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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 at to be low, no studies have previously been done to telehealth network developed nearly 20 years before quantify follow-up in this population. We hypothesised COVID-19 generated momentum for telehealth. that telemedicine specialty referral would improve time to Validation studies of ear and hearing telemedicine follow-up compared with standard primary care referral, consultations in rural Alaska demonstrated that medical thereby reducing a key rural health disparity by improving and surgical decision making were equivalent to in- access to specialty care. There is minimal evidence on the person examinations, and telemedicine reduced the accuracy of school hearing screening protocols, waiting times for specialist appointments by 8 weeks¹⁶ particularly in populations with high prevalence of ear Therefore, telemedicine has become an established infections.¹¹ Our secondary objective was to determine the mode of clinical care in rural Alaska. optimal screening methodology in this population. We

Despite being commonly used for clinical care, hypothesised that mobile health screening with telemedicine has never been used for preventive services tympanometry would be more sensitive than the school in Alaska. We report the results of the Hearing Norton screen because of increased capacity to identify infection-Sound randomised controlled trial, which evaluated a related pathology with tympanometry.¹⁸ Additional novel telemedicine specialty referral pathway for school secondary hypotheses were that prevalence of hearing hearing screening to improve timely identification of loss would be reduced, hearing-related quality of life childhood hearing loss. This trial addresses several key would improve, school performance would improve, and knowledge gaps. Rigorous telemedicine studies are few that mobile health screening and specialty telemedicine particularly randomised controlled trials that provide referral would be cost-effective. high-quality evidence that telemedicine can increase

access to care and reduce rural health disparities **Methods**

Screening programmes worldwide, including school Study design and participants

hearing screening, have substantial loss to follow-up, and We conducted a parallel, two-arm, cluster-randomised the potential of telemedicine to ameliorate this problem controlled trial over two academic years between has not been evaluated Although follow-up from school Oct 10, 2017, and March 28, 2019, in the Bering Strait screening in rural Alaska has anecdotally been reported region of northwest Alaska, USA, which spans

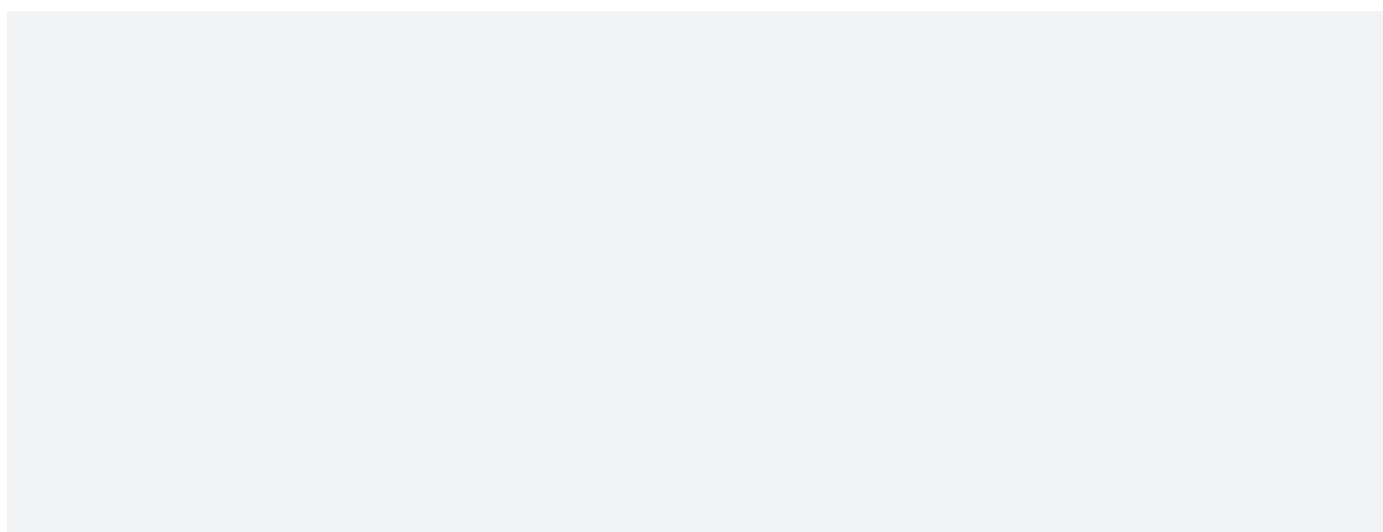
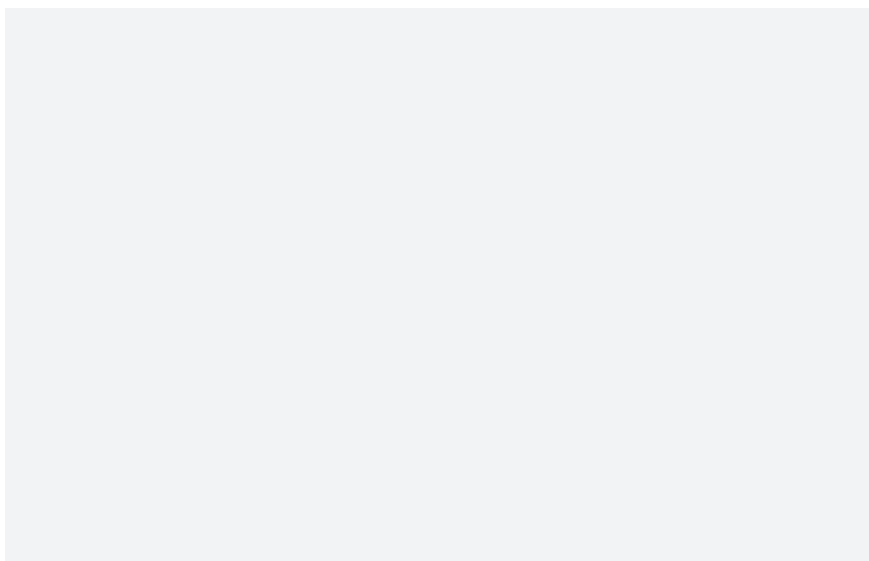


Figure 1: Map of study area (A) and referral pathways (B and C)

*Excludes Nome City school district. Reproduced with permission; copyright 2021, Duke University.

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

Trial protocols were published, and the design was health smartphone-based screen (hearX Group, South Africa) and tympanometry to assess the middle ear conducted annually in accordance with the Alaska state (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 tympanogram generated a referral. Mobile health screening day with appropriate consent were plus tympanometry screening was performed by study eligible for inclusion in the study. All eligible children who were not audiologists. Gold standard audiometric assessment was included for all children to assess the academic years. Institutional review boards of Alaska Area, NSHC, and Duke University approved the trial, air-conduction and bone-conduction audiogram at 0.5, 1, and 4 kHz with a validated tablet-based audiometer 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 negative pressure less than -200 daPa tympanogram, or findings on otoscopy (eg, occluding cerumen, retraction, acute otitis media, otorrhea, perforation, patent plugged tube, external otitis, or foreign body). Randomised referral assignments for audiologists performed the audiometric evaluation. Children who screened positive for possible hearing loss or ear disease on either screening protocol or community 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 audiologists performed the audiometric evaluation. Participants were masked to group allocation until audiometric assessment required referral. The study screening day, after which time masking referral team generated a referral list and transferred this list to assignments was not possible. Assessors remained masked throughout data collection. Study team member each community's randomised referral assignment. who provided clinical care did not read trial-related Referrals in both groups included the child's name and telemedicine referrals during the study period. Specialists the affected ear (left, right, or both), as per standard consulting on telemedicine referrals and study team practice in rural Alaska. Transfer of follow-up members performing medical record abstraction were coordination to the schools was incorporated into the masked to group allocation. Study team members study design to increase the generalisability of the performing screening or audiometric evaluations were findings. The telemedicine specialty referral intervention adapted existing telemedicine infrastructure in village clinics (figure 1B). Typical telemedicine work ow 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 should derive benefit, screening and audiometric maintained to facilitate sustainability. School leadership protocols were not randomised. All children (K–12) worked with local clinic staff, who coordinated telemedicine referrals during the study period. Specialists the affected ear (left, right, or both), as per standard consulting on telemedicine referrals and study team practice in rural Alaska. Transfer of follow-up members performing medical record abstraction were coordination to the schools was incorporated into the masked to group allocation. Study team members study design to increase the generalisability of the performing screening or audiometric evaluations were findings.

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. All children (K–12) worked with local clinic staff, who coordinated telemedicine referrals during the study period. Specialists the affected ear (left, right, or both), as per standard consulting on telemedicine referrals and study team practice in rural Alaska. Transfer of follow-up members performing medical record abstraction were coordination to the schools was incorporated into the masked to group allocation. Study team members study design to increase the generalisability of the performing screening or audiometric evaluations were findings.

Descriptions of screening and audiometric protocols have been previously published. 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 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

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

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).⁷

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

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 reflects the high burden of hearing loss and middle ear

