Telemedicine Referral to Improve Access to Specialty Care for Preschool Children in Rural Alaska: A Cluster-Randomized Controlled Trial

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Objectives: Preschool programs provide essential preventive services, such as hearing screening, but in rural regions, limited access to specialists and loss to follow-up compound rural health disparities. We conducted a parallel-arm cluster-randomized controlled trial to evaluate telemedicine specialty referral for preschool hearing screening. The goal of this trial was to improve timely identification and treatment of early childhood infection-related hearing loss, a preventable condition with lifelong implications. We hypothesized that telemedicine specialty referral would improve time to follow-up and the number of children receiving follow-up compared with the standard primary care referral.

Design: We conducted a cluster-randomized controlled trial in K-12 schools in 15 communities over two academic years. Community randomization occurred within four strata using location and school size. In the second academic year (2018–2019), an ancillary trial was performed in the 14 communities that had preschools to compare telemedicine specialty referral (intervention) to standard primary care referral (comparison) for preschool hearing screening. Randomization of communities from the main trial was used for this ancillary trial. All children enrolled in preschool were eligible. Masking was not possible because of timing in the second year of the main trial, but referral assignment was not openly disclosed. Study team members and school staff were masked throughout data collection, and statisticians were blinded to allocation during analysis. Preschool screening occurred once, and children who were referred for possible hearing loss or ear disease were monitored for follow-up for 9 months from the screening date. The primary outcome was time to ear/hearing-related follow-up from the date of screening. The secondary outcome was any ear/hearing follow-up from screening to 9 months. Analyses were conducted using an intention-to-treat approach.

Results: A total of 153 children were screened between September 2018 and March 2019. Of the 14 communities, 8 were assigned to the telemedicine specialty referral pathway (90 children), and 6 to the standard primary care referral pathway (63 children). Seventy-one children (46.4%) were referred for follow-up: 39 (43.3%) in the telemedicine specialty referral communities and 32 (50.8%) in the standard primary care referral communities. Of children referred, 30 (76.9%) children in telemedicine specialty referral communities and 16 (50.0%) children in standard primary care referral communities received follow-up within 9 months (Risk Ratio = 1.57; 95% confidence interval [CI], 1.22 to 2.01). Among children who received follow-up, median time to follow-up was 28 days (interquartile range [IQR]: 15 to 71) in telemedicine specialty referral communities compared with 85 days (IQR: 26 to 129) in standard primary care referral communities. Mean time to follow-up for all referred children was 4.5 (event time ratio = 4.5; 95% CI, 1.8 to 11.4; p = 0.045) times faster in telemedicine specialty referral communities compared with standard primary care referral communities in the 9-month follow-up time frame.

Conclusions: Telemedicine specialty referral significantly improved follow-up and reduced time to follow-up after preschool hearing screening in rural Alaska. Telemedicine referrals could extend to other preventive school-based services to improve access to specialty care for rural preschool children.

Key words: Child health, Hearing loss, Healthcare disparities, Mobile health, Rural health, School hearing screening, Telemedicine, telehealth.

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IDEA

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INTRODUCTION

Screening for hearing loss is an essential preventive service for children from birth through secondary education. Prevalence of hearing loss in school-age children is more than double that in newborns (Fortnum et al. 2001) and has significant consequences for speech and language development, educational achievement, and vocational implications if left untreated (Järvelin et al. 1997; Bess et al. 1998; Wake et al. 2004; Kennedy et al. 2006; Lieu et al. 2012). Screening programs are

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particularly critical in rural areas where children are more likely to experience infection-related hearing loss subsequent to otitis media, which can be treated or even prevented with early identification (Shaheen et al. 2012; Mukara et al. 2017).

Ear infections are most common in children younger than age 7 (Monasta et al. 2012), with infection-related hearing loss in preschool children known to cause speech and language delays that impact readiness for kindergarten compared with normal-hearing peers (Fitzpatrick et al. 2011; Ching et al. 2013, 2017). Early identification and treatment of hearing loss can improve language outcomes (Yoshinaga-Itano et al. 1998; Moeller, 2000; Kennedy et al. 2006). As a result, children enrolled in Head Start, a US federally-funded program to provide early childhood education, are required to undergo hearing screening. However, despite well-established hearing screening programs in early childhood, loss to follow-up after screening is a systemic problem (Cunningham et al. 2018; Rodriguez et al. 2018). Loss to follow-up is further exacerbated in rural areas, where limited numbers of specialists for subsequent treatment intensify barriers to care (Wong et al. 2017; Rodriguez et al. 2018; Monteiro et al. 2019).

In Alaska, where nearly 75% of communities are not connected to a hospital by road (Carroll et al. 2011), innovative telemedicine solutions were developed and implemented across the state to overcome geographic barriers and connect rural communities with specialty care. Similar to other rural areas, children in rural Alaska experience a high prevalence of ear infections and subsequent hearing loss compared with the general US population (Reed et al. 1967; Kaplan et al. 1973; Curns et al. 2002; Singleton et al. 2009). While telemedicine consultations for ear and hearing care have been validated as equivalent to in-person examination and have significantly reduced wait times for specialist appointments (Patricoski et al. 2003; Kokesh et al. 2008; Hofstetter et al. 2010), telemedicine has not yet been used for preventive services, such as follow-up for school hearing screening. We designed a randomized trial, Hearing Norton Sound, to address loss to follow-up by leveraging existing telemedicine infrastructure and generate evidence for screening protocols in kindergarten through 12th-grade schools (Emmett, Robler, Gallo, et al. 2019; Emmett, Robler, Wang, et al. 2019; Robler et al. 2020). The Hearing Norton Sound cluster-randomized controlled trial was conducted over two academic years to evaluate the effectiveness of the telemedicine specialty referral intervention in school-age children in the region. We report here an ancillary trial conducted in preschools in the region in the second year of the main trial, with the goal of evaluating telemedicine specialty referral in preschool hearing screening. We hypothesized that telemedicine specialty referral would improve time to follow-up and the number of preschool children receiving follow-up compared with the standard primary care referral.

MATERIALS AND METHODS

Study Design

Hearing Norton Sound consisted of a cluster-randomized trial designed to evaluate telemedicine specialty referral in school-aged children (Emmett et al. 2022). A cluster randomized trial is a trial in which groups of individuals are randomly allocated to a treatment arm rather than being randomized individually. Cluster randomization was chosen because the referral intervention was designed for communities as a whole. The main trial enrolled children K-12th grade over two academic years (2017–2019) in 15 rural communities served by Bering Strait School District (BSSD) and Norton Sound Health Corporation (NSHC; Emmett, Robler, Gallo, et al. 2019; Emmett, Robler, Wang, et al. 2019). The ancillary trial was added in the second year of the main trial (academic year 2018–2019) after community members, including teachers and parents, requested inclusion of early childhood education. Children were enrolled from September 2018–March 2019. Preschools were present in 14 of the 15 communities that participated in the main trial. The ancillary trial was designed to be analogous to the main trial, enrolling all children in preschool who were present on screening day and who had signed parental consent. Assignment to the telemedicine specialty referral pathway versus standard primary care referral pathway was based on the randomized assignment of the community in the main trial (see Figs 1 and 2). On the basis of community preference, all participating preschool children received both the preschool hearing screening and mobile health (mHealth) hearing screening in addition to a benchmark audiometric assessment (further description provided in procedures). The study was reviewed and approved by the Institutional Review Boards of Alaska Area, NSHC, and Duke University.

Randomization and Masking

Randomization of the telemedicine specialty referral intervention for the main trial for school-aged children occurred at the community level (with each community having one school) and was stratified by geographic location and weighted by school size for the 15 communities comprising BSSD (Emmett, Robler, Wang, et al. 2019). The randomized allocation was computer generated by one of the study statisticians (N.Y.W.) and assigned eight communities to the telemedicine specialty referral pathway and seven communities to the standard primary care referral pathway. Randomized assignment from the main trial was maintained for the preschool ancillary trial, with 14 of the 15 communities having a preschool program. Three entities in the region conduct preschool or Head Start programs. These include BSSD (n = 2), Rural Alaska Community Action Program, Inc. (RURal CAP; n = 2), and Kawerak, Inc. (n = 10). On the basis of the locations of these programs and the randomization assignment from the main trial, eight communities received the telemedicine specialty referral pathway, and six communities received the standard primary care referral pathway for the ancillary preschool trial.

All children enrolled in one of these preschool programs were eligible, and parents/guardians received study information and written informed consent as part of school paperwork. All preschool children with signed parental consent and child assent who were present at school on screening day were enrolled by the study team according to the prespecified community-level referral assignments.

For the ancillary trial, masking of the randomization assignment was not possible because preschools received the treatment assignment that had been applied to the K-12 school in each community in the previous year during the main trial. Thus, it was possible that community members and study team members had knowledge of which communities were assigned to the intervention versus comparison arm. However, randomization assignments were not openly disclosed. Furthermore, the audiologists and otolaryngologists consulting on the

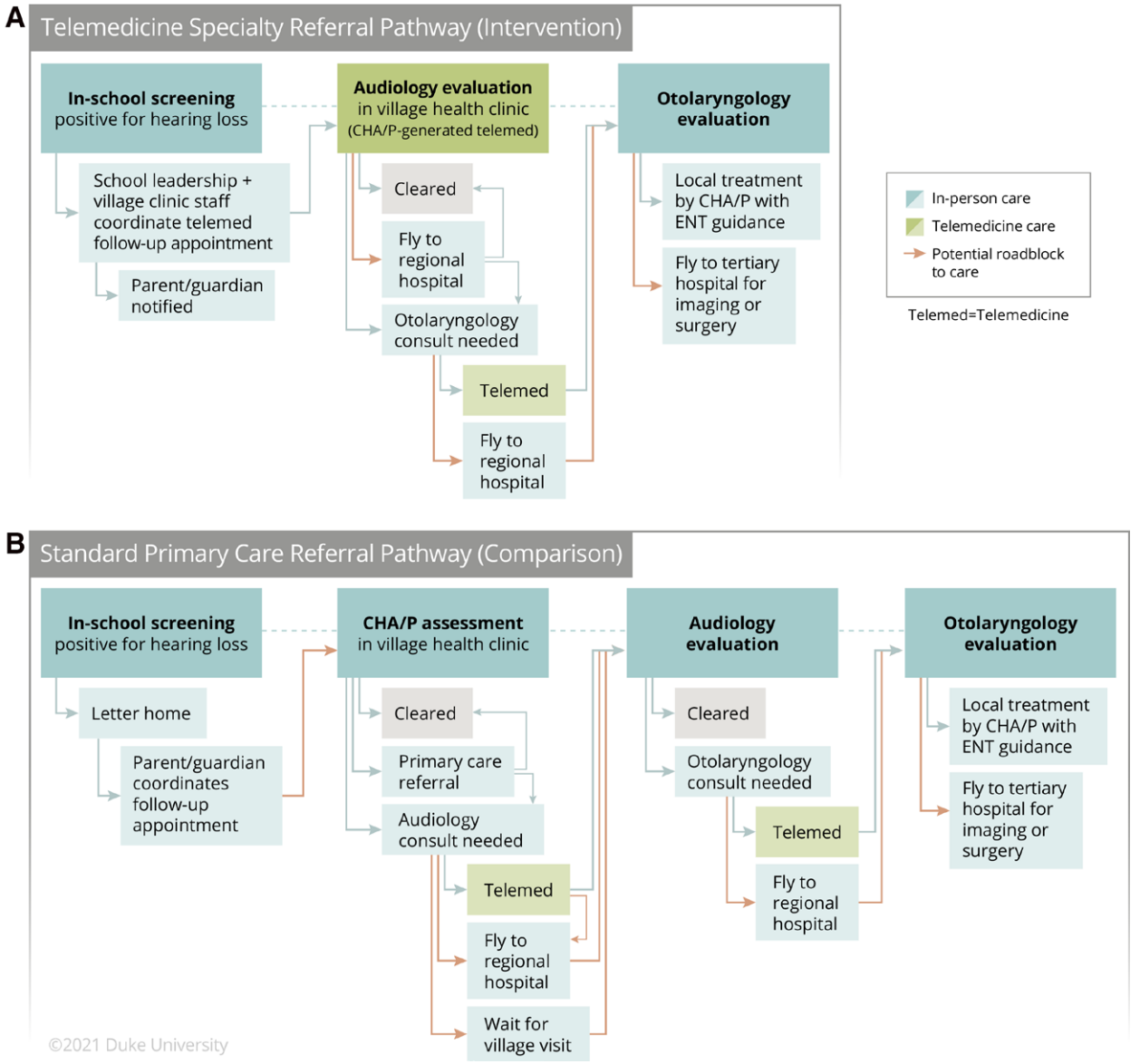

Fig. 1. Visual representation of telemedicine specialty referral and standard primary care referral pathways (similar figure previously published in Emmett et al., 2022, https://doi.org/10.1016/S2214-109X(22)00184-X, CC BY). CHA/P, Community Health Aide/Practitioner.

telemedicine referrals continued to be blinded to the intervention allocation, and clinical study team members abstained from consulting on any study-related cases for the ancillary trial as they did for the main trial. During the hearing screening data collection, all study team members, including those collecting mHealth screening and school staff collecting preschool hearing screening, were masked to other results obtained on the day of screening. Masking was achieved by not sharing screening results and spacing apart the different screening stations. Trial statisticians were not blinded to treatment allocation; however, they used the same analytic plan as was used for the main trial, which was blind to allocation.

Procedures

The preschool screen, the mHealth screen, and the benchmark audiometric assessment were all completed on the same day. Details on the screening and audiometric protocols have been previously published (Emmett, Robler, Wang, et al. 2019). Testing occurred in quiet locations in the preschool such as empty classrooms, libraries, or conference rooms. The preschool screening consisted of a distortion product of otoacoustic emissions screening using the Natus/Biologic AudX and was performed by school staff, typically teachers and support staff. Training was provided to teachers by school administration, and technical support was provided by audiology staff at the Norton Sound Health Corporation, as is standard practice. The screening protocol tested at 2 kHz, 3 kHz, 4 kHz, and 5 kHz, with pass-criteria defined as three out of four frequencies that met pre-determined criteria (6 dB SNR; Gorga et al. 1997). A referral was generated if one or both ears did not pass. The protocol did not include rescreening. The mHealth screening consisted of a validated, smartphone-based behavioral pure-tone hearing screening and a middle ear evaluation using tympanometry. This mHealth plus tympanometry screening was selected because of high rates of otitis media in the Alaska Native population (Reed et al. 1967; Kaplan et al. 1973; Curns et al. 2002; Singleton et

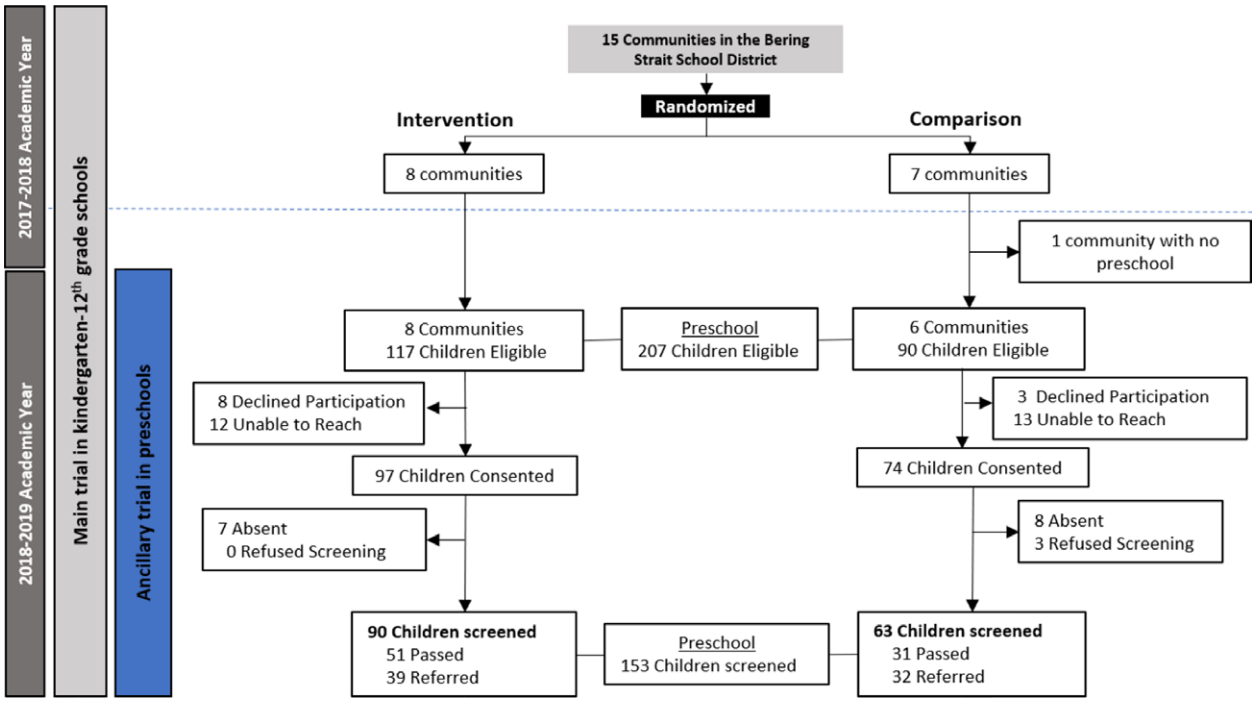

Fig. 2. The Consolidated Standards for Reporting Trials (CONSORT) diagram of participant flow during the study period.

al. 2009). The mHealth plus tympanometry screening was conducted by study team members who were not trained audiologists. These study team members received initial training from audiology study staff that included how to use the equipment, how to categorize tympanogram types, and how to perform basic troubleshooting. Screening included pure tones at 1 kHz, 2 kHz, 4 kHz, and 6 kHz at 20 dB HL using a validated android smartphone application with calibrated headphones (HearScreen by hearX Group, South Africa), and automated tympanometry to assess middle ear function (Otometrics Otoflex 100, Denmark). For the mHealth hearing screening, each participating child was conditioned to respond to a presented tone. A practice session was performed until the child demonstrated understanding of the task. If a child could not be conditioned, then results were marked as "could not be evaluated." At the end of the initial screening, frequencies without a response were rescreened. A referral for the mHealth plus tympanometry screening was generated if there was no response in either ear to any pure-tone frequency after rescreening or tympanometry resulting in either a flat (Type B) or negative pressure <-200 daPa (Type C) tympanogram (FitzZaland & Zink 1984). Children who could not be evaluated for pure-tone screening did not generate a referral but could still be referred for tympanometry, if indicated. The audiometric assessment consisted of audiometry, tympanometry, and digital otoscopy. Audiometric testing was conducted to provide a benchmark comparison for the screening protocols and was performed by trained audiologists. Diagnostic audiometry was performed using Shoebox Audiometry by Clearwater Clinical, which is an iPad-based audiometer using calibrated transducers. Air conduction thresholds were measured at 0.5 kHz, 1 kHz, 2 kHz, 4 kHz, 6 kHz, and 8 kHz for both ears, with masked bone conduction thresholds obtained for thresholds exceeding 25 dB HL to determine hearing loss type. Standard behavioral audiometry and conditioned play audiometry were used to assess hearing thresholds. If a child could not be conditioned to the task, the result was marked as "could not be evaluated." Tympanometry was performed using the Otometrics Otoflex 100 tympanometer and digital otoscopy using the Otometrics Otocam, a USB-based otoscope. A referral for the audiometric assessment was generated for pure-tone average >25 dB HL (0.5kHz, 1kHz, 2kHz) or any single frequency ≥ 30 dB HL in either ear, a flat (Type B) or negative pressure <-200 daPa (Type C) tympanogram in either ear, or digital otoscopy was positive for ear pathology as determined by the audiologist (e.g., occluding cerumen, retraction, effusion, acute otitis media, otorrhea, perforation, patent or plugged tube, external otitis, or foreign body). Children who could not be evaluated for pure-tone audiometry did not generate a referral but could still be referred for tympanometry or otoscopy, if indicated.

The telemedicine specialty referral (intervention) was the same as in the main trial (Emmett et al. 2022), which utilized existing telemedicine infrastructure in local village health clinics. The telemedicine referral was adapted to streamline documentation and clinical workflows while maintaining core billing requirements to facilitate sustainability, reducing the time required for ear/hearing-related follow-up from 60 to 90 minutes to 5 to 10 minutes. For communities randomized to telemedicine specialty referral, school leadership worked with local clinic staff to coordinate the telemedicine follow-up appointments for preschool children requiring referral. Parents were notified, typically by a phone call, that their child was referred and would be seen at the clinic for the telemedicine specialty referral. On the basis of community feedback before the trial, parents were encouraged but not required to accompany their child to the appointment, with the exception of children in grades 2 and younger (Robler et al. 2020). A Community Health Aide/Practitioner (CHA/P), who provides frontline care in local village health clinics (*Overview of the*

Alaska Community Health Aide Program, 2005), performed the asynchronous telemedicine referral to audiologists located at the regional hospital in Nome and otolaryngologists located at the tertiary hospital in Anchorage if surgical or complex medical management was required (see Fig. 1).

The standard primary care referral (comparison) represented standard practice in the region for all preschool programs and included a letter from the school sent home to parents of children who did not pass the hearing screening. The letter requested the family bring the child to the local clinic for further evaluation, at which point the child could be managed in one of several ways: referral to a primary care provider, wait for an audiologist to travel to their community for a field clinic or telemedicine consultation to audiology. Audiologists would consult otolaryngology as needed for surgical or complex medical management (see Fig. 1). As telemedicine is standard practice in this region, we did not restrict the use of telemedicine in standard primary care referral communities.

Trial Outcomes

Consistent with the main trial, the primary outcome for the ancillary trial was time to ear/hearing-related follow-up, measured in days from the date of hearing screening for referred children and collected through chart review of the multiorganizational shared electronic health record (EHR) utilized by Norton Sound Health Corporation and the tertiary referral hospital in Anchorage. This allowed us to ascertain whether any ear/hearing encounters occurred within either health system for the study population. Follow-up was defined as an ear/hearing encounter with a CHA/P, a primary care provider, audiologist, or otolaryngologist (established care pathways in rural Alaska), measured by the presence of any ear/hearing International Statistical Classification of Diseases, 10th Revision (ICD-10) diagnosis code in the EHR. A list of eligible codes can be found in a Table in Supplemental Digital Content 1, http://links.lww.com/EANDH/B125. Presence of such codes could indicate further evaluation that resulted in a child being cleared or a formal diagnosis of an ear/hearing condition. The primary outcome represents time to follow up for the two possible referral pathways: the telemedicine specialty referral or the standard primary care referral. The primary outcome of ear/hearing follow-up was determined by chart review of the EHR for 9 months (275 days) from screening date. The secondary outcome was presence of any ear/hearing follow-up from screening date to 9 months for referred children. The secondary outcome was added after trial registration to provide additional context to the primary outcome, as well as to allow comparability of study outcomes with previous screening studies which commonly report follow-up as a binary outcome.

Statistical Analysis

Preliminary evidence of shortened wait times using telemedicine included all age groups, not just school-aged children (Kokesh et al. 2004, 2011; Hofstetter et al. 2010). We therefore anticipated that the telemedicine specialty referral intervention would work the same way for preschool children as it did for older school-aged children. Assuming an intraclass correlation coefficient (ICC) of 0.05 and SD of 3 weeks, a coefficient of variation of cluster size of 0.94, and total N = 150 over 14 clusters with 40% hearing screening referrals, there would be 80% power to detect a difference in mean reduction in time to diagnosis of 2.7 weeks between arms. Power calculations were performed using PASS version 11 software (Hintze 2011).

We conducted analyses using an intention-to-treat approach based on a prespecified analysis plan. Intention-to-treat is an analytical approach used in randomized trials that evaluates outcomes by the treatment arm (in this case follow-up pathway) originally assigned at randomization, regardless of treatment received (if any; McCoy 2017). Intention-to-treat analysis was used because it preserves randomization and gives a pragmatic estimate of the treatment effect (follow-up).

Individual-level sociodemographic characteristics (self-reported from parental surveys of study participants) and community-level characteristics were compared to assess baseline balance between telemedicine specialty referral and standard primary care referral communities, with continuous variables summarized using medians with interquartile range (IQR) and means with SDs and categorical variables summarized using frequencies and percentages.

Cumulative incidence ear/hearing-related follow-up over the 9 months (275 days) from the date of screening was visualized for both the telemedicine specialty referral pathway and the standard primary care referral pathway using the Kaplan-Meier (KM) product-limit estimates (Kaplan & Meier 1958). Median time to follow-up was computed with KM-associated upper and lower confidence limits at the 50th percentile. We pre-specified 1-, 2-, and 3-months (operationalized as 30, 60, and 90 days) as endpoints of interest in addition to the full 9-month (275-day) follow-up for all analyses. A longer time period was used to evaluate ear/hearing-related follow-up as there is little evidence to define when follow-up typically occurs and work within this region of rural Alaska indicated follow-up could take multiple months (Hofstetter et al. 2010). Shorter pre-specified periods were evaluated to better understand patterns to follow-up care and because ultimately, timely ear/hearing follow-up is essential.

Because of the presence of censoring in the measurement of time to follow-up (i.e., because there is no time to follow-up for those who do not receive follow-up within 275 days) and skew in the distribution of time to follow-up for those that did receive ear/hearing-related follow-up, accelerated failure time (AFT) models using lognormal time distribution was used to estimate between-arm ratios of time to follow-up (Wang 2006; Crowther 2019). This was done for the primary time point of interest (275 days post-screening) and each of the secondary time points of interest (30-, 60-, and 90-day). Children not followed up in either arm were right-censored at 275 days (or the corresponding earlier endpoint). A value of 0.5 days was added to the time to follow-up of any child who was seen in the health system on the same day as screening. Random intercepts were used to account for within-community correlation in the outcome because of the cluster-randomized design. Unadjusted and adjusted effect estimates were computed, with unadjusted using only treatment assignment and an indicator for stratum. Adjusted analyses additionally accounted for age (range 3 to 5 years), sex, and highest education level of the primary caregiver (≤12th high school diploma or General Educational Development versus some college or a college degree). The adjusted treatment effect was the primary estimate of interest because of the concern that randomizing a small number of clusters may result in baseline imbalances in individual characteristics and to enhance precision of the estimated treatment effect (Kahan et al. 2014) with

all adjustment variables prespecified. Regression parameters were exponentiated to give the interpretation of event time ratios (ETRs), with the standard primary care pathway in the numerator for interpretability. An ETR greater than one would indicate that participants in the standard primary care referral pathway took longer to receive follow-up than those in the telemedicine specialty referral pathway, while an ETR less than one would indicate that participants in the standard primary care referral pathway were seen sooner than those in the telemedicine specialty referral pathway.

Secondary analysis estimated differences in probability of any ear/hearing-related follow-up by treatment arm during each pre-specified follow-up time point using risk ratios (RR) and risk differences (RD). RR quantifies the ratio of the probability of having ear/hearing-related follow-up for the participants in the intervention arm to the probability of follow-up in the comparison arm, while the RD quantifies the absolute difference. Widely accepted reporting guidelines recommend that both RRs and RDs be reported for trials with binary outcomes (Schulz et al. 2010), which helps guard against misinterpretation of effect sizes (Turner et al. 2021). RRs greater than one (or RDs greater than zero) indicate increased ear/hearing-related follow-up for the telemedicine specialty referral pathway versus the standard primary care referral pathway. A modified Poisson regression (Poisson distribution with log link and robust standard errors [SEs]) was used to estimate RRs (Yelland et al. 2011), while Gaussian distribution and identity link were used to estimate RDs (Huang, 2021). Binomial models or modified Poisson regressions with identity link were the preferred specifications for estimation of RDs; however, lack of convergence necessitated the use of linear probability models (i.e., Gaussian distribution with identity link). Because of concerns about model misspecification to estimate RDs, sensitivity analysis estimated RDs by applying the delta method to estimates from the modified log-Poisson model. All models were estimated using generalized estimating equations (GEE) with independent working correlation matrices to account for the cluster randomized design and robust SEs to account for model misspecification. Independent correlation structure was chosen because of variable cluster sizes (Sullivan Pepe & Anderson 1994). As with the AFT models, unadjusted and adjusted treatment effects were computed with adjusted effect considered primary.

With a relatively small number of clusters (k = 14), inflated type 1 error was a concern. Therefore, permutation tests (Heß 2017; Wang & De Gruttola 2017) were used to obtain *p* values for the primary and secondary outcomes. To obtain confidence intervals (CI) for effect estimates, Kauermann-Carroll (KC) corrections (Kauermann & Carroll 2001) were used to compute SEs for the treatment effects for all binary outcome models and the between-within adjustment of denominator degrees of freedom (Li & Redden 2015) was used for analyses of time to follow-up.

Clustering by communities for log time to ear/hearing-related follow-up was quantified as an ICC and calculated (with 95% CIs) from the variance of the random effects and scale parameter. However, with a small number of clusters on which to base the calculation, the measure is likely to be imprecise and potentially biased. The ICC and 95% CIs for the binary secondary outcome of any follow-up was computed using an analysis of variance estimator (Wu et al. 2012) on cluster-level proportions.

A subgroup analysis was prespecified to examine heterogeneity of treatment effects (HTE) according to a child's hearing management status, operationalized as one of three levels: under active management for hearing-related conditions (ear/hearing follow-up within 3 months before screening day or wearing a hearing aid on screening day), under previous management (no ear/hearing follow-up in 3 months prior but with an ear/hearing follow-up greater than 3 months and less than 5 years from the date of screening), or never managed (no ear/hearing follow-up found in the 5 years before screening).

With only one primary hypothesis and one secondary hypothesis of interest, no adjustment for multiple comparisons was made. *P* values are presented only for the primary outcome and secondary outcome. All other computed effect estimates and 95% CIs should be considered exploratory.

Analyses adhered to the Consolidated Standards for Reporting Trials guidelines for cluster trials (Campbell et al. 2012) and were performed using Stata Software version 17 (StataCorp 2021).

RESULTS

Participants

During the 2018–2019 academic year, total preschool enrollment was 207. Within this eligible population, 153 (73.9%) children were screened, including 90 in telemedicine specialty referral communities and 63 in standard primary care referral communities (Fig. 2). Of those screened, 71 (46.4%) required referral, including 39 (43.3%) in telemedicine specialty referral communities and 32 (50.8%) in standard primary care referral communities. Baseline characteristics were similar, with a slightly lower proportion of females (41.0% versus 50.0%) and a higher proportion of children with hearing loss (30.8% versus 10%) in the telemedicine specialty referral communities compared with standard primary care referral communities (Table 1).

Effects of the Intervention

Among children who received follow-up, median time to follow-up was 28 days (IQR: 15 to 71) in telemedicine specialty referral communities compared with 85 days (IQR: 26 to 129) in standard primary care referral communities. Mean time to follow-up for all referred children was 4.5-fold faster (ETR = 4.5; 95% CI, 1.8 to 11.4; $p = 0.045$) in telemedicine specialty referral communities compared with standard primary care referral communities in the 9-month follow-up time frame (Fig. 3). Of the 71 children who were referred, 30 (76.9%) of 39 in telemedicine specialty referral communities received follow-up within 9 months (275 days) compared with 16 (50.0%) of 32 in primary care referral communities (Table 2).

Children in telemedicine specialty referral communities were 57% more likely (RR = 1.57, 95% CI, 1.22 to 2.01; $p = 0.028$) than children in standard primary care communities to receive follow-up by the 9-month follow-up endpoint, corresponding to an RD of 28.4 percentage points (pp) (95% CI, 13.8 to 43.1pp) (Table 2). RRs ranged from 1.57 to 3.31 and 28.4 to 41.2 for RDs, with the size of the effect varying substantially with the timepoint for follow-up. The largest risk ratio for the proportion followed-up was at 60 days (RR = 3.31; 95% CI, 1.60 to 6.84), indicating that the majority of follow-up in telemedicine specialty referral communities occurred during

TABLE 1. Demographic and clinical characteristics at time of screening for referred children

	Comparison (N = 32)	Intervention (N = 39)
Sociodemographics*		
Age (years)		
Median (Q1, Q3)	4.0 (3.0, 4.0)	4.0 (3.0, 4.0)
Age		
3 years	12 (37.5%)	13 (33.3%)
4 years	15 (46.9%)	19 (48.7%)
5 years	5 (15.6%)	7 (17.9%)
Female	16 (50.0%)	16 (41.0%)
American Indian or Alaska Native	>90%	>90%
Highest education level of any caregiver		
High school diploma or less	23 (71.9%)	26 (66.7%)
Some college or college degree	9 (28.1%)	13 (33.3%)
Clinical characteristics		
Hearing loss severity†		
No hearing loss in either ear	18 (90.0%)	18 (69.2%)
Mild (PTA >25-40dB)	2 (10.0%)	8 (30.8%)
Missing	12	13
Middle ear disease (any ear)‡	16 (53.3%)	21 (56.8%)
Missing	2	2
Ear/hearing-related healthcare management		
Never managed	8 (25.0%)	11 (28.2%)
Previously managed§	17 (53.1%)	19 (48.7%)
Currently managed¶	7 (21.9%)	9 (23.1%)
Number of clusters and cluster size summaries		
Enrolled in Study	(n = 63)	(n = 90)
Mean cluster size (SD)	10.5 (5.9)	11.3 (6.9)
Median cluster size (Q1, Q3)	13 (5, 13)	11 (7, 14)
Analyzed	(n = 32)	(n = 39)
Mean cluster size (SD)	5.3 (3.8)	4.9 (2.9)
Median cluster size (Q1, Q3)	6 (2, 8)	5 (3, 7)
Number of clusters with enrolled children	6	8
Number of clusters with referred children	5	8

PTA: Pure-tone average.
**Self-reported by parent/guardian of participant.*
†Based on World Health Organization (WHO) definition and assessed via audiometric evaluation.
‡Assessed via tympanometry and/or otoscopy during audiometric evaluation.
§Defined as having audiology or otolaryngology encounter >3 months and <5 years from date of screening and not under active management.
¶Defined as having audiology or otolaryngology encounter within 3 months before screening day or wearing a hearing aid on screening day.

the 60-day period and follow-up became similar between the two arms after 60 days (Fig. 3). Alternative computation of RDs using the delta method can be found in Table ST1, Supplemental Digital Content 2, http://links.lww.com/EANDH/B126.

Calculation of the intracluster correlation coefficient for time to ear/hearing-related follow-up and presence of an ear/hearing-related follow-up (at 275 days) was unstable because of a low calculated variance on the random effect with estimate and 95% CI close to zero and thus are not reported. Calculated ICC for the binary outcome of any ear/hearing-related follow-up was 0.040 (95% CI, 0.000 to 0.226) (see Table ST2 in Supplemental Digital Content 2 http://links.lww.com/EANDH/B126).

Heterogeneity of Treatment Effects (HTE)

During the 9-month follow-up period, 72.7% versus 37.5% of never managed children had ear/hearing-related follow-up in telemedicine specialty referral versus standard primary care referral communities, respectively. Similar proportions for the previously managed children received follow-up, whereas nearly all currently managed children were followed up (77.8% in telemedicine specialty referral communities and 100% in standard primary care referral communities; Table ST3, Supplemental Digital Content 2, http://links.lww.com/EANDH/B126). Proportion of children followed-up by 275 days in telemedicine specialty referral communities versus proportion followed-up in standard primary care referral communities was similar between never managed (RR = 2.1; 95% CI, 0.8 to 5.3) and previously managed children (RR = 2.2; 95% CI, 1.2 to 3.9) compared with those who were actively managed (RR = 0.8; 95% CI, 0.6 to 1.2). Never-managed children in telemedicine specialty referral communities had relatively faster follow-up (ETR = 7.1; 95% CI, 1.3 to 40.2) compared with actively managed children (ETR = 2.0; 95% CI, 0.4 to 10.4) (Fig. 4). Small sample size limited the ability to test such differences with adequate precision, thus the HTE analyses must be interpreted with caution and are strictly exploratory. Treatment effect estimates by subgroup can be found in Tables ST4 and ST5, in Supplemental Digital Content 2, http://links.lww.com/EANDH/B126).

DISCUSSION

In this ancillary cluster-randomized controlled trial, telemedicine specialty referral improved time to follow-up and

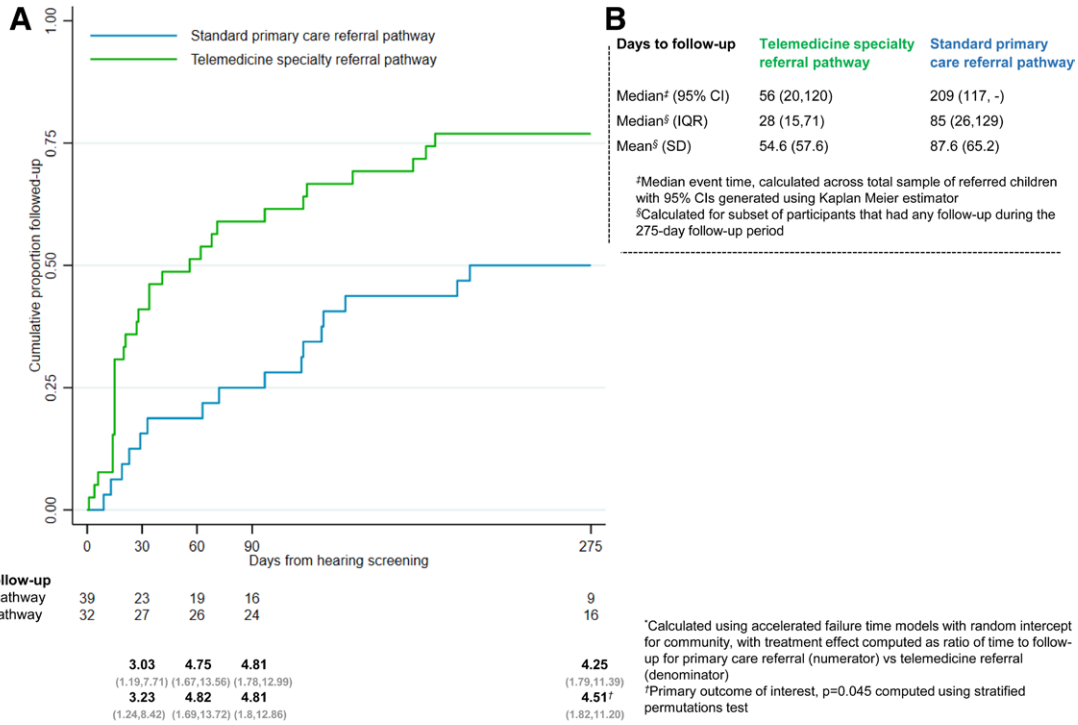

Fig. 3. Kaplan-Meier Product Limit Estimates and event time ratios (A), and summary statistics (B) for time to first ear/hearing follow-up after screening referral in telemedicine specialty referral pathway versus standard primary care referral pathway. CI, confidence interval; IQR, interquartile range.

TABLE 2. Regression estimates for difference in secondary outcome of any ear/hearing-related follow-up for children who are referred in telemedicine specialty referral pathway (intervention) versus standard primary care referral pathway (comparison) at 1-month, 2-months, 3-months, and 9-months

	Any ear/hearing-related follow-up		Any ear/hearing-related follow-up (Intervention/Comparison)		Any ear/hearing-related follow-up (Intervention-Comparison)	
	Sample Proportions, N (%)		Risk Ratio*, (95% CI)		Risk Difference†, (95% CI)	
Timepoint for Follow-Up	Comparison (n = 32)	Intervention (n = 39)	Unadjusted‡ (n = 71)	Adjusted‡ (n = 71)	Unadjusted‡ (n = 71)	Adjusted‡ (n = 71)
30 days	5 (15.6)	16 (41.0)	2.66 (0.90 to 7.87)	3.01 (1.45 to 6.25)	27.0 (-10.0 to 63.9)	28.6 (-8.1 to 65.2)
60 days	6 (18.8)	20 (51.3)	3.05 (1.75 to 5.31)	3.31 (1.60 to 6.84)	37.2 (10.5 to 63.8)	38.5 (13.2 to 63.9)
90 days	8 (25.0)	23 (59.0)	2.73 (1.72 to 4.35)	2.80 (1.85 to 4.23)	40.4 (10.1 to 70.8)	41.2 (11.0 to 71.4)
275 days	16 (50.0)	30 (76.9)	1.58 (1.15 to 2.16)	1.57 (1.22 to 2.01)§	28.7 (9.7 to 47.7)	28.4 (13.8 to 43.1)

Computed using generalized estimating equations with Poisson distribution and log link (exponentiated parameters); 95% confidence intervals incorporate Kauermann-Carroll (KC) correction to SEs.
†Computed using generalized estimating equations with Gaussian distribution and identity link; 95% confidence intervals incorporate Kauermann Carroll correction to SEs.
‡Unadjusted regressions include treatment and strata indicators only; adjusted include female sex, age (range 3–5 years), and highest educational attainment by any parent/guardian (≤high school diploma/GED or at least some college).
§Secondary outcome of interest, p = 0.028 computed using stratified permutation test.

reduced loss to follow-up compared with standard primary care referral for preschool hearing screening in rural northwest Alaska. To date, little evidence exists for the use of telemedicine for prevention in the preschool population, and there is no present evidence from rural areas. While most evidence in this area currently comes from school-based programs (Reynolds & Maughan 2015), there are a few examples of telemedicine models applied to early childhood education programs in an urban setting. One example is McConnochie et al. (2005), who evaluated the impact of telemedicine on common acute problems such as absenteeism. However, there is no literature to date that has evaluated telemedicine for preventive health services, specifically involving specialty care, in the preschool population.

This ancillary trial is the first to apply telemedicine specialty referral for hearing screening in rural preschool programs. While most preschool programs are mandated to conduct preventive health screenings such as vision and hearing, little evidence exists for how well children are subsequently identified and managed. In a handful of studies evaluating hearing screening in the preschool population, loss to follow-up ranged from 20-79% in both rural and urban settings (Allen et al. 2004; Serpanos & Jarmel 2007; Wu et al. 2014). Results from this trial found 50% of preschool children in the primary care referral pathway were lost to follow-up, compared with 23% in the telemedicine specialty referral pathway.

Similar to the main trial, the strengths of this ancillary trial included participation from all preschool programs in the region

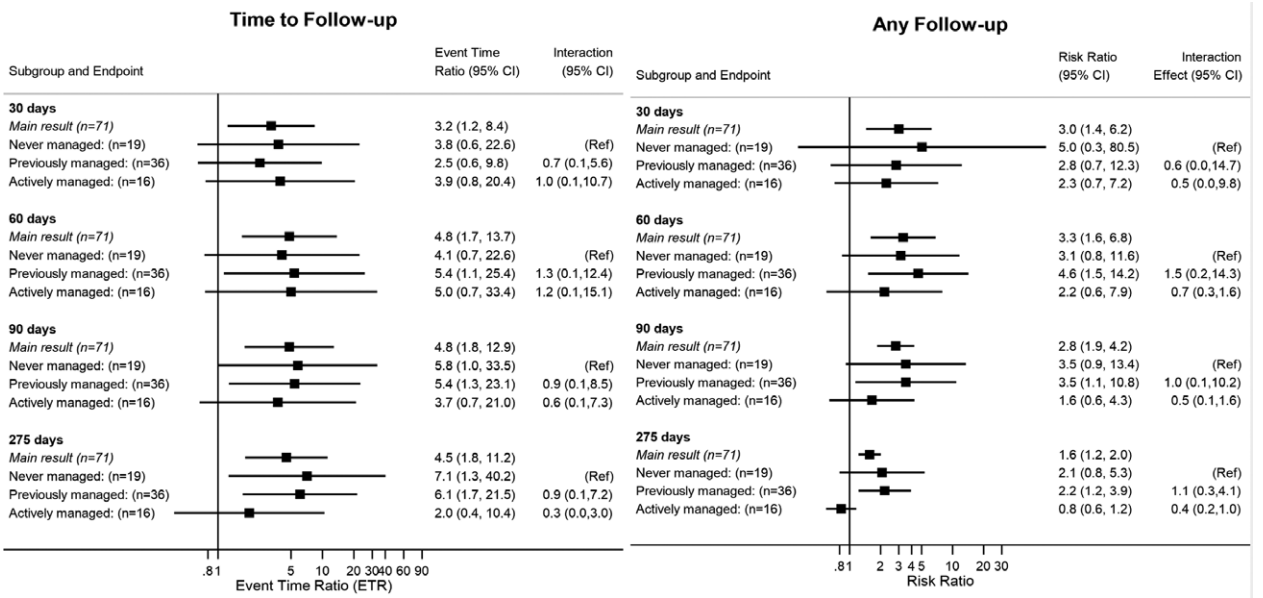

Fig. 4. Heterogenous treatment effect (HTE) estimates of event time ratios and risk ratios for time to follow-up and any ear/hearing-related follow-up for referred children in telemedicine specialty referral communities (intervention) versus standard primary care referral communities (comparison), by management status. CI, Confidence interval; ETR, event time ratio.

with strong community engagement. A focus on sustainable referral pathways coordinated through the school and clinic, as well as the use of existing infrastructure, increases the likelihood of local adoption and implementation.

This ancillary trial has several limitations. First, the sample was limited in size. Despite this, 74% of the eligible population participated, allowing for good representation of the preschool population in the region. Second, randomization for the main trial was maintained for the ancillary trial. This prevented concealment of randomization before enrollment. Third, while the use of existing telemedicine infrastructure in a Tribal healthcare setting facilitated integration of the study into routine clinical care, it limits generalizability to other settings that may include multiple healthcare organizations and more variation in insurance coverage. Lastly, use of the established telemedicine network within the clinic required coordination between staff at the local school and clinic to complete the telemedicine intervention. Many communities had to complete the preventive telemedicine specialty referrals amongst influenza outbreaks, urgent clinical care needs, and staff shortages, resulting in variations in how well the intervention worked across the region. Despite these challenges; however, results from this trial indicate the telemedicine specialty referral improved time to follow-up and reduced loss to follow-up compared with the standard primary care referral pathway. Future work should focus on integrating the telemedicine specialty referral pathway directly into the school to further reduce barriers and increase generalizability to other rural environments.

In this ancillary trial, over 20% of the sample of referred children had hearing loss and greater than 50% had middle ear disease. The effects of unmanaged hearing loss and ear disease in preschool children can be significant and are further compounded in rural areas where access to healthcare is more difficult (Idstad & Engdahl 2019; Kingsbury et al. 2022; Schuh & Bush 2022). Results from the trial suggest that the use of telemedicine for specialty referral after preschool hearing screening increased the likelihood and speed with which a child receives ear/hearing-related follow-up. Preschool programs represent an essential access point for preventive services for preschool children, particularly for those living in rural areas. This trial provides early proof-of-concept evidence for the use of telemedicine referral for specialty access after hearing screening to ensure preschool children requiring additional testing receive the necessary care.

CONCLUSIONS

Telemedicine specialty referral improved time to follow-up and reduced loss to follow-up for referred hearing screening in preschool programs in rural northwest Alaska. Telemedicine represents a mode of preventive healthcare delivery that can improve access to ear and hearing care for preschool children in rural environments.

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S.E. and S.K.R. (Principal Investigators) 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. S.D.E., A.P., E.L.T., J.J.G., A.L., N.-Y.W., and S.K.R. involved in study concept and design.

S.D.E., M.I.-J., C.D.J., P.H., and S.K.R. participated in acquisition of data. S.D.E., A.P., E.L.T., J.J.G., N.-Y.W., K.L.H., and S.K.R. participated in

analysis and interpretation of data. K.L.H., S.D.E., and S.K.R. involved in literature search. S.D.E., A.P., and S.K.R. involved in drafting the article. S.D.E., A.P., E.L.T., J.J.G., A.L., M.I.-J., C.D.J., P.H., N.-Y.W., K.L.H., and S.K.R. participated in critical revision of the article for important intellectual content. A.P. and E.L.T. involved in statistical analysis. S.D.E. and S.K.R. obtained funding. S.D.E. and S.K.R. involved in study supervision.

De-identified participant data, data dictionary, study protocol, statistical analysis plan, and informed consent form will be made available beginning 6 months and ending 5 years after article publication to researchers upon request, pending Tribal approval and possible execution of a data use agreement, as needed. Proposals should be directed to Dr. Samantha Kleindienst Robler at skleindienst@NSHCORP.ORG.

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