Prevalence of Childhood Hearing Loss in Rural Alaska

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Objectives: Childhood hearing loss has well-known lifelong consequences. Certain rural populations are at higher risk for infection-related hearing loss. For Alaska Native children, historical data on hearing loss prevalence suggest a higher burden of infection-related hearing loss, but updated prevalence data are urgently needed in this high-risk population.

Design: Hearing data were collected as part of two school-based cluster-randomized trials in 15 communities in rural northwest Alaska over two academic years (2017–2019). All enrolled children from preschool to 12th grade were eligible. Pure-tone thresholds were obtained using standard audiometry and conditioned play when indicated. The analysis included the first available audiometric assessment for each child (n = 1634 participants, 3 to 21 years), except for the high-frequency analysis, which was limited to year 2 when higher frequencies were collected. Multiple imputation was used to quantify the prevalence of hearing loss in younger children, where missing data were more frequent due to the need for behavioral responses. Hearing loss in either ear was evaluated using both the former World Health Organization (WHO) definition (pure-tone average [PTA] > 25 dB) and the new WHO definition (PTA ≥ 20 dB), which was published after the study. Analyses with the new definition were limited to children 7 years and older due to incomplete data obtained on younger children at lower thresholds.

Results: The overall prevalence of hearing loss (PTA > 25 dB; 0.5, 1, 2, 4 kHz) was 10.5% (95% confidence interval [CI], 8.9 to 12.1). Hearing loss was predominately mild (PTA >25 to 40 dB; 8.9%, 95% CI, 7.4 to 10.5). The prevalence of unilateral hearing loss was 7.7% (95% CI, 6.3 to 9.0). Conductive hearing loss (air-bone gap of ≥ 10 dB) was the most common hearing loss type (9.1%, 95% CI, 7.6 to 10.7). Stratified by age, hearing loss (PTA >25 dB) was more common in children 3 to 6 years (14.9%, 95% CI, 11.4 to 18.5) compared to children 7 years and older (8.7%, 95% CI, 7.1 to 10.4). In children 7 years and older, the new WHO definition increased the prevalence of hearing loss to 23.4% (95% CI, 21.0 to 25.8) compared to the former definition (8.7%, 95% CI, 7.1 to 10.4). Middle ear disease prevalence was 17.6% (95% CI, 15.7 to 19.4) and was higher in younger children (23.6%, 95% CI, 19.7 to 27.6) compared to older children (15.2%, 95% CI, 13.2 to 17.3). High-frequency hearing loss (4, 6, 8kHz) was present in 20.5% (95% CI, 18.4 to 22.7 [PTA >25 dB]) of all children and 22.8% (95% CI, 20.3 to 25.3 [PTA >25 dB]) and 29.7% (95% CI, 27.0 to 32.4 [PTA ≥ 20 dB]) of children 7 years and older (limited to year 2).

Conclusions: This analysis represents the first prevalence study on childhood hearing loss in Alaska in over 60 years and is the largest cohort with hearing data ever collected in rural Alaska. Our results highlight that hearing loss continues to be common in rural Alaska Native children, with middle ear disease more prevalent in younger children and high-frequency hearing loss more prevalent with increasing age. Prevention efforts may benefit from managing hearing loss type by age. Lastly, continued research is needed on the impact of the new WHO definition of hearing loss on field studies.

Key words: Alaska, Child health, Healthcare disparities, Hearing loss, Prevalence, Rural health.

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IDEA

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INTRODUCTION

Childhood hearing loss has well-known, lifelong consequences for speech and language development, school achievement, future employment opportunities, and quality of life (Järvelin et al. 1997; Bess et al. 1998; Wake et al. 2004; Khairi Md Daud et al. 2010; Emmett & Francis, 2015; Tomblin et al. 2015; Roland et al. 2016). An estimated 1.6 billion people are affected by hearing loss worldwide, including 70 million children (GBD 2019 Hearing Loss Collaborators 2021). The World Health Organization (WHO) estimates that 60% of childhood hearing loss is preventable, and this estimate rises as high as 75% in low-resource settings where infection-related hearing loss is common (Krug et al. 2016). Policymakers addressing this public health challenge need up-to-date prevalence data to quantify the burden of disease, highlight potential strategies for prevention, and guide the allocation of adequate resources for early identification and treatment.

Certain populations are at particularly high risk for infection-related hearing loss, including Alaska Native and American Indian children in the US and indigenous children in other countries (Curns et al. 2002; Singleton et al. 2009; Smith & Boss 2010; Jervis-Bardy et al. 2014). Historically, the prevalence of

hearing loss in Alaska Native children has been estimated to be up to 31%, compared to 1.7 to 5% prevalence among the general US child population (Reed et al. 1967; Mehra et al. 2009). However, Alaska prevalence data are over 50 years old, and much has changed during this time. Telehealth has become widely available for ear and hearing care in rural Alaska, expanding access to rural, difficult-to-reach areas of the state (Kokesh et al. 2004; Hofstetter et al. 2010; Kokesh et al. 2011; Carroll et al. 2011). Additionally, the introduction of pneumococcal vaccination, including the 7-valent (PCV7) and 13-valent (PCV13) pneumococcal conjugate vaccines, has reduced the frequency of otitis media across the US (Singleton et al. 2009, 2018). There is nevertheless still evidence of a higher burden of otitis media among rural Alaska Native children, with otitis media rates 3-fold higher than in the general US population despite similarly high rates of PCV13 vaccination (Singleton et al. 2018). Updated prevalence data are urgently needed to characterize the current landscape of childhood hearing loss in this high-risk population.

We report hearing loss prevalence in the Bering Strait region of rural, northwest Alaska from Hearing Norton Sound, a cluster-randomized trial (2017–2019) evaluating mHealth screening tools and telemedicine specialty referral for school hearing screening in a kindergarten-12th grade (K-12) population (Emmett et al. 2019a; Emmett et al. 2019b; Emmett et al. 2022). Based on the request of participating communities, an ancillary trial was launched in the second academic year of the main trial to include preschool-aged children. This prevalence analysis, therefore, spans both the K-12 and preschool trials, to include 1634 children ages 3 to 21 years. To enhance comparability to studies in other populations, the trials were designed and carried out using the WHO definition of hearing loss (pure-tone average [PTA] >25 dB of 0.5, 1, 2, and 4kHz) applied to either ear. However, in March 2021 the WHO published the World Report on Hearing, which reduced the cutoff for hearing loss to PTA ≥20 dB. We have therefore included both definitions in this analysis to maintain consistency with the original study design while simultaneously providing results that can be compared to future studies.

MATERIALS AND METHODS

Study Sample

The study sample was derived from two cluster-randomized school-based trials in rural Alaska with K-12 and preschool children. A total of 1634 participants, aged 3 to 21 years, were included in the sample, representing 71% of the target population of 2299 eligible children (Fig. 1).

Briefly, the Hearing Norton Sound trial was a parallel two-arm cluster-randomized, controlled trial conducted over two academic years (2017–2018, 2018–2019) designed to evaluate telemedicine specialty referral compared to standard primary care referral for school hearing screening (Emmett et al. 2019a; Emmett et al. 2019b). The trial was conducted in the 15 rural communities of the Bering Strait region of northwest Alaska. This region, which spans 23,000 square miles, is only accessible by plane. Over 95% of residents are Alaska Native, and Yup'ik, Iñupiaq, and Siberian Yup'ik are the primary heritages represented in the population. The 15 rural communities in the region are served by a single school district, Bering Strait School District (BSSD), and Norton Sound Health Corporation (NSHC), a Tribal Health Organization, is the source of healthcare. The ancillary preschool trial was added in the second academic year of the main trial (2018–2019) based on community input requesting the inclusion of early childhood education. The ancillary trial design was analogous to the main trial, with all children enrolled in preschool in the region eligible to participate. Of the 15 communities in the main trial, 14 communities had preschools and participated in the ancillary trial. Both trials were approved by the Institution Review Boards of Alaska Area, NSHC, and Duke University and were registered on Clinicaltrials.gov (NCT03309553, NCT03662256).

Measurements

Data collection for the trials has been previously described (Emmett et al. 2019b). In short, all children who were present on school screening day with signed parental consent and child assent were enrolled. Children could participate even if they were eligible in only one of the academic years of the trial. Enrolled children underwent a comprehensive audiometric assessment on screening day. These evaluations were completed by trained audiologists on the study team in a quiet room (e.g., unused classroom) in the school. Parents of enrolled children completed a sociodemographic questionnaire. If more than 10% of children in a given school were absent on screening day, a make-up day was scheduled to screen children who were initially absent.

Audiometric Protocol

The comprehensive audiometric assessment consisted of air- and bone conduction audiometry with a validated tablet-based audiometer (Thompson et al. 2015; Saliba et al. 2017) and supra-aural earphones (Shoebox, Clearwater Clinical, Canada), diagnostic tympanometry (Otometrics Otoflex 100, Denmark), and digital otoscopy (Otocam, Otometrics, Denmark). The audiometric assessment (pure-tone testing, tympanometry, otoscopy) was performed by a trained audiologist using standard methods. Air conduction audiometry was performed at 0.5, 1, 2, and 4 kHz, and thresholds were obtained using the standard Hughson-Westlake method. Children were given verbal instructions to raise their hand when they heard a beep, and a practice tone was given at 1000 Hz to confirm understanding. If a child was unable to condition to the task, particularly for children in preschool and kindergarten, the audiologist switched to conditioned play audiometry using appropriate toys found in the school (e.g., puzzle, blocks/bears with bucket). Bone conduction testing was performed for any corresponding air conduction threshold ≥25 dB. Masking was completed for both air and bone conduction testing when indicated (>40 dB difference between ears for air conduction and ≥10 dB air-bone gap for bone conduction for each frequency tested). In the second academic year of the trial (2018–2019), as well as in the ancillary trial (2018–2019), 6 and 8 kHz were added to the air conduction audiometric protocol to evaluate for high-frequency hearing loss. Audiometric equipment was calibrated annually per standard practice. Testing took place in quiet rooms during school hours, such as an empty classroom, library, or office. Rooms were marked for hearing testing, and testing was paused if noise was present. Noise levels were monitored using built-in technology that alerted the tester if excessive noise was present.

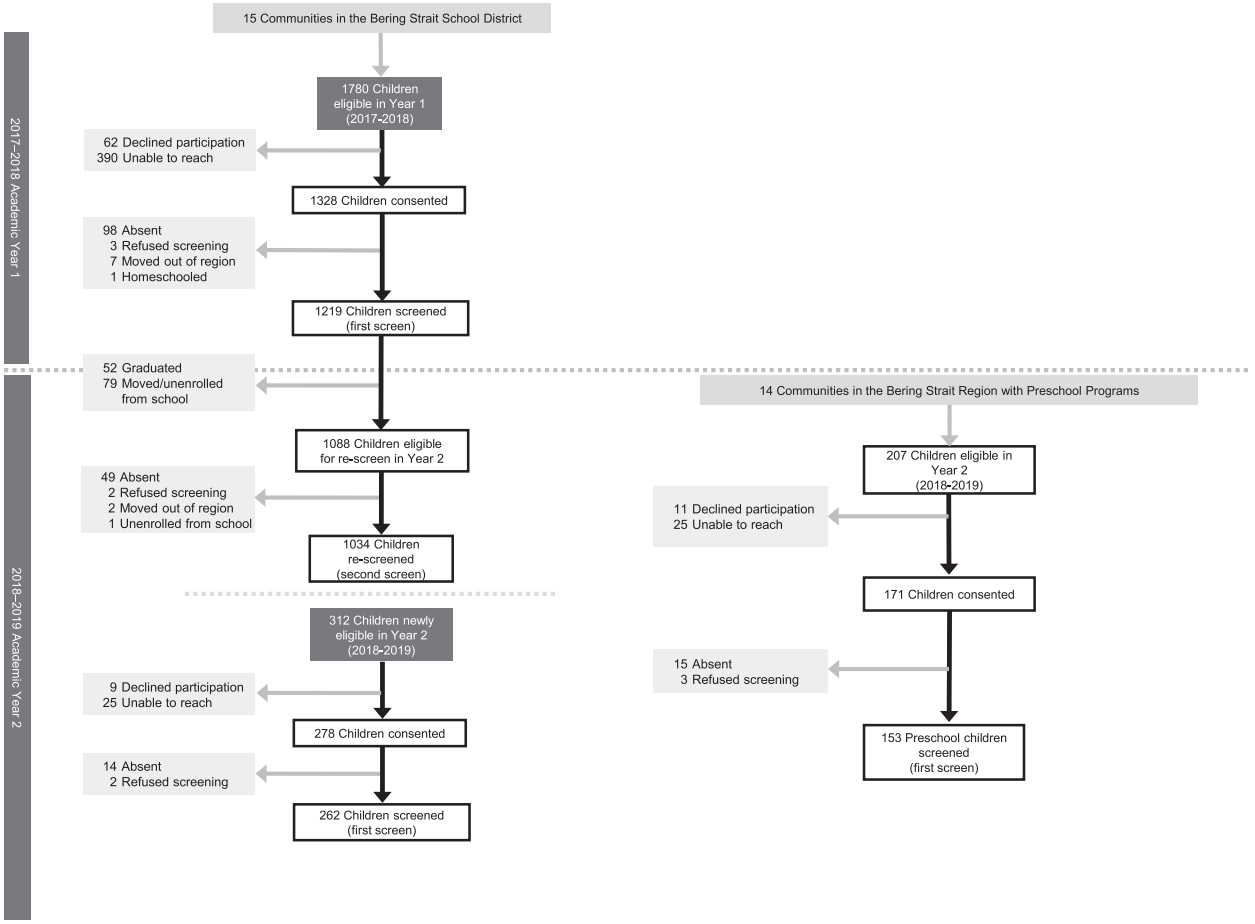

Fig. 1. Flow diagram.

Definitions

A complete list of hearing loss definitions is available in Table 1 in Supplemental Digital Content 1, http://links.lww.com/EANDH/B124. Briefly, hearing loss was defined for either ear based on the new WHO criteria of PTA (0.5, 1, 2, 4 kHz) ≥20 dB. Data were also analyzed using the former WHO criteria of PTA (0.5, 1, 2, 4 kHz) >25 dB with which the study was originally designed. High-frequency hearing loss was defined with both 20 dB and 25 dB criteria using a high-frequency PTA (4, 6, 8 kHz) or a single threshold of ≥30 dB at 6 or 8 kHz in either ear. Children that met the hearing loss criteria were categorized into sensorineural, conductive, or mixed hearing loss types for each individual frequency (0.5, 1, 2, 4 kHz) in either ear ≥25 dB. Sensorineural hearing loss was defined as an air-bone gap of <10 dB, conductive hearing loss as an air-bone gap of ≥10 dB with bone conduction ≤20 dB, and mixed hearing loss was defined as an air-bone gap of ≥10 dB with bone conduction >20 dB. Type of hearing loss was counted by frequency and ear for each child, such that each type of hearing loss found would be included (i.e., non-mutually exclusive summary measures since children with conductive hearing loss in one ear and sensorineural hearing loss or mixed loss in the other ear would be counted twice). Middle ear disease was defined as the presence of a type B tympanogram or negative pressure <−200 daPa or abnormal findings on otoscopy identified by the audiologist as requiring follow-up (retraction, effusion, acute otitis media, external otitis, otorrhea, perforation, patent tube, and plugged tube).

Statistical Analysis

All analyses were performed using Stata version 17 software. Descriptive statistics were calculated to summarize basic sociodemographic and clinical characteristics of the children used in each study sample (i.e., first available screening, year 2 screening), including age, sex, Alaska Native/American Indian race, grade, year of first screening, highest education level of any caregiver, and whether first screening was a make-up screening. Sociodemographic characteristics of age, sex, Alaska Native/American Indian race, and grade collected during the Hearing Norton Sound trial in each school year were compared to the overall distribution of such characteristics extracted from de-identified BSSD school data from the same academic years to assess the generalizability of prevalence estimates to the overall eligible population of school students in BSSD.

Point prevalence of ear/hearing conditions was calculated at the level of the child, indicating that the condition was present for either ear. In the case where a condition had differing levels of severity for each ear, the prevalence was calculated using the highest severity present for either ear. When presenting overall prevalence estimates, proportions of children with the condition were summarized overall and then quantified as having the condition in one (unilateral) or both ears (bilateral).

To minimize the effect of the Hearing Norton Sound intervention on measurements of prevalence, the first available comprehensive audiometric assessment that each child provided (i.e., before exposure to the enhanced telemedicine referral) was used to calculate WHO-defined hearing loss, type of hearing loss, and middle ear disease. Because high-frequency tones were only collected in year 2 of the study, all high-frequency hearing loss prevalence estimates used year 2 study data only.

Evaluating both the former and new WHO definitions for hearing loss using data from our study presents some caveats that require consideration. During trial data collection, we used the former WHO definition (PTA > 25 dB). In the younger children who were more difficult to test, if a threshold of 20 dB was obtained, it was considered within normal limits based on the hearing loss definition used in the protocol, and further threshold testing was not performed. This was done to reduce fatigue and increase the likelihood for acquiring more data across frequencies in the younger participants. Using the new WHO definition of hearing loss, particularly for children 7 years of age and younger, resulted in an artificially inflated number of children with hearing loss due to suprathreshold responses at 20 dB. Therefore, for this analysis, we compared both WHO definitions of hearing loss for children 7 years of age and older with complete and accurate threshold data, and limited analysis with children 3 to 6 years of age to the former WHO-defined hearing loss to best inform the prevalence of hearing loss in the younger group.

There was additional interest in the heterogeneity of prevalence of ear/hearing conditions by age. The association between age and prevalence of hearing conditions was explored descriptively and graphically using polynomial smoothing plots with 95% confidence intervals (CIs). The prevalence was also calculated at more discrete intervals using 2-year age bands, starting at age 3 and ending with children age 17 and older. Age-specific prevalence (with 95% CIs) was calculated using binomial regression. Prevalence estimates pooled over all ages were calculated using exact binomial 95% CIs.

Missing audiometry data were expected to be more prevalent in younger participants due to requirements for conditioned response. Furthermore, it is plausible that children with hearing loss and/or middle ear disease may be more likely to experience cognitive delay, resulting in the inability to condition, leading to an expected downward bias in estimates of prevalence in younger ages. For this reason, multiple imputation was used to help avoid potentially underestimating the prevalence of hearing loss in this younger age group where missing data were more common. Multiple imputation with chained equations was used to produce a second set of estimates of hearing loss, both overall and age-specific, with estimates combined using Rubin's rules (Rubin 2004; Van Buuren 2007; Lee & Carlin 2010); for additional details of the multiple imputation model specification see Table 2 in Supplemental Digital Content 1, http://links.lww.com/EANDH/B124. For prevalence measures using audiometry and a standard definition of hearing loss, multiply imputed estimates were relied upon as plausibly more accurate measures of prevalence, though are still presented side by side with complete case estimates. However, for audiometry with high-frequency hearing loss, the study team lacked reliable correlates of hearing loss in the youngest age group (age 3 to 6) to accurately estimate high-frequency hearing loss using multiple imputation. By contrast, tympanometry and otoscopy did not have age-related missing data, and thus the prevalence of middle ear disease using complete case data should reflect actual prevalence in the study sample.

RESULTS

Baseline demographics are described in Table 1. There were two cohorts included in this analysis (Fig. 1): first screening (n = 1634) and year 2 screening (n = 1449), the latter of which included high frequencies in the audiometric evaluation. The median age (first screening, 9.0 years [6.0, 13.0]; year 2, 10.0 years [7.0, 13.0]) and percentage Alaska Native (first screening, 1563 [95.7%]; year 2, 1389 [95.9%]) were similar between the two cohorts. There were proportionally more children in preschool and first grade in the first screening cohort (510 [31.2%]) compared to the year 2 cohort (383 [26.4%]). More children underwent make-up screening in the first screening cohort (77 [4.7%]) compared to the year 2 cohort (26 [1.8%]), but there were no substantial differences in baseline demographics compared for children screened initially and those screened on make-up day (see Table 3 in Supplemental Digital Content 1, http://links.lww.com/EANDH/B124).

Basic sociodemographic characteristics were comparable between BSSD as a whole and children that participated in Hearing Norton Sound, suggesting that results are generalizable to the population of school children in preschool through 12th grade in this region during both study years (see Table 4 in Supplemental Digital Content 1, http://links.lww.com/EANDH/B124). Consequently, no weighting for probability of selection was performed.

The prevalence of hearing loss and middle ear disease in the first screening cohort using both complete case data and multiple imputation can be found in Table 2. Using the former WHO definition of hearing loss, the overall prevalence was 10.5% (95% CI, 8.9 to 12.1). The severity was skewed towards mild hearing loss, and unilateral hearing loss (7.7%, 95% CI, 6.3 to 9.0) was more common than bilateral (2.8%, 95% CI, 1.9 to 3.7) with this definition. Of children with hearing loss (PTA > 25 dB), 85.6% of children had mild hearing loss and 14.4% had moderate or greater. Baseline demographics of children with and without sufficient data to estimate hearing loss can be found in Table 5 in Supplemental Digital Content 1, http://links.lww.com/EANDH/B124.

Conductive hearing loss was the most common hearing loss type in this cohort, with a prevalence of 9.1% (95% CI, 7.6 to 10.7). Sensorineural and mixed hearing loss were present in 5.9% (95% CI, 4.6 to 7.3) and 3.5% (95% CI, 2.5 to 4.6) of children, respectively (Table 2). Of children with hearing loss (PTA > 25 dB), 76.7% had conductive hearing loss, 53.5% had sensorineural hearing loss, and 33.1% had mixed hearing loss, taking into consideration that children could have more than one type of hearing loss (i.e., non-mutually exclusive).

Table 3 stratifies definitions of hearing loss by age group (3 to 6 years versus 7 years and older) to describe general patterns in older and younger children. Hearing loss was more prevalent for younger children aged 3 to 6 years (14.9%, 95% CI, 11.4 to 18.5) compared to children 7 years of age and older (8.7%, 95% CI, 7.1 to 10.4). When applying the new WHO definition to children aged 7 and older, the prevalence of hearing loss increased to 23.4% (95% CI, 21.0 to 25.8). The majority of hearing loss was mild (21.6%, 95% CI, 19.2 to 24.0), with

TABLE 1. Baseline characteristics of analytic samples

	First Screening (N = 1634)	Year 2 (N = 1449)
Child's age (yrs)	9.7 (4.1)	9.9 (4.1)
Mean (SD)	9.0 (6.0, 13.0)	10.0 (7.0, 13.0)
Median (Q1, Q3)		
Child age range (yrs)		
3–6	449 (27.8%)	346 (23.9%)
7–9	404 (25.0%)	369 (25.5%)
10–12	331 (20.5%)	317 (21.9%)
13–15	241 (14.9%)	247 (17.1%)
16+	193 (11.9%)	169 (11.7%)
Missing	16	1
Age at screening (yrs)		
3–6	449 (27.8%)	346 (23.9%)
7+	1169 (72.2%)	1102 (76.1%)
Missing	16	1
Child's sex		
Male	858 (52.5%)	777 (53.7%)
Female	776 (47.5%)	671 (46.3%)
Missing	0	1
Alaska Native/American Indian		
No	71 (4.3%)	60 (4.1%)
Yes	1563 (95.7%)	1389 (95.9%)
Grade Level		
Preschool	153 (9.4%)	153 (10.6%)
K–5	865 (52.9%)	705 (48.7%)
6–8	318 (19.5%)	304 (21.0%)
9–12	298 (18.2%)	287 (19.8%)
Grade at screening		
Preschool–1st	510 (31.2%)	383 (26.4%)
2nd–12th	1124 (68.8%)	1066 (73.6%)
Highest education level of any caregiver		
<12 grade	93 (5.8%)	76 (5.3%)
HS diploma or GED	1027 (64.4%)	915 (64.3%)
Some college	320 (20.1%)	290 (20.4%)
College degree	155 (9.7%)	142 (10.0%)
Missing	39	26
School year of screening		
2017–2018	1219 (74.6%)	0 (0.0%)
2018–2019	415 (25.4%)	1449 (100.0%)
Make-up screening?		
No	1557 (95.3%)	1423 (98.2%)
Yes	77 (4.7%)	26 (1.8%)

GED, general education development; HS: high school.

moderate or worse representing 3.3% (95% CI, 2.3 to 4.3) of the cohort. Unilateral hearing loss (13.5%, 95% CI, 11.6 to 15.5) was slightly more prevalent than bilateral (9.9%, 95% CI, 8.2 to 11.6). As expected, missing data for hearing loss were more prominent in younger children ($p < 0.001$), as were conditions such as middle ear disease ($p = 0.013$), type B/C tympanometry ($p = 0.001$), and otoacoustic emission school screening referral ($p = 0.004$) (see Table 5 in Supplemental Digital Content 1, http://links.lww.com/EANDH/B124).

Middle ear disease was present in 17.6% (95% CI, 15.7 to 19.4) of the full cohort and was evenly divided between unilateral (8.8%, 95% CI, 7.4 to 10.2) and bilateral (8.8%, 95% CI, 7.4 to 10.2) (Table 2). The majority of children with middle ear disease had type B (10.1%, 95% CI, 8.6 to 11.5) or type C (5.0%, 95% CI, 3.9 to 6.1) tympanograms. A total of 14.4% (95% CI, 12.7 to 16.1) of children had otoscopic findings consistent with middle ear disease (Table 2). Details about the pathology visualized on otoscopy can be found in Table 6 in Supplemental Digital Content 1, http://links.lww.com/EANDH/B124.

A visualization of the prevalence of hearing loss by age for complete case data and each multiply imputed dataset for the first screening cohort is provided in Figure 2. Both WHO definitions of hearing loss are provided and compared to the prevalence of middle ear disease by age. The prevalence of hearing loss was highest in the youngest ages and increased again in the late teenage years (17 and 18 years). Middle ear disease prevalence was highest in the youngest children and progressively declined with age. For the PTA >25 dB definition of hearing loss, the multiply imputed data reflected higher prevalence in the youngest children than the complete case dataset, which is consistent with the observation that young children with missing data had more pathology than those who were able to complete the audiometric evaluation. Additional visualizations of hearing loss prevalence for both WHO definitions with and without multiple imputation using discrete age intervals, along with the prevalence of middle ear disease, are provided in Figures 3 and 4. Comparing the prevalence of the standard definitions of hearing loss and middle ear disease between the two cohorts (first

TABLE 2. Prevalence of hearing conditions across all ages (first screening), with complete case and multiply imputed estimates

	Complete Case		Multiply Imputed	
Outcome	Obs	Prevalence (95% CI)	Obs	Prevalence (95% CI)
WHO defined hearing loss (PTA > 25 dB definition)*	1556	8.5 (7.2–10.0)	1634	10.5 (8.9–12.1)
WHO defined hearing loss severity				
Mild (PTA >25–40 dB)	1556	7.3 (6.1–8.7)	1634	8.9 (7.4–10.5)
Moderate+ (PTA >40 dB)	1556	1.2 (0.7–1.9)	1634	1.5 (0.9–2.1)
WHO defined hearing loss laterality*				
Unilateral	1556	6.3 (5.1–7.6)	1634	7.7 (6.3–9.0)
Bilateral	1556	2.2 (1.6–3.1)	1634	2.8 (1.9–3.7)
Any sensorineural hearing loss	1556	3.0 (2.2–4.0)	1634	5.9 (4.6–7.3)
Any conductive hearing loss	1556	6.6 (5.4–7.9)	1634	9.1 (7.6–10.7)
Any mixed hearing loss	1556	1.9 (1.3–2.7)	1634	3.5 (2.5–4.6)
Middle ear disease†	1603	17.5 (15.7–19.5)	1634	17.6 (15.7–19.4)
Middle ear disease laterality†				
Unilateral	1603	8.8 (7.5–10.3)	1634	8.8 (7.4–10.2)
Bilateral	1603	8.7 (7.4–10.2)	1634	8.8 (7.4–10.2)
Tympanometry type				
Type B	1596	9.8 (8.4–11.4)	1634	10.1 (8.6–11.5)
Type C	1596	4.9 (3.9–6.1)	1634	5.0 (3.9–6.1)
Otoscopic findings consistent with middle ear disease	1628	14.4 (12.7–16.2)	1634	14.4 (12.7–16.1)

**Based on a pure-tone average of 0.5 kHz, 1 kHz, 2 kHz, 4 kHz >25 dB on either ear.*
†Defined as type B or C tympanometry OR otoscopy findings of retraction, effusion, acute otitis media, otorrhea, perforation, presence of tympanostomy tube, or external otitis requiring healthcare follow-up on either ear.
PTA, pure-tone average; WHO, World Health Organization.

TABLE 3. Prevalence estimates with 95% confidence intervals for WHO-defined hearing loss and middle ear disease, using complete case and multiply imputed data for children by age

Outcome	Complete Case	Multiply Imputed
Children aged 3–6 yrs		
WHO defined hearing loss (PTA >25 dB definition)*	10.0 (7.1–12.9)	14.9 (11.4–18.5)
WHO defined hearing loss severity (PTA >25 dB definition)*		
Mild (PTA >25–40 dB)	9.0 (6.2–11.8)	13.9 (10.3–17.5)
Moderate+ (PTA >40 dB)	1.0 (0.0–2.0)	1.4 (0.1–2.7)
WHO defined hearing loss (PTA >25 dB definition) laterality*		
Unilateral	8.0 (5.3–10.7)	11.1 (8.0–14.3)
Bilateral	2.0 (0.6–3.4)	3.8 (1.6–6.0)
Middle ear disease†	22.9 (19.0–26.8)	23.6 (19.7–27.6)
Middle ear disease laterality†		
Unilateral	10.0 (7.2–12.8)	10.7 (7.7–13.6)
Bilateral	12.9 (9.8–16.1)	13.0 (9.9–16.1)
Children aged 7+ yrs		
WHO defined hearing loss (PTA >25 dB definition)*	8.1 (6.5–9.6)	8.7 (7.1–10.4)
WHO defined hearing loss severity (PTA >25 dB definition)*		
Mild (PTA >25–40 dB)	6.8 (5.3–8.2)	7.7 (6.1–9.3)
Moderate+ (PTA >40 dB)	1.3 (0.6–2.0)	1.5 (0.8–2.2)
WHO defined hearing loss laterality (PTA >25 dB definition)*		
Unilateral	5.7 (4.4–7.1)	6.3 (4.9–7.7)
Bilateral	2.3 (1.5–3.2)	2.4 (1.5–3.3)
WHO defined hearing loss (PTA ≥20 dB definition)‡	22.9 (20.5–25.4)	23.4 (21.0–25.8)
WHO defined hearing loss severity (PTA ≥20 dB definition)‡		
Mild (PTA 20–<35 dB)	19.7 (17.4–21.9)	21.6 (19.2–24.0)
Moderate+ (PTA 35+ dB)	3.3 (2.3–4.3)	3.3 (2.3–4.3)
WHO defined hearing loss laterality (PTA ≥20 dB definition)‡		
Unilateral	13.1 (11.1–15.0)	13.5 (11.6–15.5)
Bilateral	9.9 (8.2–11.6)	9.9 (8.2–11.6)
Middle ear disease†	15.5 (13.4–17.6)	15.2 (13.2–17.3)
Middle ear disease laterality†		
Unilateral	8.4 (6.8–9.9)	8.0 (6.5–9.6)
Bilateral	7.1 (5.7–8.6)	7.2 (5.7–8.7)

**Based on a pure-tone average of 0.5 kHz, 1 kHz, 2 kHz, 4 kHz >25 dB on either ear.*
†Defined as type B or C tympanometry OR otoscopy findings of retraction, effusion, acute otitis media, otorrhea, perforation, presence of tympanostomy tube, or external otitis requiring healthcare follow-up on either ear.
‡Based on a pure-tone average of 0.5 kHz, 1 kHz, 2 kHz, 4 kHz ≥20 dB on either ear.
PTA, pure-tone average; WHO, World Health Organization.

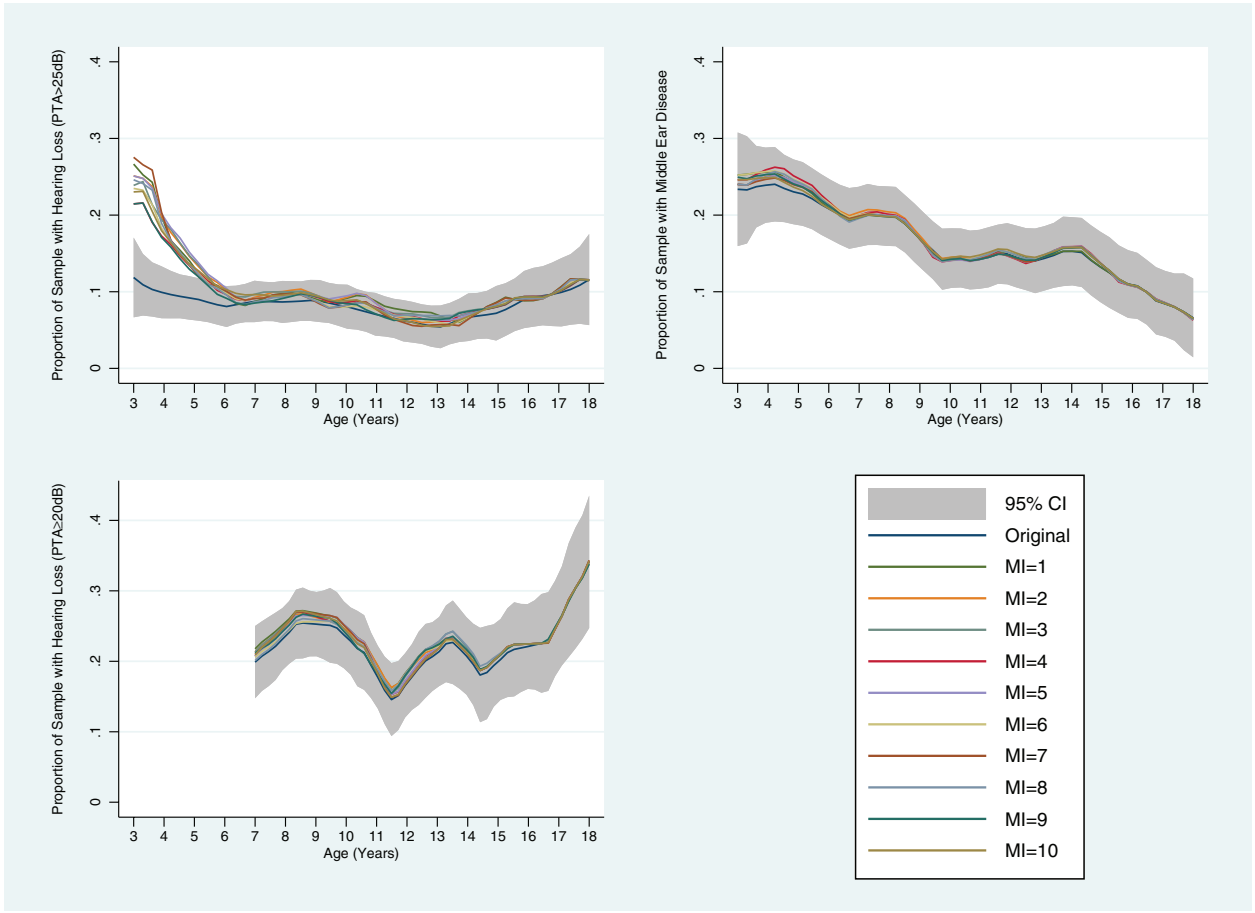

Fig. 2. Local polynomial smoothing plots of proportion with WHO-defined hearing loss and middle ear disease, by age (with 95% confidence intervals). Mean proportion from complete case data (original) presented alongside proportions computed from 10 multiply imputed datasets. CI, confidence interval; MI, multiple imputation; PTA, pure-tone average.

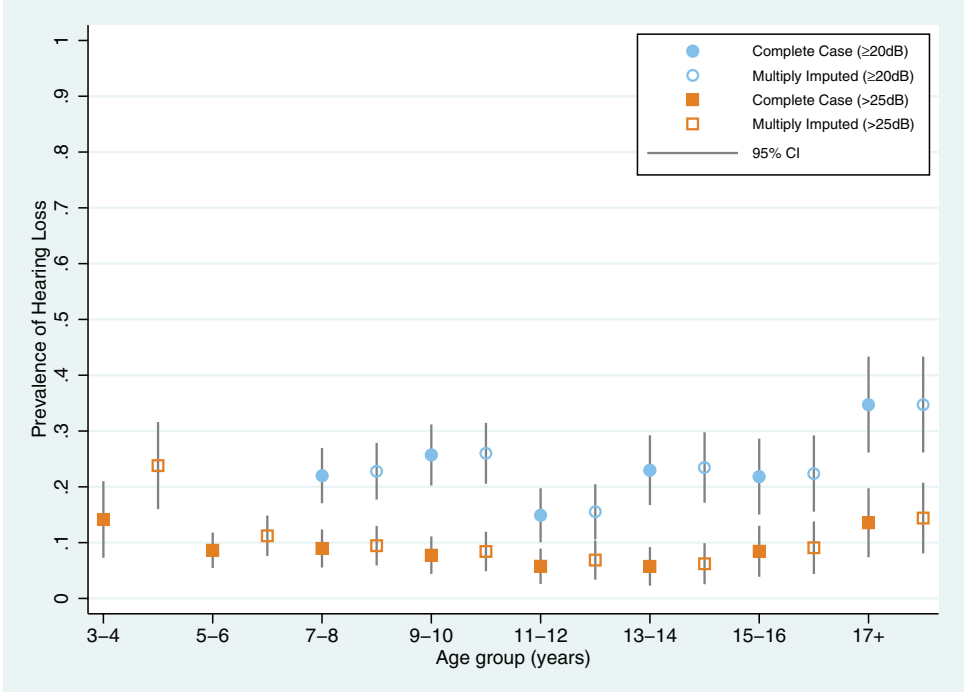

Fig. 3. Prevalence of hearing loss (both WHO definitions) by age, with 95% confidence intervals for complete case and multiply imputed data. CI, confidence interval.

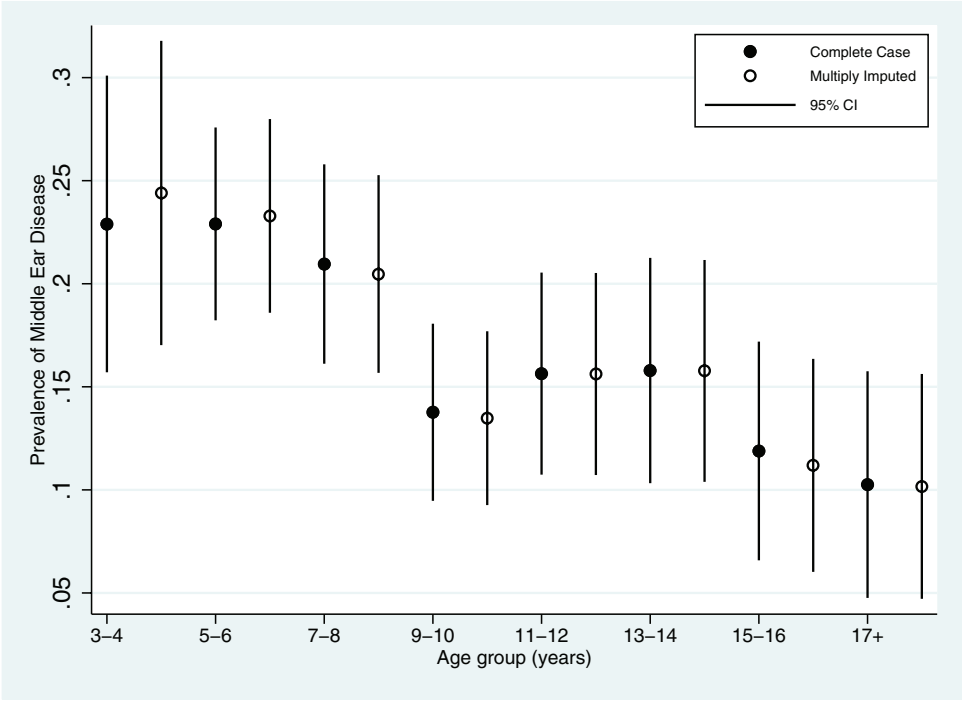

Fig. 4. Prevalence of middle ear disease by age with 95% confidence intervals for complete case and multiply imputed data. CI, confidence interval.

screening versus study years 1 and 2), the prevalence of hearing loss was marginally higher in the second year of the study compared to the first screening (see Table 7 in Supplemental Digital Content 1, http://links.lww.com/EANDH/B124).

Similar to the standard screening frequencies, high-frequency tones were difficult to obtain in children aged 3 to 6 and could not be reliably imputed due to a lack of reliable correlates of high-frequency hearing loss. Therefore, the prevalence for this measure was provided using complete case data from study year 2 for all children and for children aged 7 and older (Table 4, Fig. 5). High-frequency hearing loss (4, 6, 8 kHz), defined using a PTA >25 dB for all ages, was present in at least 20.5% (95% CI, 18.4 to 22.7) of children. High-frequency hearing loss was more prevalent for children 7 years of age and older (22.8%, 95% CI, 20.3 to 25.3 for PTA >25 dB and 29.7%, 95% CI, 27.0 to 32.4 for PTA ≥20 dB) compared to the cohort inclusive of children 3 to 6 years of age (20.5%, 95% CI, 18.4 to 22.7 for PTA >25 dB; Table 4). The opposite trend was demonstrated for middle ear disease, which decreased with age (Fig. 5).

TABLE 4. Prevalence of high-frequency hearing loss (PTA>25 dB) overall and for children aged 7 years of age and older* (PTA > 25 dB and PTA ≥ 20 dB) using year 2 data

Outcome	Complete Case
	Prevalence (95% CI)
	(n = 1362)
All children*	
Hearing loss (PTA >25 dB)†	20.5 (18.4–22.7)
High-frequency hearing loss laterality (PTA >25 dB)†	
Unilateral	12.5 (10.8–14.4)
Bilateral	8.0 (6.6–9.6)
Children aged 7+ yrs*	
High-frequency hearing loss (PTA >25 dB)†	22.8 (20.3–25.3)
High-frequency hearing loss laterality (PTA >25 dB)†	
Unilateral	13.5 (11.4–15.5)
Bilateral	9.3 (7.6–11.0)
High-frequency hearing loss (PTA ≥20 dB)‡	29.7 (27.0–32.4)
High-frequency hearing loss laterality (PTA ≥20 dB)‡	
Unilateral	16.4 (14.2–18.6)
Bilateral	13.3 (11.3–15.3)

*Age-specific prevalence only presented for age 7 years and older due to missing data in the youngest age group and lack or correlates for imputation.
†Based on a pure-tone average of 4 kHz, 6 kHz, 8 kHz >25 dB on either ear.
‡Based on a pure-tone average of 4 kHz, 6 kHz, 8 kHz ≥20 dB on either ear.
PTA, pure-tone average.

DISCUSSION

Using data from two cluster-randomized trials in the Bering Strait region of rural Alaska, we demonstrate that there continues to be a substantial burden of childhood hearing loss and middle ear disease in rural Alaska Native children. This analysis represents the first prevalence study on childhood hearing loss in Alaska in over 60 years. To our knowledge, this is the largest cohort with hearing data ever collected in rural Alaska and includes a representative sample of preschool- and school-aged children from across an entire region of the state.

The prevalence estimates are slightly reduced in this cohort compared to historical data. In 1967, Reed and colleagues found hearing loss prevalence to be 31% in a cohort of 378 children aged 3 to 5 years from rural western Alaska using criteria equivalent to the former WHO definition (PTA > 25 dB) (Reed et al. 1967). Overall, the prevalence in the current cohort was 10.5% using the former WHO definition (PTA > 25 dB). However, this includes a much larger age range (3 to 21 years), and when we focus specifically on the youngest children ages 3 to 6 years, the prevalence is 14.9%. While these two analyses used different

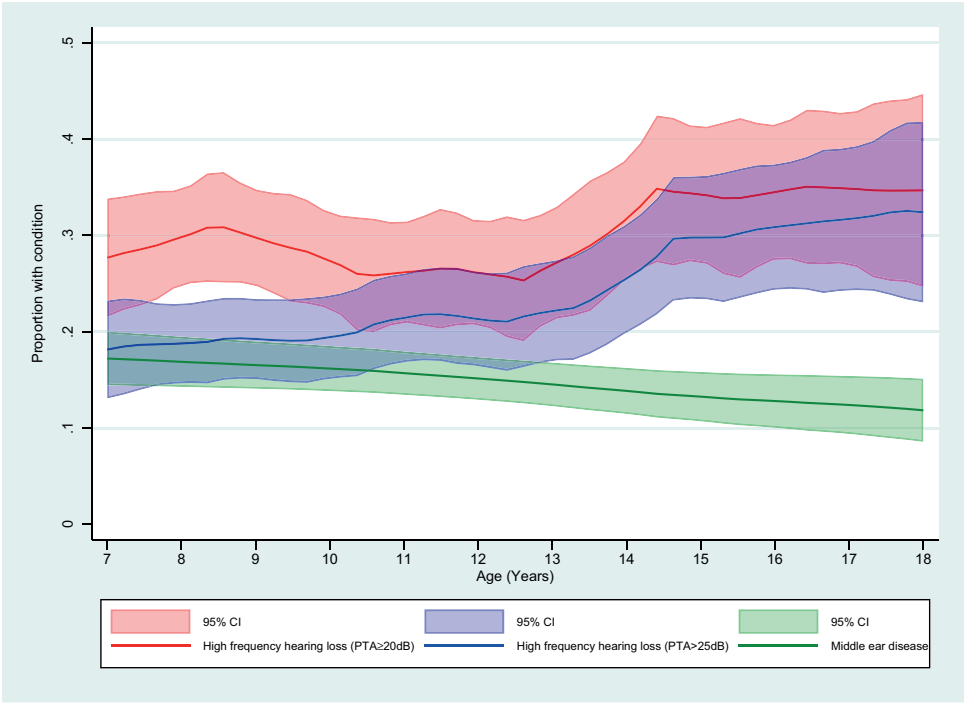

Fig. 5. Local polynomial smoothing plots of proportion with high-frequency hearing loss and middle ear disease using the year 2 sample, by age (with 95% confidence intervals). CI, confidence interval; PTA, pure-tone average.

study designs and were conducted in different regions of rural Alaska and are therefore not directly comparable, a decline in the prevalence is expected given improved access to care and widespread coverage of pneumococcal vaccination that have occurred over the past 20 years. Hearing loss prevalence for children and adolescents continues to be higher in rural Alaska than in the general US population, which has been estimated as 3.1% using a compilation of nationally representative screening studies that applied the criteria of PTA >25 dB (Mehra et al. 2009).

Similar to historical data, middle ear disease continues to be prominent in the youngest children, suggesting that infection-related hearing loss continues to be an important contributor to overall prevalence in this population. This finding is consistent with other recent studies that observed higher otitis media rates in rural regions of Alaska than in urban areas of the state or the general US (Singleton et al. 2018). Similarly, high burdens of infection-related hearing loss have been noted in other circumpolar and indigenous populations, including cohorts in Greenland and Australia (Morris 1998; Gunasekera et al. 2007; Jensen et al. 2013; Avnstorp et al. 2016).

This analysis also highlights the substantial burden of high-frequency hearing loss in this rural Alaska cohort, which may be secondary to noise exposure from traditional subsistence activities, such as hunting. There is no standard definition of high-frequency hearing loss, and thus comparisons across studies are challenging. Nevertheless, the prevalence of high-frequency hearing loss observed in this rural Alaskan cohort (23% with a PTA > 25 dB over 4, 6, and 8 kHz) is higher than the prevalence found in an NHANES study of US adolescents 12 to 19 years (4.7% with a PTA > 25 dB over 3, 4, 6, and 8 kHz) (Shargorodsky et al. 2010). The prevalence of high-frequency hearing loss increases with age in this cohort, a pattern that is best visualized with the former WHO definition (Fig. 5). Increased risk of childhood hearing loss with age was also observed in a cohort in Greenland where there is also a strong tradition of hunting (Jensen et al. 2013). The age pattern observed with high-frequency hearing loss in the current cohort contrasts with middle ear disease, which is most prevalent in the youngest children and decreased with age. These distinct etiologies contributing to the overall prevalence of childhood hearing loss in the Alaska Native population have notable policy implications and highlight the importance of preventive measures focused on ear infections in early childhood and noise exposure in adolescence.

The WHO definition of hearing loss changed shortly after the completion of the Hearing Norton Sound trials, lowering the PTA for hearing loss from >25 dB to ≥20 dB. To maintain consistency with the original study design while also providing estimates that could be compared with future studies, we chose to analyze the data using both WHO definitions. Not surprisingly, the prevalence was much higher with the new definition (23.4%) compared to the prior definition (8.7%). We noted some challenges with the new definition that should be taken under consideration for future studies. The new definition produced startlingly high prevalence estimates in the youngest children, which necessitated focusing solely on children aged 7 and older for that analysis. This dramatic difference is likely secondary to the challenges of conducting pure-tone testing with this young population, particularly outside of a sound-proof environment. In the preschool trial, study audiologists followed the protocol for the definition of hearing loss (PTA > 25 dB) to measure thresholds and characterize hearing as normal or abnormal. In the youngest children who were difficult to condition and fatigued quickly, it is possible that results were suprathreshold (e.g., reliable responses at 20 dB) for children with perceived normal hearing. The issue of suprathreshold results was not common in older children, because conditioning was more straightforward as children aged. The aberrancy in results with

the new WHO definition highlights that this lower threshold to define hearing loss may be particularly challenging to implement in field studies with young children. It will be important for future studies to begin with this definition and train testers to focus on thresholds of 15 dB or less to facilitate accurate results. While the new WHO definition is comparable to the commonly applied clinical cutoff (15 dB) for normal-hearing children, field testing is very different from testing in a sound treated environment. Minimizing ambient noise in the testing environment, as well as the future development of audiometric equipment for use outside the sound booth that is increasingly sensitive at the lowest thresholds will be important to facilitate accurate data collection with the new definition. Ultimately, the widespread adoption of a single definition of hearing loss will greatly facilitate the comparison of results across populations and regions and therefore should be promoted, albeit with the challenges noted above taken into consideration.

In the preschool trial, up to one-third of children did not condition to audiometric testing. We report multiply imputed results in this analysis to address these missing data in the youngest children. Because a full audiometric assessment was performed on every child, including objective tests such as otoacoustic emissions, tympanometry, and otoscopy, there were ample data to build a multiple imputation model. There was nevertheless uncertainty in the multiple imputation estimates, which is illustrated by the spread of multiple imputation results at the youngest ages (Fig. 2). The multiply imputed results for middle ear disease did not demonstrate the same spread, which is expected since middle ear disease can be evaluated strictly with objective testing and therefore had significantly fewer missing data (Fig. 2). It should be noted that the ability of multiple imputation to produce unbiased prevalence estimates relies on the availability of enough variables that predict missingness and missing data values themselves (Sterne et al. 2009), which in practice cannot be definitively confirmed. However, the availability of results from multiple types of screening tools, including those that did not require a behavioral response, adds confidence to the ability to produce plausible estimates for the standard measures of hearing loss, correlating both with missingness itself and with the value of the missing responses. Unfortunately, such correlates were not available for the high-frequency tones, limiting our confidence in the ability to reliably measure the prevalence of high-frequency hearing loss in children younger than seven. The prevalence of high-frequency hearing loss, most commonly due to age- or noise-related hearing loss, is expected to be low in this preschool-aged population, however.

There are limitations to this study that should be mentioned. This prevalence analysis was conducted using two randomized trial cohorts designed to evaluate the impact of mHealth school screening and telehealth referral on the identification of childhood hearing loss. To avoid potential influence from the trial intervention, we analyzed data from children's first screening over two academic years which meant point prevalence was measured over 12 to 18 months, and assumed that the incidence of the outcome did not change over time; an assumption we believe is reasonable. The influence of the intervention could not be avoided for the high-frequency analysis for children who participated in both years of the trial, as high frequencies were added in the second year of the study. Finally, because this was a field-based trial where audiometric evaluations were conducted in the school environment and not in a sound-proof booth, it is possible that noise influenced results. The tablet-based audiometer used for testing performed continuous noise monitoring and notified study audiologists if the environment was too loud.

There are several important strengths of this study. Unlike many screening studies that limit full evaluations to children who do not pass screening, all children in this cohort underwent a comprehensive audiological evaluation. Study audiologists lived and worked in the region where the study was conducted and had experience with testing Alaska Native children. Importantly, this prevalence study included approximately 71% of preschool- and school-aged children in the Bering Strait region, and demographic data were similar between the full school-aged population and the study population. Results are therefore generalizable to the Bering Strait region, which covers 23,000 square miles of rural northwest Alaska. Although not directly generalizable to the state as a whole, these results also have implications for the broader Alaska Native population in rural regions of Alaska and are consistent with other studies that demonstrate a high rate of otitis media in rural regions (Singleton et al. 2018).

There are important public health implications of this study. Hearing loss continues to be common in rural Alaska Native children, with higher prevalence than in the general US population. Infection-related hearing loss is more prevalent in younger children in this cohort, while high-frequency hearing loss is more common with increasing age. Prevention efforts may benefit from focusing on managing different hearing loss types by age. Continued research on the impact of the revised WHO definition of hearing loss would be valuable, with particular attention to the challenges of its application in young children.

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Drs. Susan Emmett and Samantha Kleindienst Robler (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.

Study concept and design: S.D.E., A.P., J.J.G., A.B.L., N.-Y.W., P.H., S.K.R.; Acquisition of data: S.D.E., M.I.-J., C.D.J., P.H., A.A.R., S.K.R.; Analysis and interpretation of data: S.D.E., A.P., J.R.E., S.K.R.; Literature Search: S.D.E., K.L.H., S.K.R.; Drafting of the manuscript: S.D.E., A.P., S.K.R.; Critical revision of the manuscript for important intellectual content: S.D.E., A.P., J.R.E., S.K.R.; Statistical analysis: A.P., J.R.E.; Obtained funding: S.D.E., P.H., S.K.R.; Study supervision: S.D.E., S.K.R.

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The data supporting the findings of this study are available upon reasonable written request to the corresponding author.

The authors have no conflicts of interest to disclose.

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