

BMJ Open Communities Helping the Hearing of Infants by Reaching Parents (CHHIRP) through patient navigation: a hybrid implementation effectiveness stepped wedge trial protocol

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ABSTRACT

Introduction As the most common neonatal sensory disorder in the USA, infant hearing loss has an incidence of 1.7 per 1000 births. The consequences of delayed diagnosis and failure to obtain timely intervention include significant communication impairment and negative socioeconomic effects. Early Hearing Detection and Intervention (EHDI) national standards dictate that all infants should be screened and diagnosed by 3 months of age and there is a need for interventions that promote adherence to timely diagnosis. Patient navigation (PN) has been shown to be efficacious to decrease non-adherence with infant hearing diagnostic care; however, PN has yet to be tested in diverse communities or implemented into real-world settings.

Methods and analysis The proposed research is a community-engaged, type 1 hybrid effectiveness-implementation trial of a PN intervention aimed at decreasing infant hearing diagnosis non-adherence after failed newborn hearing screening, delivered in state-funded EHDI clinics. Guided by our community advisory board and partners, we aim to (1) test the effectiveness of PN to decrease non-adherence to receipt of infant hearing diagnosis within 3 months after birth using a stepped-wedge trial design, (2) investigate implementation outcomes and factors influencing implementation and (3) determine the cost-effectiveness of PN from the perspective of third-party payers. The study will be conducted from April 2019 until March 2024.

Ethics and dissemination This protocol was approved by the University of Kentucky Institutional Review Board. Although all research involving human subjects contains some risk, there are no known serious risks anticipated from participating in this study. We will seek to disseminate our results in a systematic fashion to patients, key stakeholder, policymakers and the scientific community. Our results will impact the field by partnering with communities to inform the scale-up of this innovative patient supportive intervention to create efficient and effective EHDI programmes and maximise public health impact.

Trial registration number Clinicaltrials.gov (Pre-results phase): NCT03875339.

Strengths and limitations of this study

- A strength of this study is the investigation of patient navigation intervention delivered within public-funded healthcare system for hearing healthcare, which represents a meaningful step forward in developing efficient, effective and scalable paediatric hearing healthcare.
- An intervention centred in the community and informed by diverse stakeholders while targeting parents/caregivers of infants immediately after abnormal screening is a novel strategy to improve paediatric hearing healthcare efficiency and cultural sensitivity.
- The use of a type 1 hybrid effectiveness-implementation study with a stepped-wedge trial design is innovative in the field of hearing sciences.
- Limitations of a stepped-wedge design includes a delay in some participants or study sites receiving the intervention and the influences of temporal changes in policy or practice on the intervention during the course of the study.
- The delivery of preventive interventions to high risk and underserved communities is often overlooked but is an essential component of this research.

INTRODUCTION

Paediatric hearing loss constitutes a major public health problem, as the most common neonatal sensory disorder in the USA, with an incidence rate of 1.7 per 1000 births.¹ The consequences of delayed infant hearing loss diagnosis and intervention include significant delays in language, cognitive and social development with profound effects on education and employment.^{2 3} These consequences carry substantial financial burdens, as according to the Centers for Disease Control and Prevention (CDC), the lifetime medical, educational and occupational costs due to deafness for children born in 2000 are estimated at US\$2.1 billion.⁴

Early identification and treatment of infant hearing loss is essential but unfortunately delayed. According to the US Preventive Services Task Force, early diagnosis of hearing loss reduces language development problems, social and emotional challenges and learning and behaviour disorders.⁵ In conjunction with diagnosis, intervention before 6 months of age has profound effects on language expressive measures and social adjustment.^{6–9} The national Early Hearing Detection and Intervention (EHDI) programme has implemented a ‘1-3-6’ rule that is the current standard for newborn hearing testing. This rule states that all infants should be screened no later than 1 month after birth, diagnosed before 3 months of age and receive intervention within 6 months of age.^{10–14} The Joint Committee on Infant Hearing and CDC have set the benchmark that no more than 10% of infants should be non-adherent to diagnostic testing by 3 months of age,^{11 15} however; data shows that this standard is not being met across the country. In 2015, 59.4% of US infants failed to obtain a diagnosis within 3 months after an abnormal newborn hearing screening test.¹⁵ Despite efforts to document follow-ups and promote adherence to timely diagnostic testing, Kentucky has a non-adherence rate of over 30%.¹ Children of underserved rural regions, like Appalachia, are also at an increased risk of delayed diagnosis and treatment of hearing loss.^{16–18}

Timely adherence to infant diagnostic testing and hearing loss treatment is a complex process and parents face many barriers. Despite multiple streamlining initiatives, many parents find the diagnostic and treatment process for infant diagnostic testing difficult to navigate.¹⁹ Families who travel greater distances for resources, have lower levels of parental education, low socioeconomic status or have public insurance are at an increased risk of non-adherence.^{18 20–22} Families of children with hearing loss report that they lack confidence and resources needed for healthcare decision-making for their child.²³ Many parents lack role models who have been through the process of hearing loss diagnosis and intervention,²⁴ as more than 90% of deaf and hard of hearing children have hearing parents.²⁵ Consistent with the Social Cognitive Theory (SCT) model,^{26–29} multiple factors intersect to influence hearing healthcare adherence and access.

Only patient navigation (PN) has been shown to be efficacious despite other approaches to decrease non-adherence to infant hearing testing and treatment. Patient navigators (PNs) are trained individuals who assess and mitigate personal, interpersonal and environmental barriers to healthcare adherence and access, consistent with SCT-based approaches to promote healthy behaviours.^{26–29} PNs educate patients on health conditions and healthcare systems while facilitating adherence to healthcare recommendations.³⁰ Primarily implemented and studied in the cancer field, PN reduces non-adherence and hastens diagnosis and treatment.^{31–33} PN programmes are effective in assisting patients from underserved backgrounds, including rural regions.^{34–41} The positive patient-level effects of PN (ie, improved adherence with medical diagnostic testing^{42 43} and timely diagnosis

and treatment^{36 42 44}) have resulted in significant healthcare cost savings.³⁴ In infant hearing assessment, PN improves the efficiency of scheduling diagnostic testing and significantly increases parent knowledge of diagnostic testing recommendations compared with the standard of care.⁴⁵ Our recent randomised controlled trial (RCT) in an academic centre demonstrated the efficacy of PN in decreasing non-adherence in infant hearing healthcare.⁴⁵

Additionally, public-funded EHDI programmes provide infrastructure within each state to assess and track hearing in infants, providing an ideal platform for the delivery of PN targeting infant hearing testing and treatment on a community level. EHDI programmes have the capacity to target the most vulnerable patient populations in diverse community settings (eg, low levels of parental education, low socioeconomic status, public insurance), who are also at highest risk for non-adherence with recommended diagnostic testing.^{17–22} In Kentucky, public-funded community audiology clinics serving EHDI patients are administered by the Kentucky Office for Children with Special Health Care Needs (OCSHCN). By partnering with EHDI and OCSHCN to conduct this research, we can assess not only effectiveness of PN, but also implementation factors, outcomes and costs expended/averted in the settings intended to reach the most vulnerable patient populations in our state.

Guided by our Community Advisory Board, EHDI partners and hearing healthcare stakeholders, we will conduct a type 1 hybrid effectiveness-implementation trial in 10 OCSHCN clinics testing PN while simultaneously investigating implementation outcomes and measuring costs.⁴⁶ A type 1 hybrid effectiveness-implementation trial will allow investigators to test the PN intervention through an effectiveness trial while also evaluating implementation strategies of the intervention. This study will evaluate the effectiveness of PN to decrease non-adherence rates of infant hearing diagnosis within 3 months after birth while also exploring associations of implementation factors and outcomes.⁴⁷ Finally, cost-effectiveness of PN versus standard-of-care will be compared. Given prior findings of efficacy, it is hypothesised that PN will decrease non-adherence to infant hearing diagnosis within 3 months after birth compared with standard-of-care. Our findings will directly inform state-level policy and services impacting children with hearing loss and set the stage for a national multisite implementation trial to maximise public health impact.

METHODS AND ANALYSIS

Overview

The proposed study will use a stepped-wedge trial design and deliver PN sequentially in 10 state-funded Kentucky OCSHCN clinics randomised to cross from usual care to PN in steps of 6-month intervals over the project period. The study start date is 1 April 2019 and conclusion date is 31 March 2024. Prior to initiation of PN at each clinic, the control condition will be the standard of care. The overall effectiveness of PN will be tested by comparing non-adherence rates during the PN condition to those

Table 1 Study data collection activities

Study data collection activities

Aim	Data source(s)	Analysis strategy	Goal
AIM 1: Evaluate the effectiveness of the patient navigation intervention	Parent–infant dyads (1120) OCSHCN clinic data State EHDI data	Comparing non-adherence rates to follow-ups in PN condition versus standard of care	Reduce infant hearing diagnosis non-adherence
AIM 2: Assess implementation outcomes	Parent participants (40) Patient navigators (30) Clinic administrators, staff and providers (30)	Quantitative Data: Descriptive statistics and bivariable comparisons Qualitative Data: Analysing interviews for codes and themes	Identify implementation strategies to increase uptake and adoption of patient navigation
AIM 3: Conduct cost-effectiveness analysis	PN establishment Programme implementation Parent time and travel Treatment costs Non-medical costs Non-adherence costs for clinics	Estimate costs associated with an incremental change in effectiveness for each PN modality compared with standard of care using the incremental cost-effectiveness ratio	Compare net costs and net effectiveness of patient navigation to standard of care

EHDI, Early Hearing Detection and Intervention; OCSHCN, Office for Children with Special Health Care Needs; PN, patient navigation.

during the standard of care condition. Simultaneously, assessments of preliminary implementation outcomes (ie, acceptability, adoption, recruitment/retention and fidelity) and multilevel factors (drawn from the Consolidated Framework for Implementation Research (CFIR)⁴⁷) influencing implementation of PN in each clinic will occur. Lastly, a cost-effectiveness analyses will be conducted from the perspective of third-party payers. See [table 1](#) for data collection activities for each study aim. Our study will inform EHDI programming to (1) reduce infant hearing diagnosis non-adherence, (2) identify implementation strategies to increase uptake and adoption of PN and (3) contribute to the implementation science field. The study is registered with ClinicalTrials.gov and protocol data is found in [table 2](#).

Community advisory board

This study uses a community advisory board (CAB) which focuses on paediatric hearing and the psychosocial needs of families with deaf and hard of hearing (DHH) children. The CAB is comprised of parents and teachers of DHH children, audiologists, speech–language pathologists, early interventionists who provide social services or healthcare services for babies and toddlers with developmental delays and disabilities, mental health providers, the director of the Kentucky chapter of Hands and Voices and state-level programme administrators. The CAB plays an active role in identifying patient navigators involved in the study, informing the content of patient navigator training, provide resources that are communicated to participants through the patient navigators, advising recruitment methods and intervention delivery and disseminating study results.

Patient navigation intervention

PN is effective in other healthcare fields, as well as accepted and effective among rural and low socioeconomic status populations. PNs are trained individuals who assess and assist with barriers to healthcare adherence

PN intervention delivery will follow the American Cancer Society PN model.⁴⁸ Selection of PNs will occur within each of the 10 study site communities by CAB referral or from an existing pool of parents/patients who have requested to be involved in hearing healthcare research and patient advocacy (maintained by the OCSHCN). Each PN candidate will be reviewed by the CAB. Inclusion criteria: (1) age 21 years or older, (2) able to speak and read English and (3) willing to complete the PN training curriculum and deliver PN. Our PNs will work with parents to identify and address specific barriers to obtaining follow-up diagnostic hearing testing for their infants (eg, making appointments, child-care and transportation issues).⁴⁹ PNs will provide social support via supportive listening, educational materials about hearing testing and referrals for additional assistance, if needed.⁵⁰ PNs will first contact parents by telephone for an initial interview to build rapport and assess for needs and resources. PNs will then contact parents weekly using the parent's preferred method (ie, phone, text or email). Weekly PN contacts will continue until the diagnostic test has been obtained (verified by EHDI data) or until 12 weeks since birth have elapsed. The *standard of care* in OCSHCN clinics involves provision of printed educational materials and an educational video regarding infant hearing loss and EHDI services while in the hospital. Parents in the standard of care group will be given their outpatient follow-up appointment prior to discharge. Once discharged from the hospital, parents from this group will have access to discuss any questions or concerns with the OCSHCN office, but this will be parent-initiated contact outside of the phone reminder 48 hours prior to the appointment. Although the limited access to healthcare during the COVID-19 pandemic affected referrals and recruitment for the study, no adaptations had to be made to the intervention due to COVID-19 as the intervention occurs remotely.

Table 2 Trial registration data

Primary registry and trial identifying number	ClinicalTrials.gov NCT03875339
Date of registration in primary registry	14 March 2019
Secondary identifying numbers	n/a
Source(s) of monetary or material support	National Institutes of Health (National Institute on Deafness and Other Communication Disorders, National Institutes of Health Office of The Director)
Primary sponsor	National Institute on Deafness and Other Communication Disorders
Secondary sponsor(s)	National Institutes of Health Office of The Director
Contact for public queries	Matthew L Bush, MD, PhD, MBA (matthew.bush@uky.edu)
Contact for scientific queries	Matthew L Bush, MD, PhD, MBA (matthew.bush@uky.edu)
Public title	Communities Helping the Hearing of Infants by Reaching Parents (CHHIRP)
Scientific title	Communities Helping the Hearing of Infants by Reaching Parents: The CHHIRP Navigator Trial
Countries of recruitment	USA
Health condition(s) or problem(s) studied	Paediatric hearing loss
Intervention(s)	Intervention: Patient navigation to promote adherence to diagnostic hearing testing after a referred result on infant hearing screening. Control condition: Treatment/care as usual.
Key inclusion and exclusion criteria	Ages eligible for study: Up to 99 years (child, adult, older adult). Sexes eligible for study: All. Accepts healthy volunteers: Yes. Inclusion criteria: Parent-infant dyads: 1. Infant failed a hearing screening in one or both ears before postnatal hospital discharge. 2. Infant was referred for follow-up diagnostic testing at 1 of the 10 participating OCSHCN clinics. 3. Parent able to speak either English or another language using phone interpreting services. Exclusion criteria: Children and parents live outside Kentucky or who will be moving out of Kentucky within the first 3 months of life.
Study type	Interventional Allocation: Randomised intervention with parallel assignment via stepped wedge trial design. Parallel assignment masking: None. Primary purpose: Prevention.
Date of first enrolment	1 August 2019
Target sample size	2240
Recruitment status	Enrolling by invitation
Primary outcome(s)	Number of participants who do not receive diagnostic audiological testing within 3 months after a referred infant hearing screening test.
Key secondary outcomes	n/a

Aim 1: effectiveness of PN

Overview

Under a sequential rollout in steps of 6-month intervals, 10 OCSHCN clinics will be randomly allocated to implement PN over the project period. Randomisation will be stratified by clinical patient population size and conducted by a biostatistician. The highly pragmatic stepped-wedge

trial design will allow all clinics to contribute control and intervention data. All 10 clinics will be in the control condition during the first 6 months of the study. The first clinic will cross to the intervention condition following month 6 of Y1, and the 10th clinic will do so following month 6 of Y4 of the project period. PN will continue through the first half of Y5.

Study sample and recruitment

Approximately 500–600 infants are referred for diagnostic testing to the OCSHCN clinics after failed infant hearing screening which would provide a total of approximately 3000 infants over the course of the 5-year study. Based on state-level EHDI data, there will be ~1700 referrals to the 10 clinics under the PN intervention condition. We aim to enrol ~66% of all eligible referrals (N=1120). The inclusion criteria include parents whose infants fail hearing screening in one or both ears before postnatal hospital discharge and are referred for follow-up diagnostic testing at 1 of the 10 participating OCSHCN clinics. Parents who live or move outside Kentucky during the study will be excluded, because they are not eligible for services at the OCSHCN. Recruitment of all eligible parent–infant dyads in that clinic will begin once it crosses from standard of care to PN in the stepped-wedge design. Within 1 week of postnatal hospital discharge, research personnel will call the parents referred to a clinic in the PN condition to describe the study, screen for eligibility and enrol them into the study using verbal informed consent over the phone (online supplemental file 1). Uninterested parents who do not consent will undergo follow-up per standard of care.

Measures and procedures

Clinics: All data collected on non-adherence to follow-up within 3 months after birth will be at the clinical level. The primary effectiveness outcome is the proportion of non-adherent referrals for diagnostic testing at each clinic during each month of the trial. The stepped-wedge design will allow monitoring of non-adherence trends throughout the control and intervention conditions for each clinic and the state as a whole over the entire course of the study. The comparison of clinics' PN condition data to their control data will assess the effectiveness of PN to decrease non-adherence rates. Data will be shared with the CAB as well as OCSHCN administration on a quarterly basis. **Parent-infant dyads:** During the same phone call to obtain parental informed consent, research personnel will administer baseline measures on knowledge of hearing loss, self-efficacy for obtaining follow-up testing and perceived barriers to follow-up testing. Following completion of baseline measures, parents will be contacted by the PN to initiate intervention delivery. Post-test measures will be administered by phone by a research assistant 16 weeks after birth (1 month after the 3-month target deadline for diagnostic testing has passed). Parent participants will receive compensation for each set of completed measures. Aggregated data will

be shared with the CAB quarterly to obtain feedback. **Parent-infant measures:** Secondary effectiveness outcomes include parents' knowledge, self-efficacy and barriers regarding obtaining follow-up diagnostic testing for their infant. Parent knowledge will be assessed using four multiple-choice items developed from our prior RCT on diagnostic testing purpose and recommendations. Parent self-efficacy to obtain testing for their infant will be measured using a 10-item scale adapted from an existing self-efficacy measure for cancer screening.⁵¹ Each item uses a 5-point Likert-type response scale and addresses one step associated with the testing process (eg, making an appointment, transportation, payment, proceeding when worried). Parent-identified barriers will be measured using five items with 5-point Likert-type response options that tap barriers to obtaining diagnostic testing identified by parents in our preliminary studies.^{52 53} We will also obtain EHDI data for all enrolled infants until their 1 year birthday (or until data collection ends) to determine the time from birth to initial completed diagnostic assessment, number of no-show appointments and number of rescheduled appointments.

Sample size

The trial follows a stepped-wedge design consisting of data collection from 10 clinics over a 4.5-year period. To optimise power while also ensuring an adequate number of subjects within each clinic for each time period, steps will consist of 6-month periods. The number of clinics receiving PN will accumulate over time. Based on this design and with an estimated sample size of 1120 infant-parent dyads, using a two-sided test and a 5% significance level, we will have greater than 90% power to detect a difference between PN and standard of care conditions, assuming a clinically meaningful effect of PN in reducing non-adherence rates from Kentucky's current rate of 33.7% to the CDC benchmark of 10%. This power calculation accounts for clustering within clinics (ie, statistical correlation among the binary non-adherence outcomes from subjects within the same cluster, as measured by the coefficient of variation). With an extremely conservative enrolment estimate of 10 referred infants per clinic per 6-month period and the usual range of coefficients of variation from 0.15 to 0.4 to account for clustering,⁵⁴ statistical power will range from 0.91 to 0.96. Annual clinic referrals are larger than represented by this estimate, with 500–600 newborns with failed hearing screens referred each year; if even slightly larger cluster sizes of 15 are enrolled per clinic each period, power will reach at least 0.98 under all scenarios.

Statistical analysis

All tests will be two-sided and will use a 5% statistical significance level. Analyses will be conducted in SAS V.9.4 (SAS Institute). Secondary analyses will use adjusted significance levels or p values to control for Type I errors due to multiple testing using the method of Benjamini and Hochberg.⁵⁵ The primary effectiveness outcome is non-adherence rate, obtained with

clinic-level data. Analysis methods must account for any clustering within clinics (ie, statistical correlation among the binary non-adherence outcomes from patients within the same clinic). The primary intent-to-treat data analysis will use generalised estimating equations (GEE) with corrected empirical SEs in order to maintain valid inference.⁵⁶ We will fit the commonly used logistic regression model for the analysis of binary outcomes arising from a stepped-wedge design with clinics.⁵⁴ Specifically, fixed effects for trial condition and time will be included in the model. These separate time effects for each time period will be included in the statistical model, accounting for secular trends and thus ensuring a valid model. For all analyses, recommended statistical approaches will be used in the presence of missing data (eg, multiple imputation at the cluster level). Sensitivity analyses will be considered and dictated by the type(s) of missing data. Consistent with National Institutes of Health (NIH) requirements for rigour and transparency, secondary analyses will include important sociodemographic variables as covariates within the above model. Regarding additional secondary outcomes of interest, we will explore associations among parent knowledge, self-efficacy and barriers as related to (a) non-adherence, (b) time interval from birth to the initial completed outpatient auditory brainstem response (ABR), (c) number of no-show office visits and (d) number of rescheduled visits. Each of these analyses will use the same general GEE approach to account for clustering of outcomes from subjects in the same clinic. Depending on the outcome type, a marginal (population average interpretation) generalised linear model will be fit.

Aim 2: implementation factors and outcomes

Overview

Aim 2 will investigate factors associated with implementation and effectiveness outcomes across the 10 clinic sites. This aim is guided by the CFIR,⁴⁷ using implementation constructs and outcomes recommended by Proctor *et al.*⁵⁷ Sources of data for this aim will include process records as well as quantitative and qualitative data from study participants. To prepare for potential scale-up of PN, we must understand the implementation-related factors and outcomes that will maximise its public health impact. Consistent with CFIR, we will assess PN knowledge, attitudes and behaviours; inner and outer setting clinical characteristics; and six key implementation outcomes: *acceptability, adoption, appropriateness, feasibility, reach* and *sustainability*.

Study sample and recruitment

All participants under aim 2 will undergo informed consent (online supplemental file 1). **Patient navigators:** The PNs (N=30: 3 navigators per clinic) will be employed by the study and enrolled as study participants. During the selection process, research personnel will inform potential PNs the purpose of the study and will provide written informed consent to participate in study procedures. At least one PN per clinic must be bilingual (Spanish and English). Ideally, PNs will be individuals from the communities where the clinics are located, but

it is not required. Potential PNs will be identified with the help of the CAB, community member referrals and OCSHCN staff. **Clinic administrators, staff and providers:** At each clinic, one administrator, one staff member and one hearing healthcare provider (N=30) will be invited to participate in Aim 2. They must be aged 18 years or older and be able to speak and read English. **Parent participants:** All parent participants recruited in Aim 1 (N=1120) will also provide Aim 2 data during their baseline and post-test assessments. We will invite and consent a subset of approximately 40 (depending on saturation) parent participants to complete qualitative key informant interviews during their post-test assessment. These parents will represent a combination of urban versus rural communities and non-adherent versus adherent results.

Measures and procedures

Patient navigators: Baseline data collection from PNs will occur immediately following study enrolment and before PN training. At the conclusion of each training, PNs will take an examination to ensure comprehension of critical principles necessary for successful navigation. If the PN fails the examination, the study team will work with the PN to address the incorrect responses and retest the PN. If the PN continues to fail the examination, the PN will be let go from the study. For those who proceed to intervention delivery, PNs will (1) audio-record 10% of PN sessions (with parent permission) to assess fidelity, (2) complete a PN fidelity checklist⁵⁰ following each PN session and (3) maintain process logs detailing time, attendance, frequency and modes of contact with families, and other activities or expenses of PN delivery. Approximately 6 months after completing PN training, all PNs will complete post-test assessments and key informant interviews. PNs will receive compensation for completing the assessments and the semi-structured interview. In each clinic, PN will continue after the 6-month assessment time point until the end of the study. These data will be shared with the CAB quarterly to obtain feedback on PN training and delivery. **Clinic administrators, staff and providers:** Approximately 6 months after crossing to the PN condition, administrators, staff and providers from each clinic will participate in key informant interviews and complete quantitative measures. These participants will receive compensation for completing the assessments. **Parent participants:** At the Aim 1 post-test assessment, all parents will complete a PN satisfaction measure.⁵⁸ Using purposive sampling methodology,^{59 60} parents (N~40) will be selected to complete a 1-hour semi-structured key informant interview exploring parents' experiences with the PN intervention. We will seek a wide variety of parent perspectives from parents of different races, ethnicities, sociodemographic factors and receipt/response to the intervention. Parents will receive compensation for completing the interviews. Implementation outcomes of interest for this study include *acceptability, adoption, appropriateness, feasibility, reach* and *sustainability*. Measures of each implementation outcome are summarised in [table 3](#).

CFIR implementation factors of interest are under four CFIR domains: PN characteristics, inner setting characteristics (structural characteristics, networks and communication, culture, implementation climate and readiness for implementation) and outer setting characteristics (patient needs and resources, cosmopolitanism, peer pressure, external policies and incentives), also summarised in [table 3](#). Patient navigators will complete the following implementation measures 6 months after starting the intervention: Evidence-Based Practice Attitude Scale-50, Perceived Characteristics of Intervention Scale, Intervention Appropriate Measure (IAM), Feasibility of Intervention Measure (FIM), self-efficacy measures and fidelity checklists. Parents will complete the following implementation measures 1 month after the newborn's follow-up appointment: a satisfaction questionnaire and the Acceptability of the Intervention Measure. OCSHCN clinic staff will complete the following implementation measures 6 months after the intervention started at their clinic: IAM, FIM and the Program Sustainability Assessment Tool. Qualitative key informant interviews will be used to complement newly developed quantitative measures, guided by the CFIR interview tool. Key informant interviews with parents, PNs and clinic administrators/staff/providers will be audio-recorded with permission.

Sample size

Because Aim 2 is primarily exploratory, the sample sizes for this study are based on power calculations for the primary effectiveness outcome in Aim 1. With only 10 clinics participating, we may not have adequate power to detect significant associations among implementation factors and outcomes. For our qualitative analyses, the numbers of participants planned for key informant interviews are based on previous work and expectations regarding purposive sampling and the numbers needed to attain saturation.

Statistical analysis

Aim 2 analyses are exploratory in nature and intend to inform potential scale-up and multistate evaluation of implementation of PN, if effective. Aim 2 employs a convergent mixed-methods approach to interpret quantitative and qualitative findings simultaneously.^{61 62} For all quantitative measures, we will obtain descriptive statistics and conduct exploratory bivariable comparisons among the 10 clinics, examining PN, inner and outer setting characteristics that seem to be associated with effectiveness and implementation outcomes. Exploratory analyses of the quantitative data will use marginal (population average interpretation) generalised linear models. Analyses comparing patient-level data between clinics (eg, parent satisfaction) will use the same general GEE approach described in Aim 1 to account for clustering of outcomes in the same clinic. Comparisons of clinic-level data will not require accounting for clustering. For key informant interviews, specific outcomes of interest include themes generated by the study team

Table 3 Measures of implementation outcomes and implementation factors

Implementation outcome/definition	Measure/sample items	Source
Acceptability: Intervention is perceived as agreeable, palatable or satisfactory	<i>Acceptability of the Intervention Measure</i> ⁵⁸ (four items, eg, patient navigation...meets my approval, is appealing to me)	Parents PNs and clinic admins/ staff/providers
Adoption: Use of the intervention	Binary indicator of whether PN was delivered even once at each OCSHCN clinic	PN process records
Appropriateness: Perceived fit, relevance, compatibility of the intervention for a given setting	<i>Intervention Appropriate Measure</i> ⁵⁸ (four items, eg, patient navigation...seems fitting, seems suitable)	Parents PNs and clinic admins/ staff/providers
Feasibility: Extent to which the intervention can be delivered	<i>Feasibility of Intervention Measure</i> ⁵⁸ (four items, eg, patient navigation...seems possible, seems doable)	PNs and clinic admins/ staff/providers
Reach: Proportion of eligible patients receiving the intervention	In each clinic, the ratio of parent–infant dyads receiving any does of PN to the number of all potential dyads referred	PN process records EHDI records
Sustainability: Extent to which the intervention is maintained over time	Following the 6-month assessment time point, number of consecutive months (out of all months in which referrals occurred) in which PN was delivered	PN process records
CFIR domain	Implementation factors/measures	Source
PN characteristics	<ol style="list-style-type: none"> 1. Knowledge: PN examination 2. Attitudes: <i>Evidence-Based Practice Attitude Scale-50</i>⁶⁹ 3. Behaviours: Fidelity checklists, PN session audiotapes 4. Demographics: Age, sex, race, ethnicity, education level, professional experience 	PNs PN process records Qualitative interviews
Inner setting characteristics	<ol style="list-style-type: none"> 1. Number of full time employees (FTEs) employed 2. Patient population size 3. Organisational characteristics: <i>Programme Sustainability Assessment Tool</i>⁷⁰ 	Existing clinical data PNs and clinic admins/ staff/providers
Outer setting characteristics	<ol style="list-style-type: none"> 1. County population size 2. Rurality 3. Appalachian vs non-Appalachian county 4. Number of competing service providers in county 	Existing county data

CFIR, Consolidated Framework for Implementation Research; EHDI, Early Hearing Detection and Intervention; PNs, patient navigators.

regarding PN, inner setting and outer setting factors affecting implementation of the PN intervention in OCSHCN clinics across Kentucky. Digital recordings of key informant interviews will be transcribed in full. Facilitated by use of Atlas.ti,⁶³ the research team will read the text line-by-line to apply codes, which as labels that represent important or recurring themes, develop an initial codebook for each theme, and use concordant and discordant codes to refine and develop a final codebook that will contain the list of codes used for analysis. The results will be analysed for inter-rater reliability. If reliability does not reach or exceed 0.85, the raters will re-examine and revise codes and definitions accordingly until consensus is achieved. Once initial topical coding has been completed, 10% of the sample (ie, two parents and three clinic staff/administrators/ providers) will be invited to participate in a member-checking process to determine whether additional data collection is necessary and to ensure valid inferences are made through coding procedures.⁶⁴ Participants involved in member-checking will receive additional compensation for their time and effort. Following any further corrections to the coding, the investigative team will develop a summative

grid of interview themes. The summative grid of themes and quantitative results will be presented to the CAB. The convergent mixed-methods design of Aim 2 will allow us to simultaneously consider quantitative and qualitative data from multiple perspectives to contextualise and better understand key implementation factors linked with the implementation outcomes of PN.^{61 62} This process will inform the development of implementation strategies designed to enhance implementation outcomes in future scale-ups.

Aim 3: costs

Overview

Aim 3 involves incremental cost-effectiveness analyses in which net costs and net effectiveness of the intervention will be compared with that of standard of care for patients referred to OCSHCN clinics after a failed newborn hearing screen. It is hypothesised that PN *will be cost-effective compared with the standard of care from the perspective of third-party payers*. Results will be expressed as a ratio of differences in observed costs to differences in observed outcomes.

Measures and procedures

Costs: Both direct and indirect costs from PN will be included. Costs from the initial newborn hearing screen will be excluded from analyses as they are incurred for all infants regardless of outcomes. **Direct costs** include the cost of: PN establishment (recruitment and training), programme implementation (office space; PN time, travel, materials and supervision; staff turnover), parent time and travel (travel and time spent receiving diagnostic services), treatment costs (costs of a re-screen or diagnostic appointment), non-medical costs (potential cost savings due to earlier identification) and non-adherence costs for the clinics (no-show appointments). Research activity costs are not included. **Indirect costs** include opportunity costs of time (loss of productivity/wages) for the parent(s). Cost data will be collected and monitored annually throughout the study period for both the intervention and standard of care conditions. Unit programme costs will be documented as Aims 1 and 2 are implemented and sustained over the study period. Unit costs of PN and parent participant travel estimates will use the distance between patient and clinic address/zip code multiplied by the standard General Services Administration (GSA) standard mileage rate and adding any lodging expenses (if applicable). Unit costs of PN time and supervision estimates will use logs applying the hourly pay rate for PNs or supervisor. Parent participant time estimates will use the average wage rate for Kentucky from the Bureau of Labor Statistics. Unit treatment costs estimations will use administrative charge data from OCSHCN clinics. Unit cost savings from early identification will be estimated using data from the literature.⁶⁵ Unit costs of non-adherence estimates will include costs of spent PN time and staff costs from rescheduling. Loss of productivity estimates will use calculated driving estimates to determine time away from work and lost wages for one parent. **Effectiveness:** The *measured outcome of effectiveness* from cost comparison is the proportion of individuals in each group who achieve diagnosis by 3 months of age. We will derive these outcome data using clinic-level non-adherence rates reported monthly by EHDI collected in Aim 1.

Statistical analysis

The cost-effectiveness analysis will follow analytical procedures outlined by Muennig and Bounthavong.⁶⁶ The study will estimate costs associated with an incremental change (measured in percentage points) in effectiveness for each PN modality compared with standard of care. Using effectiveness and implementation data from Aims 1 and 2, probabilities and costs to each terminal event outlined will be applied to determine cost-effectiveness for each outcome. Using TreeAge Pro software,⁶⁷ results will be reported as a ratio of incremental cost to incremental effectiveness for each terminal event, comparing costs and outcomes of the PN group to the standard of care group. An incremental cost-effectiveness ratio (ICER) will be calculated for each alternative to determine the

relative difference in costs associated with a percentage change in effectiveness for each alternative. For any alternative with positive incremental cost but negative incremental effectiveness, an ICER will not be calculated. The calculated ICERs will determine which alternative may produce the best outcome without exceeding stakeholders' threshold of willingness-to-pay. Results will be presented for alternative thresholds and reported using cost-effectiveness acceptability curves to plot the probability of each alternative being cost-effective in relation to different values of willingness-to-pay.⁶⁸ One-way sensitivity analyses will be used to account for uncertainty in our parameter estimates, including the number of participating families, costs of implementation and PN effectiveness. The sensitivity analysis will estimate the expected value of PN given changes in each parameter, using the estimate derived from study data $\pm 20\%$. Probabilistic sensitivity analyses will be used to vary multiple model inputs, including the site of PN, on the expected value of PN. The results of the analysis will be presented to the Kentucky Medicaid programme and other third-party payers to determine interest and ability to reimburse for PN services.

Patient and public involvement

The patient and public involvement in this study is centred around our CAB, described above in the Methods and Analysis section (Community Advisory Board). The CAB is comprised of parents and teachers of DHH children, audiologists, speech-language pathologists, early interventionists who work with babies and toddlers with developmental delays and disabilities, mental health providers, the director of the Kentucky chapter of Hands and Voices and state-level programme administrators. Members of the CAB provided input to the study design, specifically the selection of the stepped-wedge design which ensures that the intervention is available at all clinical study sites at some point during the study. The CAB plays an active role in identifying patient navigators involved in the study, informing the content of patient navigator training, provide resources that are communicated to participants through the patient navigators, advising recruitment methods and intervention delivery and disseminating study results. The CAB meets quarterly with the investigators to assist with study-related activities, such as identification of patient navigators, review and interpret research findings, build connections to colleagues, communities and resources, assist with research barriers and disseminate information back to their communities and settings. Outside of this study, the investigators use the CAB to identify and address issues related to paediatric hearing loss to design studies that meet the communities' needs. Patients and the CAB were not involved in development of the research question or the outcome measures. The content and the burden of the intervention was evaluated by patient members of the community advisory board and provided feedback in the study design phase.

ETHICS AND DISSEMINATION

Ethical considerations

This protocol was approved by the University of Kentucky Institutional Review Board on 12 February 2019 (Protocol #47997). Although all research involving human subjects contains some risk, there are no known serious risks anticipated from participating in this study. One risk presented by this study is a potential breach of confidentiality, including the risk of Protected Health Information being seen by someone unauthorised to do so. The PNs are community-based research personnel who will be carefully trained to help patients by answering questions and guiding them through the diagnostic and therapeutic process of receiving hearing healthcare. In order to prevent miscommunication, they will not be allowed to provide medical advice but instead will facilitate contact with healthcare providers. Implementation measures collected from PNs at baseline and post-test will be considered research measures and will not affect their employment in the study. For clinic administrators, staff members or hearing healthcare providers who participate in Aim 2, the risks associated with breaches of confidentiality primarily involve risks to employment and/or professional relationships. If they share negative perceptions of their agencies, colleagues or organisational leadership, it is possible that a breach of confidentiality could put their working relationships or employment status in jeopardy. These risks will be explicitly detailed in the informed consent process with these participants.

Resource sharing and dissemination plan

Dissemination of clinical trial findings is vital in facilitating translation of research results into practice. Ultimately, scientific rigour, transparency and reproducibility are enhanced by sharing data, methods and documentation. Data will be made widely available as possible while safeguarding the privacy of participants and protecting confidential and proprietary data. Data will be publicly available immediately following the acceptance for publication of the main findings from the final data set. Our study protocol will be published and all instruments will be available to other researchers. Findings will be communicated to our CAB, patients, providers and administrators in Kentucky's OCSHCN clinics and EHDI programme, as well as to state and national EHDI stakeholders. The EHDI Advisory Board will assist with dissemination of results by reviewing the initial findings, identifying resources for extending the reach of the project outside of the OCSHCN clinics, and identifying opportunities for further dissemination. Dissemination will be built into our study methods by collecting information directly from study participants on dissemination methods. Dissemination strategies specific to each target audience will be used incorporating input from our EHDI Advisory Board. Our results will attempt to be shared in high-impact, peer-reviewed journals and national/international professional conferences. The cost-effectiveness results will be essential in the usefulness of practice

and adequate engagement of public and private sector policymakers.

Limitations

Aim 1: Recruitment: The volume of referrals for infant hearing may fluctuate and the timing of such referrals may vary. This will be monitored closely and the team will work with OCSHCN clinics to enrol all eligible subjects efficiently and effectively. **Loss to follow-up:** Those who are non-adherent and lost to follow-up may have been tested at another institution. The intent-to-treat analyses will maintain those participants in the study and assess their adherence using the EHDI database and individual follow-up attempts. **Control group:** Prior to rollout of PN, each clinic will be in a control condition and no individual-level data will be collected. If further individual-level data is needed from the control condition, recruitment of participants may occur during standard of care. **Turnover of PNs:** In the case of turnover, new PN candidates will be identified with the assistance of our CAB members. If there is an inability to recruit or maintain a PN in a particular clinic, then a PN from a neighbouring clinic will cover as contacts are expected to occur by phone. **Alternate study designs:** The stepped-wedge trial design may delay intervention delivery to potential participants and the impact of policy or practice changes that could influence study results. Each design considered for this project has limitations. An RCT with individual randomisation would add significant complexity, increase the likelihood of contamination and require additional personnel and resources to identify and train PNs simultaneously in all 10 clinics. A cluster-randomised trial would have many of the same limitations as a traditional RCT. A typical crossover design would pose logistical and ethical challenges because PN would be rolled out to five clinics simultaneously and reverting to the control condition may be unacceptable. The stepped-wedge design lends itself well to the goals of hybrid effectiveness-implementation studies and allows adequate time at each clinic to enhance the feasibility of a study of this magnitude. With this design, all clinics eventually cross to the PN condition, consistent with the preferences of community partners and stakeholders. To address the possibility of process/procedural changes during the control condition, attention will be given to monitor for policy and practice changes within study sites. **Aims 2 and 3:** Potential Aim 2 limitations include difficulties with recruitment, loss to follow-up and turnover of PNs. Our strategies to overcome these roadblocks are the same as described under Aim 1. In Aim 2, however, each of these roadblocks also relates directly to the implementation factors and outcomes under investigation. Therefore, encountering these problems and exploring their causes will inform our Aim 2 analyses and conclusions and allow for identification of strategies to improve implementation of PN. For Aim 3, if PN does not demonstrate cost-effectiveness, the study results remain useful for identifying target areas for potential cost reduction and could guide others considering implementation of similar PN



programmes. Another potential challenge in Aim 3 is accurate measurement of costs. The study requires reliable data on PN implementation and treatment costs and necessitates detailed monitoring throughout the study period. Despite the challenge of collecting and assuring validity of these data, all partners have committed to documenting and providing the cost data outlined.

Contribution of this study

Participation in the study involves minimal risk for participants in comparison to the potential benefits. Potential benefits include improvement in early detection and intervention of hearing loss in children, increased awareness of hearing loss in children and reduced community and cultural barriers between patients and the health-care system. Previous studies have positively demonstrated the efficacy of patient navigation in decreasing non-adherence to infant hearing testing in a university clinic-based setting. Under the principles of translational science, this study is critical as it seeks to assess the effectiveness, implementation factors and cost of PN to improve delivery of infant hearing healthcare within a larger state-funded clinical environment. This research has potential to inform and change health policy on a state and national level and expand across state lines in future studies. If PN is found to be effective in these settings, future research will investigate (1) the implementation of PN into multiple practice types (university-based, private, state-funded) in a multistate trial, and (2) testing effects of PN on adherence to hearing healthcare treatment. This groundbreaking research in hearing healthcare will be translatable into practice and will contribute to implementation science.

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Contributors MB is the principal investigator and directs all study activities and coordinates team members. Study design: MB, CS, LC, PW, JJ, JS and NS. Analysis design: MB, CS, PW, NS and LC. Protocol generation: MH, MS, BB, AM, MB, CS, LC, PW, JJ, JS and NS. We would like to express members of the communities involved in their study for their guidance in designing and conducting this study.

Competing interests None declared.

Patient and public involvement Patients and/or the public were involved in the design, or conduct, or reporting, or dissemination plans of this research. Refer to the Methods section for further details.

Patient consent for publication Not applicable.

Ethics approval This protocol was approved by the University of Kentucky Institutional Review Board on 12 February 2019 (Protocol #47997). Participants gave informed consent to participate in the study before taking part.

Provenance and peer review Not commissioned; externally peer reviewed.

Data availability statement De-identified participant data from the trial, including quantitative and qualitative data, will be publicly available once the manuscript containing the study's findings is accepted for publication. Other documents that will be made available include the study protocol, informed consent forms and analytical code from the study. Data will be available for 6 years after acceptance for publication of study results. The data can be used for any type of analyses. The shared data can be used for investigators whose proposed use of the data has been approved by an independent review committee identified for this purpose. Proposals to use the data should be directed to matthew.bush@uky.edu.

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