Mobile health school screening and telemedicine referral to improve access to specialty care in rural Alaska: a cluster-randomised controlled trial

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Summary

Background School-based programmes, including hearing screening, provide essential preventive services for rural children. However, minimal evidence on screening methodologies, loss to follow-up, and scarcity of specialists for subsequent care compound rural health disparities. We hypothesised telemedicine specialty referral would improve time to follow-up for school hearing screening compared with standard primary care referral.

Methods In this cluster-randomised controlled trial conducted in 15 rural Alaskan communities, USA, we randomised communities to telemedicine specialty referral (intervention) or standard primary care referral (control) for school hearing screening. All children (K–12; aged 4–21 years) enrolled in Bering Straight School District were eligible. Community randomisation occurred within four strata using location and school size. Participants were masked to group allocation until screening day, and assessors were masked throughout data collection. Screening occurred annually, and children who screened positive for possible hearing loss or ear disease were monitored for 9 months from the screening date for follow-up. Primary outcome was the time to follow-up after a positive hearing screen; analysis was by intention to treat. The trial was registered with ClinicalTrials.gov, NCT03309553.

Findings We recruited participants between Oct 10, 2017, and March 28, 2019. 15 communities were randomised: eight (750 children) to telemedicine referral and seven (731 children) to primary care referral. 790 (53·3%) of 1481 children screened positive in at least one study year: 391 (52·1%) in the telemedicine referral communities and 399 (50·4%) in the primary care referral communities. Of children referred, 268 (68·5%) in the telemedicine referral communities and 128 (32·1%) in primary care referral communities received follow-up within 9 months. 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 (adjusted event-time ratio 17·6 [95% CI 6·8–45·3] for all referred children). There were no adverse events.

Interpretation Telemedicine specialty referral significantly improved the time to follow-up after hearing screening in Alaska. Telemedicine might apply to other preventive school-based services to improve access to specialty care for rural children.

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Introduction

Childhood hearing loss has well known, profound implications for language development, school achievement, and future employment opportunities.[1–4] Some populations experience a disproportionately high burden of childhood hearing loss, including rural Alaska Native children, among whom there is a prevalence of up to 31% compared with 1·7–5% in the general US population.[5,6] Similar to low-resource settings globally, the majority of hearing loss in Alaska is related to infection, with otitis media 4–5 times more prevalent in rural Alaska Native children despite pneumococcal vaccination.[4,5,7–9]

WHO estimates that 60% of all childhood hearing loss is preventable, and 75% is preventable in low-income and middle-income countries, where infection-related causes are common.[10] School-based health programmes often provide the only access to preventive services for underserved children who live in rural areas, and school hearing screening is mandated in Alaska. However, loss to follow-up from school hearing screening is a persistent problem. Follow-up has been reported to range from 10% to 65% in school screening programmes worldwide, and scarcity of specialists in rural areas is frequently highlighted as a major barrier to care.[11] For example, in the USA, urban areas have 263 specialists per 100 000 people, whereas rural areas have only 30 specialists per 100 000 people.[12] A recent survey of US rural health clinics highlighted too few specialty providers as the most frequent reason for difficulty establishing specialist referrals, followed by poor appointment availability, and distance to travel.[12]

Telemedicine has become integral to health care in the current era. The Alaska Tribal Health System addressed

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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

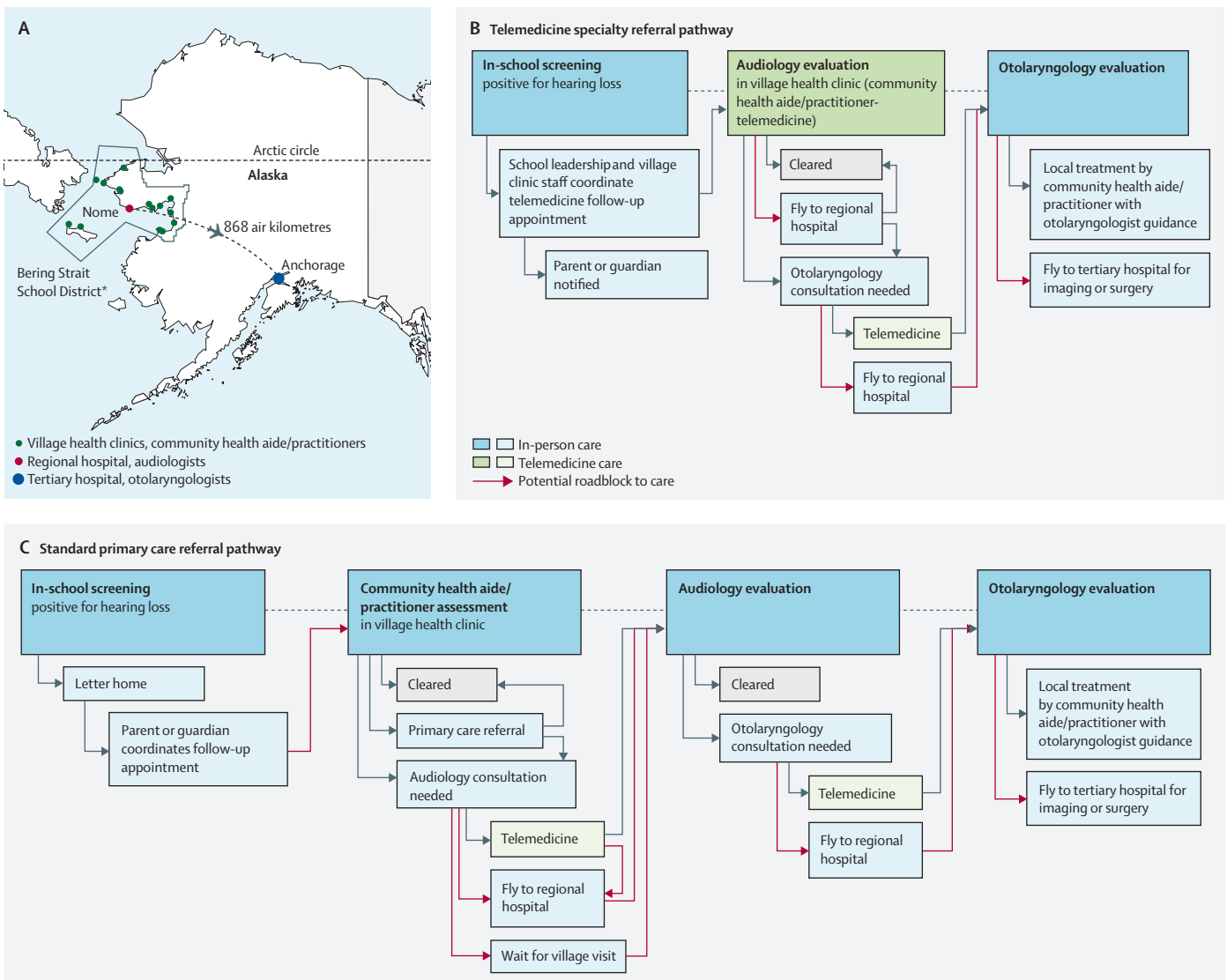

Figure 1: **Map of study area (A) and referral pathways (B and C)**
*Excludes Nome City school district. Reproduced with permission; copyright 2021, Duke University.

23 000 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.[19] 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

(figure 1C). Schools sent a letter home to families of children requiring referral, requesting they bring their child for evaluation at the village health clinic. Once a child presented for evaluation, three possible treatment pathways could occur: wait for an audiology field clinic (held every 3–4 months); a telemedicine consultation to audiology using the standard 60–90 min telemedicine workflow; or referral to a primary care provider. Audiologists requested otolaryngology consultation as per standard practice. Schools also provided referral lists to the NSHC Department of Audiology. Audiology staff contacted families to schedule appointments during the next field clinic. Children in communities assigned to the intervention but who did not enrol in the trial received the standard primary care referral pathway.

All communities participating in the study had telemedicine capabilities within village health clinics; community health aide/practitioners routinely use this technology for management of all health concerns. We did not restrict the use of telemedicine in primary care referral communities.

On school screening day, participating children completed the school hearing screen, mobile health screen, gold standard audiometric evaluation, and the Hearing Environments and Reflection on Quality of Life (HEAR-QL) questionnaire. The Bering Strait School District administered AIMSweb mathematics and reading assessments (a validated measure of academic performance used by Bering Strait School District) three times annually. For children who required referral, the electronic health record was monitored by chart review for follow-up for 9 months (275 days) from the hearing screening date (primary outcome).

Outcomes

The primary outcome was time to follow-up after a positive hearing screen, measured in days from the date of screening (for children receiving their first study referral). Follow-up was defined as an ear or hearing encounter with a community health aide/practitioner, primary care provider, audiologist, or otolaryngologist, measured by the presence of any ear or hearing International Statistical Classification of Diseases, 10th Revision (ICD-10) diagnosis code in the electronic health record. The presence of such codes could indicate further evaluation that resulted in a child being cleared or a formal diagnosis of an ear or hearing condition. Eligible diagnoses included all ear or hearing ICD-10 codes listed in the trial protocol (appendix 1; p B1–9).

Secondary outcomes included the binary outcome of any follow-up, the differences in prevalence of hearing loss, hearing-related quality of life, and school performance among children who required referral from screening in year 1. These secondary outcomes were limited to the subset of children who participated in both years of the trial to allow for within-person change. Prevalence, defined with WHO criteria (pure tone audiometry >25 dB at 0·5, 1, 2, and 4 kHz in either ear) was measured by audiometric evaluation.[23] Hearing-related quality of life was evaluated with the validated HEAR-QL questionnaire, and school performance was assessed with national percentile scores in mathematics and reading from AIMSweb.[24] Most secondary outcomes were limited in age ranges and grades that could be assessed (HEAR-QL for children aged ≥7 years, and AIMSweb for grades 1–8, aged approximately 6–13 years). Other outcomes included sensitivity and specificity of screening, measured with screening and gold standard audiometric data from the trial, and cost-effectiveness of screening and referral pathways, assessed with a Markov model. Detailed definitions of all outcomes are in appendix 2 (p 18–19).This was a minimal risk study, and no serious adverse events were anticipated.

See Online for appendix 2

Statistical analysis

The estimated sample size was 1500 across 15 schools (one per community) with assumed equal cluster sizes and an intraclass correlation of 0·25, a two-sided type 1 error rate of 5%, and 90% retention. This yielded 81% power to detect a reduction in time to follow-up of 3 weeks (SD 3·0) between groups.[16,20]

Analyses used an intention-to-treat approach according to a prespecified statistical analysis plan, with key elements summarised here. Cumulative incidence of follow-up over 9 months (275 days) from screening date was visualised using Kaplan-Meier curves. We prespecified 30, 60, and 90 days as additional timepoints of interest.

Primary outcome analysis calculated between-group ratios of time to follow-up, estimated using accelerated failure time models with random intercepts for school, assuming log-normal time distribution. The primary outcome was expressed as an event-time ratio, representing a ratio of mean time to follow-up. Children lost to follow-up were right censored at 275 days, and 0·5 days were added to event times for same day follow-up after screening. Unadjusted analyses included indicators for treatment group and randomisation strata. Treatment effects estimated from covariate adjusted models were considered primary and included age, sex, and the highest education level of any caregiver (all prespecified).

Secondary continuous outcomes (quality of life and school performance) were analysed using linear regression with random intercepts for school. Secondary binary outcomes were analysed using generalised estimating equations (GEE) with independence working correlation matrices and robust standard errors, with Poisson distribution and log link for risk ratios (RRs) and Gaussian distribution with identity link for risk differences (RDs).[25] Sensitivity analysis used the delta method to approximate RDs from RR models. Due to the small number of clusters and possibility of inflated type I error, permutation tests were used to obtain p values for primary and secondary outcomes,[26] and CIs were based

See Online for appendix 1

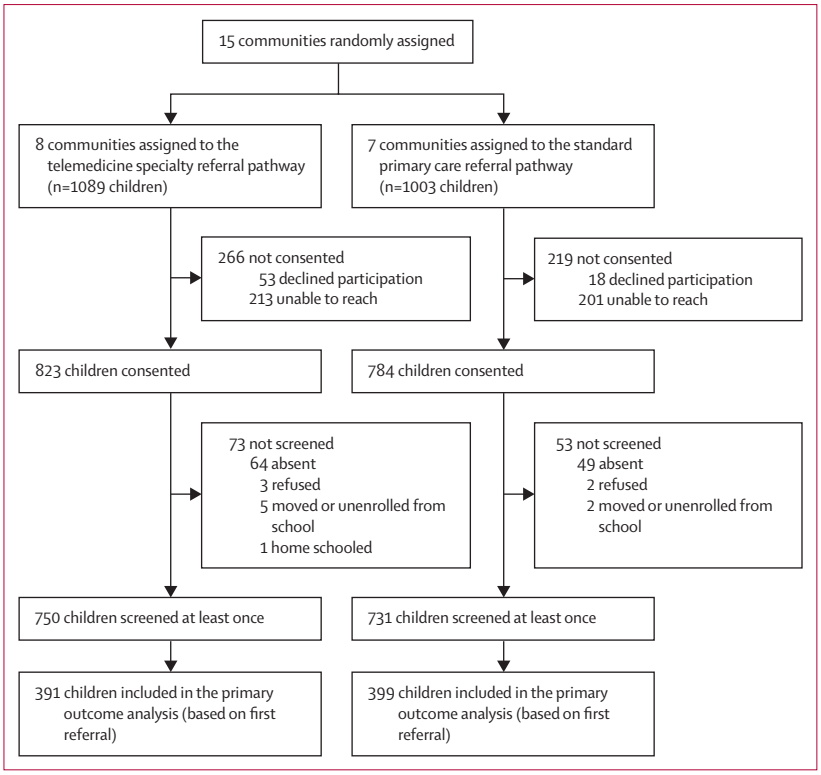

Figure 2: **Trial profile**

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).[27]

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 Project. All costs were reported in 2018 US$. Costs and QALYs were discounted 3%. The analysis estimated an incremental cost-effectiveness ratio measured as dollars per QALY and compared the four pathways. A willingness-to-pay threshold of $50 000 per QALY was assumed to determine the cost-effectiveness of the results.[28] Deterministic and probabilistic sensitivity analysis characterised model uncertainty. Full analysis details are in appendix 2 (pp 7–9, 13–14, 16, 34–35). Analyses adhered to cluster trial CONSORT guidelines and used Stata (version 16). The trial was registered with ClinicalTrials.gov, NCT03309553. The principal investigators were responsible for ensuring participant safety and secure data management. An independent Data Safety Monitoring Board oversaw trial conduct.

Role of the funding source

The funder of the study had no role in study design, data collection, data analysis, data interpretation, or writing of the report.

Results

We randomised 15 communities, eight (n=1089) to the telemedicine specialty referral pathway and seven (n=1003) to the standard primary care referral pathway. From Oct 10, 2017, to March 28, 2019, 823 children in telemedicine referral communities gave consent and 750 received screening (71·8% of the eligible population). In primary care referral communities, 784 children consented and 731 received screening (75·5% of the eligible population; figure 2; appendix 2 p 17). Of 1481 children screened, 790 (53·3%) required referral in at least one study year, including 391 (52·1%) in telemedicine referral communities and 399 (50·4%) in primary care referral communities. Baseline

	Study entry		First referral	
	Standard primary care referral pathway (n=731)	Telemedicine specialty referral pathway (n=750)	Standard primary care referral pathway(n=399)	Telemedicine specialty referral pathway (n=391)
Sociodemographic characteristics*				
Age, years	10·0 (7·0–13·0)	10·0 (7·0–13·0)	10·0 (7·0–13·0)	10·0 (7·0–14·0)
Age range, years				
4–6	149/727 (20·5%)	147/745 (19·7%)	80/395 (20·3%)	68/379 (17·9%)
7–9	206/727 (28·3%)	198/745 (26·6%)	112/395 (28·4%)	106/379 (29·0%)
10–12	167/727 (23·0%)	164/745 (22·0%)	85/395 (21·5%)	82/379 (21·6%)
13–15	109/727 (15·0%)	132/745 (17·7%)	68/395 (17·2%)	72/379 (19·0%)
≥16	96/727 (13·2%)	97/745 (13·0%)	50/395 (12·7%)	58/379 (15·3%)
Sex				
Female	333 (45·6%)	362 (48·3%)	147 (36·8%)	167 (42·7%)
Male	398 (54·4%)	388 (51·7%)	252 (63·2%)	224 (57·3%)
American Indian or Alaska Native	702 (96·0%)	716 (95·5%)	385 (96·5%)	376 (96·2%)
Grade level				
K–5	434 (59·4%)	431 (57·5%)	227 (56·9%)	218 (55·8%)
6–8	158 (21·6%)	160 (21·3%)	88 (22·1%)	85 (21·7%)
9–12	139 (19·0%)	159 (21·2%)	84 (21·1%)	88 (22·5%)
Highest education level of any caregiver				
<12th grade	38/721 (5·3%)	53/739 (7·2%)	24/377 (6·4%)	40/374 (10·7%)
High school diploma or GED	480/721 (67·6%)	446/739 (60·4%)	272/377 (72·1%)	249/374 (65·6%)
Some college	132/721 (18·3%)	153/739 (20·7%)	63/377 (16·7%)	67/374 (18·0%)
College degree	59/721 (8·2%)	81/739 (11·0%)	30/377 (8·0%)	24/374 (6·4%)
Study year				
Year 1	602 (82·4%)	617 (82·3%)	281 (70·4%)	278 (71·1%)
Year 2	129 (17·6%)	133 (17·7%)	118 (29·6%)	113 (28·9%)
Clinical characteristics				
Hearing loss severity†				
No hearing loss in either ear	669/720 (92·9%)	666/735 (90·6%)	332/388 (85·6%)	308/371 (83·0%)
Mild, PTA >25–40 dB	45/720 (6·3%)	50/735 (6·8%)	50/388 (12·9%)	54/371 (14·6%)
Moderate, PTA >40 dB	6/720 (0·8%)	14/735 (1·9%)	6/388 (1·5%)	14/371 (3·8%)
Middle ear disease‡	142/722 (19·7%)	99/741 (13·4%)	150/387 (38·9%)	108/373 (29·0%)
Ear or hearing management status§				
Never managed	158/340 (46·5%)	127/336 (37·8%)	197 (49·4%)	156 (39·9%)
Previously managed	125/340 (36·8%)	145/336 (43·2%)	140 (35·1%)	167 (42·7%)
Currently managed	57/340 (16·8%)	64/336 (19·0%)	62 (15·5%)	68 (17·4%)
Cluster sizes				
Mean	104·4 (65·4)	93·8 (36·0)	57·0 (40·2)	48·9 (23·9)
Median	89 (50–160)	87 (67–127)	62 (16–93)	46 (29–65)
Community characteristics¶				
Distance to regional centre, km‖				
Mean	183·1 (59·0)	184·7 (72·4)	..	..
Median	195·5 (113·4–214·9)	186·6 (127·4–218·3)	..	..
Population**				
Mean	342·3 (191·6)	401·1 (208·4)	..	..
Median	345·0 (162·0–574·0)	371·5 (248·0–558·0)	..	..

Data are n (%), n/N (%), median (IQR), or mean (SD) unless otherwise indicated. GED=Graduate Educational Development. PTA=pure-tone average. *Self-reported by parent or guardian of participant. †Based on WHO definition of PTA >25 dB (0·5, 1, 2, 4 kHz) in either ear and assessed via audiometric evaluation. ‡Assessed via tympanometry and otoscopy during audiometric evaluation. §Based on electronic health record query before screening date. Previously managed was defined as having an audiology or otolaryngology encounter >3 months and <5 years from date of screening and not under active management. Currently managed was defined as having an audiology or otolaryngology encounter within 3 months of screening day or wearing a hearing aid on screening day. ¶All communities are predominantly Alaska Native, have one school and one health clinic, none are accessible by road and require travel by plane, all have the same screening services and telehealth access, and none of the schools have school nurses. ‖Distance calculated from census designated place centroid of each of the 15 communities to place centroid of Nome (where the regional hospital is located). **Using estimates from the 2010 Census.

Table 1: **Baseline characteristics of children at trial entry and first referral**

	Standard primary care referral pathway (n=399)	Telemedicine specialty referral pathway (n=391)	Unadjusted analysis* (95% CI)	Adjusted analysis† (95% CI)
Primary outcome				
Median days to follow-up	82 (28–133); n=128	16 (4–51); n=268	..	..
Mean days to follow-up	92·0 (75·8); n=128	41·5 (55·7); n=268	ETR 17·1 (6·7 to 43·8)	ETR 17·6 (6·8 to 45·3)‡
Secondary outcomes§				
Followed up within 275 days	128 (32·1%)	268 (68·5%)	RR 2·25 (1·75 to 2·89)	RR 2·32 (1·41 to 3·80)
Hearing loss in year 2, among referred¶	45/241 (18·7%)	52/222 (23·4%)	RR 1·18 (0·74 to 1·89)	RR 1·20 (0·09 to 15·32)‖
Hearing loss in year 2, among all screened¶	50/512 (9·8%)	56/511 (11·0%)	RR 1·08 (0·58 to 2·01)	RR 1·12 (0·04 to 31·36)‖
Child HEAR-QL in year 2	76·7 (17·4); n=110	72·7 (18·0); n=106	–2·25 (–7·36 to 2·86)	–2·24 (–7·52 to 3·04)‖
Adolescent HEAR-QL in year 2	90·0 (13·7); n=52	82·6 (17·4); n=68	–3·47 (7·89 to 0·96)	–3·32 (–8·09 to 1·45)‖
Percentile score, mathematics in year 2	27·1 (25·5); n=189	28·3 (27·8); n=164	1·88 (–8·34 to 12·10)	2·11 (–11·96 to 16·18)‖
Percentile score, reading in year 2	16·9 (20·4); n=188	18·5 (22·6); n=164	5·40 (–3·25 to 14·06)	5·74 (–2·58 to 14·06)‖

Data are n (%), n/N (%), mean (SD), or median (IQR) unless otherwise indicated. ETR=event time ratio. RR=risk ratio. HEAR-QL=Hearing Environments and Reflection on Quality of Life. PTA=pure tone average. *Unadjusted models include fixed effects for treatment group and strata only. †Adjusted models include fixed effects for treatment group, strata, age, sex, and highest level of education of any caregiver. Analytic sample size is lower due to missing data for age and highest education of any caregiver. ‡Primary outcome of interest, days to follow-up (within 275 days) p=0·002. §Secondary outcome of interest: follow-up within 275 days p=0·002, hearing loss prevalence among referred p=0·616; hearing loss prevalence among screened p=0·883, Child HEAR-QL p=0·379, Adolescent HEAR-QL p=0·125, percentile score for mathematics p=0·623, percentile score for reading p=0·110. All p values calculated from stratified cluster permutation test. ¶Based on World WHO definition of PTA >25 dB (0·5, 1, 2, 4 kHz) in either ear. ‖Secondary outcomes are based on year 2 data because they evaluate changes in hearing loss, hearing-related quality of life, and academic performance among children who were referred in year 1.

Table 2: **Primary and secondary outcomes**

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 under the receiver operator characteristic curve (AUC) of 0·829 (95% CI 0·809–0·849) relative to the gold standard audiometric evaluation. Mobile health plus tympanometry outperformed the school screen by 17·6 percentage points (95% CI 12·6–22·5) on sensitivity and 0·097 points on AUC (95% CI 0·073–0·122; table 2). Corresponding results for positive and negative predictive values and differences between tools are in appendix 2 (p 24)

Each screening and telemedicine referral intervention was cost-effective compared with the standard of care willingness-to-pay threshold of $50 000 per QALY. The mobile health plus tympanometry screen with standard primary care referral was cost-effective at $2783 per QALY, and mobile health plus tympanometry screen with telemedicine specialty referral was cost-effective at $4504 per QALY (table 4). There were no adverse events.

Discussion

In this cluster-randomised controlled trial, telemedicine specialty referral significantly reduced the time to follow-up after school hearing screening in 15 rural Alaska Native communities compared with standard primary care referral. The proportion of children receiving follow-up increased more than two times in the telemedicine specialty referral group. This is the first randomised trial to demonstrate that telemedicine can reduce a key rural health disparity by improving access to specialty care. Telemedicine is integral to health-care delivery in the COVID-19 era, and this trial extends its applicability from clinical care to rural, school-based, preventive health programmes. This study has implications for improving

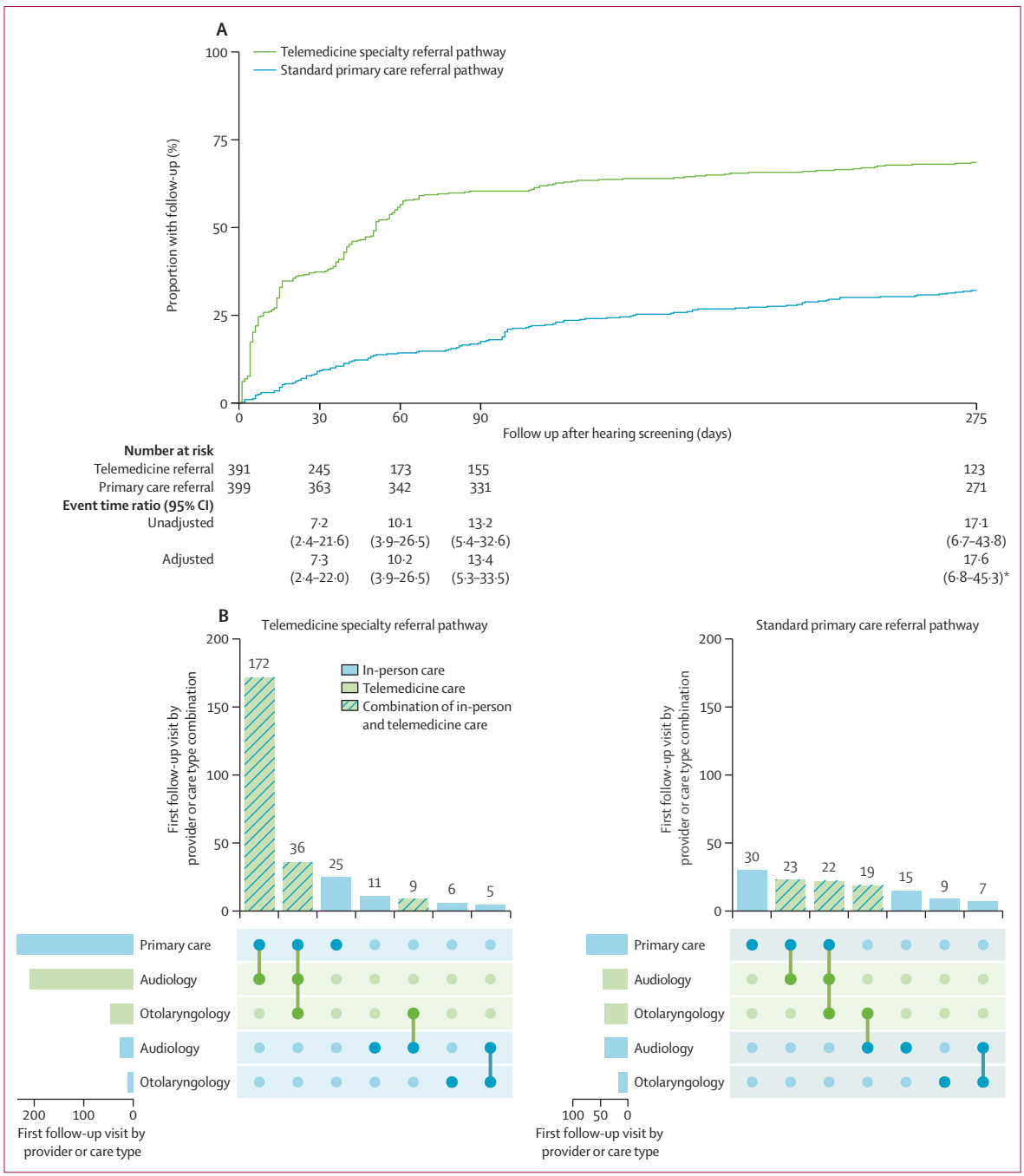

Figure 3: **Primary outcome analysis results**
(A) Kaplan-Meier curves for time to follow-up; (B) provider and care type (telemedicine *vs* in-person) for first follow-up visit (connected dots indicate care from multiple providers). *Primary outcome.

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 the enhanced ability to detect infection-related middle ear pathology with tympanometry. Both the mobile health plus tympanometry screen and novel specialty telemedicine referral pathway were cost-effective using a conservative conventional willingness-to-pay threshold of $50 000 per QALY.[28]

More than 50% of participating children required referral in at least one of the study years. Although some

	Number included*	Number referred	Sensitivity (95% CI)	Specificity (95% CI)	AUC (95% CI)
Screening tools					
School screen	2400	555 (23·1%)	59·5 (54·7 to 64·2)	86·9 (85·2 to 88·6)	0·732 (0·709 to 0·754)
Mobile health	2414	477 (19·8%)	57·9 (53·4 to 62·4)	90·9 (89·5 to 92·3)	0·744 (0·722 to 0·766)
Mobile health plus tympanometry	2367	600 (25·3%)	77·0 (73·1 to 80·9)	88·8 (87·3 to 90·4)	0·829 (0·809 to 0·849)
Difference in diagnostic accuracy					
Mobile health plus tympanometry *vs* school screen	2388	..	17·6 (12·6 to 22·5)†	1·9 (–0·1 to 3·9)†	0·097 (0·073 to 0·122)
Mobile health plus tympanometry *vs* mobile health	2367	..	19·1 (15·5 to 22·8)†	–2·1 (–2·8 to –1·4)†	0·085 (0·068 to 0·103)

Data are n (%) unless indicated otherwise. AUC=area under the receiver operator characteristic curve. *Sample sizes for diagnostic accuracy metrics indicate the total number of observations available for analysis—ie, those with non-missing values for both index tool (mobile health or school screen) and gold standard (audiometric evaluation). †Percentage point differences.

Table 3: **Other outcomes**

	Incremental cost-effectiveness ratio				Costs and QALYs	
	1	2	3	4	Costs	QALYs
School screen with standard primary care referral	..	$2783	$4085	$4504	$262	9·81
Mobile health screen plus tympanometry with standard primary care referral	..	..	$10 872	$10 491	$550	9·91
School screen with telemedicine specialty referral	..	..	..	$9728	$766	9·93
Mobile health screen plus tympanometry with telemedicine specialty referral	..	..	..	..	$863	9·94

Currency shown is US dollar. QALY=quality-adjusted life-year. 1=school screen with standard primary care referral. 2=mobile health screen plus tympanometry with standard primary care referral. 3=school screen with telemedicine specialty referral. 4=mobile health screen plus tympanometry with telemedicine specialty referral.

Table 4: **Cost-effective analysis**

referrals were due to false positives, this large proportion reflects the high burden of hearing loss and middle ear disease in this rural Alaska Native population and is consistent with previous studies demonstrating higher otitis media visit rates in rural Alaska Native children than urban Alaska Native children.[4,5,7–9] We hypothesised that the prevalence of hearing loss would improve in the telemedicine group because the majority of hearing loss in this rural Alaska Native population is infection-related, and interventions such as tympanostomy tube placement and tympanoplasty can improve this type of hearing loss. However, we found no differences in hearing loss prevalence between the groups. We also found no differences in hearing-related quality of life or school performance between groups. Although there are no comparable randomised trials that measured hearing-related quality of life or academic performance after a health-care delivery intervention, observational studies have shown improvements in school performance and mixed effects on hearing-related quality of life with hearing loss treatment.[29,30] Our ability to measure differences in these secondary outcomes was limited by the short length of the trial and the reduced sample size due to age restrictions on school performance testing and hearing-related quality of life questionnaires. Future studies with longer follow-up periods are needed to fully evaluate these important hearing-related outcomes.

Strengths of this study included a pragmatic trial design that included all children (K–12) in an entire school district over two academic years, with excellent recruitment and retention. This design allowed measurement of outcomes at the population level, which is necessary for policy decisions. Community engagement in design of the intervention and transfer of the referral coordination process to school staff increased sustainability beyond the trial period.[22]

There are limitations to this study. We designed the trial to use existing telemedicine infrastructure in a region of Alaska where the Tribal Health System is the sole source of health care. This allowed us to evaluate telemedicine specialty referral for school-based preventive services in an environment where telemedicine technology is already routinely used for clinical care and it was feasible to incorporate the entire care continuum from village health clinic to tertiary centre. Although essential for proof of concept, this trial was conducted in a unique environment within the Alaska Tribal Health System where patients receive federally covered health care and the electronic health record is part of a shared consortium; therefore, the findings might not be generalisable to other rural areas where multiple health-care entities and more variation in insurance coverage exist. Future research would benefit from expansion into other environments beyond tribal health.

This trial has notable broad public health implications. 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. This trial focused on school hearing screening, but the model of specialty telemedicine referral is applicable to other preventable health conditions. Importantly, this novel telemedicine model promotes early access to specialists in an effort to decrease health disparities.

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 drafting the manuscript. SDE, AP, ELT, JJG, ABL, SMI, CDJ, HEP, N-Y W, KLH, JRE, PFH, MY, JB, and SKR critically appraised the manuscript for important intellectual content. AP and ELT did the statistical analysis. SDE and SKR obtained funding and supervised the study. All authors had full access to deidentified data. SDE and SKR had full access to all the data in the study and take responsibility for the integrity of the data and the accuracy of the data analysis.

Declaration of interests

SKR has received payment for telehealth lectures by the American Academy of Audiology and Audiology Today. All other authors declare no competing interests.

Data sharing

Deidentified participant data, the data dictionary, study protocol, statistical analysis plan, and informed consent form will be made available beginning 6 months and ending 5 years following article publication to researchers upon request, pending tribal approval, and possible execution of a data use agreement. Proposals should be directed to Samantha Kleindienst Robler at skleindienst@nshcorp.org.

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