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Research in context

Evidence before this study

School screening is an accepted public health intervention for early identification and treatment of childhood hearing loss. However, there are multiple gaps in the evidence around school screening. Loss to follow-up is a major concern in screening programmes globally, and the potential role of telehealth to improve follow-up from school screening has not been evaluated. There is no consensus on screening protocols, and data are not available on the optimal protocol to identify children with infection-related hearing loss common in low-resource settings. We searched PubMed using the terms, "telehealth or telemedicine," "school screening," "rural," and "specialist." We found no clinical trials evaluating telehealth as an intervention to address loss to follow-up from screening or to improve access to specialist care in rural settings. New Zealand, which has a high prevalence of infection-related hearing loss in the Maori population, has incorporated tympanometry into national screening protocols. However, these protocols are only applied to preschool and new entrant school children. A screening protocol incorporating pure-tone screening and tympanometry previously tested in kindergarten and first grade students in British Columbia, Canada, where infection-related hearing loss is common, was selected for this trial in Alaska.

Added value of this study

To our knowledge, we report the first randomised trial to demonstrate that telemedicine can reduce rural health disparities in access to specialty care, with a mean time to follow-up that is 17·6-times faster (95% CI 6·8–45·3; p=0·002) in communities randomised to telemedicine specialty referral compared with standard primary care referral. This study also demonstrates that mobile health screening with tympanometry outperforms the school screen in a K-12 population (aged 4–21 years) with high prevalence of ear infections. Both telemedicine specialty referral and mobile health screening with tympanometry were found to be cost-effective.

Implications of all the available evidence

Rural schools represent an essential access point for preventive services for children worldwide, yet loss to follow-up from school screening programmes and scarcity of specialists exacerbate barriers to care in rural communities. Telemedicine specialty referral can improve follow-up and reduce time to follow-up after school screening in rural communities. This model could be applicable to other preventable health conditions and represents an intervention that can promote access to specialists to reduce rural health disparities. Mobile health screening with tympanometry can improve identification of childhood hearing loss in populations where infection-related aetiologies are common. Additional research is needed to test implementation of these interventions in low-resource settings globally.

geographical barriers to specialty care through a telehealth network developed nearly 20 years before COVID-19 generated momentum for telehealth.[13] Validation studies of ear and hearing telemedicine consultations in rural Alaska demonstrated that medical and surgical decision making were equivalent to in-person examinations, and telemedicine reduced the waiting times for specialist appointments by 8 weeks.[14–16] Therefore, telemedicine has become an established mode of clinical care in rural Alaska.

Despite being commonly used for clinical care, telemedicine has never been used for preventive services in Alaska. We report the results of the Hearing Norton Sound randomised controlled trial, which evaluated a novel telemedicine specialty referral pathway for school hearing screening to improve timely identification of childhood hearing loss. This trial addresses several key knowledge gaps. Rigorous telemedicine studies are few, particularly randomised controlled trials that provide high-quality evidence that telemedicine can increase access to care and reduce rural health disparities. Screening programmes worldwide, including school hearing screening, have substantial loss to follow-up, and the potential of telemedicine to ameliorate this problem has not been evaluated.[11] Although follow-up from school screening in rural Alaska has anecdotally been reported to be low, no studies have previously been done to quantify follow-up in this population. We hypothesised that telemedicine specialty referral would improve time to follow-up compared with standard primary care referral, thereby reducing a key rural health disparity by improving access to specialty care. There is minimal evidence on the accuracy of school hearing screening protocols, particularly in populations with high prevalence of ear infections.[11] Our secondary objective was to determine the optimal screening methodology in this population. We hypothesised that mobile health screening with tympanometry would be more sensitive than the school screen because of increased capacity to identify infection-related pathology with tympanometry.[17,18] Additional secondary hypotheses were that prevalence of hearing loss would be reduced, hearing-related quality of life would improve, school performance would improve, and that mobile health screening and specialty telemedicine referral would be cost-effective.

Methods

Study design and participants

We conducted a parallel, two-arm, cluster-randomised controlled trial over two academic years between Oct 10, 2017, and March 28, 2019, in the Bering Strait region of northwest Alaska, USA, which spans

Figure : Map of study area (A) and referral pathways (B and C)
*Excludes Nome City school district. Reproduced with permission; copyright 2021, Duke University.

23000 square miles and includes 15 small rural communities and the regional hub of Nome, all accessible only by plane (figure 1A). Each community has a school enrolling children from age 4 to 21 years. A cluster-randomised design was chosen because the referral intervention was designed for communities as a whole. This region was selected because of the high burden of infection-related hearing loss and the presence of well established telemedicine infrastructure in daily use for clinical care. More than 95% of these communities' residents are Alaska Native, primarily of Yup'ik, Iñupiaq, and Siberian Yupik heritage. The Bering Strait School District serves all 15 rural communities. The sole source of health care is Norton Sound Health Corporation (NSHC), a tribal health organisation that is part of the Alaska Tribal Health System, which provides health-care services to the Alaska Native population on the basis of treaties previously signed with the US Government. In rural areas such as the Bering Strait region, where the Tribal Health System is the sole health-care entity, all community members are eligible for services. Local health care is provided by community health aide/practitioners, supported by the NSHC regional hospital in Nome and the tribal tertiary hospital for the state, Alaska Native Medical Center, Anchorage, USA.[10] Telemedicine infrastructure in village health clinics is routinely used for clinical management of ear and hearing problems, using asynchronous telemedicine consultations from local community health aide/practitioners to audiologists in Nome and otolaryngologists in Anchorage.

Trial protocols were published, and the design was informed by community guidance.[20–22] Screening was conducted annually in accordance with the Alaska state mandate. Written informed consent was obtained from the parent or guardian of participants, and child assent was also required. All children (grades K-12; aged 4–21 years) enrolled in the 15 schools who were present on hearing screening day with appropriate consent were eligible for inclusion in the study. All eligible children could participate even if they were eligible in only one of the academic years. Institutional review boards of Alaska Area, NSHC, and Duke University approved the trial, and the review boards of Alaska Area and NSHC represented Alaska Native tribal interests.

Randomisation and masking

Communities were randomised to telemedicine specialty referral (intervention) or standard primary care referral (control) for school hearing screening. Randomisation of communities occurred within four strata, which were based on a combination of location (ie, north, middle, south, and island) and school size (ie, more than or less than 100 students). Randomised referral assignments for communities were computer generated by one of the study statisticians (N-YW), using SAS (version 9.4). Participants were masked to group allocation until screening day, after which time masking referral assignments was not possible. Assessors remained masked throughout data collection. Study team members who provided clinical care did not read trial-related telemedicine referrals during the study period. Specialists consulting on telemedicine referrals and study team members performing medical record abstraction were masked to group allocation. Study team members performing screening or audiometric evaluations were masked to the other results during screening. Statisticians were masked to group allocation during data analysis.

Procedures

School hearing screening occurs annually in the Bering Strait School District in accordance with the Alaska mandate. Based on community feedback that all children should derive benefit, screening and audiometric protocols were not randomised.[22] All children (K–12) underwent the school hearing screen, mobile health screen plus tympanometry, and a gold standard audiometric evaluation.

Descriptions of screening and audiometric protocols have been previously published.[20] The school hearing screening consisted of distortion product otoacoustic emission screening at 2, 3, 4, and 5 kHz (Natus/Bio-Logic, USA) using pass or refer criteria, in which three of four frequencies must meet predetermined response conditions. This automated protocol did not include rescreening. Teachers performed the school screen, as per standard practice. Mobile health screening included pure tones at 1, 2, and 4 kHz at 20 dB, with a validated mobile health smartphone-based screen (hearX Group, South Africa) and tympanometry to assess the middle ear (Otometrics, Denmark). If a child did not respond to a tone, rescreen at that frequency was performed. Absence of a response to a tone at any frequency in either ear or a type B (flat) or negative pressure less than –200 decapascal (daPa) tympanogram generated a referral. Mobile health plus tympanometry screening was performed by study staff who were not audiologists. Gold standard audiometric assessment was included for all children to assess sensitivity and specificity of screening protocols using an air-conduction and bone-conduction audiogram at 0·5, 1, 2, and 4 kHz with a validated tablet-based audiometer (Shoebox, Clearwater Clinical, Canada), diagnostic tympanometry (Otometrics, Denmark), and digital otoscopy (Otocam, Otometrics, Denmark). Referral was generated for pure-tone average more than 25 dB or a threshold more than 30 dB at a single frequency, type B or negative pressure less than –200 daPa tympanogram, or findings on otoscopy (eg, occluding cerumen, retraction, effusion, acute otitis media, otorrhea, perforation, patent or plugged tube, external otitis, or foreign body). Audiologists performed the audiometric evaluation.

Children who screened positive for possible hearing loss or ear disease on either screening protocol or audiometric assessment required referral. The study team generated a referral list and transferred this list to school leadership to coordinate follow-up according to each community's randomised referral assignment. Referrals in both groups included the child's name and the affected ear (left, right, or both), as per standard practice in rural Alaska. Transfer of follow-up coordination to the schools was incorporated into the study design to increase the generalisability of the findings.

The telemedicine specialty referral intervention adapted existing telemedicine infrastructure in village clinics (figure 1B). Typical telemedicine workflow for clinical care, including documentation in two electronic health systems, was streamlined from 60–90 min to 5–10 min for the intervention by reducing documentation to a single electronic system with core billing requirements maintained to facilitate sustainability. School leadership worked with local clinic staff, who coordinated telemedicine follow-up appointments for children who required referral. Chaperones or parents transported children from school to clinic for telemedicine appointments. Based on community feedback before the trial, parents were encouraged but not required to attend, except for children in grades 2 (aged approximately 7 years) and younger.[22] Community health aide/practitioners in village clinics performed telemedicine consultations to audiology.[19] Audiology providers requested otolaryngology telemedicine consultation for surgical and medical management, as per standard practice in rural Alaska.

The standard primary care referral previously used for school screening in northwest Alaska was the control

on the t-distribution using Kauermann-Carroll corrected standard errors (for GEE) and the between-within denominator degrees of freedom approach (for random-effects models, including the primary outcome).[37]

Baseline characteristics for secondary outcomes, RDs for primary and secondary outcomes, as-treated and complier average causal effect estimation, heterogeneity of treatment effects, intraclass correlation coefficient estimation, Standards for the Reporting of Diagnostic Accuracy Studies diagram and confusion matrices for sensitivity and specificity analysis, missing data analysis, and cost-effectiveness analysis are in appendix 2 (pp 10–35). With only one primary hypothesis of interest, no adjustment for multiple comparisons was applied. p values were computed only for the primary and secondary adjusted treatment effect estimates. All other computed estimates and 95% CIs are considered exploratory.

The cost-effectiveness analysis compared four hypothetical screening and referral combinations: school screen with the standard primary care referral pathway; mobile health screen plus tympanometry with the standard primary care referral pathway; school screen with the telemedicine specialty referral pathway; and mobile health screen plus tympanometry combined with the telemedicine specialty referral pathway. This comparison used a Markov model with an embedded decision tree (appendix 2 p 15) that simulated the expected health effects and costs incurred over a 12-year time horizon for a hypothetical cohort of 1000 children. The model was constructed using Microsoft Excel (2013).

The decision tree represented the pathway for children to be screened, diagnosed, and treated. The Markov model represented the three distinct health states that children could be in at any given time: healthy, hearing loss, and treated states. The initial population, comprised of children aged 5 years, transitioned through the model using 1-month timesteps. The transition probabilities were derived from the trial data and were supplemented with published literature. The health utilities for each health state were derived from the published literature and enabled the effects to be measured as quality-adjusted life-years (QALYs). The analysis used a health system perspective; therefore, the costs represented health-care resource utilisation. The intervention costs were estimated using a micro-costing approach on the basis of the study. Hearing treatment costs were derived from published literature and estimates from the Healthcare Cost and Utilization

characteristics upon first referral were similar, with a slightly higher proportion of girls in the telemedicine referral communities (42·7%) than the primary care communities (36·8%; table 1)

Among children who received follow-up, mean time to follow-up was 41·5 days (SD 55·7) in the telemedicine referral communities and 92·0 days (75·8) in the primary care referral communities (table 2). The adjusted event-time ratio for mean days to follow-up for all referred children was 17·6 (95% CI 6·8–45·3; p=0·002; figure 3A). Of the 790 children who were referred, 268 (68·5%) of 391 in telemedicine referral communities received follow-up for their first study referral within 9 months (275 days) of the screening date, compared with 128 (32·1%) of 399 in primary care referral communities (table 2). The adjusted RR for follow-up within 9 months was 2·32 (95% CI 1·41–3·80; p=0·002; table 2). Telemedicine versus in-person care and provider type for first follow-up are described in figure 3B. The majority of children in the telemedicine referral communities received follow-up consisting of a combination of in-person primary care (from a health aide) and audiology via telemedicine, whereas the most common pattern observed in the standard primary care referral group was in-person follow up with primary care only. There were no meaningful differences by study group in hearing loss prevalence, hearing-related quality of life, or school performance (table 2).

Mobile health plus tympanometry screening had a sensitivity of 77% (95% CI 73·1–80·9; table 3), a specificity of 88·8% (95% CI 87·3–90·4), and an area access to specialty care in low-resource settings globally, where specialists are typically located in cities and are not easily accessible in rural communities. This trial also demonstrated that the mobile health plus tympanometry screen was more accurate than the school screen in this rural Alaska Native population, probably because of

referrals were due to false positives, this large proportion
re ects the high burden of hearing loss and middle ear

Contributors

SDE, AP, ELT, JJG, ABL, N-YW, JB, and SKR contributed to study concept and design. SDE, SMI, CDJ, HEP, and SKR contributed to acquisition of data. SDE, AP, ELT, JJG, N-Y W, KLH, JRE, PFH, MY, JB, and SKR contributed to analysis and interpretation of the data. SDE and KLH did the literature search. SDE, AP, ELT, and SKR contributed to