Hearing Impairment in the Extended High Frequencies in Children Despite Clinically Normal Hearing

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Objectives: Pediatric hearing impairment, regardless of degree and type, has a detrimental effect on speech perception, cognition, oral language development, academic outcomes, and literacy. Hearing assessment in the clinic is limited to 8 kHz although humans can hear up to 20 kHz. Hearing impairment in the extended high frequencies (EHFs > 8 kHz) can occur despite clinically normal hearing. However, to date, the nature and effects of EHF hearing impairment in children remain unknown. The goals of the present study were to determine the effects of EHF hearing impairment on speech-in-noise recognition in children and to examine whether hearing impairment in the EHFs is associated with altered cochlear functioning in the standard frequencies.

Design: A volunteer sample of 542 participants (4 to 19 years) with clinically normal audiograms were tested. Participants identified with EHF impairment were assigned as cases in a subsequent case-control study. EHF loss was defined as hearing thresholds greater than 20 dB in at least one EHFs (10, 12.5, or 16 kHz). Speech recognition thresholds in multi-talker babble were measured using the digit triplet test. Distortion product otoacoustic emissions (f_2 = 2, 3, 4, and 5 kHz) were measured to assess cochlear functioning.

Results: Thresholds in the EHFs were as reliable as those in the standard frequency range. Thirty-eight children had EHF hearing impairment regardless of a clinically normal audiogram. A linear mixed-effects model revealed that children with EHF hearing impairment had higher (poorer) mean speech recognition threshold than children with normal EHF sensitivity (*estimate* = 2.14 dB, 95% CI: 1.36 to 3.92; effect size = small). The overall magnitude of distortion product otoacoustic emissions was lower for children with EHF impairment (*estimate* = –2.47 dB, 95% CI: –4.60 to –0.73; effect size = medium). In addition, the pure-tone average for standard audiometric frequencies was relatively higher for EHF-impaired children (*estimate* = 3.68 dB, 95% CI: 2.56 to 4.80; effect size = small).

Conclusions: Hearing impairment in the EHFs is common in children despite clinically normal hearing and can occur without a history of otitis media. EHF impairment is associated with poorer speech-in-noise recognition and preclinical cochlear deficits in the lower frequencies where hearing thresholds are normal. This study highlights the clinical need to identify EHF impairments in children.

Key words: Digit triplets, Extended high frequency, Hidden hearing loss, Otitis media, Otoacoustic emissions, Speech-in-noise recognition.

(Ear & Hearing 2022;43;1653–1660)

INTRODUCTION

Hearing impairment is a common sensory disorder that affects 1 in every 5 children by 18 years (Lieu et al. 2020). Even slight or minimal hearing impairment during childhood has detrimental effects on auditory resolution, speech-in-noise recognition, language acquisition, educational and social/emotional aspects of functional status (Bess et al. 1998; Moore et al. 2020; Le Clercq et al. 2020). In general, studies that examined the effects of childhood hearing impairment have considered hearing loss in the standard audiometric frequency range (0.25 to 8 kHz). However, the upper-frequency range of human hearing extends up to 20 kHz. Frequencies above 8 kHz are designated as extended high frequencies (EHFs). While the consequences of clinical audiometric deficits are well established, there is a gap in knowledge regarding the adverse effects of EHF hearing impairment.

Hearing thresholds measured in humans over a substantial portion of the life span (5 to 90 years) suggest that age-related decline in hearing is evident at EHFs earlier than at standard audiometric frequencies (Lee et al. 2012; Rodríguez Valiente et al. 2014). Compared with adults, children have superior hearing in the EHFs (10 to 16 kHz) (Rodríguez Valiente et al. 2014; Lee et al. 2012). However, a recent study showed that nearly 20% of typically developing children (6 to 14 years) have EHF hearing loss (Hunter et al. 2021). To date, literature on EHF hearing in children remains sparse and generally focused on hearing sensitivity. EHF hearing impairment can occur in children due to several causes, such as pressure equalization (PE) tube surgery for treating otitis media (OM) (Gravel et al. 2006; Margolis et al. 2000; Hunter et al. 1996; Hunter et al. 2021), ototoxicity from cystic fibrosis treatment (Al-Malky et al. 2015; Blankenship et al. 2021; Caumo et al. 2017) or from heavy metal exposure (Shargorodsky et al. 2011).

Several studies demonstrate the role of EHF hearing in complex listening situations for adults (Best et al. 2005; Trine & Monson 2020; Motlagh Zadeh et al. 2019; Mishra et al. 2021; Hunter et al. 2020). Emerging evidence suggests that EHF hearing could contribute to speech-in-noise recognition in children as well (Flaherty et al. 2021; McCreery & Stelmachowicz 2013; Hunter et al. 2020; Nakeva von Mentzer 2020). Of relevance here, Blankenship et al. (2021) reported that children with clinically normal hearing treated with ototoxic drugs exhibit a significant positive relationship between EHF hearing and speech-in-noise recognition performance. Similarly, Petley et al. (2021) found a significant interaction between EHF hearing and Listening in Spatialized Noise—Sentences talker advantage scores for children. However, Hunter et al. (2021) did not find a direct relationship between EHF hearing and listening disorders in children, identified via a caregiver-questionnaire.

Hearing impairment in the EHFs can occur despite a normal audiogram. Characterizing pediatric EHF hearing impairment is of basic and clinical interest as it represents a form of hidden hearing loss that is not visible on the clinical audiogram. Currently, the potential consequences of EHF impairment in children remain largely unknown. Considering that EHF energy in speech is useful for children in complex listening environments (Flaherty et al. 2021), it was hypothesized that reduced EHF hearing would impact children's speech-in-noise perception. In addition, EHF hearing impairment can be considered as

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<zdoi; 10.1097/AUD.0000000000001225>

an early marker of subclinical hearing deficits in the lower frequencies (e.g., Hunter et al. 2020b). It is plausible that cochlear factors that reduce EHF hearing likely also influence hearing in the standard frequencies. Such subtle deficits may be evident in otoacoustic emissions (OAEs). Indeed, a relationship between EHF hearing acuity and distortion product otoacoustic emissions (DPOAEs) recorded at standard frequencies has been reported for adults (Arnold et al. 1999; Dreisbach et al. 2008). Likewise, Hunter et al. (2020a) showed that DPOAEs were lower in the standard frequencies in addition to the EHFs for children with a history of tympanostomy who also had elevated EHF hearing thresholds relative to the controls. However, the potential effects of age as a covariate were not adjusted in these studies.

The purpose of the present study was to investigate the nature of EHF hearing impairment in children. In a cohort of children with clinically normal audiograms, we aimed to (1) determine age-dependent variations in EHF hearing, (2) examine the effect of EHF hearing impairment on speech-in-noise recognition, and (3) evaluate if children with EHF impairment have pre-clinical cochlear deficits in the lower frequencies, as revealed by OAEs.

MATERIALS AND METHODS

Study Population

Participants aged 4 to 19 years volunteered for the study (n = 542; boys = 314). For treating age as a continuum through the onset of adulthood and for referencing to adults, eight 19-year olds were included. In order to determine the effects of EHF hearing impairment, participants identified with EHF impairment were assigned as cases in a subsequent case-control study.

EHF impairment was defined as the hearing thresholds greater than 20 dB HL for at least one EHF (10, 12.5, or 16 kHz) for either ear in the presence of a normal clinical audiogram (0.25 through 8 kHz). Inclusion criteria for the study were normal clinical audiogram, normal middle ear function assessed by 'A'-type tympanogram, and no recent history of otitis media within the past year. Participants were speakers of Indian English. Parents or older participants answered two specific questions related to speech-language development or other behavioral concerns (yes/no) and history of OM (yes/no). Audiometry and speech-in-noise recognition were tested in all participants. Otoacoustic emissions and threshold test-retest measurements were completed in a limited sample depending on the time availability in a single test session. All audiologic tests were conducted in a sound-attenuating booth. The study protocol was approved by the Institutional Review Board.

Pure-Tone Audiometry

Air conduction hearing thresholds were measured using pure-tones according to a modified Hughson-Westlake procedure for standard frequencies (0.25, 0.5, 1, 2, 4, 6, and 8 kHz) and EHFs (10, 12.5, and 16 kHz) via a MA 42 audiometer (MAICO Diagnostics GmbH, Germany) with Sennheiser HDA 300 headphones. The audiometer was calibrated according to the ANSI S3.6 2010 standards. For examining test-retest reliability, threshold measurements were repeated in both ears after headphone removal and replacement within the same session for a limited sample (n = 27 children or 54 ears, boys = 15).

Speech-in-Noise Recognition

The digit-in-noise recognition test was implemented via a custom software Angel Sound, similar to those described in previous studies (Oba et al. 2011; Mishra & Boddupally 2018; Mishra et al. 2022). Speech recognition thresholds (SRTs) were measured using a closed-set task, one-up and one-down, adaptive procedure converging on the signal-to-noise ratio (SNR) that produced a 50% correct score. Stimuli for the digit-triplet test included single digits "zero" through "nine" produced by one male speaker. The sampling frequency was 22,050 Hz. During testing, three digits were randomly concatenated and presented in a sequence (e.g., "five-two-six") in the presence of six-talker babble (3 males and 3 females). Stimuli were presented at 65 dB SPL using HDA 300 headphones. Participants were required to respond by clicking on response boxes labeled "zero" through "nine" and displayed on the laptop screen or by typing in the numbers on the keyboard. Correct identification of the entire sequence in digit-triplets reduced the SNR by 2 dB, while the SNR increased by 2 dB if the whole sequence in digit-triplets was not correctly identified. Each test run consisted of 25 trials. All SNR reversals were included for computing the SRT. Each child received two familiarization runs.

Distortion Product Otoacoustic Emissions

Distortion-product OAEs were recorded from both ears in 48 children (boys = 31) using a clinical system (ERO•SCAN, MAICO Diagnostics GmbH, Germany). Stimuli were two primary tones (f_1 and f_2; f_2/f_1 = 1.22) with f_2 frequencies 2, 3, 4, and 5 kHz and primary level (l_1 = 65 and l_2 = 55 dB SPL). The recording of the ear canal signals for a given f_2 continued until the noise floor was less than –10 dB SPL, or 4 sec of artifact-free averaging was completed. DPOAE level and noise floor estimates were obtained. DPOAE measurement was accepted if the SNR was ≥6 dB.

Statistical Analyses

The test-retest repeatability of thresholds was analyzed using intra-class correlation coefficients. The pure-tone averages for standard frequencies (PTA_{SF}) and EHFs (PTA_{EHF}), SRTs, and DPOAEs were modeled using separate linear mixed-effects models. The mixed-effects model allows probing the effect of a target variable while adjusting for the effects of other variables (Yang et al. 2014). For example, the effect of EHF impairment on SRT can be determined while adjusting for age effects, that is, the effect of EHF loss on SRT is adjusted for any correlation between age and SRT. The model included fixed effects for age (log-transformed), EHF group (EHF-normal and EHF-impaired), and OM history (positive and no). A random intercept was included to consider the variability between participants. Specific models planned are summarized in Table 1. In addition, to specifically examine the age-related variations, a single breakpoint in the response versus age function was estimated for PTA_{SF}, PTA_{EHF}, and SRT following a segmented regression approach called change-point analysis (Muggeo 2003; Muggeo 2016). Recognizing that threshold variability at EHFs could potentially impact the categorization of EHF groups, PTA_{EHF} was used as a predictor for most models. All tests were two-sided, and the significance threshold was set at p = 0.05. For significant effects from the mixed models, effects sizes were computed using partial ω^2 and are interpreted in

TABLE 1. Planned statistical models tested

A Priori Hypotheses or Models	Predictors	Response Variables
Effect of OM history on hearing sensitivity	Age and OM history	PTA_{SF} and PTA_{EHF}
Relationship between hearing sensitivity in the standard frequencies and EHFs	Age and PTA_{SF}	PTA_{EHF}
Effect of age on hearing sensitivity and speech-in-noise recognition (change-point analyses)	Age	$PTA_{SF,}$ PTA_{EHF}, and SRT
Speech-in-noise recognition	Model 1: age, OM history, EHF group; Model 2: age, EHF group, PTA for speech frequencies (0.5–4 kHz), PTA_{EHF}	SRT
Moderation effect of age on the relationship between EHF hearing and speech-in-noise recognition	Age, PTA_{EHF}, and age × PTA_{EHF}	SRT
Effect of EHF loss on DPOAEs	Age, OM history, and EHF group	overall DPOAEs
Relationship between DPOAEs and speech-in-noise recognition	Age and overall DPOAEs	SRT

DPOAE, distortion product otoacoustic emissions; EHF, extended high frequency (10, 12.5, and 16 kHz); OM, otitis media; PTA_{EHF}, pure-tone average for EHFs; PTA_{SF}, pure-tone average for standard frequencies (0.25 through 8 kHz); SRT, speech recognition threshold.

the specific sections. Analyses were performed using the R (R Project for Statistical Computing).

RESULTS

Thirty-eight out of 542 participants had EHF hearing impairment (boys = 23). Nineteen children had bilateral, and 19 children had unilateral (right ears = 11) hearing loss. The mean PTA_{EHF} was 5 (SD = 5) and 18 dB HL (SD = 4) for EHF-normal and EHF-impaired children, respectively. Among the three EHFs, 16 kHz followed by 12.5 kHz was more frequently affected. The 38 participants were categorized as EHF-impaired group, and the remaining 504 participants served as controls or the EHF-normal group; note, both groups had clinically normal audiograms. Five out of 38 children with EHF impairment had a history of OM. The mean ages of EHF-normal children (9 years, SD = 3) and EHF-impaired children (11 years, SD = 3) were significantly different (Welch's $t_{42.29}$ = –3.54, 95% CI: –3.23 to –0.88, $p < 0.001$).

The intra-class correlation coefficients show good test-retest reliability for individual EHFs, $PTA_{SF,}$ and PTA_{EHF} (Table 2). More than 95% of individual ears had test-retest threshold differences within ±10 dB for individual EHFs, which are highly comparable to standard frequency threshold variability and clinically accepted norms, and are consistent with the literature (Beahan et al. 2012).

Figure 1 plots the mean hearing thresholds as a function of frequency. It also shows the histogram of age distribution of children with and without EHF impairment. Compared with

TABLE 2. Test-retest reliability of EHF thresholds (n = 54 ears)

Frequency (kHz)	ICC	95% CI (ICC)	Number of Individual Ears in Different Test-Retest Difference (dB) Categories: 0	5	10	15	20
10	0.76	0.61–0.85	36	12	3	3	0
12.5	0.83	0.72–0.90	25	22	6	1	0
16	0.80	0.68–0.88	28	17	7	1	1
PTA_{EHF}	0.87	0.79–0.92					
PTA_{SF}	0.86	0.77–0.92					

ICC indicates intra-class correlation coefficient.
PTAEHF, Pure-tone average for extended high frequencies (10, 12.5, and 16 kHz).
PTA_{SF}, pure-tone average for standard frequencies (0.25 through 8 kHz).

the controls, EHF-impaired children had 3.68 dB higher PTA_{SF} and 13.45 dB higher PTA_{EHF} (PTA_{SF}: 95% CI: 2.56 to 4.80, $p < 0.001$, partial $\omega^2 = 0.07$; PTA_{EHF}: 95% CI: 11.73 to 15.16, $p < 0.001$, partial $\omega^2 = 0.30$). Forty-two children had a history of OM, consistent with the 5 to 9% prevalence of OM reported for the same geographic region (Chadha et al. 2014; Rupa et al. 1999). However, OM history had no significant effect on PTA_{SF} or PTA_{EHF} (PTA_{SF}: β= –0.06, 95% CI: –1.14 to 1.01, p = 0.91; PTA_{EHF}: β = 0.27, 95% CI: –1.65 to 2.17, p = 0.79). An additional mixed-effects model revealed that age and PTA_{SF} significantly predicted PTA_{EHF} (age: β = 3.08, 95% CI: 0.68 to 5.50, p = 0.01, partial ω^2 = 0.01; PTA_{SF}: β = 0.92, 95% CI: 0.82 to 1.02, $p < 0.001$, partial ω^2 = 0.32). This significant relationship between PTA_{SF} and PTA_{EHF} may suggest common underlying mechanisms driving pure-tone sensitivity.

The age-dependent variations in PTA with significant breakpoints are shown in Figure 2. Change-point analysis revealed significant breakpoints in PTA versus age functions (PTA_{SF}: breakpoint = 0.75 log units, 95% CI: 0.69 to 0.82, slope 1 = –14.61, slope 2 = 3.50, p = 0.03, age = 5.6 years; PTA_{EHF}: breakpoint = 0.97 log units, 95% CI: 0.60 to 1.04, slope 1 = –0.98, slope 2 = 13.42, p = 0.001, age = 9.4 years). A significant breakpoint in the SRT-age function was also observed (breakpoint = 1.01 log units, 95% CI: 0.99 to 1.04, slope 1 = –18.51, slope 2 = 2.53, $p < 0.001$, age = 10.2 years; Figure 2).

For the EHF-normal group, the mean SRTs by age groups with data from relevant literature are presented in Table 3. The mean values are consistent with relevant normative data from previous studies (Koopmans et al. 2018; De Sousa et al. 2018). The SRT was higher (poorer) for the EHF-impaired than the EHF-normal group (EHF loss: β = 2.14, 95% CI: 1.36 to 3.92, $p < 0.001$, partial ω^2 = 0.05) when significant age effects were adjusted (age: β = – 12.20, 95% CI: –13.49 to 10.91, $p < 0.001$, partial ω^2 = 0.39). Otitis media history had no significant effect on SRT (β = –0.46, 95% CI: –1.20 to 0.28, p = 0.22). An additional mixed-effects model revealed significant effects of age, PTA_{EHF}, PTA for speech frequencies (0.5 to 4 kHz) and EHF loss (age: β=–12.34, 95% CI: –13.61 to –11.09, p<0.001, partial ω^2=0.40; speech frequencies: β = 0.06, 95% CI: 0.02 to 0.10, p = 0.005, partial ω^2 = 0.007; PTA_{EHF}: β = 0.06, 95% CI: 0.03 to 0.09, p = 0.0001, partial ω^2 = 0.01; EHF loss: β = 1.17, 95% CI: 0.32 to 2.02, p = 0.007, partial ω^2 = 0.01). The variance inflation factor was less than 2 for all predictors confirming the

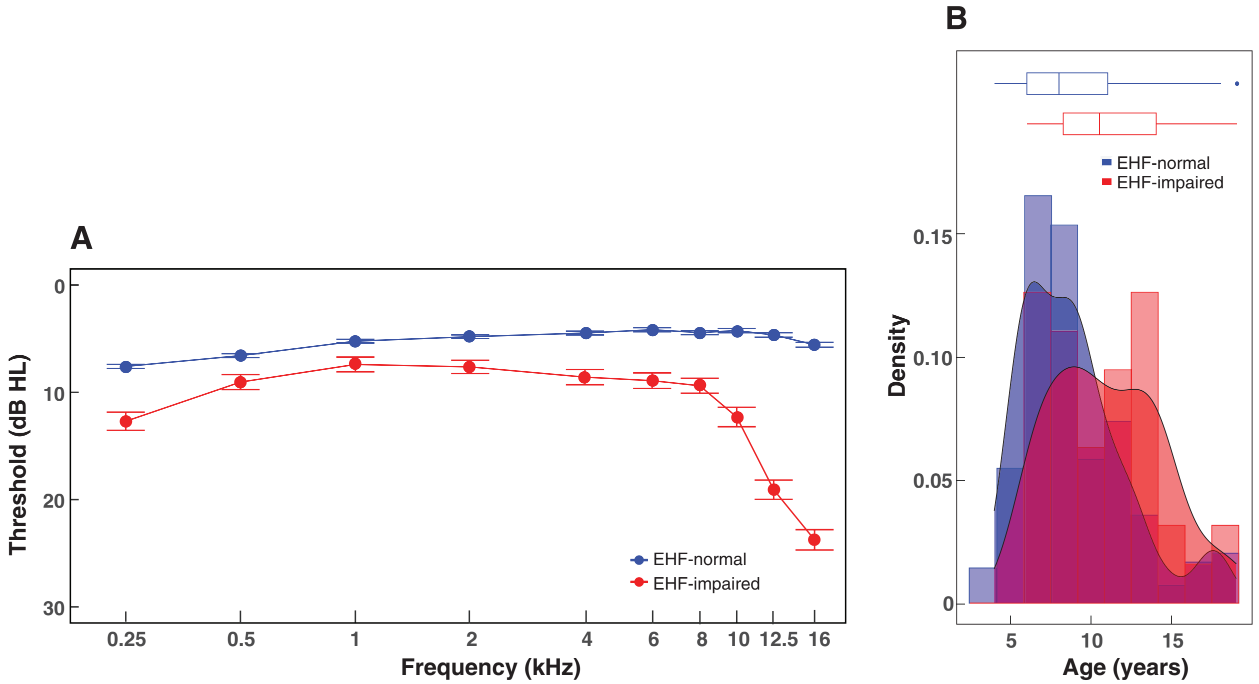

Fig. 1. Characteristics of EHF-impaired children. A, Mean hearing thresholds as a function of frequency (log-scaled) for children with and without EHF impairment. Error bars represent ±standard errors of the mean. B, Age distribution for EHF-normal and EHF-impaired children fitted using Kernel density functions. Box and whisker plots for EHF-normal and EHF-impaired groups are shown on the top. EHF indicates extended high frequency.

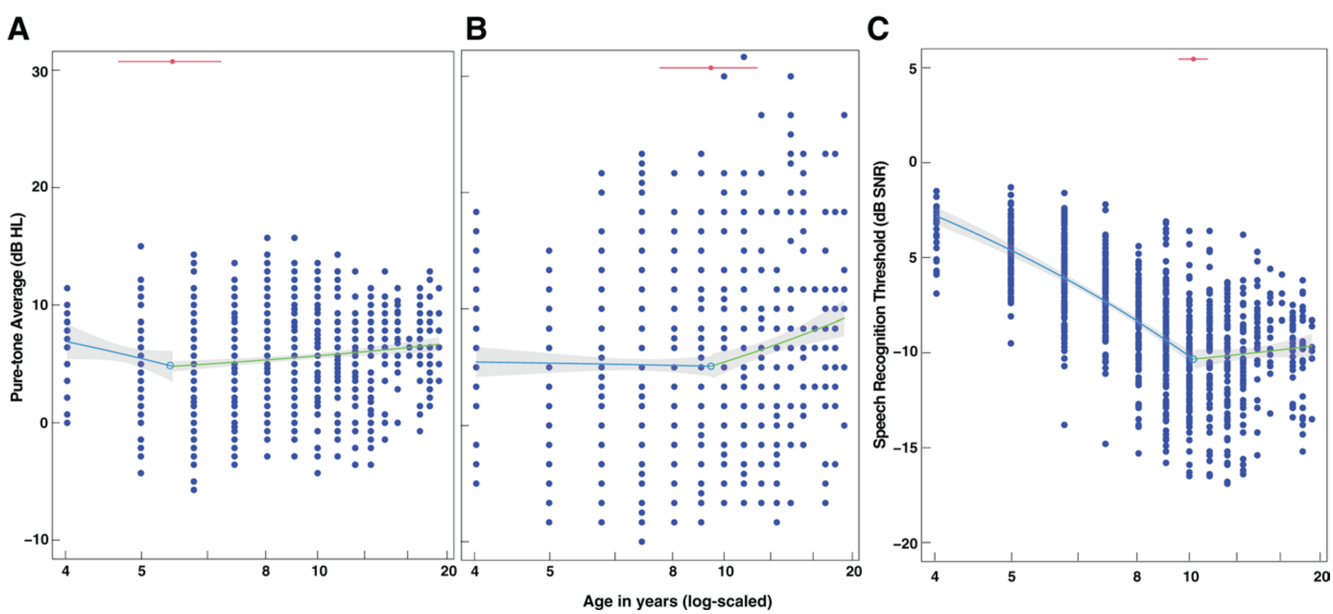

Fig. 2. Age-dependent variations. A, Pure-tone averages for standard frequencies as a function of age with a breakpoint at 5.6 years (0.75 log units). B, Pure-tone averages for EHFs as a function of age with a breakpoint at 9.4 years (0.97 log units). C, Speech recognition thresholds as a function of age with a breakpoint at 10.2 years (1.01 log units). For all plots, the gray-shaded areas mark the corresponding 95% confidence intervals for the line fit, and the horizontal error bars on the top (in red) show the breakpoints with their 95% confidence intervals.

lack of multicollinearity. The model outcomes show that EHF hearing loss is associated with higher (poorer) SRT even when the effects of other variables are adjusted. Figure 3 shows age-adjusted SRTs for EHF-normal and EHF-impaired groups. The age-dependent relationship between PTA_{EHF} and SRT is also depicted in Figure 3. The relationship between PTA_{EHF} and SRT was moderated by the age of the child (age: $\beta = -13.38$, 95% CI: –15.01 to –11.74, $p < 0.001$, partial $\omega^2 = 0.26$; PTA_{EHF}: $\beta = -0.10$, 95% CI: –0.26 to 0.05, $P = 0.19$; age×PTA_{EHF}: $\beta = 0.20$, 95% CI: 0.04 to 0.36, $p = 0.014$, partial $\omega^2 = 0.005$). To elucidate the significant interaction, SRT as a function of PTA_{EHF} (–10 to 35 dB HL) was computed for three distinct ages (lower and upper quartiles and median) using the moderation model outcome [i.e., Mean SRT = 3.71 − (13.38 × log(age))

TABLE 3. Mean speech recognition thresholds by age groups with relevant data from the literature

Age Group (yrs)	Present Study	Koopmans et al. (2018)	De Sousa et al. (2018)
4–5	–4.58 (1.49)	–5.8 (1)	—
6–8	–7.11 (2.37)	–7.4 (0.9)	—
9–12	–9.96 (2.80)	–8.4 (0.3)	—
5–15	–8.29 (3.22)	—	–7.2 (5.4)

Values in parenthesis show standard deviations.

– (0.10 × PTA_{EHF}) + (0.20 × log(age) x PTA_{EHF}]. Note, any age can be input into the equation; however, these specific ages (6.03, 8.91, and 10.96 years) show different patterns of the interaction (Figure 3). Although the magnitude of the interaction effect was small, the increase in SRT with increase in PTA_{EHF} is higher for older children (e.g., 8.91 and 10.96 years) compared with younger children (e.g., 6.03 years).

Out of 48 children, DPOAEs were absent in both ears of one child with bilateral EHF impairment. Figure 4 shows box-whisker and violin plots for DPOAEs for the remaining participants (n = 47 or 94 ears; ears with EHF loss = 26). The overall DPOAE magnitude was significantly lower for the EHF-impaired group relative to the EHF-normal group (β = –2.47, 95% CI: –4.60 to –0.73, p < 0.001, partial ω^2 = 0.13). The effect of age or OM history was not significant (age: β = 3.65, 95% CI: –4.83 to 11.93, p = 0.38; OM history: β = –1.83, 95% CI: –6.02 to 2.39, p = 0.38). DPOAEs also predicted SRTs with adjustments for age effects (age: β = –9.94, 95% CI: –14.28 to –5.60, p < 0.001, partial ω^2 = 0.29; DPOAEs: β = –0.17, 95% CI: –0.29 to –0.04, p = 0.006, partial ω^2 = 0.11; n of ears = 94). For every unit decrease in the overall DPOAEs, the SRT worsens by 0.17 units when age effects are adjusted.

DISCUSSION

The main findings are (1) of 542 participants, 38 children had EHF impairment despite normal audiograms; (2) significant breakpoints in age versus pure-tone averages and SRT functions were observed; (3) despite clinically normal hearing, EHF-impaired children performed more poorly on the digits-in-noise recognition task relative to the EHF-normal group (effect

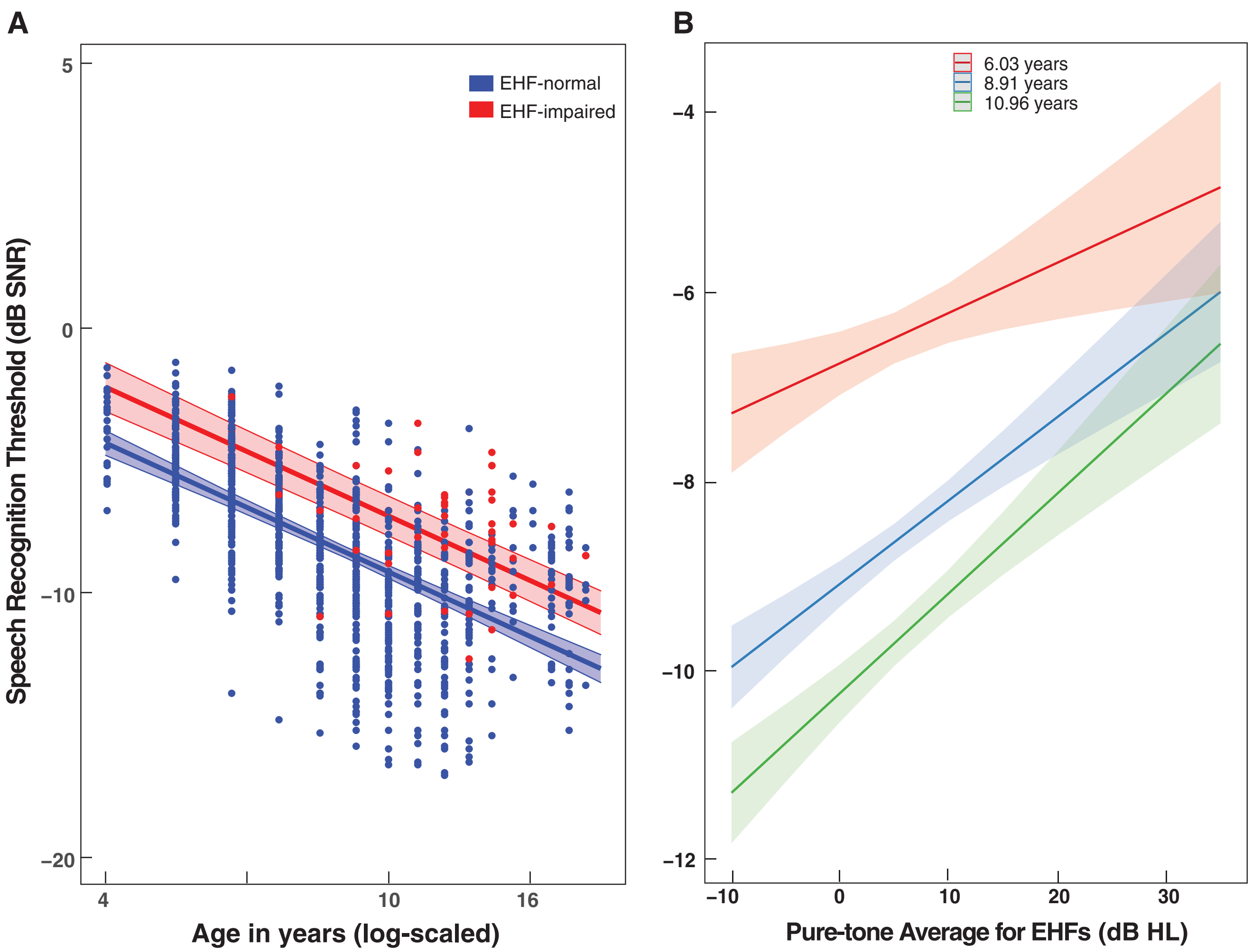

Fig. 3. Age-adjusted speech recognition thresholds. A, SRTs as a function of age with linear fits comparing NH and EHF-impaired children (SRT = 2.95 – 12.17 × log(age) + 2.11 × (g), where g is 0 for NH and 1 for EHF-impaired children). B, The relationship between pure-tone average for EHFs and SRT for three different ages (0.78, 0.95, and 1.04 in log units); these ages are representative of the distinct relationship patterns; mean SRT = 3.71 – (13.38 × log(age)) – (0.10 × PTA_{EHF}) + (0.20 × log(age) × PTA_{EHF}. For all plots, the shaded areas mark the 95% confidence intervals for corresponding lines. EHF indicates extended high frequency; SRT, speech recognition threshold.

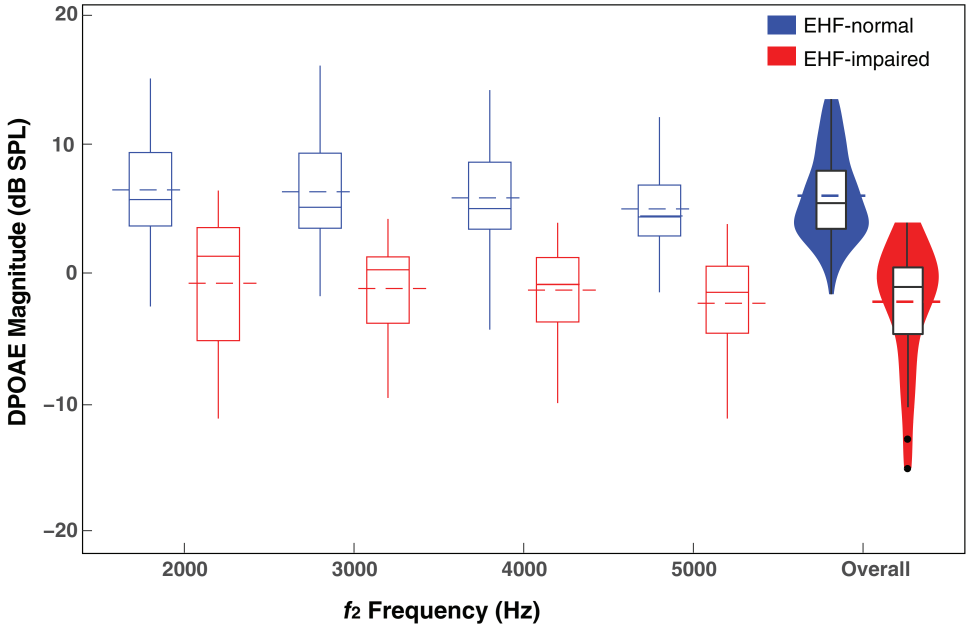

Fig. 4. Distortion product otoacoustic emissions. Box and whisker plots showing DPOAE magnitudes for various f_2 frequencies for EHF-normal and EHF-impaired children. The boxes span the interquartile ranges, whiskers show the ranges, the solid and broken horizontal bars indicate the median and mean, respectively. For overall DPOAE magnitudes, violins show the probability density smoothed by a kernel density estimator. DPOAE indicates distortion product otoacoustic emission; EHF, extended high frequency.

size = small); and (4) DPOAEs in the standard frequencies for EHF-impaired children were lower compared with the EHF-normal group, and DPOAEs predicted speech-in-noise recognition (effect size = medium).

Age-Dependent Variations in Hearing

EHF hearing sensitivity remained relatively unchanged until about 9 years of age and slowly deteriorated thereafter. This observation is consistent with the onset of elevated EHF thresholds at around 10 years reported by Trehub et al. (1989). In contrast, improvements in hearing thresholds for the standard frequencies are evident up to 6 years of age, after which there was a gradual change in sensitivity until adulthood. These findings are broadly consistent with previous studies showing that the hearing sensitivity for standard frequencies matures later than the EHFs (Trehub et al. 1988; Trehub et al. 1989). For instance, hearing thresholds for 2 and 4 kHz mature at around 8 years, whereas maximal sensitivity for 10 kHz is achieved by 4 or 5 years (Trehub et al. 1988). It is difficult to ascertain the mechanisms responsible for these frequency-dependent developmental changes in hearing sensitivity. Since young children are involved, it is intuitive to consider factors such as attention, motivation, cognitive processes, and internal noise as the primary source for developmental changes (e.g., Buss et al. 2006). These factors would likely exert similar changes across frequency instead of the frequency-dependent changes observed here, although it is not unreasonable to expect that different frequencies may impose different attentional/cognitive demands. Likewise, the contribution of certain peripheral auditory mechanisms, for example, immature sound transmission via the middle ear (Mishra et al. 2017), to the differential changes in hearing sensitivity cannot be entirely ruled out.

Speech understanding in noise follows a prolonged developmental process, and this process is even longer when the masker is competing speech (Brown et al. 2010; Leibold & Buss 2019). Although auditory factors, such as central auditory maturation, and non-auditory factors, such as selective attention, contribute to the age-related improvement in performance, the digit-triplet test is less dependent on non-auditory factors and requires minimum linguistic skills (Smits et al. 2013; Kaandorp et al. 2016). Children as young as 4- or 5-year old can perform the digit triplet test (Koopmans et al. 2018; De Sousa et al. 2018; Moore et al. 2019). As expected, age had a significant effect on the SRT with a large effect size. The SRT improved with age to reach adult-like performance at around 10 years of age, similar to the prolongation of maturation of SRT for the digit triplet test reported by Koopmans et al (2018).

EHF Hearing Impairment and Speech-in-Noise Recognition

The most striking finding is that the onset of EHF hearing loss (i.e., EHF hearing thresholds > 20 dB HL), without obvious risk factors such as PE tubes or ototoxicity, can be observed as early as 6 years of age in typically developing children. Among the sampled population with clinically normal audiograms, 7% of children had EHF hearing impairment and the EHF loss count increased with age. Compared with the present study, a higher percentage of prevalence of EHF loss has been reported for 13- to 19-year olds (10%) (Peñaranda et al. 2020) and for 6- to 14-year olds with suspected listening disorders (32%) (Hunter et al. 2021). Although 5 out of 38 (13%) EHF-impaired children had a history of OM, unlike previous studies (Gravel et al. 2006; Margolis et al. 2000; Hunter et al. 1996), we did not find a significant effect of the history of OM on EHF hearing (PTA_{EHF}). The present study was not prospectively designed to examine the effects of OM on EHF hearing; however, an important possibility for the discrepancy with previous studies is that no children with OM history in the study sample were treated with PE tubes.

EHF hearing impairment was associated with poorer speech-in-noise performance (higher SRT). The effect of EHF impairment on SRT remained significant even after adjusting for age and other effects such as PTA for speech frequencies. Petley et al. (2021) reported a weak but significant interaction (Spearman $\rho = 0.27$) between EHF hearing and the Listening in Spatialized Noise—Sentences Talker Advantage scores. Likewise, Flaherty et al. (2021) showed that access to EHFs improves speech-in-speech recognition in NH children. We found that age moderates the relationship between EHF hearing and speech-in-noise recognition for children. The worsening of SRT with a decline in EHF hearing was higher for older children relative to younger children. This may suggest that younger children are inefficient in using EHF cues to the extent older children can for speech-in-noise recognition.

The functional consequences of EHF hearing loss can be serious for children, although the SRT was only 2 dB poorer for the EHF-impaired group. The effect may be especially relevant for children in real-world listening situations, for example, classroom, where the acoustic environment is suboptimal (Crandell & Smaldino 2000). In addition, the lack of EHF-related SRT advantage could exacerbate the adverse effects of other factors, such as inattentiveness, on learning in the classroom. Besides, the 2-dB corresponds to ~15% change in speech recognition score based on the digit psychometric functions for children (Moore et al. 2019).

Cochlear Function in Lower Frequencies

Children with EHF impairment had significantly lower DPOAEs even for the standard frequencies, consistent with a recent study (Hunter et al. 2021). The effect remained stable even after adjusting for effects of age and OM history. A similar effect was also observed for hearing thresholds for the standard frequencies, wherein EHF-impaired children had poorer hearing sensitivity (~4 dB) relative to the EHF-normal group. Reduced DPOAEs in the 2 to 5 kHz region associated with EHF hearing impairment could indicate subtle outer hair cell dysfunction at the standard frequencies or that DPOAEs reflect cochlear mechanisms over a broad range of frequencies. It is unlikely that DPOAEs received contributions from far-basal sources along the basilar membrane that encode frequencies one octave or higher than the highest f_2 frequency tested (5 kHz). In humans, the far-basal (high-frequency) source of DPOAEs has been shown to be 1/3rd octave higher than the f_2 frequency (Martin et al. 2009; Martin et al. 2011). Thus, it is possible that the pathology that caused EHF hearing loss is also associated with subclinical cochlear dysfunction in the standard frequencies. In addition, cochlear functioning in the standard frequencies predicted speech-in-noise performance with a medium effect size even after adjusting for age effects. Nevertheless, the consequences of such subtle cochlear insults on the auditory resolution during development remain unknown.

Clinical Implications

Currently, there is no clinical intent to identify, monitor, and/or manage pediatric EHF hearing impairment. This is perhaps due to the assumption that hearing impairment for frequencies above 8 kHz has no adverse consequences. We demonstrated that EHF-impairment in children is associated with higher standard frequency hearing thresholds, reduced DPOAEs in lower frequencies and higher (poorer) SRTs. The effect sizes were small to medium. We also found that the test-retest variability of hearing thresholds in the EHFs for children was within ± 10 dB, suggesting acceptable clinical reliability of EHF thresholds for pediatric applications. These findings highlight that including EHF audiometry in pediatric hearing assessment could provide invaluable hearing health information. For example, EHF hearing could indicate early signs of deterioration in the overall hearing health.

The enormous enthusiasm in hidden hearing loss among researchers and clinicians alike has led to the frequent use of cochlear synaptopathy and hidden hearing loss terms interchangeably. Recent reports (Hoben et al. 2017; Kohrman et al. 2020) suggest that other mechanisms such as outer hair cell deficits in addition to cochlear synaptopathy could contribute to the pathogenesis of hidden hearing loss in humans. Present findings support this emerging notion that hidden cochlear impairments, in the form of altered DPOAEs and elevated EHF thresholds, can occur despite a normal audiogram.

Limitations

The prevalence rate of EHF impairment may not be generalized to other populations. It is possible that the digit triplet test underestimated the effects of EHF hearing impairment that would be optimally assessed using a spatial hearing test, for example, Petley et al. (2021).

CONCLUSIONS

Children as young as 6 years can have EHF hearing impairment despite normal audiograms and without obvious risk factors. EHF hearing impairment in children is associated with relatively poorer speech-in-noise recognition, and suboptimal cochlear function and elevated hearing thresholds in the standard frequencies. This study highlights the role of EHFs in hearing in children and supports the consideration of subclinical cochlear deficits in defining auditory impairments that are not visible on the standard audiogram. Future studies are warranted to determine the effects of pediatric EHF hearing impairment on auditory, language, and communication development.

ACKNOWLEDGMENTS

The authors have no conflicts of interest to disclose.

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Received October 12, 2021; accepted March 12, 2022; published online ahead of print April 25, 2022.

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