Title: Hearing threshold shifts among 11 − 35 year olds with early hearing impairment.

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Appendix is attached to this manuscript. [Supplemental digital content is available for this article. Direct URL citations appear in the printed text and are provided in the HTML and text of this article on the journal's Web site (www.ear-hearing.com)].

Definitions

* Unless stated otherwise, 'dB' refers to dB HL.

†In this article, *hearing loss* refers to the condition in which individual hearing threshold levels (HTLs) differ from a recognized [normative population] standard, while *HTL shift* refers to deviation from an individual's specified baseline audiogram.

‡Leisure noise refers to sounds at or above 80 dB (A), encountered during recreational activities (e.g., music in nightclubs or exercise classes, gunshots, racing vehicle noise) or during domestic (nonwork) activities (e.g., using power tools).

ABSTRACT

Objectives: Data obtained from the clinical records of selected 11-to 35-year-olds with preadult onset hearing impairment were analyzed with two primary aims: (1) to determine the incidence of hearing threshold level (HTL) shift in this cohort and, (2) to examine the relationship between HTL shift, whole-of-life noise exposure and other factors.

Design: Cross-sectional cohort study [erratum: Retrospective cohort study with longitudinal follow-up]. Retrospective HTL + survey data for a sample of 237 young Australians receiving hearing (re)habilitation services were obtained. From these data, two subsets, (A) n = 127 and (B) n = 79, were analyzed. Participants with risk factors for progressive hearing loss (other than noise exposure) were excluded from both subsets. Subset (A) additionally excluded cochlear implant recipients, and subset (B) excluded cases with diagnosis of hearing loss after age 5 years. Using subset (A) data, the differences between final (recent) and specified baseline (initial) HTLs at 250, 500, 1000, 2000, and 4000 Hz were calculated and three criteria for HTL shift were applied. Correlations between reported noise exposure and HTL shift were calculated (Mann-Whitney *U* test). Using subset (B) data, relationships between high frequency (HF) HTL shift and exposure, and other personal and extrinsic factors were examined (Cox Regression model). Survival analyses (Kaplan-Meier) were performed to reveal the temporal pattern of HF shift. The magnitude of HF shifts at 5, 10 and 15 years post-initial audiogram (i.e., specified baseline) were also calculated.

Results: For subset (A), HTL shift (≥ 15 dB any frequency, and/or ≥ 10 dB * at two adjacent frequencies) was observed in 46.5% of cases examined. HF shift (≥ 15 dB at 2000 and/or 4000 Hz; one or both ears) was observed in 33.1% of cases. There was no relationship between HTL shift and reported whole-of-life exposure. For subset (B), no relationship was found between HF shift and noise exposure, nor 9 of 10 personal or extrinsic covariates tested. HF shift was significantly associated with HTL ≥ 70 dB at 2000 and/or 4000 Hz at initial audiogram. Survival analysis also illustrated that HF shift was more frequent, and occurred earlier, when HF hearing loss was ≥ 70 dB at initial audiogram. Median HF shifts at 15 years after initial audiogram were in the magnitude of 5 to 10 dB, and at the 90th percentile shifts were 25 to 30 dB.

Conclusions: HTL shift was observed in almost 50% of cases without predisposing factors for progressive hearing loss. The magnitude of HF shift increased gradually over time. While no relationship was found between HTL shift and noise exposure, the interpretation of this finding is restrained by the small spread of whole-of-life noise exposures, within a relatively conservative range. Nevertheless, this is the first direct examination of the relationship between HTL shift and noise exposure in young people with preadult hearing impairment.

INTRODUCTION

Hearing impairment (HI) is one of the most common disabling conditions (Borchgrevink, 2003). At the time of writing, the Australian Federal Government, via the statutory authority Australian Hearing® (AH), was the sole provider of fully-subsidized hearing (re)habilitation services to its citizens from birth to 26 years of age. As of December 2014, 21,968 Australians less than 26 years of age were receiving AH services (Australian Hearing, 2015).

Childhood hearing loss[†] has the potential to impair language development, literacy, educational, and vocational opportunities (Access Economics, 2006). Adjusting to the diagnosis of permanent hearing loss is challenging for parents of affected children (Watson et al., 1990), and prognostic information may be valued during this adjustment process. To date, there has been little scientific evidence from which to inform parents, particularly when the etiology of the hearing loss cannot be medically determined.

Information counselling may consequently focus on the configuration of the child's preliminary audiogram. Unfortunately, however, HI is not always a stable condition. Hearing threshold level (HTL) shifts were scientifically observed as early as the first decade of the 20th century (Barr & Wedenberg, 1965) and hearing deterioration was described in a number of subsequent publications (Macrae, 1968; Reilly et al., 1981; Newton & Rowson, 1988; Parving, 1988; Levi et al., 1993; Brookhouser et al., 1994; Berrettini et al., 1999; Pittman & Stelmachowicz, 2003). Hearing deterioration can be very significant, both physically and psychologically (Meyerhoff et al., 1994) and can add to parental/family stress. It is important that progressive loss is quickly identified so that any possibilities for medical treatment can be investigated (Meyerhoff et al., 1994). If unremediated, HTL shift can increase hearing disability – by degrading communication fluency, social interaction and educational progress. HTL shift, therefore, places a burden on (re)habilitation management programs, as changes to devices, educational and communication strategies must be made to optimize outcomes. The clinical significance of progressive hearing loss in childhood and adolescence is acknowledged in current Australian and US pediatric audiology protocols (e.g., King, 2010; American Academy of Audiology, 2013) and the clinical monitoring of HTLs is emphazised.

Estimates of the incidence of progressive sensorineural (SN) hearing loss in young people are limited, and vary in the previous literature, according to (1) the population of interest (e.g., age, etiology), (2) the HTL shift considered to be a "significant" deterioration (criterion) and, (3) the observation period (time between the initial and most recent audiograms; Newton & Rowson, 1988). A list of relevant research is provided in the Appendix [Appendix in Supplemental Digital Content 1 http://links.lww.com/EANDH/A340]. A wide range in the estimated incidence has been reported: 2 to 33% (Meyerhoff et al., 1994), 4 to 30% (Berrettini et al., 1999). Based on a review of studies examining the likelihood and rate of deterioration in HTLs based on etiology, Meyerhoff et al. (1994) also concluded that 25 to 50% of children born with genetic, SN hearing loss will experience HTL shift (given sufficient residual hearing at initial assessment for HTL shift to be measured).

Brookhouser (2002) stated that audiological management decisions should ideally consider any predictable pattern of HTL behavior. In practice, the possibility of future HTL shift is more likely to be emphasized where a specific risk factor for progression of hearing loss is medically understood. Etiologies associated with progressive SN hearing loss include Alports, Ushers, and Waardenburgs syndromes, and various presentations of inner ear dysgenesis, for example, Mondini dysplasia (Meyerhoff et al., 1994) and "enlarged vestibular aqueduct syndrome" (Madden et al., 2003; Oyler, 2007). However, progressive hearing loss also occurs in cases of nonsyndromic etiology (Berrettini et al., 1999), and an Israeli study of 92 children with bilateral SN hearing loss found no relationship between etiology and hearing deterioration (Levi et al., 1993).

Descriptions of progressive hearing loss in the literature are relatively few, and typically involve case reports of families with hereditary SN hearing loss (Newton & Rowson, 1988). Barr and Wedenberg (1965) commented that progressive hearing loss was sometimes characterized by irregularity, where for long periods the HTLs would be stable, but then suddenly deteriorate. Deterioration could be unilateral or bilateral. Newton and Rowson (1988) also observed that deterioration can be rapid or, conversely, so gradual that HTL variation may be attributed to test-retest error, particularly if only consecutive audiograms were examined. It has also been observed that HTL shifts tend to occur simultaneously in the same direction at adjacent audiometric frequencies, rather than at individual frequencies (Macrae, 1988), while Newton and Rowson (1988), suggested that patterns of progression do not occur in a uniform manner across the frequency range. Newton and Rowson also described a tendency for deterioration to be greater if initial hearing loss is milder in degree. Berretini et al. (1999) emphasized that hearing deterioration can occur at any age. In some cases HTLs may also fluctuate (i.e., deteriorate and then recover) over time (Brookhouser, 2002).

Excessive hearing aid amplification (Macrae, 1995) and/or the use of hearing aids in high noise environments (Dolan & Maurer, 1996), have been implicated in the occurrence of HTL shift, particularly in children with HI. Ching et al. (2013) conducted a modeling study, in which the asymptotic threshold shifts associated with contemporary hearing aid technology (including automatic gain control) were predicted. It was concluded that individuals with more severe hearing loss will be affected by amplification-related HTL shift, even when non-linear hearing aids are fitted according to recognized prescription procedures, and particularly if hearing aids are used in loud environments.

Overall, there is a dearth of contemporary scientific evidence concerning progressive hearing loss in young people. The need for more research, to ascertain the incidence of progressive hearing loss in larger populations of children and to determine the associated factors, was highlighted more than 20 years ago (Levi et al., 1993). There have, however, been few studies of this kind since that time (see Appendix) [Appendix in Supplemental Digital Content 1, http://links.lww.com/EANDH/A340].

Leisure noise exposure[‡] has also been associated with pure-tone hearing loss and there is an extensive body of relevant literature (Carter et al., 2014). However, this literature, refers exclusively to young people with “normal” (non-impaired) hearing pre-exposure and, overall, the risk has tended to be overstated (Schlauch & Carney, 2012; Carter et al., 2014; Williams et al., 2015). There is, however, evidence that a proportion of young people are exposed to sufficient noise for HTL shift to feasibly result (Tambs et al., 2003; Zhao et al., 2010; Beach et al., 2013). Before the current investigation, there has been no commentary on the leisure noise exposure of young people with early hearing impairment, although there is a clinical perception that it may be a contributing factor in HTL shift. As a duty of care, clinicians are encouraged to advise that hearing aid wearers “avoid prolonged exposure to high noise levels” (Dillon, 2012, p. 333).

This study aimed to address these broad gaps in knowledge. There were two main research questions:

1. In the absence of specific risk factors for progressive SN hearing loss, what proportion of young people with HI experience HTL shift?

2. Is there is a relationship between whole-of-life noise exposure and HTL shift, whereby greater exposure is associated with increased HTL shift?

MATERIALS AND METHODS

HTL data were collected during the latter part of a two-phase study of the hearing health, attitudes and behavior of young Australians, initiated in the context of community concern that leisure noise exposure is causing pure-tone HTL shift in an increasing number of young people. The details of the first study phase (in which most participants had "normal" hearing) have been described previously (Carter, 2011; Williams et al., 2014; Carter, et al. 2015; Williams et al., 2015; Carter et al., 2016a).

Ethics

Protocols were approved by the Australian Hearing Human Research Ethics Committee and the Human Research Ethics Committee, University of Sydney. Participation was voluntary and there were no incentives for taking part.

Participant recruitment

The present study involved 268 adolescents and young adults with preadult onset HI. Most participants were recruited via AH, however, hearing-related consumer groups and some private audiology practices also promoted participation. Personalized invitation packages (including survey forms) were distributed to potential participants during AH appointments at 15 New South Wales (NSW) hearing centers. Age was the only specific inclusion criterion; however, AH audiologists occasionally withheld invitations (e.g., in cases of severe client disability, health or family issues). Approximately 80% of participants lived in greater metropolitan Sydney, and the remainder in regional/country NSW. Hearing loss was SN in nearly all cases. As shown in Table 1, the degree of hearing losses varied. Most participants were diagnosed with HI before school age, and almost all by adolescence. Most were fitted with, and wore, bilateral hearing aids.

Table 1. Participant information.

Total survey responses: >10 < 36 years	n = 268			
A) Incidence analysis subset:	n = 127			
B) Survival analysis subset:	n = 79			
Mean participant age (A):	19.4			
(B):	18.9			
Participant gender (A):	Female: 84 (66%)	Male: 43 (34%)		
(B):	Female: 50 (63%)	Male: 29 (37%)		
Degree of pure tone hearing loss (better ear 4 $FAHL_{500, 1000, 2000, 4000 Hz}$)				
	Mild (21−39 dB)	Moderate (40−59 dB)	Severe (60−89 dB)	Profound (90+ dB)
A)	37 (29.1%)	48 (37.8%)	25 (19.7%)	2 (1.6%)
B)	15 (18.9%)	31 (39.2%)	22 (27.8%)	8 (10.1%)
Reported device use				
	Hearing aids only	Cochlear implant	Devices no longer used	
A)	121 (95.0%)	0	4 (3.1%)	
B)	67 (84.8%)	12 (15.2%)	0	

Two participants used assistive listening device only.
4 FAHL, four frequency average hearing level.

Data collection

Detailed clinical information (e.g., audiograms, hearing aid data and case histories) were collected from the participants' AH clinical files by research audiologists and/or were provided by subsequent hearing service providers. As noted, national protocols are followed by AH, therefore it is assumed that HTLs were measured in accordance with relevant standards for calibration and test procedure.

HTL data

In most cases, pure-tone audiometry (PTA) records from age at hearing loss diagnosis to age at study participation (or age of loss of eligibility for AH if prior to participation) were available. As AH pediatric protocols prescribe regular monitoring of HTLs, the number of audiograms per participant record was often large. To constrain the scale of data, audiograms at specific target ages were collected. For 11- to 17-year-olds, target audiogram ages were; 5, 7, 8, 9, 10 and 15 years; for 18 - to 25-year-olds target audiogram ages were 5, 8, 9, 10, 15 and 20 years and; for 26 - to 35-year-olds, target audiogram ages were 5, 10, 15, 20, 25 and 30 years. The audiogram closest to the target age (and without indication of test unreliability) was copied. In most cases a serial ("continuous") audiogram was available for the audiometric frequencies 250, 500, 1000, 2000, 3000, and 4000 Hz and this record was also obtained. HTL values were entered into a purpose-designed database and de-identified audiograms were stored electronically for quality assurance. For participants above the eligibility age for AH services, a checklist requesting audiometric information (if available) was sent to the participant/their hearing services provider. In a few cases, data for analysis were a composite of AH and post-AH assessments.

The target age for initial audiogram was 5 years. As audiometry is usually truncated in younger children (re; developmental stage/concentration span), the initial HTL data were often limited to 250, 500, 1000, 2000, and 4000 Hz. Reliable audiograms were not always available at the exact target age, for reasons including, unreliable responses or middle ear dysfunction at target age, or diagnosis later than the target age. Of 79 cases with diagnosis by 5 years of age, 65 (82.3%) had a reliable audiogram recorded by age 5 years, eight cases by age 6 years, and the remaining seven cases by age 7 years.

Survey data

The development of participant surveys used in this study has been described previously (Carter et al., 2016a) and surveys are publicly available (Carter et al., 2016a, b). The surveys included a large number of items pertaining to hearing health, leisure behavior and attitudes towards noise exposure. Demographic and participant health items for under 18-year-olds were included in a complementary parent survey. A key element of the questionnaire was a concise, but detailed, measure of lifetime activity participation (focused on noisy activities) referred to as the "leisure table", plus additional questions relating to music listening habits (including personal stereo player [PSP] use). The response format for the leisure table is illustrated in Figure 1. Response options for the additional question, "on average, how often have you attended nightclubs, dance parties, gigs, etc.?" were: less than once a year/once or twice a year/once every few months/1 to 3 times a month/1 to 3 days a week/4 to 5 days a week/6 to 7 days a week. Regarding frequency of PSP use, response options were: less than once a month/1 to 3 times a month/1 to 3 days a week/4 to 5 days a week/6 to 7 days a week.

Surveys were returned by 253 individuals with HI in the target age range. In several cases, however, incomplete responses prevented estimation of the individual's whole-of-life noise exposure.

Go to **live music** (e.g., bands, concerts, musicals) at **smaller venues** (e.g., **hall, theatre, club** etc.)			
About how much have you done this activity? ☐ Never → go to next activity ☐ Less than once a year ☐ 1-2 times a year ☐ Once every few months ☐ Once a month ☐ Once every 2-3 weeks ☐ Once a week ☐ More than once a week	**About how long do (did) you do the activity each time?** ______hours each time **How old were you when you first did this activity?** ________years of age **How many times a week?** _______ times	**How many years altogether have you done this activity?** _______years in total **Hearing aids/CI worn?** *(switched ON)* ☐ **Never** ☐ **Sometimes** ☐ **About half the time** ☐ **Most of the time** ☐ **Always**	**Hearing protection used?** ☐ **Never** ☐ **Some of the time** ☐ **Mostly/always** Type____________

Figure 1. Activity participation response format.

Data analysis

Data selection

HTL + comprehensive survey data were provided in 237 cases. From these, two subsets were analyzed, with the primary exclusion criterion of presence of risk factor(s) for progressive hearing loss (other than noise exposure), as noted previously. In particular, data for individuals with structural abnormalities of the inner ear, other conditions known to be associated with progressive SN hearing loss, and fluctuating/recurrent conductive hearing loss were excluded. Participant data were excluded when one, or both, ears had been subject to such conditions. All exclusion criteria are listed in Table 2. The choice of exclusion criteria was based on a review of literature pertaining to causes of childhood hearing loss and the first author’s relevant clinical expertise. It is acknowledged that the risk factors adopted may not be exhaustive, and there may be other factors yet to be scientifically identified.

Table 1. Criteria for inclusion/exclusion of audiometric test results in analysis.

Etiology / Factor	***n* = (% of 234)***	***Subset (A)***	***Subset (B)***
Unknown/inconclusive	99 (42)	√	√
Prematurity/birth complications (without consequent chronic conditions)	14 (6)	√	√
Infection in utero	2 (1)	√	√
Genetic (medical report and/or strong family history)	34 (14.5)	√	√
Meningitis	5 (2)	√	√
Significant disability/health condition *(e.g., cerebral palsy, epilepsy, cancer, developmental disability, cranial abnormality, tracheostomy, cystanosis, brain injury, multiple conditions)*	28 (12)	X	X
Syndrome/genetic disorder *(e.g., Ushers, CHARGE, Alports, Down, Pierre Robon, Turners, Beckwith Weidermann, achondroplasia, tuberous sclerosis, trisomy 3)*	14 (6)	X	X
Chronic otological disorders *(e.g., infection/discharge, tympanic membrane perforation, cholesteatoma, microtia, mastoiditis, total cochlear loss)*	14 (6)	X	X
Auditory neuropathy (no other conditions)	2 (1)	X	X
Cochlear/vestibular abnormality *(e.g., "fragile" cochlea, Meniere's disease, enlarged vestibular aqueduct syndrome, sudden onset hearing loss, vertigo, Mondini dysplasia)*	14 (6)	X	X
Frequent, early middle ear pathology later resolved; permanent conductive loss without otological symptoms	2 (1)	X	X
Other (e.g., palate abnormality, type 1 diabetes+connexion 26, suspected "stiff" ossicles, hypercholesteromolaemia)	3 (1.5)	X	X
Hearing loss diagnosed before age 6 years	162 (69)	√	√
Hearing loss diagnosed at, or after, age 6 years	72 (31)	√	X
Cochlear implant recipient	36 (15)	X	√ †

√ = *data included;* X = *data* *not* *included.*

* *Cases in which both survey and sufficient retrospective HTL data were provided.*

† *Shifts relating to cochlear implantation were not regarded as "events" in survival analysis.*

Data for individuals with multiple disabilities and/or severe health problems were excluded from all analyses. Data were also excluded where whole-of-life noise exposure could not be estimated (i.e., missing or incomplete survey), and in cases where all HTL data were at, or beyond, measurable limits at the initial audiogram. In several instances HTLs in one ear only exceeded measurable limits, in which case data for the ear with measurable hearing were still analyzed. As noted, cochlear implant recipients were excluded from subset (A) data analyses (which focused on differences between initial and final audiograms) on the basis that implantation surgery accounted for at least some of the shift observed during (or before) the observation period. Subset (B) excluded participants with a diagnosis of hearing loss after age 5 years. Specific analyses of subset (B) data included survival analysis (explained below), and determination of the magnitude of shift over time.

Definitions of HTL shift

Various definitions of significant HTL shift have been reported in the literature.

For these analyses three criteria were applied:

a) A downward shift (i.e., increase) in HTL of ≥ 15 dB at one or more of the following air conduction frequencies: 250, 500, 1000, 2000, or 4000 Hz.
b) A downward shift in HTL of ≥ 10 dB at any of the adjacent air conduction frequency pairs: 250/500, 500/1000, 1000/2000, and 2000/4000 Hz.
c) A downward shift of ≥ 15 dB at 2000 and/or 4000 Hz in either or both ears (referred to as 'high frequency shift').

The first two criteria reflect those used in AH protocols (Australian Hearing, 2000) and are based on the extensive work of Macrae (1988, 1989). Importantly, this work considered the need to control for spurious identification of HTL shift. The minimum HTL shift that must be observed, to be confident that a variation is not simply a function of inherent behavioral test-retest reliability, was determined (i.e., for a 5 dB step size; 10, 15, or 20 dB depending on frequency). The third criterion was chosen on the basis that high frequency (HF) HTL shift may be more significant in terms of its effects on speech intelligibility (Parving, 1988), and may be more closely associated with noise exposure. As a point of comparison, standards for occupational noise management recognise a downward shift of ≥ 10 dB for the frequencies 3000 and 4000 Hz, ≥ 15 dB for 500, 1000, 1500, 2000, and 6000 Hz, and ≥ 20 dB at 8000 Hz (re; Waugh & Macrae, 1980).

Estimation of whole-of-life noise exposures

Leisure, music listening, and work data were used to calculate a whole-of-life exposure in Pa^2 h (Pascal squared hours) according to a method described by Williams (2008). This method adapts techniques described in International Organization for Standardization 1999 (International Organization for Standardization, 2013).

An "acceptable" whole-of-life exposure criterion for age, calculated (in Pa^2h) by multiplying life years (age) by 222.2 Pa^2h, and used in previous studies (Beach et al. 2013, Carter et al., 2016a), was adopted. For a single life year, 222.2 Pa^2h represents the defined action level, or agreed acceptable exposure standard (85 dB for 8 working hours, or 1.01 Pa^2h), for continuous workplace noise in many workplace health and safety jurisdictions around the world (Williams, 2008; Williams, et al., 2015). It is important to note that these standards assume no exposure to damaging noise (i.e., leisure or nonwork sources > 75 dB) in the remaining 16 hours per day, and were based on data for adult populations.

At the time of writing, no specific model for acceptable noise exposure in children or adolescents was available.

Statistical techniques

For subset (A), initial HTLs-final HTLs (at 250, 500, 1000, 2000, and 4000 Hz) were calculated and the three criteria for HTL shift were applied. Correlations between whole-of-life noise exposure and HTL shift were tested (Mann-Whitney *U* test). For subset (B), in which age at diagnosis/initial audiogram was fairly homogeneous, relationships between HF shift and other personal and extrinsic factors were examined using χ^2 tests and Cox's Regression model.

Selection of factors

Factors were selected with respect to existing knowledge about noise exposure relating to (1) hearing aid amplification (Humes & Jesteadt, 1991; Macrae, 1991a, b, 1992, 1994; Ching et al., 2013) and (2) amplified music (Hanson & Fearn, 1975; West & Evans, 1990; Meyer-Bisch, 1996; Mostafapour, 1998; Peng, 2007; Zhao et al., 2010), as explained below.

Kaplan-Meier survival analyses were performed to investigate the temporal occurrence of HF shift among cases with diagnosis by age 5 years. Survival analysis is a statistical technique used for analyzing the time to one or more "events". In most examples of its use, it is applied to determine time to death, or time to disease diagnosis or disease recurrence (Collett, 1994). In the current analysis, the "event" = HF shift, according to criterion (c). The age at occurrence of the first "event" was determined by first plotting the series of HTLs at 2000 and 4000 Hz for the left and right ears for each case, at each of the target age points. Where an event was identified, the continuous audiogram data were used (wherever available) to more exactly determine the age at which the event occurred. Where a shift according to the criterion was observed, but subsequently recovered (i.e. fluctuation occurred), this was not accepted as a true event for the purposes of the survival analysis. HTL shifts post-cochlear implantation, similarly, were excluded as events. The magnitude of HTL shifts after specific observation periods were also examined.

RESULTS

Based on the number of invitations delivered to AH, the overall take-up rate was ~14%, while the participation rate (i.e., survey return) for those who consented was ~92%. The range in participant age, combined with the range in age at diagnosis, resulted in a variety of individual observation periods (i.e., age at final audiogram-age at initial audiogram), from a minimum of 3 months to a maximum of 29.5 years, for subset (A). The median observation period (initial to final audiogram) was 10.2 years (mean = 10.9 years). A bar chart illustrating the observation periods for subset (A) is shown in Figure 2. For subset (B), age was distributed as follows: < 18 years = 37 (46.8%), ≥ 18 years = 42 (53.2%).

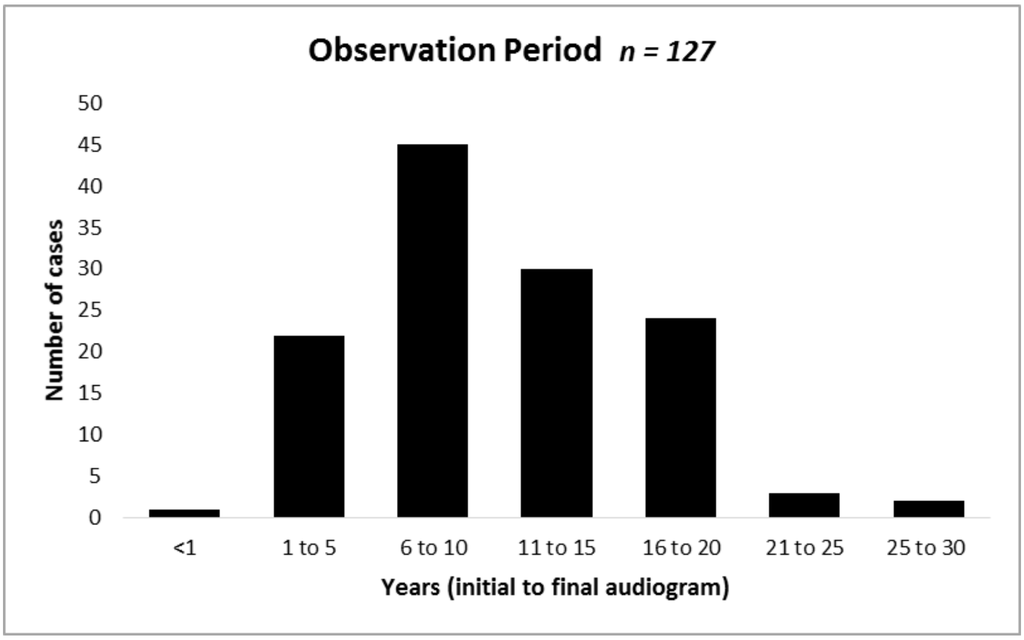

Figure 2. Observation periods, analysis subset (A).

HTL shift: final-initial audiogram

Subset (A). Applying both criterion (a) and criterion (b), HTL shift was observed in 59/127 cases (46.5%). Table 3 provides a breakdown of the number of cases of shift, according to criterion and by test ear. With respect to HTL shift, no material difference between the left and right ears was observed. Applying criterion (c), HF shift in an individual ear was observed in 31 cases (left 24.4%; right 24.8%) and, in either ear, in one third of cases; 42/127 (33.1%).

Subset (B). According to criterion (c), HF shift was observed in 39.2% of cases.

Table 2: Count of HTL shift by ear, according to criterion (a) and (b): subset (A).

Shift criterion (a) ≥ 15 dB at one or more individual frequencies (initial to final audiogram)								
Hz	LT 500	LT 1000	LT 2000	LT 4000	RT 500	RT 1000	RT 2000	RT 4000
Shift n	11	20	23	20	13	15	20	26
Shift %	8.7	15.7	18.1	15.7	10.4	12.0	16.0	20.8
valid n	127	127	127	127	125	125	125	125
Shift criterion (b) ≥ 10 dB at one or more adjacent frequency pairs (initial to final audiogram)								
Hz	LT 250/500	LT 500/1000	LT 1000/2000	LT 2000/4000	RT 250/500	RT 500/1000	RT 1000/2000	RT 2000/4000
Shift n	14	15	27	23	14	18	20	23
Shift %	11.1	11.8	21.3	18.1	11.6	14.4	16.0	18.4
valid n	127	127	127	127	125	125	125	125

Correlation between HTL shift, noise exposure and other factors

Subset (A). Only 10/127 (8%) of cases had exposure above the "acceptable" criterion for their age. Very few participants reported any significant work-related noise exposure. The distribution of whole-of-life exposure was examined using a one-sample Kolmogorov-Smirnov test. Exposure was not normally distributed, therefore non-parametric (Mann-Whitney *U*) tests were applied. No correlation between whole-of-life noise exposure and occurrence of shift was shown: Criterion (a) and/or (b) $z = -1.489$; sig. = 0.137; criterion (c) $z = -0.974$, sig. = 0.330.

Subset (B). A Cox regression model was applied to test for correlations between HF shift (according to criterion (c)) and ten categorical factors (listed in Table 4). Chi-squared testing was used to select factors for input to the Cox's regression analysis, adopting an inclusion criterion of $p < 0.25$. As illustrated in Table 4, five factors met the criterion for inclusion.

Table 3. Chi-squared results by factor: subset (B), n = 79

Factor	Factor present	df	Asymp. Sig. (2-sided)
Gender (F= 63.2%; M= 36.7%)		1	0.767
HTL ≥ 70 dB at 2, 4 kHz (initial)	41.8%	1	<0.001 *
4FAHL ≥ 70 dB either ear (initial)	22.5%	1	0.031 *
High hearing aid volume (> 3)	14.0%	1	0.226 *
Hearing aid use ≥ 8 hours/day	67.0%	1	0.192 *
Life exposure above median	49.4%	1	0.748
Frequent PSP use †	60.8%	1	0.181 *
Play musical instrument †	46.8%	1	0.429
Frequent dance ‡	12.7%	1	0.262
Frequent music at venues ‡	15.2%	1	0.852

* *Meets inclusion criterion:* $p < 0.25$
† *Participation ≥ once per week.*
‡ *Participation ≥ once per month.*

Table 5 shows the results of the Cox's regression analysis. Only one factor, HTL ≥ 70 dB at 2000 and/or 4000 Hz at initial audiogram, was found to be significantly associated with the occurrence of HF shift.

Table 4. Cox regression analysis results by factor

	df	Sig	95% CI
HTL ≥ 70 dB at 2000 and/or 4000 Hz (initial)	1	0.011 *	0.120-0.759
4FAHL ≥ 70 dB (initial)	1	0.263	0.640-5.136
High hearing aid volume	1	0.566	0.269-2.050
Hearing aid use ≥ 8 hours/day	1	0.610	0.311-1.984
Frequent PSP use	1	0.516	0.587-2.887

* *Statistically significant.*

Survival analysis: Subset (B)

A Kaplan-Meier survival analysis was performed, and survival functions were plotted, for cases with HTL ≥ 70 dB versus cases with HTL < 70 dB at 2000/4000 Hz in the initial audiogram (Figure 3). Cumulative survival (*Y* axis) refers to the proportion of cases for which, to the time indicated on the *X* axis (approximate age in years), an event has not been observed. Vertical lines on the plot indicate "censored" cases. Cases were censored at the final audiogram age for each individual with no event in the observation period. To illustrate, for the group with HF HTLs < 70 dB at initial audiogram (plotted in black), the oldest participant in the group is just over 25 years old. At this age point, 40% of cases in the group have experienced an event according to the criterion (HF shift ≥ 15 dB). At the same age point, 80% of participants in the group with HF HTLs ≥ 70 dB at (plotted in grey) have experienced an event.

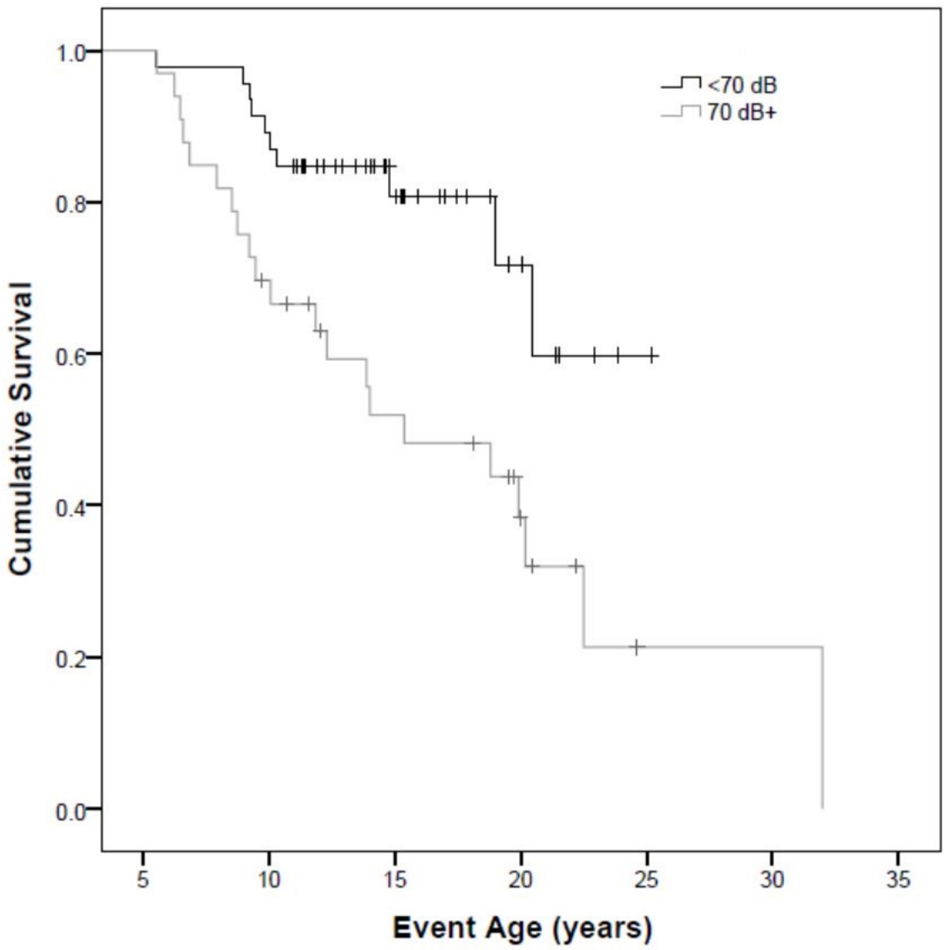

Figure 3. Kaplan-Meier survival functions for HTL 2000/4000 Hz at initial audiogram.

┼ = censored cases.

Magnitude of HTL shift: subset (B)

The magnitude of HF shifts for each ear, 5, 10 and 15 years post initial audiogram (i.e., approximate ages 10, 15 and 20 years) were determined. Table 6 presents the amount of shift (in dB HL) after 3 observation periods: 5 years, 10 years, and 15 years. At 15 years post initial audiogram (approximate age 20 years) at the 50th percentile, observed shifts were in the magnitude of 5 to 10 dB. Shifts at the 90th percentile were in the range of 25 to 30 dB. At the 10th percentile, an improvement (decrease) in HTLs in the order of 5 dB was evident

Table 5. Magnitude of HTL shift over time.

Subset (B) n = 79: Initial HTLs obtained at ~5 years of age															
Period	5 years (Age = 10 years)					10 years (Age = 15 years)					15 years (Age = 20 years)				
Percentile	10	25	50	75	90	10	25	50	75	90	10	25	50	75	90
LT 2000 Hz Shift, dB	-10	-5	0	5	10	-5	0	0	10	20	-5	0	10	15	27
n	59					47					32				
LT 4000 Hz Shift, dB	-9.5	-5	0	5	10	-5	0	5	11	16.5	-4	0	5	15	24
n	60					46					31				
RT 2000 Hz Shift, dB	-10	-5	0	5	10	-6	-5	0	10	15	-5	0	10	15	28
n	60					47					31				
RT 4000 Hz Shift, dB	-10	-5	0	5	10	-10	-5	5	10	21.5	-5	0	5	20	30
n	58					46					31				

A positive value represents a deterioration (downward shift) in HTL, while a negative value represents and improvement (upward shift) in HTL.

HTL, hearing threshold level.

DISCUSSION

The first research question, "what proportion of young people with early HI, without known risk for progressive hearing loss, experience HTL shift?" was addressed by analyses of subset (A) data. Applying both criterion (a) and (b) simultaneously, HTL shift was observed in almost 50% of cases. Because of the detailed nature of the survey and clinical file review, and the well-defined characteristics of participants in the analysis subsets, the generalizability of the results is unambiguous. Apart from the applicability of the main findings to parent/client counselling, these results usefully inform future studies of HTL shift in individuals with preadult onset hearing loss.

Observation period

A notable strength of the current study was the wide participant age range, which provided satisfactory observation periods in a reasonable proportion of cases. As Figure 2 illustrates, 58 cases (46%) had a final audiogram > 10 years after initial audiogram, and 28 cases (22%) > 15 years after initial audiogram. The median observation period was approximately 10 years. The age ranges and observation periods (where stated) of previous studies can be compared in the Appendix [Appendix in Supplemental Digital Content 1http://links.lww.com/EANDH/A340)]. It is evident that the observation periods previously used have typically been < 10 years.

Initial audiogram age has also varied among previous studies (see Appendix), and has not always been stated. Newton and Rowson (1988) noted observations starting from < 1 year of age in one analysis, and reported evidence of HTL shift prior to the age of 5 years. It is acknowledged that the current study may have overlooked shift events before the selected initial audiogram age. However, the decision to exclude audiograms obtained at preschool age was made to increase the consistency and reliability of the initial audiogram data, and to ensure availability of HTLs at four or more audiometric frequencies.

Comparison with previous research

As noted previously, variation in methods and exclusion criteria make it difficult to compare the findings of different studies. The early Swedish study by Barr and Wedenberg (1965; see Appendix), however, adopted a similar shift criterion to the current investigation. A shift in 22/40 (55%) cases was reported, which is very similar to the current finding of 59/127 (46.5%) (subset (A)).

Whole-of-life noise exposure and HTL shift

With respect to the second research question, "is there is a relationship between whole-of-life noise exposure and HTL shift?," there was no statistically significant relationship between HTL shift and reported whole-of-life noise exposure, or other factors that potentially relate to noise exposure (e.g., regular PSP use). However, the interpretation of the findings is restricted by the small spread of whole-of-life noise exposures, largely within the "acceptable" criterion for age.

A clear association between HF shift and the presence of HF HTL (2000 and/or 4000 Hz) ≥ 70 dB at initial audiogram was observed. At (approximate) age 15 years, almost 50% of cases with a HF HTL ≥ 70 dB at initial audiogram had experienced a HF shift ≥ 15 dB, compared with just 1 in 5 cases in which HF HTLs were ≤ 70 dB at initial audiogram. This could relate to hearing aid amplification, given the high gain levels required to achieve speech audibility of speech in cases of severe or profound hearing loss. It is also plausible that physiological differences in individuals with severe, early onset loss may account for increased susceptibility to shift.

In the Australian context, the hearing of young people is systematically monitored up to the age of 26 years. As illustrated in Table 6, the present study revealed evidence of increasing shift with increasing observation period. The current data also suggest that HTL shift may typically be minimal until after the age of 20 years. The very slow progression of hearing loss over adolescence may undermine vigilance in the audiological management of young adults. If the trend towards deterioration observed in this study continues into the third decade, HTL shift may reach significance (in functional terms) during early working life, when the support services typically available to children/students have ceased.

Recruitment of participants between ages 26 and 35 (i.e., above the AH eligibility age) was challenging, as there appeared to be relatively few adults in their 20s and 30s among private audiology caseloads. The high cost of hearing aids and the absence of substantial reimbursements from health insurance schemes may deter young people from continuing to monitor their hearing and upgrading devices. As noted, the failure to make appropriate technical and strategic adjustments as hearing changes will contribute to increased hearing disability in functional terms. In addition, HTL shifts combined with concomitant advances in available technologies, may move some hearing aid wearers into candidacy for cochlear implantation, or benefit from other assistive listening devices. Without ongoing audiological monitoring, some young adults may miss out on significant benefits of changing device/strategy. On a more positive note, the finding that approximately half the participants in the current study showed no evidence of HTL shift is also significant. It may be reassuring for parents to know that, in the absence of known risk factors for progressive loss, hearing deterioration during childhood and adolescence is not inevitable. Furthermore, the magnitude of HF shifts observed in this study was typically small (e.g., in the order of only 5 to 10 dB at the 50th percentile at age 20 years).

The present study revealed a slight improvement in HTLs at the 10th percentile. It has been observed that even typically developing children may become more attuned and listen more attentively to soft sounds over time (Brookhouser, 2002; Buss et al., 2016) which may be a contributing factor in observed HTL improvements. Pittman and Stelmachowicz (2003) also reported hearing improvement in 5% of observed cases.

Limitations of the study

The number of participants was reasonably large, however, the application of the exclusion criteria reduced the size of the analysis datasets. The response rate (in terms of the number of invitations distributed to AH) was relatively low. Recruiting young people with disability for research is inherently difficult. Liamputtong (2007, p.3) classified both individuals with disabilities and adolescents as "vulnerable" and "difficult-to-access" research populations. Considerable effort was made to ensure that a representative sample was achieved (e.g., including participants in country and regional locations). The length and complexity of surveys may also have affected participation rate, by discouraging individuals with lower literacy levels or with English as a second language. This was unavoidable, as sufficient information about noise exposure history was needed to make a reasonable estimation of individual risk. Possibly, the greatest uncertainty arises from the use of self-reported data in estimating noise exposure. Difficulty and/or inaccuracy in recalling events could be an influencing factor, as noted by Williams et al. (2015). As mentioned, the main limitation in interpreting the findings was the small number of participants estimated to be at risk for noise-induced HTL shift.

Continuation of surveillance

Currently available AH data could be used to expand the findings of this study. At the time of writing, however, Government funded hearing services in Australia were transitioning to new arrangements, including a National Disability Insurance Scheme. Subsidized hearing services may be extended to some individuals between 26 and 65 years of age. Establishment of a national database, conserving at least audiometric and demographic data, would allow the surveillance of HTLs to be continued under a new decentralized model. National infrastructure, centralized record keeping and cooperation among service providers would be required to achieve this goal. The extension of subsidized hearing services to some adults in mid-life may also create a new opportunity to investigate the interaction between early hearing loss and presbycusis. Meanwhile, the current results offer new insight into the probability of HTL shift in the time between school age and early adulthood.

CONCLUSIONS

This is the first time the relationship between HTL shift and whole-of-life noise exposure has been directly examined in a homogeneous group of young people with early HI. Whole-of-life noise exposure was not associated with HTL shift for this cohort; however few participants reported high levels of noise exposure. Nevertheless, almost 50% of selected participants had experienced HTL shift. As the hearing health care system continues to evolve, more systematic surveillance of HTL shift, particularly through mid-life, will further increase our understanding of the prognosis of early onset hearing loss and the likely impact of noise exposure over the longer term.

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Appendix. Summary of HTL shift in young people with hearing impairment literature.

Authors, date	Country	Participants (age)	*n* =	Shift criterion	Observation period	Incidence/Conclusions
Guild, 1950	US	8 – 13 years ENT surgical patients	4	Case studies	2 – 6 + years	Progression of loss in case studies 'gradual'. 'High-tone' loss can progress markedly during childhood
Kinney, 1961	US	6 – 16 years Aided children	178	Increased 'average' loss of ≥10 dB in aided ear	126= monaurally aided	13/126 (10.3%) cases (less powerful hearing aids) 19/39 (48.8%) (higher powered aids)
Barr & Wendenberg, 1965	Sweden	Pre-play audiometry - ? Deaf and severely HI children	84	Deviation of ≥ 10 dB at two frequencies, or ≥ 15 dB at one frequency 15 dB = 'significant'	3 – 15 years	22/40 (55%) with 'heredity genesis' 0/23 with maternal rubella 0/15 with 'perinatal accidents' 6/6 with meningitis
Macrae, 1968	Australia	5 – 18 years Aided children	134	Mean change reported	1 – 12 years Mean= 5.25 years	Mean change in the order of 6 dB in aided ears 'Small' change in unaided ears
Reilly et al., 1981	US	Children with HI; most aided	90	≥ 15 dB at two or more frequencies from first to final audiogram	At least 2 years Baseline= first 'reliable' audiogram (3 – 17 years)	25/90 (28%) 'Progressive hearing loss is not uncommon in children'
Böhme, 1985 [GER]	Germany	5 – 23 years	44		Mean= 6 years	One third

Newton & Rowson, 1988	UK	6 – 10 years Bilateral SN loss; some aided	Gp 1 =177 Gp 2 =27	≥ 15 dB at any frequency; between first and last HTLs & not attributable to ME* or NOHL†	Gp 2 0 – 12 years	Gp 1; 16/177 (9%), 12 before the age of 5 years 'likely to be a minimum figure'
Parving, 1988	Denmark	7 – 17 years (median= 13) Children with HI	138	Difference from 1st to 2nd investigation of ≥ i.15 dB at 2kHz or 4kHz ii. average of 2 & 4kHz, or average of 0.5, 1 & 2 kHz Mean of right & left HTLs used	5 years	2 – 6 % depending on criterion used Deterioration > 15 dB; i. 8/132 (6%) ii. 5/132 (4%) iii. 3/132 (2%)
Ruben et al., 1982	US	Aided children with moderate to severe HI Lower SES‡	72	≥ 10 dB	3 – 10 years (4 cases) Group average follow up time 6.7 years (n= 70); 'many 1 – 2 years'	4/70 (6%)
Levi et al., 1993	Israel	3 years + (born 1967-1982) Mild to severe HI Excluded where loss ≥ 90 dB	92	i. ≥ 15 dB at average of 0.5, 1, 2 & 4 kHz ii. ≥ 15 dB at average of 0.5, 1 & 2 kHz iii. ≥ 15 dB at average of 2 & 4 kHz iv. ≥ 20 dB only at 0.5 & 1 kHz	Up to 15 years	21/92 (22.8%) 13 bilateral, 8 unilateral Nil in cases with mild hearing loss (< 40 dB HL) No gender difference

Savastano et al., 1993		0 – 14 years SN loss	42	Compared cases with earlier and later onset of loss	Examined rapidity of deterioration and changes in audiogram configuration. Etiology= unknown	
Brookhouser, et al., 1994	US	1 – 19.9 years at 1st audiogram Mild or greater SN loss	1108	≥10 dB at 0.25, 0.5, 1, 2, 4 or 8 kHz	Mean= 4.9 years (for 229 cases identified with progressive loss)	240/1108 (22%) cases showed progressive loss; 11 cases- insufficient data 22/365 ears (6%)= purely progressive 208/365 ears (57%) fluctuating & gradually progressive
Berrettini et al., 1999	Italy	Bilateral SN loss	178	Mean worsening ≥ 20 dB recorded 'on at least two frequencies' in the range 0.5 – 4 kHz	Excluded syndromic genetic cases. Included one case of LVAS and one of CMV	11/178 (6.2%)
Pittman & Stelmachowicz, 2003	US	6 – 14 years	132	≥ 15 dB at two or more frequencies	8 years maximum	17/132 (13%) 7/132 (5%) improved
Current study		11 – 35 years	127	≥ 10 dB at two adjacent frequencies, or ≥ 15 dB at one or more frequencies. 0.25 – 4 kHz.	<1 to 30 years maximum Mean= 11 years Median= 10 years Initial= ~5 years	59/127 (46.5%)