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ORIGINAL RESEARCH

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Hearing-related quality of life in children and adolescents in rural Alaska

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Abstract

Objective: This study evaluated the Hearing Environments and Reflection on Quality of Life (HEAR-QL) questionnaire in rural Alaska, including an addendum crafted through community feedback to reflect the local context. The objectives were to assess whether HEAR-QL score was inversely correlated with hearing loss and middle ear disease in an Alaska Native population.

Methods: The HEAR-QL questionnaires for children and adolescents were administered as part of a cluster randomized trial in rural Alaska from 2017 to 2019. Enrolled students completed an audiometric evaluation and HEAR-QL questionnaire on the same day. A cross-sectional evaluation of questionnaire data was utilized.

Results: A total of 733 children (ages 7–12 years) and 440 adolescents (ages ≥13 years) completed the questionnaire. Median HEAR-QL scores were similar among children with and without hearing loss (Kruskal–Wallis, $p = .39$); however, adolescent HEAR-QL scores significantly decreased with increasing hearing loss ($p < .001$). Median HEAR-QL scores were significantly lower in both children ($p = .02$) and adolescents ($p < .001$) with middle ear disease compared with those without. In both children and adolescents, the addendum scores were strongly correlated with total HEAR-QL score ($\rho_{Spearman} = 0.72$ and 0.69, respectively).

Conclusions: The expected negative association between hearing loss and HEAR-QL score was observed in adolescents. However, there was significant variability that could not be explained by hearing loss, and further investigation is warranted. The expected negative association was not observed in children. HEAR-QL scores were associated with middle ear disease in both children and adolescents, making it potentially valuable in populations where the prevalence of ear infections is high.

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KEYWORDS
Alaska, hearing loss, HEAR-QL, middle ear disease, quality of life

1 | INTRODUCTION

Childhood hearing loss adversely affects school performance, family dynamics and relationships, and vocational opportunities.[1–5] The prevalence of childhood hearing loss is thought to be disproportionately higher in rural Alaska, where the population is primarily Alaska Native, compared with the general U.S. child population.[6–8] Alaska Native children have a higher rate of ear infections, and the majority of hearing loss in rural Alaska is infection related.[6] However, hearing-related quality of life (QOL) has not been assessed in this population.

The goal of QOL assessment tools is to quantify the relationship between a disease process and a person's subjective experience.[9] Such assessment is one of the first steps needed to determine functional impact of a condition, and, if an intervention needs to be made, to assess the impact of an intervention. The Hearing Environments and Reflection on Quality of Life (HEAR-QL) questionnaire is the only validated tool to assess hearing-related quality of life in children[10] and adolescents.[11] HEAR-QL quantifies responses on a scale of almost always (0) to never (4) to estimate the impact of hearing loss on quality of life, assessing areas of communication related to environment, social activity, school difficulties, and feelings.

The child HEAR-QL was developed and validated in an English-speaking, mostly white, school-aged population in Missouri,[10] while the adolescent HEAR-QL was validated in a population of teens from mostly white, middle-to-higher income families, with highly educated parents.[11] An important aspect of cross-cultural research is creating tools and solutions that are culturally relevant and applicable across populations. Thus, there is a need to assess the cultural relevance and generalizability of this tool in evaluating hearing-related QOL in rural Alaska.

The primary objective of this study was to characterize the relationship between HEAR-QL scores and hearing loss in a rural Alaska population. This includes a regional addendum developed through community feedback to more accurately reflect the environment and context of rural Alaska that was appended to the end of each of the HEAR-QL questionnaires. We hypothesized that if the HEAR-QL tool accurately quantifies the subjective experience of children with hearing loss, reported QOL will be lower than that of children without hearing loss. This relationship was observed for both HEAR-QL tools in the populations in which they were validated. Further, we expected to observe an inverse relationship between HEAR-QL score and pure tone average (PTA)—a continuous measure of hearing loss severity. The absence of such a relationship may be evidence that a different tool is needed to assess hearing-related quality of life in this population.

A secondary objective of this study was to characterize the relationship between HEAR-QL scores and middle ear disease. We are unaware of any study that has used HEAR-QL to examine the effect of infection-related hearing loss on QOL. Due to the unusually high burden of ear infections in this population, we examined the degree to which the subjective experience of children or adolescents with middle ear disease may differ from those with and without hearing loss, and thus whether this tool could be used to assess the impact of future interventional strategies to treat middle ear disease in this population. We hypothesized that children and adolescents with middle ear disease would experience diminished hearing-related QOL compared with those without middle ear disease, and that children with both hearing loss and middle ear disease would experience the lowest quality of life.

2 | MATERIALS AND METHODS

2.1 | Study overview

Hearing Norton Sound was a mixed-method community randomized trial evaluating a new hearing screening and telemedicine referral pathway in 15 communities in the Norton Sound region of Northwest Alaska. Full details of the trial and qualitative components are available elsewhere.[12–14] A 4-question addendum was created based on community input during focus groups held prior to the start of the trial and appended to the end of the original HEAR-QL questionnaires (Table TABLE A1). These questions were included to reflect environments and activities common in this rural Alaskan region.

2.2 | HEAR-QL Administration

Eligibility for trial enrollment included all school-aged children within the Bering Strait School District, kindergarten through 12th grade with signed parental consent and child assent. HEAR-QL administration was limited to children 7 years of age and older with the child HEAR-QL validated in subjects aged 7–12 years, and adolescent HEAR-QL validated for subjects 13–21 years. The 4-item supplement was added at the end of both the original child and adolescent HEAR-QL questionnaires.

Once enrolled, each student completed the appropriate HEAR-QL version based on their age for each year of the trial. Questionnaires were completed on hearing screening day using REDCap software on Apple iPads. Data were cross-checked for missing responses at the

time of completion and de-identified prior to storage. For the child HEAR-QL, questions were read to each participant if reading assistance was required.

2.3 | Audiometric assessment

An audiometric assessment was performed on all enrolled students present on hearing screening day. Air conduction pure-tone audiometry was performed with a validated and calibrated tablet audiometer (SHOEBOX Audiometry Pro, SHOEBOX, Ltd, Canada), using supraural headphones at 0.5, 1, 2, 3, and 4 kHz bilaterally. Digital otoscopy was obtained using a USB digital otoscope (Otocam, Otometrics, Denmark) and used to determine presence of pathology at the discretion of the audiologist. Tympanometry was performed using a Bluetooth digital tympanometer (Otoflex 100, Otometrics, Denmark). Presence of middle ear disease was determined by Type B (flat) or Type C tympanogram (< −200 daPa) or positive finding on otoscopy. Hearing loss was defined as a PTA >25 dB (0.5, 1, 2, and 4 kHz) in either ear. PTA of 26–40 dB was categorized as mild hearing loss, and PTA > 40 was categorized as moderate or worse. Audiometric assessment was performed on the same day as HEAR-QL administration.

2.4 | Statistical analysis

The study was conducted over 2 years, and participants could have contributed data at two time points. However, the analytic sample included only the first available age-appropriate HEAR-QL score for each enrolled participant.

The distribution of HEAR-QL domain and addendum scores were described using the median and interquartile range. The nonparametric Kruskal–Wallis test was used to compare the distribution of total HEAR-QL scores, as well as scores from each HEAR-QL domain and the addendum, by hearing loss severity using a two-sided *p*-value to evaluate the null hypothesis that the mean ranks of the scores were the same by severity level. The same procedure was used to compare scores by middle ear disease status. To control for possible Type I errors, *p*-values were adjusted for multiple comparisons using the Šidák correction, which is conservative in the case of positively dependent tests. The correction was applied separately for comparisons by hearing loss severity and by middle ear disease since they are testing different hypotheses. In addition, the relationship between total HEAR-QL, domain and addendum scores and PTA were evaluated graphically using scatterplots with loess regression and quantified using Spearman correlations and associated 95% confidence intervals (CIs).

All quantitative analysis was conducted in SAS 9.4 (SAS Institute, Cary, North Carolina). The study was reviewed and approved by the Institutional Review Boards of Alaska Area, Norton Sound Health Corporation, and Duke University.

TABLE 1 Demographic characteristics for children and adolescents

	Children (*N* = 733)	Adolescents (*N* = 440)
Age in years, median (IQR)	9 (3)	15 (3)
Min–max	7–12	13–21
Female, *n* (%)	342 (46.7)	208 (47.3)
American Indian or Alaska Native, *n* (%)	702 (95.8)	426 (96.8)
Grade level, *n* (%)		
K-5	555 (75.7)	
6–8	178 (24.3)	142 (32.3)
9–12		298 (67.7)
Highest education level of any caregiver, *n* (%)		
<12 grade	48 (6.5)	26 (5.9)
high school diploma or GED	455 (62.1)	282 (64.1)
Some College	136 (18.6)	88 (20.0)
College Degree	74 (10.1)	33 (7.5)
Missing	20 (2.7)	11 (2.5)
Hearing loss severity, *n* (%)		
Normal Hearing	675 (92.1)	401 (91.1)
Mild (PTA >25–40 dB)	49 (6.7)	31 (7.0)
Moderate or worse (PTA >41 dB)	9 (1.2)	8 (1.8)
Hearing loss laterality, *n* (%)		
Normal hearing	675 (92.1)	401 (91.1)
Unilateral	47 (6.4)	25 (5.7)
Bilateral	11 (1.5)	14 (3.2)
Middle ear disease (either ear), *n* (%)		
No	602 (82.1)	378 (85.9)
Yes	125 (17.1)	56 (12.7)
Missing	6 (0.8)	6 (1.4)

Abbreviations: GED, general educational development; HEAR-QL: Hearing Environments and Reflection on Quality of Life; IQR, interquartile range; PTA, pure tone average.

3 | RESULTS

The age-appropriate HEAR-QL questionnaire with the addendum was completed by 733 children aged 7–12 years and 440 adolescents aged 13 years and older. Demographic characteristics of the sample are shown in Table 1. A total of 1076 students had normal hearing, 80 had mild hearing loss (PTA >25–40 dB HL), and 17 had moderate or worse hearing loss (PTA >41 dB HL) based on audiograms performed the same day as HEAR-QL administration. Of the children and adolescents with hearing loss, 74% had unilateral hearing loss and 26% had bilateral hearing loss. A total of 125 children and 56 adolescents had middle ear disease based on tympanometry and physical exam on the day of testing. For those with middle ear disease, 66% had normal hearing, 28% had mild hearing loss, and 6% had moderate or worse hearing loss (see Table 2).

Among adolescents, HEAR-QL scores were significantly lower with increasing severity of hearing loss for total scores, with a lower HEAR-QL indicating worse quality of life. This pattern was also observed for each domain and addendum (all adjusted *p*-values < .01). Median total adolescent HEAR-QL scores were higher for those with normal hearing (94.64) compared with those with mild hearing loss (87.95) and for those with moderate or worse hearing loss (60.27). In contrast, there were no significant differences in hearing loss severity among the child HEAR-QL scores after multiple comparison adjustment (Table 3). Median total child HEAR-QL scores were slightly higher for those with normal hearing (80.77) compared with those with mild hearing loss (76.92); however, no difference was noted in comparison with those with moderate or worse hearing loss (80.77). Of note is that children with normal hearing demonstrated a wide range of QOL scores (Figure 1A), whereas most adolescents with normal hearing had high QOL (Figure 1B).

TABLE 2 Middle ear disease by hearing loss severity for children and adolescents

	Middle ear disease[a]	
	Yes	No
Hearing loss severity, *n* (%)		
Normal Hearing	119 (11.2)	946 (88.8)
Mild (PTA >25–40 dB)	51 (63.8)	29 (36.3)
Moderate or worse (PTA >41 dB)	11 (68.8)	5 (31.3)

[a]*N* = 12 missing values for middle ear disease.
Abbreviation: PTA, pure tone average.

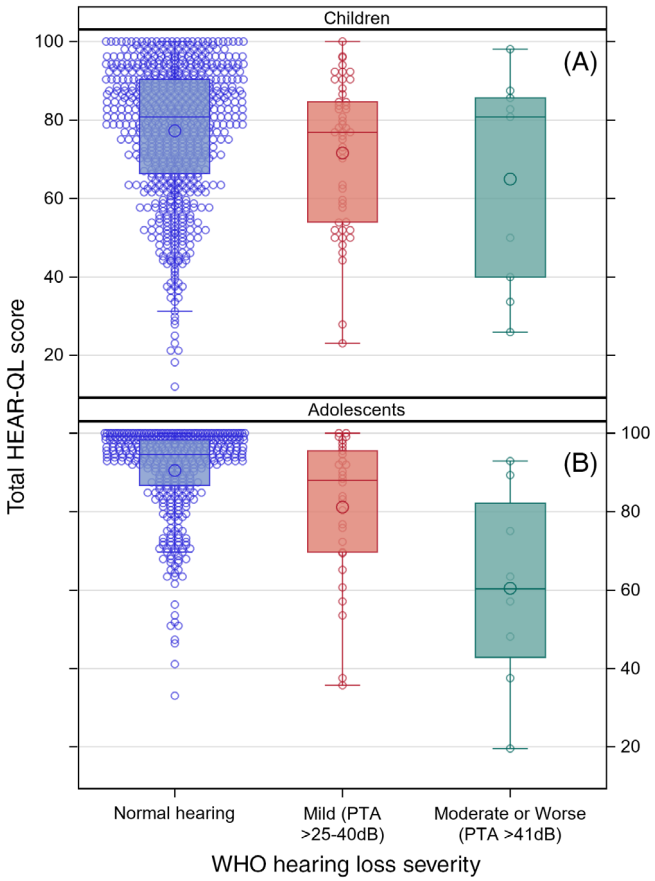

FIGURE 1 Total HEAR-QL Scores by WHO hearing loss severity for children (A) and adolescents (B). HEAR-QL, Hearing Environments and Reflection on Quality of Life; PTA, pure tone average

TABLE 3 HEAR-QL scores by WHO grade of hearing loss for children and adolescents

	WHO hearing loss grade			Kruskal–Wallis *p*-value	
	Normal hearing	Mild (PTA >25–40)	Moderate or worse (PTA >41)	Raw	Šidák adjusted
Child HEAR-QL	(*N* = 675)	(*N* = 49)	(*N* = 9)		
Environment score	76.92 (28.85)	71.15 (30.77)	73.08 (36.54)	*P* = .05	*P* = .46
Activities score	90.83 (33.33)	83.33 (33.33)	83.33 (37.50)	*P* = .10	*P* = .68
Feelings score	85.71 (28.57)	78.57 (28.57)	85.71 (57.14)	*P* = .03	*P* = .29
Total score	80.77 (24.04)	76.92 (30.62)	80.77 (45.58)	*P* = .04	*P* = .39
Addendum score	87.50 (31.25)	87.50 (37.50)	87.50 (50.00)	*P* = .32	*P* = .99
Adolescent HEAR-QL	(*N* = 401)	(*N* = 31)	(*N* = 8)		
Hearing situations score	87.50 (16.67)	77.08 (29.17)	62.50 (57.50)	*P* = .001	*P* = .01
Social interactions score	100.00 (5.36)	100.00 (21.43)	85.71 (32.14)	*P* = .001	*P* = .01
School difficulties score	98.21 (14.29)	87.50 (39.29)	46.43 (42.86)	*P* < .001	*P* < .001
Feelings score	96.88 (12.50)	87.50 (32.59)	50.00 (37.50)	*P* < .001	*P* = .003
Total score	94.64 (11.61)	87.95 (25.89)	60.27 (39.32)	*P* < .001	*P* = .001
Addendum score	100.00 (6.25)	93.75 (18.75)	68.75 (28.13)	*P* < .001	*P* = .004

Note: Values reported are the median (IQR).
Abbreviations: HEAR-QL: Hearing Environments and Reflection on Quality of Life; IQR, interquartile range; PTA, pure tone average; WHO, World Health Organization.

Both child and adolescent HEAR-QL scores had significant but mild negative correlations with PTA (Table 4 and Figures 2 and 3). Child correlations ranged from −0.16 (−0.23, −0.09) for total scores to −0.10 (95% CI: −0.17, −0.03) for addendum scores. Adolescent correlations ranged from −0.24 (−0.33, −0.15) for school difficulties scores to −0.17 (−0.26, −0.07) for hearing situations scores.

The results of the comparisons by middle ear disease are shown in Table 5. Adolescents with middle ear disease had lower median HEAR-QL scores for each domain except for the addendum, compared with adolescents without middle ear disease. The median total HEAR-QL score in adolescents with middle ear disease was 8.93 points lower than adolescents without middle ear disease. Children with middle ear disease also had lower HEAR-QL scores for each domain, compared with children without middle ear disease. Median total child HEAR-QL score was 4.81 points lower in children with middle ear disease than those without middle ear disease. There was more variation in HEAR-QL scores for children without middle ear disease (Figure 4A) compared with adolescents without middle ear disease (Figure 4B).

The 4-question addendum was administered with both the child and adolescent HEAR-QL questionnaires. In both children and adolescents, the addendum scores strongly correlated with the total HEAR-QL score ($\rho_{Spearman} = 0.72$ and 0.69, respectively).

TABLE 4 Spearman correlations (95% CI) between HEAR-QL domain scores and PTA

	Correlation (95% CI) with PTA
Child HEAR-QL	
Environment score	−.14 (−0.21, −0.07)
Activities score	−.15 (−0.22, −0.08)
Feelings score	−.14 (−0.21, −0.07)
Total score	−.16 (−0.23, −0.09)
Addendum score	−.10 (−0.17, −0.03)
Adolescent HEAR-QL	
Hearing situations score	−.17 (−0.26, −0.07)
Social interactions score	−.22 (−0.31, −0.13)
School difficulties score	−.24 (−0.33, −0.15)
Feelings score	−.20 (−0.29, −0.11)
Total score	−.21 (−0.30, −0.11)
Addendum score	−.22 (−0.31, −0.12)

Abbreviations: 95% CI, 95% confidence interval; HEAR-QL: Hearing Environments and Reflection on Quality of Life; PTA, pure tone average.

4 | DISCUSSION

This was the first study to evaluate hearing-related quality of life in children and adolescents in rural Alaska. A region-specific addendum was also included with both the child and adolescent HEAR-QL questionnaires based on community feedback to reflect the culture and environment of the region. Adolescents with hearing loss had lower HEAR-QL scores compared with adolescents with normal hearing. This difference, however, was not found in children. Both children and adolescents with middle ear disease demonstrated lower HEAR-QL scores than their peers without middle ear disease.

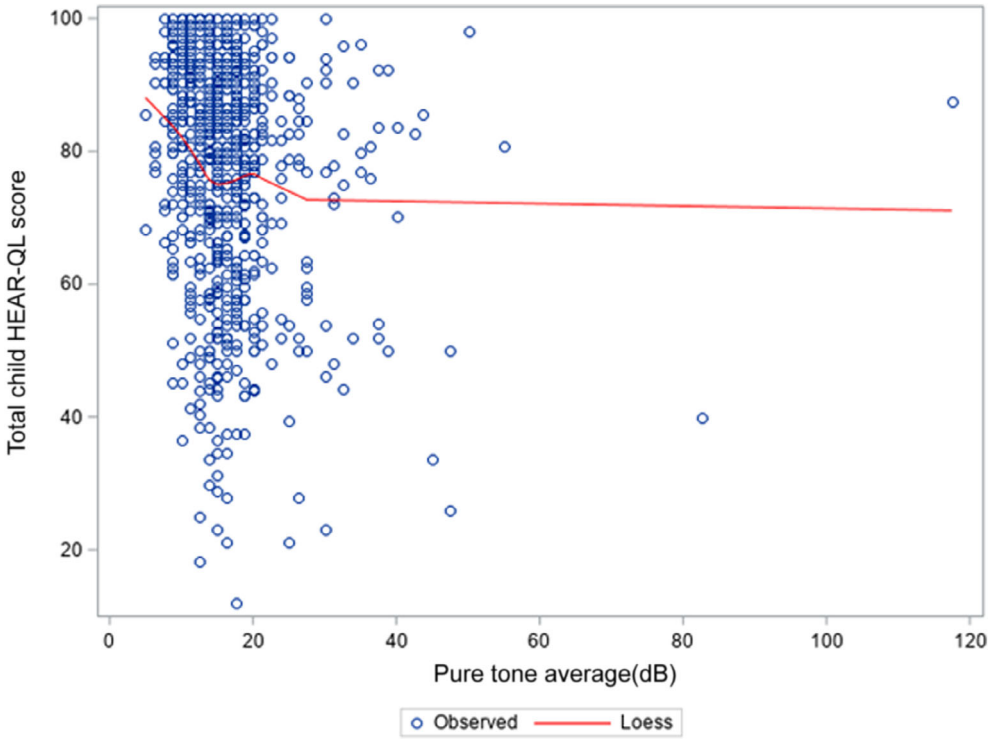

FIGURE 2 Total child HEAR-QL scores by pure tone average. HEAR-QL, Hearing Environments and Reflection on Quality of Life

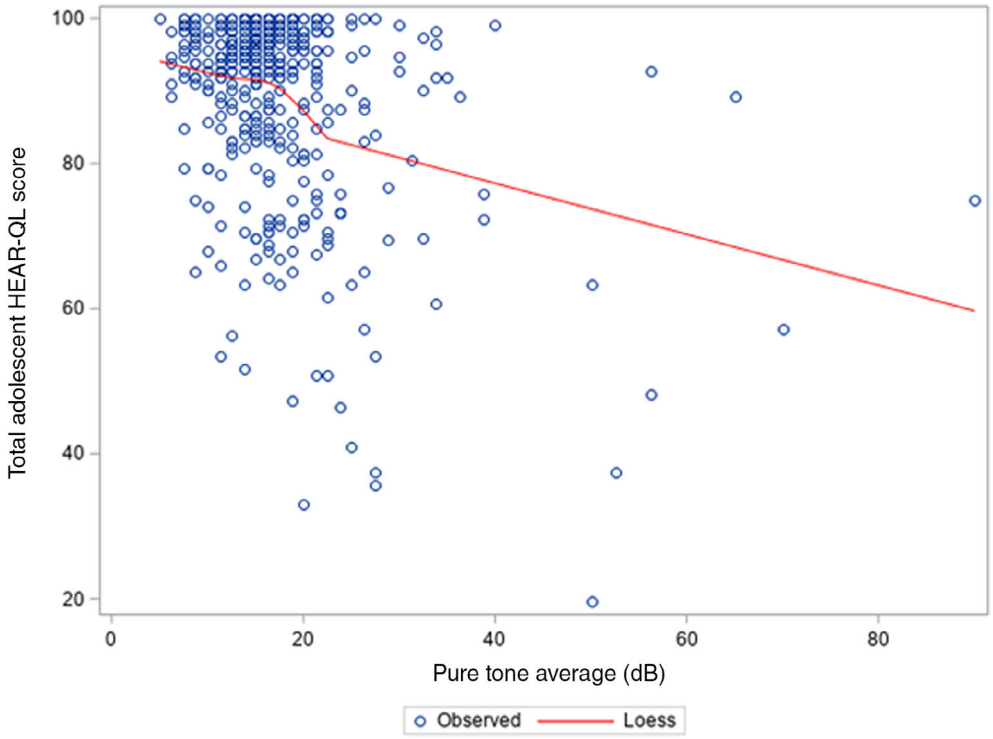

FIGURE 3 Total adolescent HEAR-QL Scores by pure tone average. HEAR-QL, Hearing Environments and Reflection on Quality of Life

TABLE 5 HEAR-QL scores by middle ear disease for children and adolescents

	Middle ear disease		Kruskal–Wallis *p*-value	
	No	Yes	Raw	Šidák adjusted
Child HEAR-QL	(*N* = 602)	(*N* = 125)		
Environment score	78.85 (26.92)	73.08 (32.69)	*P* = .01	*P* = .09
Activities score	91.67 (29.17)	83.33 (33.33)	*P* < .001	*P* = .003
Feelings score	85.71 (28.57)	78.57 (32.14)	*P* = .01	*P* = .15
Total score	81.73 (24.04)	76.92 (26.92)	*P* = .002	*P* = .02
Addendum score	87.50 (25.00)	75.00 (37.50)	*P* = .01	*P* = .12
Adolescent HEAR-QL	*(N = 378)*	*(N = 56)*		
Hearing situations score	87.50 (16.67)	79.17 (25.00)	*P* < .001	*P* = .003
Social interactions score	100.00 (3.57)	96.43 (21.43)	*P* < .001	*P* = .001
School difficulties score	98.21 (14.29)	85.71 (32.14)	*P* < .001	*P* = .001
Feelings score	96.88 (12.50)	87.50 (21.88)	*P* < .001	*P* < .001
Total score	94.64 (10.71)	85.71 (25.00)	*P* < .001	*P* < .001
Addendum score	100.00 (6.25)	100.00 (18.75)	*P* = .08	*P* = .62

Note: Values reported are the median (interquartile range).
Abbreviations: HEAR-QL, Hearing Environments and Reflection on Quality of Life.

The expected negative association between hearing loss and HEAR-QL observed for adolescents aligns with other studies, which found an inverse relationship between degree of hearing loss and HEAR-QL score in adolescents.[15,16] However, despite this association, the adolescent HEAR-QL scores in this study exhibited significant variability not explained by the PTA.

We found weaker evidence that the child HEAR-QL is associated with degree of hearing loss in Alaska Native children. We observed a trend of decreasing median QOL scores with increased level of hearing loss; however, our estimates were relatively imprecise, and any differences in median QOL scores may have been due to chance alone. We observed a high degree of variability in the HEAR-QL scores of children, both with and without hearing loss. This degree of variability could be attributed to lack of understanding of survey questions, lack of insight and ability to rank on a 5-point Likert scale, and a child's mood on the day of testing.[17] Our results do not provide the same level of evidence seen in other studies, which show that children with sensorineural hearing

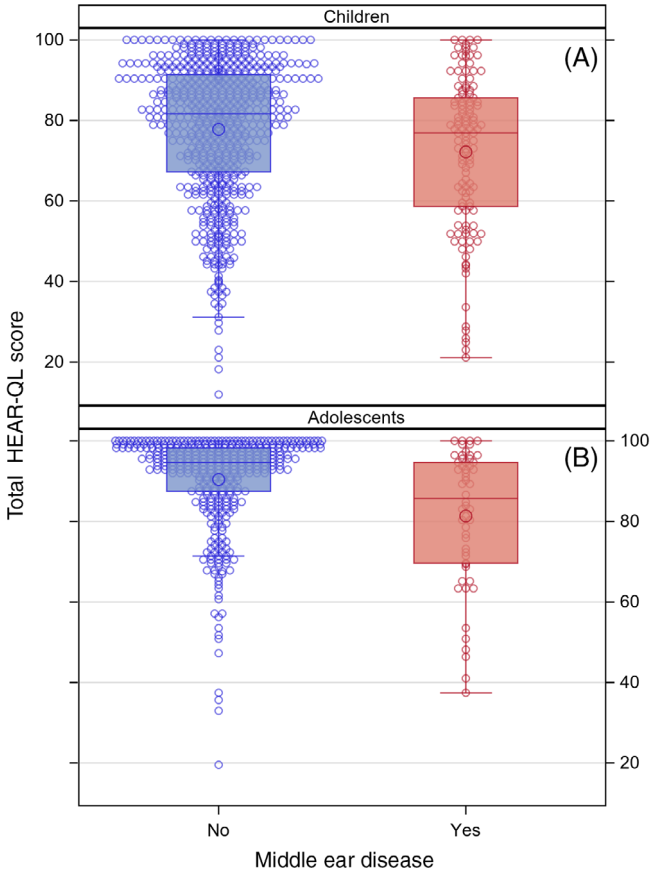

FIGURE 4 Total HEAR-QL scores by middle ear disease for children (A) and adolescents (B). HEAR-QL, Hearing Environments and Reflection on Quality of Life

loss score lower on the child HEAR-QL than their peers with normal hearing.[10]

Interestingly, we found that this tool distinguished children and adolescents with and without middle ear disease. While this tool has not previously been used to evaluate QOL in those with middle ear disease, the ability to evaluate the QOL of rural populations with higher prevalence of ear infections and infection-related hearing loss is important. Other questionnaires exist to measure quality of life in patients with middle ear disease, such as the ZCMEI-21,[18] and the OM-6,[19] both of which indicate that children with middle ear disease experience a lower quality of life than their peers without ear disease. Symptoms of middle ear disease, such as ear pain, fever, and otorrhea, can contribute to general discomfort, sleep disturbances, and even decreased appetite, all of which can contribute to a lower QOL.[20,21]

A four-item addendum was created based on community feedback about relevant culture and lifestyle not captured in the current HEAR-QL. This addendum was appended to the end of the original HEAR-QL to better assess QOL specifically in rural Alaska. Visiting a movie theater or a restaurant (items included in the original instrument) are not particularly relevant in rural Alaska, and therefore stakeholders made suggestions for questions about the effect of hearing loss on hunting, fishing, and attending dancing and drumming to capture QOL more accurately in this population. We found that the total HEAR-QL scores with the addendum exhibited a moderately strong correlation with standard HEAR-QL scores. Individually, however, addendum questions did not correlate with degree of hearing loss, similar to many items in the standard questionnaire. The benefit of validated and culturally relevant questions is well documented.[22–24] Future work to improve assessment tools for hearing-related QOL in rural Alaskan communities is therefore warranted.

Measuring hearing-related QOL in children can be done directly or indirectly. The child HEAR-QL is a tool completed by the child, thus directly measuring their own perceptions. While parent and teacher involvement are important in the workup and treatment of hearing loss, many studies have indicated that parent perceptions are poor predictors of their children's QOL.[25–28] However, younger children completing the child HEAR-QL did experience some difficulty completing the questionnaire on their own. In many instances, a study staff member had to read items to the child to complete the 5-point scale ranging from "Never" to "Always." In future studies to improve hearing-related quality of life assessment for children, it may be valuable to consider a reduced scale for response choices.

In general, hearing-related QOL is inherently difficult to quantify using a 'Never' to 'Always' scale. Additionally, we know that in other areas, such as self-reporting pain, children struggle with the abstractness of a numerical rating system.[29,30] In our study, it is possible that variability in scores could reflect a student's mood on day of testing, their current health or social state, as well as overlapping anxiety or depression[17,31] that may be independent of their hearing status. Recent work has demonstrated that visual scales are generally easier for young children to use for self-reporting.[30] Creation of a new tool using pictures, visual analog scales, or an alternative ranking system should be considered to accurately capture QOL in this age group.

A strength of our study was the large, representative sample of a rural Alaskan population. HEAR-QL has been validated in a small population in Missouri, but not in a sample size this large. An additional strength of our study was the direct self-reporting of HEAR-QL, instead of relying on a parent or guardian for answers.[26–28,32] Furthermore, all enrolled students of the target population participated in the study, which should minimize the potential for selection bias in estimating the difference in median quality of life score by hearing loss, regardless of how strongly students felt about the effect of hearing loss on their quality of life.

Our study was subject to several limitations. Since our estimated associations were exploratory and unadjusted, we cannot rule out confounding as an explanation for any associations observed between HEAR-QL score and hearing loss and middle ear disease. For example, certain living conditions may be a common factor for both infection-related hearing loss and self-reported lower hearing-related quality of life in children and adolescents. Our study also included only a small number of participants with moderate to severe hearing loss, which limited the precision of our estimates. We also observed large variability in HEAR-QL scores in this population. While this is not necessarily a limitation of our study, it does suggest that hearing-related QOL may be a multifactorial construct and the HEAR-QL instrument may

not be the best method of measuring hearing-related QOL in this population. Last, younger children in our study frequently needed the HEAR-QL read to them. While staff were trained to provide only reading assistance and to provide this support consistently across child participants completing the HEAR-QL, it is possible this process influenced HEAR-QL scores. Future studies should investigate the creation of a new tool to measure hearing-related QOL in children both with hearing loss and middle ear disease. Adaptations and variations to administration should be considered for use in diverse populations that may require different levels of administrative support to complete the tool. Item response choices that may be more understandable to younger children should be considered. The tool should also take into consideration lifestyle and environments that are either more generalizable or designed specifically for rural regions and Indigenous groups.

5 | CONCLUSION

The expected inverse association between hearing loss and HEAR-QL score was observed in adolescents; however, there was significant variability in the scores that cannot be explained by hearing loss. The expected negative association was not observed in children. Further investigation into factors responsible for this variability in HEAR-QL score across all age groups is warranted. HEAR-QL scores were associated with middle ear disease in both children and adolescents, suggesting the impact that middle ear disease may be contributing to quality of life in rural populations with higher rates of ear infections.

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APPENDIX A

TABLE A1 Community Specific Addendum to HEAR-QL

	Never	Almost Never	Sometimes	Often	Almost Always
1. Is it hard for you to hear when out in the country?					
2. Is it hard for you to hear at drumming and dancing or events at the gym?					
3. Do you spend less time hunting, fishing or berry picking because of your hearing?					
4. Do you ride snowmachines or 4-wheelers less because of your hearing?					

Abbreviation: HEAR-QL, Hearing Environments and Reflection on Quality of Life.