing Detection and Intervention
2016; 1(2): 63-71

Infant Diagnostic Evaluation via Teleaudiology Following Newborn Screening in Eastern North Carolina

Andrew Stuart

Department of Communication Sciences and Disorders
East Carolina University

Abstract

Universal newborn hearing screening in North Carolina began in 2000 under the auspices of the North Carolina Department of Health and Human Services Early Hearing Detection and Intervention Program (NC-EHDI). Despite initial success, lost to follow-up/lost to documentation for diagnostic testing was problematic. To address this, the NC-EHDI received U.S. Department of Health and Human Services Health Resources and Services Administration lost to follow-up funding to fund, in part, a pilot "Teleaudiology Project" in 2010 to provide services for infants in eastern North Carolina. This part of the state is a traditionally underserved area. The project involved a partnership with East Carolina University. The project's goals were to provide infant diagnostic evaluations in rural eastern counties and to establish a coordinated system for the delivery of audiological evaluations for infants whose families experience economic and geographic barriers to service. Project planning preparation and preliminaries, project service model, and outcome data are presented. From 2011 to 2015, outcome data provide positive proof-of-concept for a teleaudiology model in meeting national recommendations for providing diagnostic testing of infants following screening referral in a timely manner. In addition, the endeavor provides graduate audiology students with a unique didactic and clinical experience in teleaudiology.

Acronyms: ABR = auditory brainstem response; BRI = basic rate interface; CSDI = Department of Communication Science and Disorders; DPOAE = distortion product otoacoustic emissions; ECU = East Carolina University; EHDI = Early Hearing Detection and Intervention; HIPPA = Health Insurance and Portability Act; HL = hearing loss; ISDN = Integrated Services Digital Network; LFU = lost to follow-up; LTD = lost to documentation; NC = North Carolina; TM = telemedicine

Acknowledgements: This work was presented in part at the 2012 American Speech-Language-Hearing Association Annual Convention, Chicago, IL; 2015 American Speech-Language-Hearing Association Annual Convention, Denver, CO; and the Early Hearing Detection and Intervention 2012 Annual Meeting, St. Louis, MO. The assistance of Kathleen Watts, Program Manager (retired), North Carolina, Early Hearing Detection and Intervention, and Gloria Jones, Clinical Telehealth Manager (retired), East Carolina University Telemedicine Center, was invaluable toward this project.

Corresponding author's contact information: Andrew Stuart, Ph.D., Department of Communication Sciences and Disorders, Mail Stop 668, East Carolina University, Greenville, NC 27858-4353, USA. Email: stuarta@ecu.edu

Introduction

The North Carolina General Assembly passed the General Statutes Chapter 130A-125 (Screening of newborns for metabolic and other hereditary and congenital disorders) in the fall of 1999. Implemented on August 1, 2000, it mandated a newborn screening program and universal newborn hearing screening in the state of North Carolina. Specifically, it authorized each newborn to undergo physiological screening in each ear for the presence of permanent hearing loss. Presently, the North Carolina's Early Hearing Detection and Intervention Program (NC-EHDI) provides screen-rescreen-diagnosis-intervention. NC-EHDI is organizationally located in the North Carolina Department of Health and Human Services, Division of Public Health, Women's and Children's Health Section, Children and Youth Branch as part of the state Title V Maternal and Child Health Services Program.

Initial newborn hearing screening rates across North Carolina have been excellent. For example, 98.2% of infants born in 2006 were screened for the presence of permanent hearing loss (Williams, Alam, & Gaffney, 2015). In 2012, the percentage of newborns receiving hearing screening remained high (i.e., 99.1%). From those that received diagnostic testing, prevalence of permanent hearing loss per 1000 screened was estimated as 1.8 and 1.6 in 2006 and 2012, respectively.

Despite initial success with universal hearing screening of newborns, lost to follow-up (LFU)/lost to documentation (LTD) for diagnostic testing following the screening phase was problematic. For example, 53.7% (808 of 1,505) of infants, who did not pass the newborn hearing screening and were referred in 2006, were LFU/LTD and did not undergo audiological diagnostic testing (Williams et al., 2015). Although improved in 2012, a similar pattern of performance was evidenced in 2012: More than one-third of 854 newborn infants referred following newborn hearing screening (37.8%, $n = 323$) were LFU/LTD and did not complete a diagnostic evaluation.

To address the LFU/LTD for diagnostic testing, the NC-EHDI sought and received U.S. Department of Health and Human Services Health Resources and Services Administration lost to follow-up funding in September 2009. A portion of the funds was used to develop a pilot Teleaudiology Project in 2010 to provide services for infants in 38 counties in the eastern part of North Carolina. The targeted eastern North Carolina catchment area[1] is unique

[1]The catchment counties included: Beaufort, Bertie, Bladen, Camden, Carteret, Chowan, Craven, Currituck, Dare, Duplin, Edgecombe, Franklin, Gates, Greene, Halifax, Hertford, Hyde, Johnston, Jones, Lenoir, Martin, Nash, Northampton, Onslow, Pamlico, Pasquotank, Pender, Perquimans, Pitt, Robeson, Sampson, Tyrell, Warren, Washington, Wayne, and Wilson

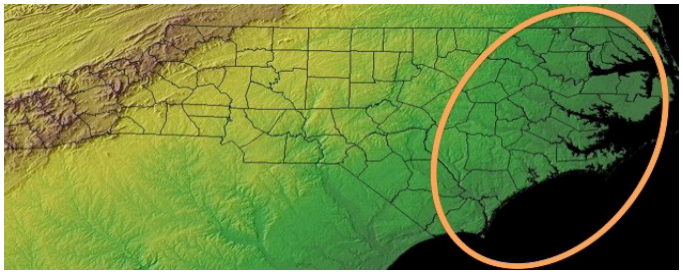

Figure 1. Topographic Map of North Carolina. The Oval Identifies the Eastern Part of the State Served by the Teleaudiology Project. David Walbert Created the Underlying Topographical Map for Learn NC (retrieved from http://www.learnnc.org/lp/editions/mapping/6413).

relative to the rest of the state (see Figure 1). Eastern North Carolina is primarily rural farmland. Traditionally, the population in these eastern counties has a median income lower than that of the rest of the state and a larger percentage of people living below the poverty level (http://quickfacts.census.gov/qfd/maps/north_carolina_map.html). In addition to poverty, this area has an unusually high teen pregnancy rate, greater percentage of Medicaid births, higher percentage of mothers who have not completed high school, and a larger percentage of minority births in comparison to the remainder of the state. Geographically, many inland bodies of water complicate travel over much of the region as well as travel from the Outer Banks to the mainland. Travel for diagnostic audiologic services can be as long as five hours and involve marine routes.

The goal of the NC-EHDI Teleaudiology Project was to provide infant diagnostic evaluations in rural eastern counties and to establish a coordinated system for the delivery of audiological evaluations for infants whose families experience economic and geographic barriers to service. The driving objectives were to reduce the number of infants in eastern North Carolina who are LFU/LTD for diagnostic testing or have delayed follow-up after referring on a hearing rescreen and to provide diagnostic evaluations. These objectives were in tune with the 1-3-6 Plan Joint Committee on Infant Hearing (2007) recommendation of providing comprehensive audiological evaluations no later than three months of age. Diagnostic testing began in June 2011. What follows is a description of the project development and implementation. Outcome data from June 2011 to July 1, 2015 is also presented.

Method

Participants

The catchment area included 22 of 98 birthing centers in North Carolina. Approximately 15% of live births (n = 68,494) in the state occurred at these birthing centers. Of those infants, 98.9% were screened prior to hospital discharge. Following initial screening, 2.3% (n =1559) were referred for rescreen. Prior to discharge, the parent(s)/caregiver(s) was/were given information and an appointment for outpatient rescreen. Of those

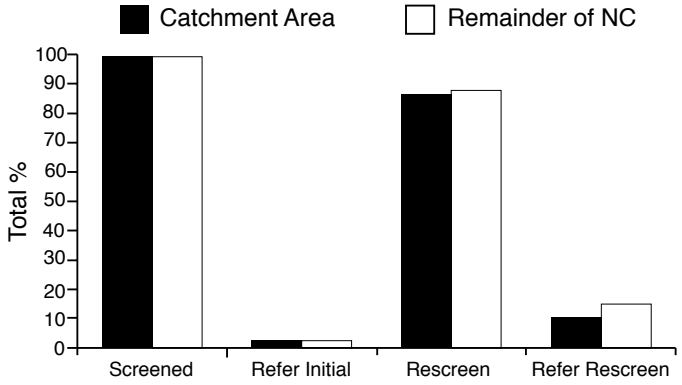

Figure 2. Percentage of Infants Screened, Rescreened, and Referred in Catchment Area and Remainder of North Carolina (NC).

referred for rescreening, approximately 86% were evaluated n = 1339). Rescreening tests were conducted following discharge at the birth hospital or at North Carolina Division of Public Health (NCDPH) local county health clinics. The percentage of infants in the catchment area that were screened and referred for a diagnostic

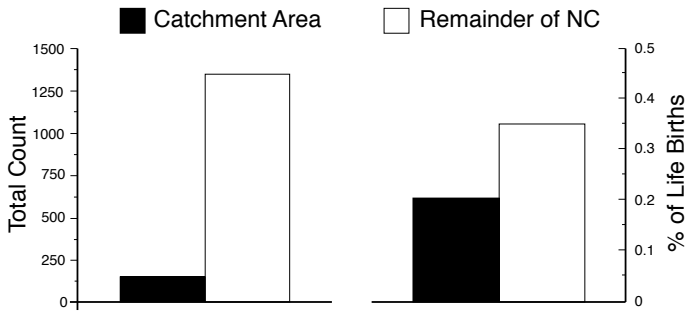

Figure 3. Total Count and Percentage of Infants Re Live Births Referred for Diagnostic Testing in Catchment Area and Remainder of North Carolina (NC).

test was similar to those in the rest of the state (see Figure 2).

Approximately 12% of those rescreened were referred for further diagnostic testing (n = 157). This represented approximately 0.2% of live births (see Figure 3) and was similar to the rest of the state. The total number of infants referred for diagnostic testing was also similar to those in the rest of the state (see Figure 3).

One hundred and fifty-seven infants were referred for diagnostic testing in the catchment area. Parent(s)/guardian(s) of 18 infants declined diagnostic testing. Approximately 29% of the remaining infants (n = 40) were evaluated through the Teleaudiology Project. Of the infants referred for diagnostic testing, 40% were female. Slightly more than one-half (i.e., 56%) were referred for unilateral diagnostic testing.

Materials and Procedures

Project planning preparation and preliminaries. Preparation and implementation of preliminary project processes by NC-EHDI took 18 months. The project initiation involved establishing a partnership with East Carolina University (ECU). This was a logical first step as the university had an established Telemedicine Program (ECU-TM) and experienced infant audiological diagnosticians in the Department of Communication Science and Disorders (ECU-CSDI). In addition, ECU-CSDI was an early pioneer in examining the provision of audiology services in a telehealth environment (Givens et al., 2003). In fact, the term "teleaudiology" was coined at ECU (Givens & Elangovan, 2003). A contract for services was developed and put in place between NC-EHDI and ECU. Following consultation with ECU-CSDI, necessary diagnostic audiologic equipment (i.e., evoked potential/otoacoustic emission systems and middle ear analyzers) was purchased. NC-EHDI developed protocols, guidelines, and training for their staff. It was also necessary to seek approval from North Carolina Board of Examiners for Speech-Language Pathologists and Audiologists for telepractice (North Carolina Administrative Code Title 21 64.0219; effective July 1, 2010).

The ECU-TM has been in continuous operation since its inception in 1992, making it one of the longest running clinical telemedicine operations in the world. The Clinical Telehealth Manager of ECU-TM initially undertook a number of preliminary steps such as determining equipment/network needs, defining technical and user support, transfering protected health information, medical records, establishing lines for referring, defining scheduling responsibilities and coordination, deciding and establishing immediate assistance protocol for teleaudiology delivery, and establishing a call center for field assistance. The ECU-TM also configured both patient end units at remote sites (See Figure 4) and the ECU-CSDI provider site. The remote sites established the ECU-TM network, which comprises heterogeneous communications links, including full and fractional T-1 (1.54 Mbps) and Integrated Services Digital Network (ISDN), which is typically aggregated at 3 Basic Rate Interface (BRI; 384 kbps). The remote sites were examination rooms in regional hospitals and typically equipped with a general view camera with pan, zoom, and tilt capability mounted on a mobile cart. These units used Advanced Encryption Standard encryption for Health Insurance and Portability Act (HIPAA) compliancy, video switching for auxiliary inputs, content sharing with laptop/audiology test equipment, user profile synchronization provisioned for wireless, and network/power connectivity. The ECU-CSDI provider site was equipped with a Polycom HDX 4000 HD video conferencing system. This system allowed for pan, zoom, and tilt camera far-end control in the remote site room; picture-in-picture layout control; directory dialing; up to 6 Mbps calls; mobile desktop capability; and dual audio/microphone capability.

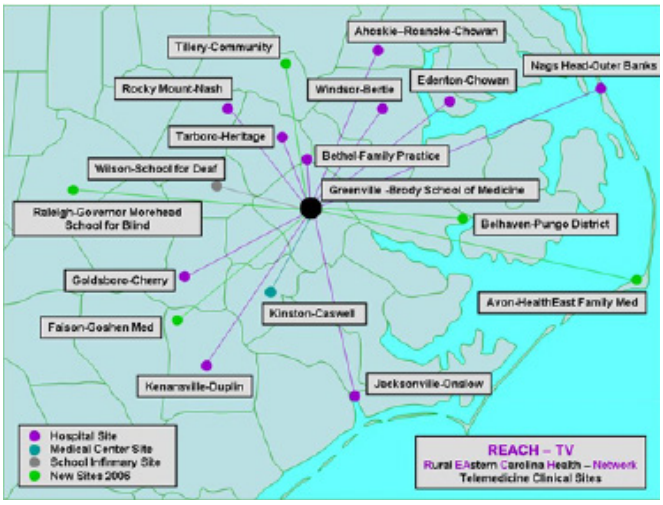

Figure 4. Map of Eastern Carolina University Telemedicine Remote Sites Accessible to the North Carolina-Early Hearing Detection and Intervention Teleaudiology Project (retrieved from http://www.ecu.edu/cs-dhs/telemedicine/telehealthnetwork.cfm).

The implementation of the diagnostic audiologic component of this project was the responsibility of the lead audiologist at ECU-CSDI. It was a four-fold process that included test protocol development, training NC-EHDI site staff, dual site preparation (i.e., remote test site and ECU-CSDI), and continuing evaluation/changes of protocol. The development of the diagnostic protocol was consistent with existing guiding diagnostic principles (American Speech-Language-Hearing Association, 2004; Joint Committee on Infant Hearing, 2007; Ontario Infant Hearing Program, 2008; British Columbia Early Hearing Program, 2008). The objective was to determine the presence or absence of permanent childhood hearing impairment with a target impairment of hearing threshold ≥ 30 dB HL in 500 to 4000 Hz range. The diagnostic protocol included patient history, cursory otoscopy, middle-ear analysis, distortion product otoacoustic emissions (DPOAEs), and auditory brainstem response (ABR). Onsite training at ECU-CSDI for NC-EHDI staff testing at remote sites was undertaken. This training included classroom instruction for test equipment and diagnostic protocols, provision of a protocol handbook, and lab instruction and exercises with test equipment and diagnostic protocols. Continued consulting support was ongoing with audiologists/technicians and the lead audiologist at ECU. Site preparation began at ECU with consultation between the lead audiologist and the Clinical Telehealth Manager at the ECU-TM. Equipment setup and training on Polycom systems was foremost. The remote audiologists/technicians in conjunction with the ECU-CSDI lead audiologist undertook site preparation at the remote sites.

Teleaudiology project service model. The Teleaudiology Project's diagnostic service delivery is a hybrid model. That is, it uses synchronous services to clients in real time and asynchronous store-and-forward

of audiometric data. Initial communication with parent(s)/caregiver(s) was with direct telephone contact by NC-EHDI staff. Diagnostic testing options via teleaudiology or at a diagnostic test center closest to their geographic location was offered. When parent(s)/caregiver(s) chose the teleaudiology option, an appointment was arranged by NC-EHDI staff. The family was informed of test date, time, and site location. Information was provided about preparations for the testing and the length of test as well. The day prior to the test, a reminder call was made to confirm the appointment, review the preparation instructions, and answer any last-minute questions.

NC-EHDI staff arrived at the remote site to set equipment up and connect with the ECU-CSDI lead audiologist prior to testing. After arrival of the infant and family, at the beginning of the testing session, an introduction to the lead audiologist at the provider site occurred via the Polycom video hardware. The infants were prepared for testing by NC-EHDI staff and the lead audiologist at the ECU-CSDI site who oversaw testing once the infant was settled.

The diagnostic test battery was consistent with the Joint Committee on Infant Hearing (2007) position statement. The protocols were consistent with the Ontario Infant Hearing Program (2008) and the British Columbia Early Hearing Program (2008). The main goal of assessment was to determine the presence or absence of permanent childhood hearing impairment. The nominal target permanent childhood hearing impairment includes any hearing threshold ≥ 30 dB HL at any frequency in the range of 500 to 4000 Hz, in either ear. The target permanent childhood hearing impairment includes conductive impairment associated with structural anomalies of the ear but does not include impairment attributable to non-structural middle ear conditions. The target also includes auditory neuropathy/auditory dys-synchrony.

All testing was attempted while the infant was in natural sleep or resting quietly. Wherever feasible, bilateral assessment included all of the procedures listed in Table 1. Except for the initial otoscopy, the order of procedures was discretional. The order of testing proceeded on the basis of obtaining the most important/most useful information first, the next most important next, et cetera for diagnostic, management, and parent/caregiver information purposes. The sequence-of-testing within a procedure (e.g., within ABR assessment) follows the same underlying principle—thus, most infants would undergo the same sequence. DPOAE and ABR testing was conducted with a GSI Audera AEP system (Version 2.67). Middle-ear analysis

Table 1. Diagnostic Test Protocol Components.

1. History taking.
2. Cursory otoscopy.
3. DPOAE amplitude and noise floor measurements at f2 frequencies of 1500, 2000, 3000 and 4000 Hz. The f2/f1 ratio was 1.2, with L1 and L2 levels of 65 and 55 dB SPL (Gorga et al., 1997).
4. Middle-ear analysis, which will include admittance tympanometry using a probe frequency of 1000 Hz and ipsilateral middle-ear muscle reflex testing using a 2000 Hz stimulus with a probe frequency of 1000 Hz (Margolis et al., 2003).
5. ABR threshold estimation by air conduction at 2000 Hz and 500 Hz with tonal stimuli. If time permits it would be desirable to also obtain ABR threshold estimation at 4000 Hz and 1000 Hz (Stapells, Gravel, & Martin, 1995).
6. Tonal stimulus ABR threshold estimation by bone conduction, where indicated, at 500 Hz and 2000 Hz (British Columbia Early Hearing Program, 2008).
7. In special circumstances, where indicated, high-intensity click-ABR measurement for auditory neuropathy/auditory dys-synchrony, including cochlear microphonic potentials and stimulus artifact analysis (British Columbia Early Hearing Program, 2008).

Note. DPOAE = Distortion Product Otoacoustic Emission ; ABR = Auditory Brainstem Response

[2]Parent(s)/guardian(s) were advised their infant be sleep deprived and arrive at the test site hungry. This means that the night prior to testing, the infant should not be allowed to get his/her normal amount of sleep. Also, it is normally appropriate to deny sleep and food for at least an hour before testing unless medically contraindicated. If the child is being brought to the test by car, it is important that every reasonable effort be made (consistent with safety) to keep the child awake on the journey. Because of the soporific effect of car journeys on infants, it was advised another person in addition to the driver is usually necessary.

was conducted with a GSI 39 Auto Tymp system. Detailed test protocols are presented in the Appendix. In the cases of unilateral referrals, the referred ear was tested first. If the infant was cooperative, the other ear was also tested. Four infants were recalled when diagnostic testing was not completed.

Diagnosis of infant hearing status was based on a general approach of audiologic inference with an integration and critical evaluation of all test findings (British Columbia Early Hearing Program, 2008). An infant was considered as audiometrically normal if air-conduction estimated hearing thresholds were 25 dB HL or better for all frequencies and/or DPOAE amplitudes exceeded the 5th percentile of the normal population and the 95th percentile of the impaired population at all frequencies (Gorga et al., 1997). An infant was considered to have a sensorineural impairment if air-conduction estimated hearing thresholds were > 25 dB HL; ABRs to bone-conducted stimuli exceeded the minimum test levels (i.e., elevated threshold); and/or DPOAE amplitudes fell below the 5th percentile of the normal population and the 95th percentile of the impaired population at all frequencies (Gorga et al., 1997) with normal peak compensated static acoustic admittance. An infant was considered to have a conductive hearing loss (abnormal middle ear function) if air-conduction estimated hearing thresholds were > 25 dB HL; ABRs to bone-conducted stimuli were present at the minimum test levels (i.e., elevated threshold); and/or DPOAE amplitudes were absent; and/or peak compensated static acoustic admittance fell below the 5th percentile of the normal population (Margolis, Bass-Ringdahl, Hanks, Holte, & Zapala, 2003). Auditory neuropathy/dys-synchrony was considered if the infant presented with OAEs and cochlear microphonics, abnormal ABRs, and absent middle ear acoustic reflexes.

Following the assessment, the parent(s)/caregiver(s) was/were counseled, via video, regarding test results by the lead audiologist. In the case where test results were pending, due to offline analysis in detail following asynchronous store-and-forward of audiometric data, parent(s)/caregiver(s) were contacted via telephone. The lead audiologist at ECU also reported diagnostic outcomes and recommendations via mail to the primary care physician/referring source within five business days of the diagnostic assessment. In addition, diagnostic outcomes and recommendations were entered into the North Carolina Division of Public Health Woman and Children Services Web (Hearing Link) website within five business days of the diagnostic assessment for state data tracking of hearing screening/diagnostic outcomes. Infants that presented with conductive hearing loss/abnormal middle ear function were referred to an otolaryngologist and recommended for retest following any medical management. Infants that presented with sensorineural hearing loss were also referred to an otolaryngologist, as well as back to the NC-EHDI staff for habilitation referral and family support services.

Results

The proportion of diagnostic tests performed via teleaudiology is illustrated in Figure 5. Diagnostic outcomes proportions are illustrated in Figure 6. The degree of sensorineural hearing loss ranged from mild to profound. Five percent ($n = 2$) of infants had an undetermined status (i.e., testing was incomplete to determine etiology). Boxplots of age of infants at screening, rescreening, and diagnostic test are shown in Figure 7.

The mean ages of infants at each test were 8.8 ($SD = 27.6$), 27.4 ($SD = 25.5$), and 73.3 ($SD = 47.3$) days for screening, rescreening, and diagnostic tests, respectively. The median ages of infants at each test were 1, 21, and

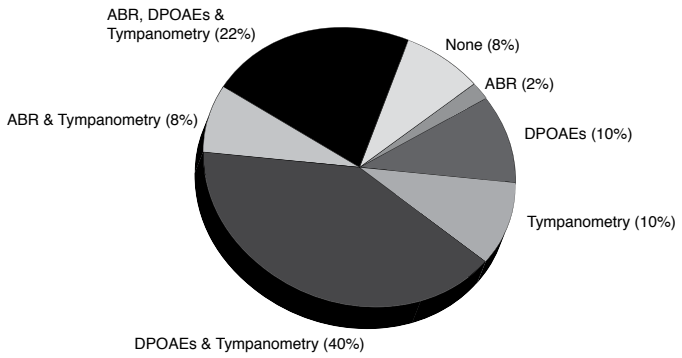

Figure 5. The Proportion of Diagnostic Tests Performed on the Infants Seen in the Teleaudiology Project. DPOAE = Distortion Product Otoacoustic Emissions; ABR = Auditory Brainstem Response.

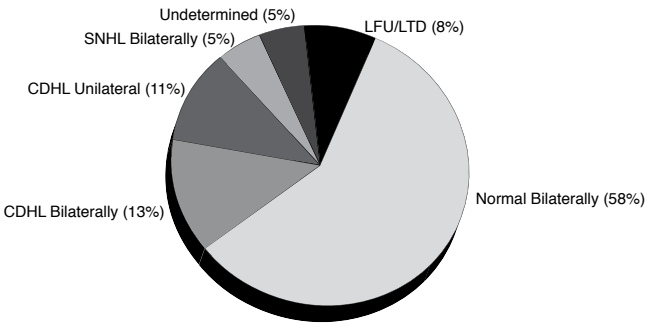

Figure 6. The Proportion of Diagnostic Outcomes with Infants Seen in the Teleaudiology Project. CDHL = Conductive Hearing Loss; SNHL = Sensorineural Hearing Loss; and LFU/LTD = Lost to Follow-up/Lost to Documentation.

60 days for screening, rescreening, and diagnostic tests, respectively. Two infants who spent considerable time in the neonatal intensive care unit prior to hospital discharge mainly drove the variability in the distributions. Those two infants did not receive their initial screening until 89 and 154 days. All other infants received their initial screening in their first month. One infant relocated out of the state after the rescreen referral and before diagnostic testing could be completed. Approximately 77% of infants referred for diagnostic testing were evaluated in the first three months after birth.

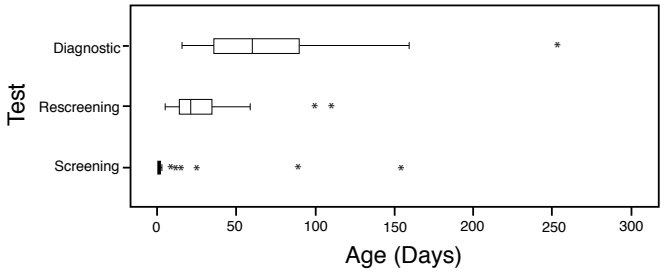

Figure 7. Box Plots of Age as a Function of Test. The top, bottom, and line through the middle off the box denote the 75th percentile, 25th percentile, and 50th percentile (median) respectively. The whiskers extend to 1.5 times the interquartile range or if no case has a value in that range, to the minimum or maximum values. The asterisks denote outliers.

Discussion

Joint Committee on Infant Hearing (2007) endorses early detection of and intervention for infants with hearing loss. Their proposed "1-3-6" plan suggests

> all infants should be screened at no later than 1 month of age (p. 898). Those who do not pass screening should have a comprehensive audiological evaluation at no later than 3 months of age. Infants with confirmed hearing loss should receive appropriate intervention at no later than 6 months of age. (p. 898)

The Teleaudiology Project developed jointly by the NC-EHDI and ECU has demonstrated a positive proof-of-concept that teleaudiology is a feasible means of meeting the recommendations for providing diagnostic testing of infants following screening referral. Specifically, approximately three-quarters of infants referred to the Teleaudiology Project for diagnostic testing were evaluated in the first three months after birth. The longest time for a diagnostic test was approximately eight months. In this case the child received the initial hearing screening after approximately five months in the neonatal intensive care unit. The encouraging results of timely diagnostic testing are particularly important in a rural area like eastern North Carolina. The catchment area presents with a number of socioeconomic challenges including poverty, lower education level, high teen pregnancies and Medicaid births, and a large percentage of minority births in comparison to the remainder of the state. Additional geographical challenges compound the socioeconomic challenges including complicated and lengthy travel over much of the catchment area.

Another positive of the project has been the involvement of audiology graduate students in training. Students were involved in the program setup from the beginning including: observation/direct participation in protocol development, dual site preparation (i.e., remote test site and teleaudiology clinic), and continuing evaluation/changes of protocol. Students placed in the teleaudiology "clinic block" also gain a unique clinical experience. That is, there are few opportunities for students to participate in teleaudiology clinical placements. For example, the Telepractice Special Interest Group of the American Speech-Language-Hearing Association (2014) in a recent survey found that of 52% of audiologists who provide services through telepractice, only 11% are in college/university facilities. Wilson and Seal (2015) reported that less than one-half of current program directors, which responded to a survey of telepractice in university AuD programs, reported they offer teleaudiology course work or clinical training. Finally, only 4% of training programs used this technology to deliver audiology services (Grogan-Johnson, Meehan, McCormick, & Miller, 2015). The Teleaudiology Project's experience and outcome data are also included in the AuD didactic curriculum for diagnostic testing models for infant hearing.

As with all programs, there remain some discouraging observations. First, there still remain a number of infants LFU/LTD for diagnostic testing (see Figure 6). Approximately 13% of infants referred were LFU/LTD. The issue of infants LFU/LTD has been identified in numerous programs (Alam, Gaffney, & Eichwald, 2014; Cockfield, Garner, & Borders, 2012; Krishnan, 2009; Liu, Farrell, MacNeil, Stone, & Barfield, 2008; Nikolopoulos, 2015; Spivak, Sokol, Auerbach, & Gershkovich, 2009). It remains a continuing concern for clinicians and program administrators. There are also a number of parent(s)/caregiver(s) whose infants were referred for diagnostic testing who declined. Unfortunately, 11% of parent(s)/guardian(s) declined diagnostic hearing testing for their infant in this catchment area. The audiologic status of these infants is unknown. The reason(s) for the parental/guardian decline is unknown. This parental/guardian noncompliance is similar to that found following preschool hearing screening referrals in the same catchment area (Allen, Stuart, Everett, & Elangovan, 2004). These findings point to the necessity of hearing health care professionals to improve public education, for both parent(s)/guardian(s) and physicians, concerning the importance of identification and habilitation of hearing loss. Of those referred for diagnostic testing, approximately 29% were seen via teleaudiology. The status of the remaining 71% is unknown. It is likely that some were LFU/LTD and did not undergo audiological diagnostic testing as was previously found in North Carolina (Williams et al., 2015). It is speculated that the majority of these infants were seen at the major birthing facilities located in the higher population areas/cities (e.g., Greenville and Jacksonville, NC).

Numerous studies have demonstrated the technical and clinical feasibility of providing audiologic services via teleaudiology. They include audiometric testing (Givens & Elangovan, 2003; Givens et al., 2003; Margolis, Killion, Bratt, & Saly, 2016), hearing screening (Krumm, Huffman, Dick, & Klich, 2008; Lancaster, Krumm, Ribera, & Klich, 2008), hearing aid fitting (Blamey, Blamey, & Saunders,

[3]The diagnostic outcomes are from the initial test conducted via teleaudiology. A "final outcome," in cases of conductive hearing loss where a medical referral and retest were recommended, is not reported.

2015; Penteado, Bento, Battistella, Silva, & Sooful, 2014), cochlear implant candidacy assessment (Aiello & Ferrari, 2015), and cochlear implant programming (Hughes et al., 2012). To date, however, there are no studies that have looked at an economic evaluation of teleaudiology services including the provision of infant diagnostic testing following newborn hearing screening. Remarkably, more than a decade ago, Suri, Dowling, Laxminarayan, and Singh (2005) presented a framework for an economic evaluation of telemedicine services both in terms of clinical effectiveness and cost-benefit. They identified a number of challenges for economic assessment including technological changes, sustainability of applications, availability of outcomes and other patient data, and generalizability of evaluation results. These same challenges face teleaudiology and specifically infant diagnostic testing following newborn hearing screening. As with their example in teleradiology, a significant barrier is the absence of a solid model for telemedicine cost analysis (i.e., how do you compare between two alternatives of teleaudiology and conventional service) and a lack of credible data sets with sufficient sample sizes. In addition, there is the need for randomized clinical trials of telemedicine. Suri et al. (2005) pointed out that studies might be driven by "technology push" rather than "clinical pull". Studies should focus on three fundamental aspects: define what services are provided and the speed of such services; identify whom the clinical service is benefiting (i.e., the clinician or the patient); and determine what outcome measures (e.g., patient and/or parent/guardian satisfaction, compliance, and outcomes) should be used.

In summary, the Teleaudiology Project developed jointly by the NC-EHDI and ECU has demonstrated positive proof-of-concept for teleaudiology in meeting the recommendations for providing diagnostic testing of infants following screening referral in a timely manner. In addition, with the project located at a university site that provides clinical training of graduate audiology students, it provides a distinctive opportunity for curriculum and clinical experiences in teleaudiology and stays current with developments in the field of audiology. Future studies are needed to evaluate the economic impact of teleaudiology services including the delivery of infant diagnostic testing following newborn hearing screening.

References

Aiello, C. P., & Ferrari, D. V. (2015). Teleaudiology: Efficacy assessment of an online social network as a support tool for parents of children candidates for cochlear implant. *Codas*, 27, 411–418.

Alam, S., Gaffney, M., & Eichwald, J. (2014). Improved newborn hearing screening follow-up results in more infants identified. *Journal of Public Health Management and Practice*, 20, 220–223.

Allen, R. L., Stuart, A., Everett, D., & Elangovan, S. (2004). Preschool hearing screening: Pass/refer rates for children enrolled in a head start program in Eastern North Carolina. *American Journal of Audiology*, 13, 29–38.

American Speech-Language-Hearing Association. (2004). Guidelines for the audiologic assessment of children from birth to 5 years of age [Guidelines]. Retrieved from http://www.asha.org/members/deskref-journals/deskref/default

American Speech-Language-Hearing Association. (2014). 2014 SIG 18 telepractice services survey results. Retrieved from www.asha.org

British Columbia Early Hearing Program (BCEHP) Diagnostic Audiology Advisory Group (2008). Diagnostic audiology protocol (PROTOCOLS DX100 – 1000). Retrieved from http://www.phsa.ca/NR/rdonlyres/06D79FEB-D187-43E9-91E4-8C09959F38D8/40115/aDAAGProtocols1.pdf

Blamey, P. J., Blamey, J. K., & Saunders, E. (2015). Effectiveness of a teleaudiology approach to hearing aid fitting. *Journal of Telemedicine and Telecare*, 21, 474–478.

Cockfield, C. M., Garner, G. D., & Borders, J. C. (2012). Follow-up after a failed newborn hearing screen: A quality improvement study. ORL - Head and Neck Nursing, 30, 9–13.

Givens, G. D., Blanarovich, A., Murphy, T., Simmons, S., Blach, D., & Elangovan, S. (2003). Internet-based tele-audiometry system for the assessment of hearing: A pilot study. , 9, 375–378.

Givens, G. D., & Elangovan, S. (2003). Internet application to tele-audiology -"nothin' but net". *American Journal of Audiology*, 12, 59–65.

Gorga, M. P., Neely, S. T., Ohlrich, B., Hoover, B., Redner, J., & Peters, J. (l997). From laboratory to clinic: A large scale study of distortion product otoacoustic emissions in ears with normal hearing and ears with hearing loss. *Ear and Hearing*, 18, 440–455.

Grogan-Johnson, S., Meehan, R., McCormick, K., & Miller, N. (2015). Results of a national survey of preservice telepractice training in graduate speech-language pathology and audiology programs. *Contemporary Issues in Communication Science and Disorders,* 42, 122–137.

Hughes, M. L., Goehring, J. L., Baudhuin, J. L., Diaz, G. R., Sanford, T., Harpster, R., & Valente, D. L. (2012). Use of telehealth for research and clinical measures in cochlear implant recipients: A validation study. *Journal of Speech Language and Hearing Research*, 55, 1112–1127.

Joint Committee on Infant Hearing (2007). Year 2007 position statement: Principles and guidelines for early hearing detection and intervention programs. *Pediatrics*, 120, 898–921.

Krishnan, L. A. (2009). Universal newborn hearing screening follow-up: A university clinic perspective. *American Journal of Audiology*, 18, 89–98.

Krumm, M., Huffman, T., Dick, K., & Klich, R. (2008). Telemedicine for audiology screening of infants. *Journal of Telemedicine and Telecare,* 14, 102–104.

Lancaster, P., Krumm, M., Ribera, J., & Klich, R. (2008). Remote hearing screenings via telehealth in a rural elementary school. *American Journal of Audiology*, 17, 114–122.

Liu, C. L., Farrell, J., MacNeil, J. R., Stone, S., & Barfield, W. (2008). Evaluating loss to follow-up in newborn hearing screening in Massachusetts. *Pediatrics*, 121, e335–e343.

Margolis, R. H., Bass-Ringdahl, S., Hanks, W., Holte, L., & Zapala, D. (2003). Tympanometry in newborn infants - 1 kHz norms. *Journal of the American Academy of Audiology,* 14, 386–395.

Margolis, R. H., Killion, M. C., Bratt, G. W., & Saly, G. L. (2016). Validation of the Home Hearing Test™. J*ournal of the American Academy of Audiology,* 27, 416–420.

Nikolopoulos, T. P. (2015). Neonatal hearing screening: What we have achieved and what needs to be improved. *International Journal of Pediatric Otorhinolaryngology*, 79, 635–637.

Ontario Infant Hearing Program. (2008). Audiologic Assessment Protocol Version 3.1. Available at http://www.mountsinai.on.ca/care/infant-hearing-program/documents/IHPAudiologicAssessmentProtocol3.1FinalJan2008.pdf

Penteado, S. P., Bento, R. F., Battistella, L. R., Silva, S. M., & Sooful, P. (2014). Use of the satisfaction with amplification in daily life questionnaire to assess patient satisfaction following remote hearing aid adjustments (telefitting). *JMIR Medical Informatics*, 2, e18.

Spivak, L., Sokol, H., Auerbach, C., & Gershkovich, S. (2009). Newborn hearing screening follow-up: Factors affecting hearing aid fitting by 6 months of age. *American Journal of Audiology,* 18, 24–33.

Stapells, D. R. (2000). Frequency-specific evoked potential audiometry in infants. In R. C. Seewald (Ed.), *A Sound foundation through early amplification* (pp. 13–32). Phonak AG.

Stapells, D. R., Gravel, J. S., & Martin, B. A. (1995). Thresholds for auditory brainstem responses to tones in notched noise from infants and young children with normal hearing or sensorineural hearing loss. *Ear and Hearing*, 16, 361-371.

Stuart, A., Yang, E. Y., & Stenstrom, R. (1990). Effect of temporal area bone vibrator placement on auditory brainstem response in newborn infants. *Ear and Hearing*, 11, 363-369.
Suri, J. S., Dowling, A., Laxminarayan, S., & Singh, S. (2005). Economic impact of telemedicine: *A survey. Studies in Health Technology & Informatics*, 114, 140-156.
Williams, T. R., Alam, S., & Gaffney, M. (2015). Progress in identifying infants with hearing loss—United States, 2006–2012, *Morbidity and Mortality Weekly Report*, 64, 351–356.
Wilson, N. J., & Seal, B. C. (2015). Telepractice in university Au.D. programs: Survey of program directors. *Perspectives on Telepractice*, 5, 27–37.
Yang, E. Y., & Stuart, A. (1990). A method of auditory brainstem response testing with infants using bone-conducted clicks. *Journal of Speech-Language-Pathology and Audiology*, 14(4), 69–76.
Yang, E. Y., Stuart, A., Mencher, G. T., Mencher, L. S., & Vincer, M. J. (1993). Auditory brain stem responses to air- and bone-conducted clicks in the audiological assessment of at-risk infants. *Ear and Hearing*, 14, 175–182.

Appendix

The distortion product otoacoustic emissions (DPOAE) protocol followed that of Gorga et al. (1997). Primary tones had an f_2/f_1 ratio of 1.22. L_1, L_2 levels were 65, 55 dB SPL. The f_2 frequencies were 1500, 2000, 3000, 4000, and 6000 Hz. A sequential signal presentation and time domain averaging was employed for data collection. The minimum and maximum averages that were acquired for each data point were 10 and 375, respectively. Frame rejection ensued if L_1 and L_2 were out of tolerance by ±5 dB and/or ambient noise levels exceeded 25 dB SPL. DPOAE collection terminated when either of the following occurred: test time exceeded 32 s or 1500 frames; 30% occurrences of frame rejection due to excessive ambient noise; and/or 20 occurrences of L_1 or L_2 being out of tolerance. The test was accepted when 32 frames were averaged and the average noise level was less than -12 dB SPL plus either of the following conditions were met: the DPOAE was 3 dB above the noise floor or the absolute noise level was less than -20 dB SPL.

For admittance tympanometry, the pressure sweep began at the starting pressure of +200 daPa and proceeded to -400 daPa at a rate of 600 daPa/s. The probe frequency was 1000 Hz. Peak compensated static acoustic admittance was determined from the negative tail at -400 daPa (Margolis et al., 2003). Ipsilateral middle-ear muscle reflex testing employed a 2000 Hz evoking stimulus. Reflex stimulus level should begin at 85 dB hearing level (HL) and increase in 5 dB steps up to no greater than 100 dB HL.

For behavioral hearing threshold estimation, ABR stimuli were air- and bone-conducted linear ramped 2-1-2 tone bursts. In the case of suspected auditory neuropathy/auditory dys-synchrony, 75 dB nHL 100 µs air-conducted clicks were used at a rate of 8.7/s. A total of 1026 samples were averaged and replicated. Tone bursts were centered at 500, 1000, 2000, and/or 4000 Hz. Stimuli were presented through a GSI TIP-50 insert earphone or a Radioear B-71 bone vibrator at a rate of 37.7/s. A total of 2014 samples were averaged and replicated. Reference threshold levels for air- and bone-conducted clicks were adopted from Yang, Stuart, Mencher, Mencher, & Vincer (1993). Reference threshold levels for tone burst stimuli were adopted from Stapells (2000). An ipsilateral recording montage was used with the noninverting electrode on the high-forehead (F_{pz}), inverting electrode on the ipsilateral postauricular area ($M_{1/2}$), and one common to the contralateral inferior postauricular area ($M_{2/1}$). Interelectrode impedances were maintained below 5000 Ω. The recorded electroencephalogram was amplified 105 and bandpass filtered (30 to 3000 Hz). Electroencephalogram samples exceeding ± 25 μV were rejected. Analysis times were 13 ms post-stimulus for click and 25 ms post-stimulus for tone bursts. The bone vibrator was placed in a supero-posterior temporal position during bone conducted stimuli delivery (Stuart, Yang, & Stenstrom, 1990). An elastic band with Velcro was used to hold the bone vibrator with a coupling force of 425 ± 25 g (Yang & Stuart, 1990). Coupling force was verified with a spring scale (Ohaus 8014) that manually pulled the bone vibrator away from the skull by a nylon monofilament attached to the bone vibrator. The coupling force was measured at the point the vibrator cleared and became flush with the scalp.