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Authors

Naugle, Kendyl
Stephans, Jihyun
Lazar, Ann
et al.

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Teletherapy to address language disparities in deaf and hard-of-hearing children: study protocol for an inclusive multicentre clinical trial

Kendyl Naugle,[1,2] Jihyun Stephans,[1] Ann Lazar,[3] Joy M Kearns,[1,4] Sarah Coulthurst,[1,4] Kathleen P Tebb,[5] Dylan K Chan [1]

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KN and JS contributed equally.

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For numbered affiliations see end of article.

Correspondence to
Dr Dylan K Chan;
dylan.chan@ucsf.edu

ABSTRACT

Introduction Children who are deaf or hard-of-hearing (DHH) are at risk for speech and language delay. Language outcomes are worse in DHH children from lower socioeconomic backgrounds, due in part to disparities in access to specialised speech-language therapy. Teletherapy may help improve access to this specialised care and close this language gap. Inclusion of diverse DHH children in prospective randomised clinical trials has been challenging but is necessary to address disparities and pursue hearing health equity. Stakeholder input regarding decisions on study design elements, including comparator groups, masking, assessments and compensation, is necessary to design inclusive studies. We have designed an inclusive, equitable comparativeness effectiveness trial to address disparities in paediatric hearing health. The specific aims of the study are to determine the effect of access to and utilisation of speech-language teletherapy in addressing language disparities in low-income children who are DHH.

Methods and analysis After stakeholder input and pilot data collection, we designed a randomised clinical trial and concurrent longitudinal cohort trial to be conducted at four tertiary children's hospitals in the USA. Participants will include 210 DHH children aged 0–27 months. 140 of these children will be from lower income households, who will be randomised 1:1 to receive usual care versus usual care plus access to supplemental speech-language teletherapy. 70 children from higher income households will be simultaneously recruited as a comparison cohort. Primary outcome measure will be the Preschool Language Scales Auditory Comprehension subscale standard score, with additional speech, language, hearing and quality of life validated measures as secondary outcomes.

Ethics and dissemination This study was approved by the Institutional Review Boards of the participating sites: the University of California, San Francisco (19-28356), Rady Children's Hospital (804651) and Seattle Children's Hospital (STUDY00003750). Parents of enrolled children will provide written informed consent for their child's participation. Professional and parent stakeholder groups that have been involved throughout the study design will facilitate dissemination and implementation of study findings via publication and through national and regional organisations.

Trial registration number NCT04928209.

STRENGTHS AND LIMITATIONS OF THIS STUDY

⇒ Stakeholder-informed, patient-centred study design ensures that the study is relevant to populations of interest and will have dissemination potential.
⇒ Intentional inclusion of a diverse population of deaf and hard-of-hearing children, a significant gap in the literature on paediatric hearing healthcare outcomes.
⇒ Comprehensive evaluation of hearing, speech, language and quality of life outcomes.
⇒ Absence of masking imparts some risk of contamination.

INTRODUCTION

Disparities in paediatric hearing healthcare

One in 500 children are born deaf or hard-of-hearing (DHH),[1] putting them at a significant risk for permanent delays in speech and language. These delays can be mitigated by early identification of hearing levels and appropriate intervention before six months of age,[2–7] which includes clinical management with hearing aids (HA) and cochlear implantation (CI)[8–11] and early intervention (EI) services, including speech and language therapy.[12] Numerous studies have demonstrated disparities in access to care, service utilisation and paediatric hearing health outcomes[13]: low socioeconomic status, underinsurance status, living in a rural area and coming from an underserved racial/ethnic group contribute to significant delays in identifying and intervening for hearing, including CI, and worsened auditory outcomes after CI.[14–17] The factors underlying these disparities among children who are DHH, particularly in speech and language, are complex but have been proposed to relate to differences in access to care.[13] In particular, children who are DHH have very specific needs that benefit from specialised support by a provider with experience in aural habilitation[18]; however,

equal access to specialised, linguistically matched EI services is poor. Inequitable access to specialised hearing healthcare, non-English home language, low-income status and public insurance are also risk factors for decreased utilisation of therapy services and delays in auditory and language function.[19–21]

EI and teletherapy for children who are DHH

EI services to support language development for children who are DHH are a collaboration between the medical and educational systems.[22 23] For children who are DHH, EI services are provided initially through an Individualised Family Service Plan,[12] and then at age three transition to an Individualised Education Programme or 504 Plan, together with their clinical care team. Exact services vary considerably within and across states, giving rise to considerable inequity in services depending on locally available resources and the ability of families to access them.

Teletherapy (TT) has been proposed to reduce disparities related to geographical access to specialised care.[24] TT sessions are virtual visits that use a parent coaching model to help parents develop their abilities to interact with their children in ways that support their child's development. In small randomised studies with 30–50 children, TT improved language outcomes in DHH children compared with controls, who received usual, in-person, care.[25 26] These studies demonstrate the efficacy of TT in a general population of children who are DHH; however, there is a critical need for research with populations who experience disparities.

Addressing disparities in paediatric hearing healthcare: specific aims and hypotheses

A child who is DHH is characterised both by demographic and clinical attributes, both of which contribute to hearing health outcomes (figure 1). Despite the broad and highly significant sociodemographic disparities in hearing outcomes for DHH children, previous large prospective studies on language outcomes in DHH children have focused primarily on white, English-speaking, well-educated, affluent families.[9 27 28] There is a critical need for rigorous studies that are intentionally inclusive of more diverse families, explicitly account for these sociodemographic factors, and are designed to be equitable and ethical to the communities that are engaged. To address the specific evidence gap in TT as well as this broader gap in inclusive research engaging diverse populations of DHH children, we have designed a prospective randomised clinical trial to test the effectiveness of access to supplemental specialised speech-language therapy to address income-based language disparities in DHH children.

The overall aim of this study is to evaluate the effectiveness of supplemental speech-language TT to address income disparities in language outcomes for DHH infants and toddlers. We designed two studies: (1) a randomised clinical trial study and (2) a concurrent prospective observational cohort study. The study design was informed by patient and provider stakeholders to optimise the ability to recruit, enrol and retain a diverse group of study participants. Our specific aims and hypotheses follow.

Specific aim 1: evaluate the effectiveness of access to supplemental TT for children who are DHH to address income-based disparities in language outcomes

Specific aim 1A

We will perform a randomised clinical trial (figure 2A) to compare language outcomes at 18 months between low-income children receiving usual care (UC) versus UC plus access to supplemental speech-language TT.

Specific aim 1B

In addition to comparing language outcomes between the two randomised groups of lower income children, we will perform a longitudinal prospective observational cohort study, in which we will concurrently accrue higher income patients receiving UC and measure the language gap between higher and lower income children. We will then evaluate whether TT can close the language outcomes gap between lower and higher income families. We hypothesise that supplemental access to specialised speech-language TT will improve language outcomes in lower income DHH children and close the language

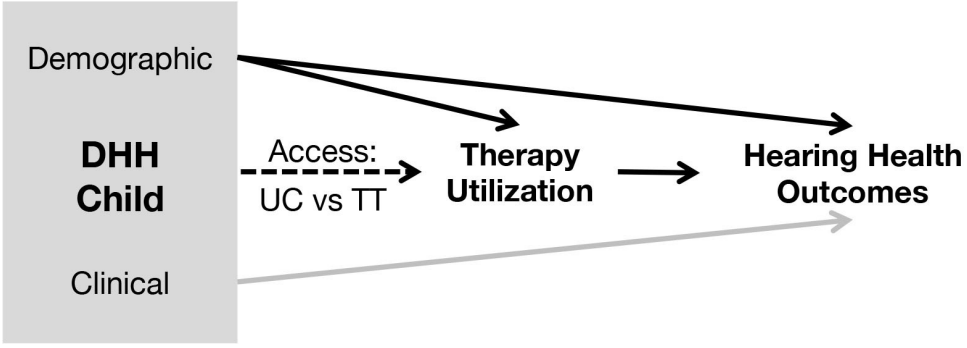

Figure 1 Theoretical model. A deaf or hard-of-hearing (DHH) child is characterised both by clinical and demographic attributes, both of which contribute directly to hearing health outcomes. Demographic factors also affect outcomes indirectly, mediated by therapy utilisation. This study seeks to evaluate whether directly intervening on access to care, by providing access to teletherapy (TT) versus usual care (UC), can improve hearing health outcomes through this utilisation pathway.

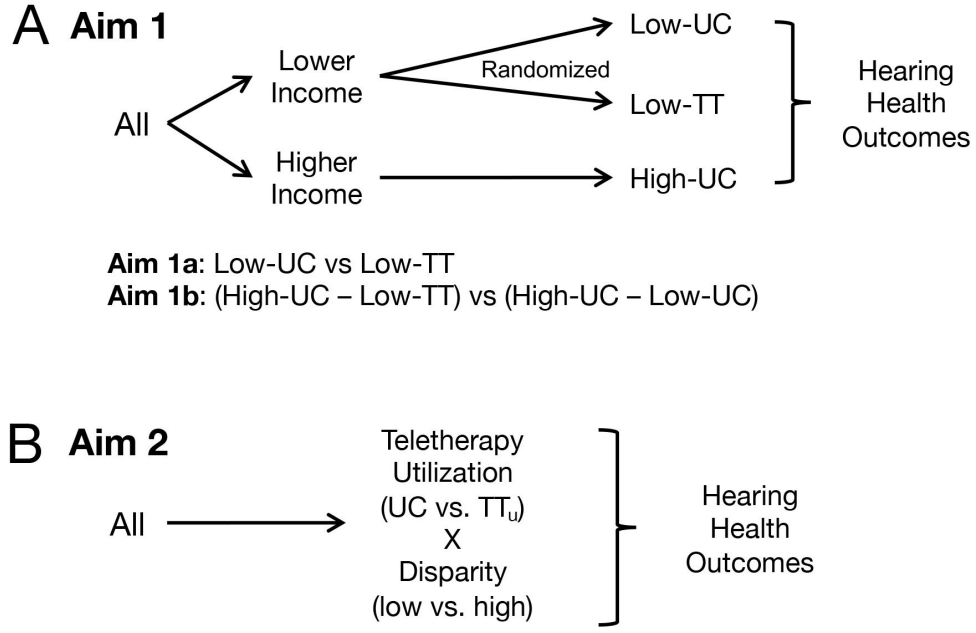

Figure 2 Study design. (A) In specific aim 1, deaf or hard-of-hearing (DHH) children are identified as lower or higher income. Lower income children are randomised to receive usual care (low-UC) or usual care plus access to supplemental speech-language teletherapy (low-TT). Higher income children receive UC and are followed as a prospective comparator cohort (high-UC). Hearing health outcomes are measured in all children identically. In specific aim 1A, a randomised clinical trial, we test differences in outcomes between low-UC and low-TT groups. In specific aim 1B, a prospective longitudinal observational cohort study, we test differences in the outcomes gap between higher income and lower income groups. (B) In specific aim 2, data obtained from this study will be used secondarily to perform a heterogeneity of treatment effects analysis on the interaction between teletherapy utilisation (TT_u) and sociodemographic disparities (dichotomised as low (greater risk for worse outcomes) and high (lesser risk for worse outcomes)), as they relate to the same hearing health outcomes.

outcomes gap between higher and lower income DHH children.

Specific aim 2: evaluate the effectiveness of utilisation of supplemental TT for children who are DHH to address sociodemographic disparities in language outcomes

Whereas specific aim 1 measures the effectiveness of access to supplemental speech-language TT, actual utilisation of TT may vary considerably. Therefore, we will perform a secondary analysis of data accrued from aim 1 to evaluate the effectiveness of utilisation of TT in children who are DHH experiencing sociodemographic disparities (figure 2B). We hypothesise that utilisation of speech-language with be associated with improved language outcomes in DHH children experiencing each of these sociodemographic disparities.

METHODS AND ANALYSIS

Study population and setting

We have designed a randomised, unblinded, multicentre trial with a concurrent observational cohort study to address our specific aims. The randomised trial will enrol children from lower income households, defined as household income less than 266% of the federal poverty level, the eligibility threshold for Medi-Cal for children. These lower income children will be randomised to UC (low-UC), or to UC plus access to supplemental TT (low-TT). In parallel with this randomised trial for lower income children, we will simultaneously, prospectively recruit a comparator cohort of higher income DHH children receiving UC (high-UC). All children will be followed prospectively according to the assessment schedule described below.

Overall, the target enrolment will be 210 children, with 70 recruited for the high-UC group and 140 for the lower income group, of which 70 will be randomised each to low-TT and low-UC. Participants will be recruited from four tertiary academic paediatric centres. Study recruitment began on 1 July 2021; enrolment was completed on 30 June 2024; and data collection is anticipated to be completed on 31 December 2025.

Study interventions

All participants will receive UC, defined as the standard care that a child who is DHH at one of the enrolling centres would receive through the managing clinical site, Part C of the Individuals with Disabilities Education Act EI programme, and additional sources to which they have access, not including supplemental specialised speech-language TT. All forms of hearing-directed care received through UC will be quantified for all children.

Access to an 18-month course of TT will be provided as a supplement to UC for children randomised to the low-TT group. Technology needs assessments will be performed and a device and/or connectivity plan will be provided if applicable. All TT providers will be licensed

speech-language pathologists (SLPs) and/or credentialed teachers of the deaf (TODs) with specialised training to work with children who are DHH and who are fluent in the child's primary home language (English or Spanish). The exact details of the therapy course will be determined based on individual family needs according to the teletherapist's standard clinical care. Utilisation of TT is not a requirement for remaining in the study. Additionally, if a study participant in the low-UC or high-UC group receives supplemental speech-language TT outside of the study after enrolment, they may remain in the study, as this TT is considered part of their UC.

Stakeholder engagement

Parents of DHH children and professional stakeholders were engaged in the study design and will continue to be engaged in the ongoing study and dissemination/implementation activities through biannual meetings of Professional and Parent Advisory Groups. Professional stakeholders include individuals across the USA, representing SLPs, TODs, audiologists, educators, researchers, policymakers, payors and social workers. We have also engaged with 12 English and Spanish-speaking sets of parents of DHH children in all aspects of this study. Stakeholders have informed the choice of primary and secondary outcome measures, refined definitions of comparators and interventions, helped identify clinical and sociodemographic covariates and confirmed the use of a non-blinded randomised study design.

Study procedures

Inclusion criteria

Children are eligible to participate in the study if they are 0–27 months old; from English or Spanish-speaking families; have intention for developing spoken language; have no medical conditions that would significantly preclude the development of spoken language; and have permanent hearing loss (HL) determined by auditory brainstem response (ABR) or behavioural audiometry as follows:

1. Bilateral sensorineural, mixed or permanent conductive HL with better ear pure tone average (PTA) of 0.5–4 kHz>20 dB.
2. Single-sided deafness (unilateral sensorineural hearing loss (SNHL) with PTA>70 dB).
3. Unilateral complete aural atresia or partial aural atresia with air conduction PTA of >60 dB HL.
4. Bilateral auditory neuropathy spectrum disorder (ANSD), as determined by ABR.

Children must also have adequate access to sound to participate in the study; children with bilateral PTA >20 dB must be fit with a clinically appropriate hearing device. T_0 is defined as the date after enrolment when 'access to sound' is achieved; for children with access to sound already at time of enrolment, T_0=date of enrolment.

Exclusion criteria

Children will be excluded from study participation if the family does not have intention to pursue any listening and spoken language for their child; the child has known moderate to severe global developmental delay, or syndrome typically associated with this level of delay (such as Down syndrome) at time of enrolment; the child is receiving supplemental speech-language TT through a clinical provider outside of this study at time of enrolment; or the child has poor prognosis for access to sound, defined as bilateral severe-to-profound SNHL plus either medical contraindication for CI or temporal bone abnormalities associated with poor CI outcomes, including cochlear nerve deficiency.

Study timeline

On informed consent obtained by study personnel (online supplemental data 1), group assignment and T_0 will be performed based on the participant's enrolment site, hearing level and hearing device status, age, home language, household income and household size, determined based on parent report and medical record review. Baseline evaluation will be performed within three months of T_0, and nine and 18-month evaluations within a window±three months around the target dates.

Randomisation

Lower income children will be randomised 1:1 to low-UC or low-TT using an automated permuted block randomisation. The randomisation will be performed with stratification for hearing level and to study site as follows:

1. Better ear hearing level (better ear PTA (0.5–4 kHz)): (1) 0–20 dB HL; (2) 21–70 dB HL; (3) >70 dB HL or bilateral ANSD.
2. Site of enrolment (UCSF Benioff Children's Hospital, San Francisco; UCSF Benioff Children's Hospital, Oakland; Rady Children's Hospital, San Diego; and Seattle Children's Hospital).

Data collection

The following forms will be used to collect baseline demographic and clinical data (online supplemental data 2):

1. Family demographics: gender, race, ethnicity, primary and other home languages, number of adults and children in household, birth order, siblings with HL, household annual income, highest education level of parents, country of birth, zip code.
2. Hearing history: screening, identification and intervention for HL, medical and developmental comorbidities.
3. Clinical data: hearing levels and aetiological testing results.

Study assessments will be performed at baseline, nine months and 18 months after T_0, and consist of formal, validated, outcome measures (table 1) together with measurements of intervention utilisation. The primary study endpoint is Preschool Language Scales, Fifth Edition (PLS-5)[29] Auditory Comprehension (AC) subscale standard score at 18 months. All other measures, including nine-month PLS-5 AC, and nine and 18-month PLS-5 Expressive Communication subscale and

Table 1 Outcome measures

Assessment tool	Domain	Description	Ages	Administered by	Secondary outcomes
Preschool Language Scales, Fifth Edition (PLS-5)[29]	Language	Standardised, norm-referenced measure of receptive and expressive language skills. Subscales: ► Auditory Comprehension (AC) ► Expressive Communication (EC) ► Total Language (TL)	0–7:11	SLP	**1–3**: PLS-5 AC SS (9 months, changes from baseline to 9 and 18 months) **4–7**: PLS-5 EC SS* **8–11**: PLS-5 TL SS*
Receptive Expressive Emergent Language Test–Fourth Edition (REEL-4)[32]	Language	Standardised, norm-referenced measure of verbal and non-verbal communication skills, including expressive and receptive language.	0–3:0	SLP	**12–15**: REEL-4 (receptive) SS* **16–19**: REEL-4 (expressive) SS* **20–23**: REEL-4 (language) SS*
Goldman-Fristoe Test of Articulation–Third Edition (GFTA-3)[33]	Speech	Standardised, norm-referenced measure of the articulation of consonant sounds in Standard American English or Spanish.	2:0–21:11	SLP	**24–27**: GFTA-3 (sounds in words) SS* **28–31**: GFTA-3 (sounds in sentences) SS*
MacArthur Bates Communicative Development Index (MBCDI)[34]	Vocabulary	Standardised, norm-referenced measure of young children's language and communication skills.	0:8–3:9	Caregiver	**38–41**: MBCDI age equivalent*
LittlEars Auditory Questionnaire (LEAQ)[35]	Auditory skills	Validated measure of auditory function. Critical versus normal limits.	0–2:0	Caregiver	**32–33**: LEAQ score at 9 and 18 months
Parent Evaluation of Aural/Oral Performance of Children (PEACH)[36]	Auditory skills	Validated measure of effectiveness of a child's use of hearing in real-world environments.	0–5:0	Caregiver	**34–37**: PEACH total score*
Hearing-Related Infant/Toddler and Parent Quality of Life (HIP-QL)[37]	Quality of life	Validated measure of dyad quality of life of a DHH child and their caregivers.	0–3:6	Caregiver	**42–45**: HEAR-QL total score*
Scale of Parental Self-Efficacy (SPISE)[38]	Self-efficacy	Validated measure of caregiver comfort level in supporting their DHH child's needs and communication development. A subset of questions is used for this study to reduce survey burden.	0–18:0	Caregiver	**46–49**: SPISE total score*
Family Outcomes Survey (FOS)[39]	Quality of life	Validated measure of caregiver perceptions of both caregiver and EI provider's ability to support their child's needs. Part B subscale, 'Communicating your child's needs'.	0–18:0	Caregiver	**50–53**: FOS total score*

All validated outcome measures administered at each assessment timepoint (baseline, 9 months and 18 months) are shown. The primary outcome is PLS-5 AC standard score (SS) at the 18-month timepoint. All other secondary outcomes are indicated in the rightmost column and will be measured at the indicated timepoints.
*Outcome will be measured at 9 months, 18 months, change from baseline to 9 months and change from baseline to 18 months.
DHH, deaf or hard of hearing; EI, early intervention; SLP, speech-language pathologist.

Total Language scores, are secondary endpoints. These outcome measures, which are all norm-referenced, validated measures available in English and Spanish, were selected to provide a comprehensive assessment of the auditory, speech and language development of DHH children. Assessments will only be administered to children within the validated chronological age range at time of the assessment. They will be conducted by an SLP and TOD, or completed by the participant's caregiver, with assistance as necessary from a study coordinator, in the parents' native home language, making use of a professional medical interpreter when necessary.

The interventions that we are testing (UC and TT) are complex. Therefore, we have implemented the following measurements (online supplemental data 3) aimed at assessing the type, amount, quality and parent perception of interventions:

1. Interval history—family: access, utilisation and parent perception of therapy services.
2. Interval history—clinical: hearing interventions, including aided audiological and device utilisation measures.
3. TT enrolment (low-TT subjects only): teletherapist characteristics.
4. TT utilisation (low-TT subjects only): TT session characteristics.

Baseline and intervention data will be obtained through a combination of parent report and electronic medical record review. When there is a discrepancy, the electronic medical record will be used. We will document this and assess whether any patterns occur relating to sociodemographic factors.

Incentive structure

Families will receive a $150 VISA gift card for their participation in each of the three assessments, as well as a $50 gas card for assessments completed in person, for a maximum of $600.

Withdrawal

Family request to withdraw from the study after randomisation, failure to complete a baseline assessment within three months of enrolment, failure to undergo CI or HA fitting within six months of enrolment (for children with bilateral >20 dB PTA) or failure to complete the 18-month PLS-5 within six months will constitute non-completion of the study. For aim 1A, withdrawal occurs after randomisation. Randomisation occurs after T_0 or baseline assessment.

Analyses

For aim 1A, we will test the hypothesis. Ho: $\theta1=0$ vs HA: $\theta1\neq0$, where $\theta1$ is the difference in PLS-AC between low-TT and low-UC at 18 months. For aim 1B, we will test Ho: $\theta2=0$ vs HA: $\theta2\neq0$, where $\theta2$ is the difference at 18 months in AC between low-income children receiving TT and higher income children receiving UC; and hypotheses III: Ho: $\theta3=0$ vs HA: $\theta3\neq0$, where $\theta3$ is the difference at 18 months in AC between low-income children receiving UC and higher income children receiving UC. Sample size was determined based on preliminary pilot data (table 2). For aim 1A, a total of 140 lower income children will be randomised 1:1 to low-TT or low-UC. For aim 1B, an additional 70 high-UC children will be recruited, for a total sample size of 210. These sample sizes were adjusted for drop-out of 20%. For specific aim 2, the association between AC, primary outcome and each of six sociodemographic disparities (table 3) at 18 months will be assessed. Heterogeneous treatment effects (HTEs) will be assessed using the standard HTE approach, an interaction between utilisation group and each disparity. A detailed analytical plan is provided in online supplemental data 4. We designed the study in accordance with the Patient-Centered Outcomes Research Institute Methodology Standards.[30]

Patient and public involvement

Parents of DHH children were directly involved in study design, including defining the primary outcome of the study, and will continue to be involved in study conduct and dissemination/implementation (see the 'Stakeholder engagement' section). As this is an open study, all study assessments performed on a study participant will be shared in their entirety with those patients' parents and their clinical care teams. At the end of the study, the final study results will be shared with all study participants.

ETHICS AND DISSEMINATION

This study protocol, Version 1.6 (21 September 2023), was approved by the Institutional Review Boards at UCSF (19-28356), RCH (804651) and SCH (STUDY00003750), and registered at ClinicalTrials.gov (NCT04928209). Protocol modifications are communicated to each institution and the trial funder. As this is a minimal risk trial with behavioural assessments and an intervention that is within standard clinical care, adverse event reporting and monitoring will not be performed, and a Data Safety and Monitoring Board will not be convened. A Data Safety and Monitoring Plan has been developed that addresses subject accrual and protocol compliance, data acquisition and completeness, and confidentiality and privacy (online supplemental data 5). Written informed consent will be obtained from a parent or legal guardian of each study participant. Assent will not be obtained, as all study participants will be 0–27 months of age at enrolment. Dissemination of study findings will be performed by publication in peer-reviewed journals and presentation at national conferences of the primary and secondary findings from this study. Implementation of these findings will be supported by members of the Professional and Parent Advisory Groups, which include leadership in local, regional and national organisations that are directly engaged in policy, training, and funding of speech-language TT. The full study protocol, participant-level deidentified dataset and statistical code will be made publicly available on request. We used the Standard Protocol Items: Recommendations for Interventional Trials checklist when writing our report.[31]

DISCUSSION

Families of DHH children experience varied challenges, especially those who are from lower income backgrounds, speak a language other than English, live in rural areas or are publicly insured.[13] Access to EI and speech-language therapy are the cornerstone of DHH children's care, and speech and language therapy offered virtually

Table 2 Pilot data

	n	AC (SS)	P value	EC (SS)	P value	TL (SS)	P value
All	38	84.4±21.9	N/A	83.2±18.8	N/A	81.6±20.2	N/A
Insurance							
Commercial	13	97.1±16.7	0.0072	92.1±16.1	0.0343	95.0±17.9	0.0026
Public	25	77.6±21.5		78.6±18.8		75.0±18.1	
Language							
English	24	90.4±20.8	0.0298	86.9±18.1	0.1126	87.0±19.3	0.0263
LOTE	14	74.6±20.7		76.8±19.1		72.1±18.8	
ACI							
≤3	14	92.5±15.4	0.0005	86.9±15.0	0.0093	89.5±16.5	0.0016
>3	13	67.2±17.3		69.6±16.9		66.5±17.2	
Teletherapy pre-post							
Intake	11	75.2±13.1	0.0112	79.6±17.5	0.022	75.2±18.6	0.0268
18 months	11	95.5±22.5		94.5±16.7		91.1±17.5	
Teletherapy							
TT	16	91.8±21.3	0.0849	91.8±16.2	0.0147	88.7±17.2	0.0671
No TT	22	79.4±21.3		77.0±18.5		76.6±21.0	
Public insurance only							
TT	12	87.0±21.3	0.0415	89.2±15.4	0.004	83.8±14.6	0.0204
No TT	13	69.6±19.0		68.8±16.4		67.5±17.8	
LOTE only							
TT	6	81.5±20.6	0.302	88.5±12.7	0.0412	80.2±16.6	0.205
No TT	8	69.5±20.6		68.0±18.9		67.0±19.3	
ACI>3 only							
TT	4	83.8±21.2	0.0137	84.0±16.5	0.0334	82.8±19.9	0.015
No TT	9	59.9±9.2		63.2±13.3		59.2±10.2	

Language outcomes (Preschool Language Scales, Fifth Edition (PLS-5) Auditory Comprehension (AC), Expressive Communication (EC) and Total Language (TL) subscale standard scores (SS)) for DHH children with the indicated sociodemographic characteristics are shown. P value for comparisons using unpaired, two-tailed, independent t-tests.
ACI, Access Challenge Index; DHH, deaf or hard-of-hearing; LOTE, language other than English; TT, teletherapy.

has been shown to be similarly efficacious to in-person services.[25 26] This comparative effectiveness study aims to assess whether providing access to TT for lower income families will improve outcomes and reduce linguistic, income, insurance and access disparities. Demonstration of the real-world effectiveness of supplemental

Table 3 Sociodemographic disparities

Factor	Dichotomisation	Lower risk	Higher risk	References
Income	±266% FPL	Higher income	Lower income	16
Insurance	Public versus commercial/mixed	Commercial/mixed	Public	17 19 20
ACI	±Median	Lower ACI	Higher ACI	40
Distance to hearing healthcare centre	±Median	Closer	Farther	13 15
Home language	English versus LOTE	English	LOTE	19 20
Highest level of parental education	±High school completion	More than high school completion	High school completion or less	41

Sociodemographic factors to be considered as confounders for prospective cohort trial (specific aim 1B) and HTE analysis (specific aim 2).
ACI, Access Challenge Index; FPL, federal poverty level; HTE, heterogeneous treatment effect; LOTE, language other than English.

speech-language TT may provide needed evidence to support improved and more widespread access to this resource for DHH children.

The current study was intentionally designed to be inclusive of more diverse participants than previous studies.[9 27 28] Primary outcome, study assessments and schedule, and recruitment, enrolment and retention plans were developed on advisement from a large and multidisciplinary group of stakeholders, including parents of DHH children who were representative of the target study population. Specific study details intended to enhance inclusion include: (1) selection of language as the primary outcome measure; (2) targeted assessment battery, selected to be relevant for clinical care and acceptable in length; (3) absence of masking between groups as well as with the clinical care team, so that study assessments can be used explicitly for clinical care; (4) intent-to-treat design in which the intervention is access, rather than utilisation, of supplemental speech-language TT; and (5) compensation plan designed to incentivise participants and overlap between research coordinator and family navigation services to coordinate clinical and research activities. Overall, the significant overlap and alignment between study activities and direct clinical care was determined across all stakeholders to be essential to enable robust and equitable study participation.

Limitations of the study include risk of contamination due to absence of masking and sharing of research assessments with clinical care teams, which was necessary to support the inclusion of diverse participants. Utilisation of a randomised study design will help mitigate this limitation. Despite our efforts to retain study participants, the study population is at significant risk for loss to follow-up. We have accounted for a large (20%) drop-out rate to buffer against this threat to study power. Finally, the sample size calculation was based on retrospective pilot data comparing DHH children who used supplemental speech-language TT. In the pilot data, the effect of the TT intervention was significant; however, utilisation of TT is subject to significant bias even among the groups experiencing disparities—these families who successfully used TT may be biased towards those who would have done well anyway regardless of their access to TT. Therefore, the actual effect size in the proposed prospective randomised clinical trial for access to TT may be smaller than that seen in the pilot data for utilisation of TT.

Despite these limitations, this study addresses a major disparity in paediatric hearing health by testing the effectiveness of TT to improve access to specialised care and language outcomes. Beyond this specific aim, successful completion of a clinical trial that is inclusive of a diverse population of families of DHH children will provide a valuable template for similarly equitable research on paediatric hearing health.

Author affiliations
[1]Department of Otolaryngology-Head and Neck Surgery, University of California San Francisco, San Francisco, California, USA
[2]School of Medicine, University of California San Diego, La Jolla, California, USA
[3]Department of Epidemiology and Biostatistics, University of California San Francisco, San Francisco, California, USA
[4]Department of Audiology, Benioff Children's Hospital, Oakland, California, USA
[5]Department of Pediatrics, Division of Adolescent and Young Adult Medicine, University of California San Francisco, San Francisco, California, USA

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ORCID iD
Dylan K Chan http://orcid.org/0000-0003-0836-3730

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