The Impact of Occupational Noise Exposure on Hyperacusis: a Longitudinal Population Study of Female Workers in Sweden

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Objectives: The aim was to assess the risk of hyperacusis in relation to occupational noise exposure among female workers in general, and among women working in preschool specifically.

Design: A retrospective longitudinal study was performed. Survey data were collected in 2013 and 2014 from two cohorts: randomly selected women from the population in region Västra Götaland, Sweden, and women selected based on having received a preschool teacher degree from universities in the same region. The final study sample included *n* = 8328 women born between 1948 and 1989. Occupational noise exposure was objectively assigned to all time periods from the first to the last reported occupation throughout working life, using the Swedish Job-Exposure Matrix (JEM) with three exposure intervals: <75 dB(A), 75 to 85 dB(A), and >85 dB(A). The JEM assigns preschool teachers to the 75 to 85 dB(A) exposure interval. The outcome hyperacusis was assessed by self-report using one question addressing discomfort or pain from everyday sounds. In the main analysis, a hyperacusis event was defined by the reported year of onset, if reported to occur at least a few times each week. Additional sensitivity analyses were performed using more strict definitions: (a) at least several times each week and (b) every day. The risk (hazard ratio, HR) of hyperacusis was analyzed in relation to years of occupational noise exposure, using survival analysis with frailty regression modeling accounting for individual variation in survival times which reflect, for example, noise exposure during years prior to onset. Occupational noise exposure was defined by the occupation held at year of hyperacusis onset, or the occupation held at the survey year if no event occurred. Models were adjusted for confounders including age, education, income, family history of hearing loss, and change of jobs due to noise.

Results: In total, *n* = 1966 hyperacusis events between 1960 and 2014 were analyzed in the main analysis. A significantly increased risk of hyperacusis was found among women working in any occupation assigned to the 75 to 85 dB(A) noise exposure group [HR: 2.6, 95% confidence interval (CI): 2.4–2.9], compared with the reference group <75 dB(A). The risk was tripled among preschool teachers specifically (HR: 3.4, 95% CI: 3.0–3.7), with the crude Kaplan-Meier curve showing a higher rate of onset early in the working life in preschool teachers compared with all the other exposure groups. The risk was increased, but not statistically significant in the main analysis, for the highest exposure group >85 dB(A), where only six hyperacusis events were identified (HR: 1.4, 95% CI: 0.6–3.1). In the sensitivity analysis, where hyperacusis was defined as occurring every day, the HR was significant also in the highest exposure group (HR: 3.8, 95% CI: 1.4–10.3), and generally slightly higher in the other exposure groups compared to the main analysis.

Conclusions: This study indicates increased risk of hyperacusis already below the permissible occupational noise exposure limit in Sweden (85 dB $L_{Aeq,8h}$) among female workers in general, and in particular among preschool teachers. Prospective studies and less wide exposure intervals could confirm causal effects and assess dose–response relationships, respectively, although this study at present suggest a need for risk assessment, improved hearing prevention measures, and noise abatement measures in occupations with noise levels from 75 dB(A). The results could also have implications for management of occupational disability claims.

Key words: Audiology, Epidemiology, Hazard ratio, Hearing disorder, Hyperacusis, Incidence, Job-exposure matrix, Longitudinal study, Occupational medicine, Occupational noise exposure, Preschool, Survival analysis, Teacher, Women.

Abbreviations: AFS = The Swedish Work Environment Authority's Statute Book; AIC = Akaike Information Criterion; CI = confidence interval; dB HL = decibel hearing level; dB $L_{Aeq,8h}$ = A-weighted 8-hour Equivalent Decibel Sound Pressure Level; dB SPL = Decibel Sound Pressure Level; dB(A) = A-weighted Decibel Sound Pressure Level; HPD = hearing protection device; HQ = hyperacusis questionnaire; HR = hazard ratio; IR = incidence rate; ISCO = The International Standard Classification of Occupations; JEM = job-exposure matrix; NIOSH = National Institute for Occupational Safety and Health; NYK = Nordic Occupational Classification system; SD = standard deviation; ULL = uncomfortable loudness level.

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INTRODUCTION

Hyperacusis is a symptom of loudness perception disorder or sound intolerance disorder (Fagelson & Baguley 2018). Several definitions exist, often describing abnormal intolerance, hypersensitivity, loudness discomfort, or pain from everyday sounds even at moderate sound levels (Fackrell et al. 2017; Tyler et al. 2014). Hyperacusis can be understood as separate from conditions such as phonophobia (fear of sounds) and misophonia (dislike or hatred of specific sounds), and recruitment of loudness (a narrow dynamic range because of hearing loss) (Fackrell et al. 2019; Jastreboff & Jastreboff 2002; Tyler et al. 2014), although they may coincide for a particular individual. In a review, Tyler et al. (2014) proposed that different symptom characteristics could distinguish four subtypes of hyperacusis: loudness, pain, annoyance, and fear.

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Studies of mechanisms mainly relate to loudness hyperacusis (Tyler et al. 2014). One dominant model argues that hyperacusis is the perceptual outcome of neural hyperactivity or increased gain in the central auditory pathways resulting from neural plasticity and adaptation, initiated by a peripheral hearing loss (Auerbach et al. 2014; Eggermont 2018; Knipper et al. 2013; Pienkowski et al. 2014; Sheppard et al. 2020). While some support for a central gain mechanism has been found in a neuroimaging study in humans (Gu et al. 2010), there are still concerns as to whether the neurophysiological changes observed in animals can explain hyperacusis in humans (Aazh et al. 2014). Since studies in humans also find hyperacusis in subjects without apparent hearing loss (Anari et al. 1999; Tyler et al. 2014), noise-induced cochlear synaptopathy (hidden hearing loss) has also been suggested as a possible peripheral initiating factor (Hickox & Liberman 2014; Liberman et al. 2016).

Whether noise exposure causes hyperacusis is one of the top-ranked research questions (Fackrell et al. 2019). Although hyperacusis can be manifested in several diagnoses with different etiologies, noise exposure has been suggested by many researchers as a predominant cause (Aazh et al. 2014; Anari et al. 1999; Axelsson & Hamernik 1987; Pienkowski et al. 2014; Tyler et al. 2014). Noise is commonly used in animal studies to elicit hyperacusis-like reactions (Pienkowski et al. 2014; Radziwon et al. 2020). Hyperacusis has also been commonly noted following acoustic incidents with sudden intense sounds (acoustic shock), often followed also by otalgia (ear pain) (McFerran & Baguley 2007; Parker et al. 2014). Interestingly, studies suggests that cochlear type-II neurons may act as pain receptors reacting to noise-induced tissue damage (Flores et al. 2015; Liu et al. 2015), possibly explaining pain hyperacusis. Interestingly, a recent study indicate that this mechanism could be activated also at lower noise exposure levels, 80 dB SPL, of broadband noise (Weisz et al. 2021).

The general population prevalence of self-reported hyperacusis has been reported at 8% to 9% among Swedish adults (Andersson et al. 2002; Paulin et al. 2016), whereas a Polish study reported a 15% prevalence (Fabijanska et al. 1999). Paulin et al. (2016) reported hyperacusis to be more common among women compared with men, opposite to Fabijanska et al. (1999). Prevalence has been found to increase with age (Andersson et al. 2002; Paulin et al. 2016). When defining hyperacusis as discomfort or pain from everyday sounds occurring at least a few times each week, we recently found a prevalence of 39% among female preschool teachers, which was more than twice as high as the prevalence of a randomly selected Swedish female population (18%) (Fredriksson et al. 2019). A high prevalence among preschool personnel has been shown in other studies, reporting that 45% experience hyperacusis sometimes or quite often (Sjödin et al. 2012). Studies have also indicated that teachers and childcare workers are common occupations among hyperacusis patients (Anari et al. 1999; Jüris et al. 2013).

One possible explanation to the increased risk in preschool personnel is occupational noise exposure. When asked about the sound environment at work, a vast majority, 72%, of preschool teachers report having been exposed to children screaming in their ears several times, and an additional 25% reported exposure to screaming a few times (Fredriksson 2018). Sound levels in preschool are high and intermittent, registering around 80 dB L_{Aeq} (A-weighted equivalent sound level) during indoor activity, with many occurrences throughout the day reaching above 85 dB L_{Aeq} (Gerhardsson & Nilsson 2013; Persson Waye et al. 2009; Sjödin et al. 2012). These sound levels could be harmful for hearing. In Sweden, the Swedish Work Environment Authority, AFS 2005:16, regulate an 8-hour lower action value of 80 dB $L_{Aeq,8h}$ and a permissible limit of 85 dB $L_{Aeq,8h.}$

While previous studies indicate that sudden intense sounds may be particularly harmful and may lead to hyperacusis, there is a lack of studies on the effects of long-term occupational noise exposure, of dose–response relationships and of causal effects. There is also a lack of studies using more objective exposure data in larger study samples, rather than self-reported exposure alone, as well as a lack of comparisons of the effect of noise exposure from working in preschool compared to other occupations. This study intends to fill in some of these knowledge gaps. Hence, the aim of this study was to examine the risk of hyperacusis in relation to occupational noise exposure during working life, assessed objectively using a Job-Exposure Matrix (JEM), in a large group of randomly selected women from the general Swedish population and a large group of preschool teachers.

MATERIALS AND METHODS

Study Design and Study Population

This is a retrospective longitudinal study with outcome and occupational history collected using self-administered postal questionnaires sent out in 2013 and 2014. In total, 25,756 individuals were sent the questionnaire; half (n = 14,524) were randomly selected women from the Swedish Population and Tax Agency Register, born between 1943 and 1989, living in region Västra Götaland, Sweden. The other half (n = 11,232) were selected from university registries based on their preschool teacher degree being issued between 1980 and 2012 from one of the five universities in the same region of Sweden. The overall response rate was 43% (n = 11,167), 38% in the population cohort and 51% among preschool teachers. The population cohort did not include any preschool teachers. The data collection and study population have been reported in detail elsewhere (Fredriksson 2018; Fredriksson et al. 2019).

The final study sample included n = 8328 women born 1948 to 1989 (age 24 to 65 years at the time of the survey). As shown in Figure 1, n = 8840 women were eligible for the current analysis, but participants with missing data on the outcome or the exposure variables (n = 503) and those with onset of hyperacusis before the first reported occupation (n = 9) were excluded.

The Ethics Committee of Gothenburg Sweden approved this study (060-13). All participants received written information and gave their consent by returning a completed questionnaire. Participants could contact the first or last author if they wanted to withdraw their consent or if they had questions about the study. There was no other interaction between study participants and the authors. Participants did not receive any compensation for participating.

Hyperacusis Outcome (Event)

In this study, we aimed to assess mainly loudness and pain hyperacusis in relation to non-specific sounds, rather than conditions relating to specific sounds or sound sources. Thus, we defined hyperacusis as "sound sensitivity, a feeling of discomfort or pain from everyday sounds." Hyperacusis was assessed

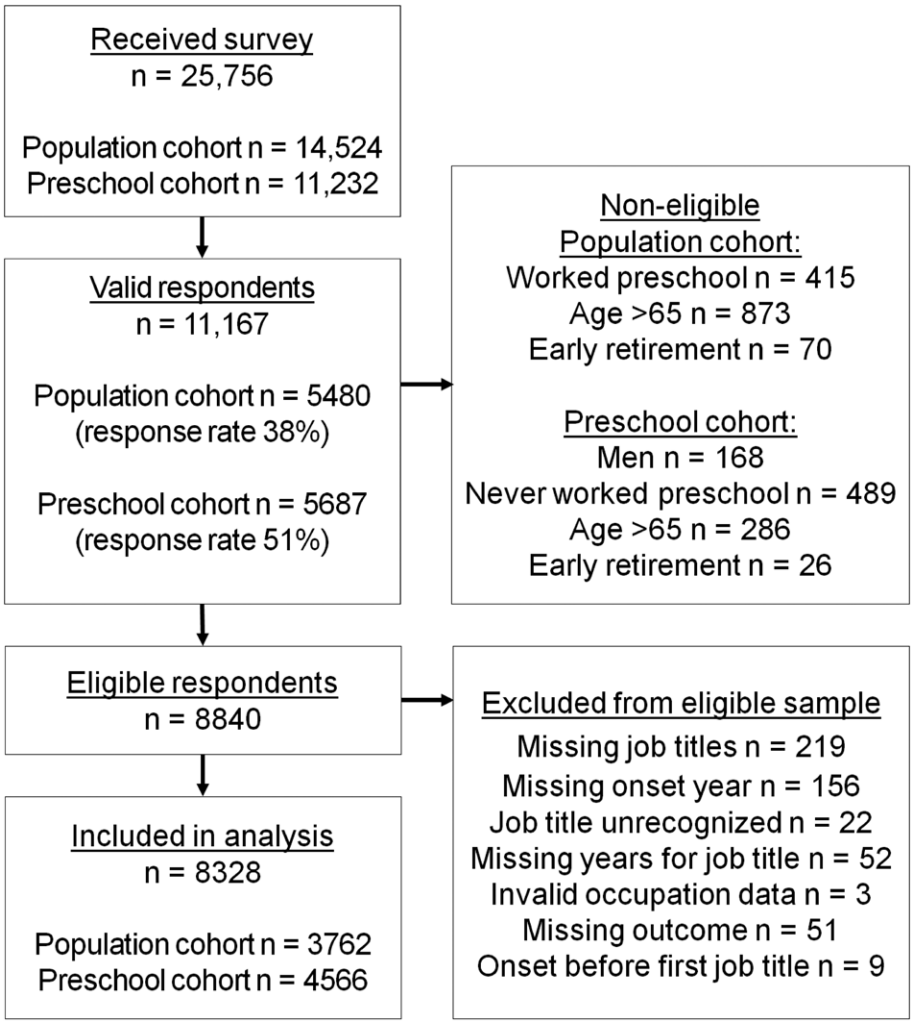

Fig. 1. Flowchart of study population, exclusion criteria, and eligible numbers.

using the self-report question: "*Do you consider yourself to be sound sensitive (feel discomfort or pain from everyday sounds)?*" The response scale had six alternatives: "no", "no, but I have previously been," "yes, a few times each month or less often," "yes, a few times each week," "yes, several times each week," and "yes, every day". In the main analysis we defined hyperacusis as occurring at least a few times each week. Additional sensitivity analyzes were performed using more strict definitions: a) at least several times each week, and b) every day. The year of first onset was retrospectively reported using the question: *"When did you first notice that you were sound sensitive?*", with free-text responses of age or calendar year. The response "no, but I have previously been" (reported by $n = 146$) was considered a "no" because the year of hyperacusis resolving was unavailable, which prevented inclusion in the time-dependent statistical analysis. Hence, a participant could only have one event. Only onset during working life were considered, as the analysis required information about the occupation (i.e. noise exposure) at year of onset. Thus, only the first, non-resolved, onset during working life was considered a hyperacusis event.

Occupational Noise Exposure

Coding Occupational History • Occupational history was collected by asking participants to report, in free text, every occupation they had ever held for more than six months throughout their working life, and the year in which they had started and ended each occupation. The responses were assigned a 5-digit occupational code (job title) according to the Nordic Occupational Classification system (NYK) from 1983 used in the Swedish census in 1985. The less specific 3-digit NYK code (job family) was used in combination with the JEM. The NYK system was originally based on the International Standard Classification of Occupations 1958. Two research assistants and one occupational hygienist coded the occupational history in collaboration with the first author (see text in Supplemental Digital Content 1, http://links.lww.com/EANDH/A995 for further details on occupational coding of job titles).

Exposure Assessment Using a Job-Exposure Matrix for Occupational Noise • Noise exposure was assessed using the Swedish JEM for occupational noise constructed by Sjöström et al. (2013). The JEM is based on 569 sound level measurements in 129 job families, in combination with a standard method of

consensus judgments between three experienced occupational hygienists for job families where noise measurements were unavailable. The JEM assigns each job family one of three noise level intervals: <75 dB(A), 75 to 85 dB(A), and >85 dB(A) estimated 8-hour time-weighted average, for separate 5-year periods from 1970 to 2004. For example, the JEM assigns preschool teachers to the 75 to 85 dB(A) exposure interval.

In this study, noise exposure was assigned to each time period with a reported and NYK-coded occupation using the JEM. The reported time period was considered when applying the JEM to occupations where the noise exposure differed depending on calendar year (Sjöström et al. 2013). In the analysis, noise exposure was assessed in two ways. First, we assessed the risk of noise exposure based on number of years in the occupation held at the year of hyperacusis onset (event), or the occupation held at the end of the study (survey year) if no event occurred during the study period. In this way, each participant was classified into one of the noise exposure groups according to the JEM, and assessing the risk in the exposed groups compared to the reference group. Occupations (NYK-coded job titles) relating to work in preschool were considered specifically in subanalyses. Second, noise exposure during years preceding onset of hyperacusis was also considered, by using the NYK codes for each preceding occupation in relation to the JEM, and analyzed as frailty, as described in the statistical analysis.

Time at Risk

In addition to assigning noise exposure to reported occupations, we also considered number of years within each occupation, in order to calculate time at risk in the survival analysis. As data on occupation was the basis for the analysis of risk from noise exposure, we defined the time at risk as person-years in any occupation with exposure assigned by the JEM, starting at the first reported year in an occupation until onset of hyperacusis (event), or if no event occurred, until the survey year. Years with missing or inadequate data were not included. A small number of participants (n = 30) reported more than one occupation during the same year, most likely due to part-time work or change of work within a year. This number increased to n=67 in the additional sensitivity analyzes. These periods were treated independently, and both were included in the calculation of person-years. Only seven events in the main analysis, and four and one event respectively in the sensitivity analyses, occurred in an overlapping period where occupations were assigned to different exposure groups. These events were assigned to the higher of the two exposure groups, in this case the 75 to 85 dB(A) interval. Because of the overlapping years, the time at risk was slightly extended mathematically and, hence, does not represent the study period (follow-up) in calendar years.

Statistical Analysis

STATA/SE (v. 14.2, 15.1, and 16.1) for Windows (StataCorp LCC, College Station, TX, USA) was used to derive the analytical and descriptive statistics. Total time at risk and incidence rates per 1000 person-years with 95% confidence intervals (CIs) were calculated for hyperacusis onset using the *stptime* STATA command. Hazard ratio (HR) with 95% CI for hyperacusis onset was analyzed in relation to years in occupational noise exposure using Cox regression in conjunction with the frailty function using the *streg* STATA command (Balan & Putter 2020). While we acknowledge the technical differences between the terms "risk" and "hazard", we have used "risk" in place of HR in interpretation and discussion of the results. The exposure groups were assigned using noise intervals from the JEM at the year of hyperacusis onset. In our survival analysis, the relevant type of data distribution was assessed using the Akaike information criterion (AIC) applying the *estat ic* STATA command. The distribution that performed best, as indicated by a lower AIC value, was retained as a main distribution model (Weibull in model 1 and 3, and Gompertz in model 2). Participants who had not developed hyperacusis were "censored" in the survival analysis at the end of the study (survey year 2013 or 2014), as is customary.

In Cox regression, individuals are assumed to have equivalent survival times and the hazard function is dependent on the observed risk variables (the covariates). Usually though, not all risk variables are known, measured or included in the regression model, although they may affect an individual's survival time and hence the hazard. In such a case, one would have a so-called unobserved heterogeneity in survival data, and the resulting survival estimates could be incorrect. A frailty regression model can in contrast to the Cox model detail the hazard at both population and individual levels and can thus inform whether there is heterogeneity. The frailty regression will yield parameters on the variance of the frailty (the unobserved heterogeneity), which in our case would lead to rejecting the null hypothesis, and infer that there is unobserved heterogeneity affecting the model. Alternatively, if the null hypothesis is not rejected, one would infer that there is no heterogeneity and interpret the hazard function as from an ordinary Cox regression. In our survival analysis, we assessed shared frailties, which in addition to accounting for heterogeneity, can generate dependency between the survival times of conditionally independent individuals. In other words, the shared frailty is relevant to event times of related individuals and repeated measures, of which repeated measures are assumed to share the same frailty. Hence the survival times are assumed conditionally independent with respect to the shared (common) frailty. An individual identifier was therefore used in the frailty model to account for the unobserved heterogeneity and the random effect in survival times.

The main analysis included one model where the two cohorts were assessed together, and a second model separating the preschool cohort from the population cohort within the 75 to 85 dB(A) exposure group. We also performed an exposure sensitivity analysis including a third model where childcare workers from the population cohort were assessed together with the preschool teachers. Additional outcome sensitivity analyses of model one and two were also performed, using the more strict definitions of the outcome previously described. The final multiple regression models were adjusted for relevant covariates (i.e. possible confounders) chosen beforehand and initially assessed in a directed acyclic graph. We assessed the following possible confounders, measured at the time of survey: age, highest attained education level, household monthly income, family history of hearing loss before age 55, history of recurrent ear infections, smoking (ever smoked daily for at least 1 month), and reporting ever having changed jobs due to noise at work. The best-fit multivariable model was chosen based on AIC using the *estat ic* STATA command. Sensitivity analyses were performed using the same adjustments as the main models. The questionnaire items and definitions have been reported in detail elsewhere (Fredriksson et al. 2019).

RESULTS

As seen in Table 1, there were in total $n = 1966$ hyperacusis events in the main analysis with symptoms occurring at least a few times each week and with onset during working life in occupations reported between the years 1960 and 2014. More than half of the events occurred during years working as a preschool teacher ($n = 1090$), an occupation assigned to the noise exposure group 75 to 85 dB(A), while only six events occurred among women working in an occupation assigned to the highest exposure group, >85 dB(A). The exposure groups were similar in terms of background factors such as age, with the reference group, <75 dB(A), being slightly older. A larger difference was found for education level because the preschool teacher occupation requires >3 years of university education.

As shown in Table 2, the risk (HR) of hyperacusis was significantly increased, more than double, among women working in occupations assigned to the 75 to 85 dB(A) noise exposure group compared with the reference group (model 1). The most common occupations in the reference group were elementary school teacher, retail sales worker, nursing assistant, office secretary, and nurse. The HR in the highest exposure group, >85 dB(A), was increased but not statistically significantly in the main analysis, and the CI was wide. The six hyperacusis events found in this group were woodworking machine setters and operators ($n = 4$), a musician ($n = 1$), and a fish butcher (among canners and preservers) ($n = 1$).

When preschool teachers were evaluated specifically (model 2), the HR was shown to substantially increase, being three times higher than the reference group. This substantial risk remained in an exposure sensitivity analysis (model 3), where childcare workers from the population cohort were assessed along with the preschool teachers, rather than with other occupations in the general 75 to 85 dB(A) exposure group. There were $n = 64$ events among childcare workers, in addition to the $n = 1090$ events found among preschool teachers. The same sensitivity analysis (model 3) also confirmed a significant, albeit lower, HR of hyperacusis in non-preschool occupations within the 75 to 85 dB(A) exposure group, excluding occupational codes that could not differentiate between preschool workers and non-preschool workers. The non-preschool occupations within the 75 to 85 dB(A) group included a variety of commonly occurring occupations among women in Sweden. The ones with the highest number of hyperacusis events were worker in a kitchen or restaurant ($n = 14$), waitress or bartender ($n = 9$), fitter-assembler and machine erector ($n = 8$), dentist ($n = 7$), and cleaner ($n = 6$). The HRs did not change in either model when adjustment for smoking was added.

As seen in Figure 2, the crude survival estimates in relation to time at risk for the four exposure groups analyzed in model 2 were plotted in a Kaplan-Meier curve. The Kaplan-Meier curve highlights the considerably higher rate of cases over time and the early onset among preschool teachers compared with the other exposure groups as well as the reference group. This is evident also in the high incidence rates shown in Table 2.

As seen in Table 3, the additional outcome sensitivity analyses confirmed the main analysis and showed generally slightly increased HRs, and most notably a significantly increased HR in the highest exposure group >85 dB(A) compared with the reference group when using the most strict outcome definition

TABLE 1. Characteristics of study participants categorized by occupational noise exposure assigned by the Swedish Job-Exposure Matrix for noise (as analyzed in model 2) based on the occupation held at year of onset of hyperacusis at least a few times each week (event) or at the end of the study if no event

	Occupational Noise Exposure														
				75–85 dB(A)											
	<75 dB(A)			Not Preschool Teachers			Preschool Teachers*			>85 dB(A)			Total Sample		
	%	*n*	Mean (SD)	%	*n*	Mean (SD)	%	*n*	Mean (SD)	%	*n*	Mean (SD)	%	*n*	Mean (SD)
Subjects (row %)	51	4260		13	1073		35	2953		0.5	42		100	8328	
Initial population cohort	71	3030		65	696		0	0		86	36		45	3762	
Initial preschool cohort	29	1230		35	377		100	2953		14	6		55	4566	
Hyperacusis events (row %)	32	630		12	240		55	1090		0.3	6		100	1966	
Age at time of survey (yrs)			48 (11)			46 (11)			44 (10)			43 (11)			46 (11)
Highest education level															
University ≥3 yrs	72	3062		57	602		100	2953		41	17		80	6634	
University <3 yrs	28	1174		43	460		0	0		59	24		20	1658	
Household income															
≥30,000 SEK/mo	80	3364		67	707		81	2360		64	27		79	6458	
<30,000 SEK/mo	20	841		33	352		19	558		36	15		21	1766	
Family history of hearing loss															
Yes	37	1546		38	402		23	686		33	14		32	2648	
No	64	2690		62	665		77	2261		67	28		68	5644	
Changed job due to noise															
Yes	4	153		5	51		6	163		0	0		4	367	
No	96	4054		95	1010		94	2778		100	42		96	7884	

Proportions are column percentages (%), if not stated otherwise.
**Nordic Occupational Classification code NYK: 036.10 preschool teacher, with noise exposure 75–85 dB(A) assigned by the job-exposure matrix.*
dB(A), A-weighted decibel level; SD, standard deviation.

TABLE 2. Hazard ratio (HR) of hyperacusis at least a few times each week (event) among women in relation to occupational noise exposure assigned by the Swedish Job-Exposure Matrix for noise based on the occupation held at year of onset of hyperacusis, or at the end of the study if no event, with frailty regression modeling accounting for individual variation in survival times, reflecting for example exposure during years prior to hyperacusis onset

Occupational Noise Exposure	Hyperacusis Events, n	Person-Years At Risk‡	Incidence Rate per 1000 Person-Years IR	(95% CI)	Crude Model HR	(95% CI)	Adjusted Model HR	95% CI
Model 1†								
<75 dB(A)	630	91024	6.9	(6.4–7.5)	(reference)		(reference)	
75–85 dB(A)	1330	78307	17.0	(16.1–17.9)	3.2*	(2.9–3.6)	2.6*	(2.4–2.9)
>85 dB(A)	6	947	6.3	(2.8–14.1)	1.4	(0.6–3.2)	1.4	(0.6–3.1)
Model 2, separating preschool teachers§								
<75 dB(A)	630	91,024	6.9	(6.4–7.5)	(reference)		(reference)	
75–85 dB(A)								
Preschool teachers (036.10)	1090	47,504	22.9	(21.6–24.3)	4.3*	(3.9–4.8)	3.4*	(3.0–3.7)
All other occupations within 75–85 dB(A)	240	30,803	7.8	(6.9–8.8)	1.5*	(1.3–1.8)	1.6*	(1.3–1.8)
>85 dB(A)	6	947	6.3	(2.8–14.1)	1.4	(0.6–3.1)	1.3	(0.6–3.0)
Model 3, exposure sensitivity analysis¶								
<75 dB(A) (excluding 039.20, 153.90)	630	90944	6.9	(6.4–7.5)	(reference)		(reference)	
75–85 dB(A)								
Preschool teachers and childcare workers (including 036.10, 153.10)	1154	55007	21.0	(19.8–22.2)	4.0*	(3.6–4.4)	3.6*	(3.2–4.0)
Non-preschool occupations within 75–85 dB(A) (excluding 036.90)	120	19451	7.6	(6.5–8.8)	1.2	(0.9–1.5)	1.3*	(1.1–1.6)
>85 dB(A)	6	947	6.3	(2.8–14.1)	1.4	(0.6–3.1)	1.6	(0.7–3.6)

Statistical significance ($p < 0.05$).

†Best-fit model 1 adjusted for age, the highest education attained, household income, and family history of hearing loss.

‡Person-years at risk was defined as years exposed to occupational noise until onset of hyperacusis (event) or time of survey if no event occurred, including overlapping years in different occupations for n = 30 subjects.

§Best-fit model 2 adjusted for age, highest education attained, household income, family history of hearing loss, and ever changed jobs because of noise at work.

¶Best-fit model 3 adjusted for the highest education attained, household income.

CI, confidence interval; HR, hazard ratio; IR, incidence rate.

"every day". The sensitivity analyses resulted in decreased incidence rates in all exposure groups, because of fewer events and a longer time at risk being accumulated for those who were no longer considered to have onset.

DISCUSSION

Main Findings

The key result in this study is the significantly increased risk of hyperacusis among women working in occupational noise exposure in the interval 75 to 85 dB(A), assigned objectively using a JEM, compared with women working in <75 dB(A). Notably, a more than three times increased risk was found among women working in preschool, and an increased risk of at least 30% in other occupations within the same noise exposure interval, 75 to 85 dB(A). The significant results were confirmed in an exposure sensitivity analysis with a strict division of preschool workers and non-preschool workers. The main results were confirmed in additional sensitivity analyzes using more strict outcome definitions of hyperacusis at least several times each week, or hyperacusis every day. The risk was also increased, but the HR not statistically significantly in the main analysis, among the relatively few women who were exposed to noise >85 dB(A). However, the sensitivity analysis using the most strict outcome definition every day showed a significantly increased HR also in this highest exposure group. Thus, our study showed significant risk of hyperacusis already below the current 8-hour permissible noise exposure limit, 85 dB $L_{Aeq,8h}$, regulated by the Swedish Work Environment Authority in order to prevent noise-induced hearing damage, mainly hearing loss, and also a significant risk for more frequently occurring symptoms above the exposure limit.

One explanation of the substantially increased risk of hyperacusis among preschool workers may be infrequent use of hearing protection devices (HPDs) at work. A previous study found that fewer than 10% of preschool teachers compared with about 30% of women in other occupations reported frequent use of HPDs, when simultaneously reporting exposure to high sound levels at work (Fredriksson et al. 2019). The non-use of HPDs in noise-exposed human service occupations such as health care, social service, and education has also been reported in a large National Institute for Occupational Safety and Health study in the United States (Tak et al. 2009). This is likely partly explained by perceived detrimental effects of hearing protection devices use on speech perception. Preschool personnel may also find it inappropriate to wear HPDs in the presence of parents and may consider that wearing HPDs reduces their ability to fulfill their teaching duties (Koch et al. 2016). According to the Swedish Work Environment Authorities regulation on noise at work, the use of HPDs is a hearing prevention measure taken when other noise abatement measures have failed, such as replacing or isolating noisy machines. However, reducing the noise at the source may prove difficult in occupations where human activity, central to the work, is the main source of noise. Intervention studies in preschool have shown small reductions in sound levels after acoustic measures, such as installation of sound-absorbing ceilings or dining tables, although the reduction has often not been shown to be statistically significant (Persson Waye et al. 2009; Sjödin et al. 2014). A majority of preschool teachers are repeatedly exposed to loud screaming into their ears (Fredriksson 2018), such as when comforting a crying child. This particular work

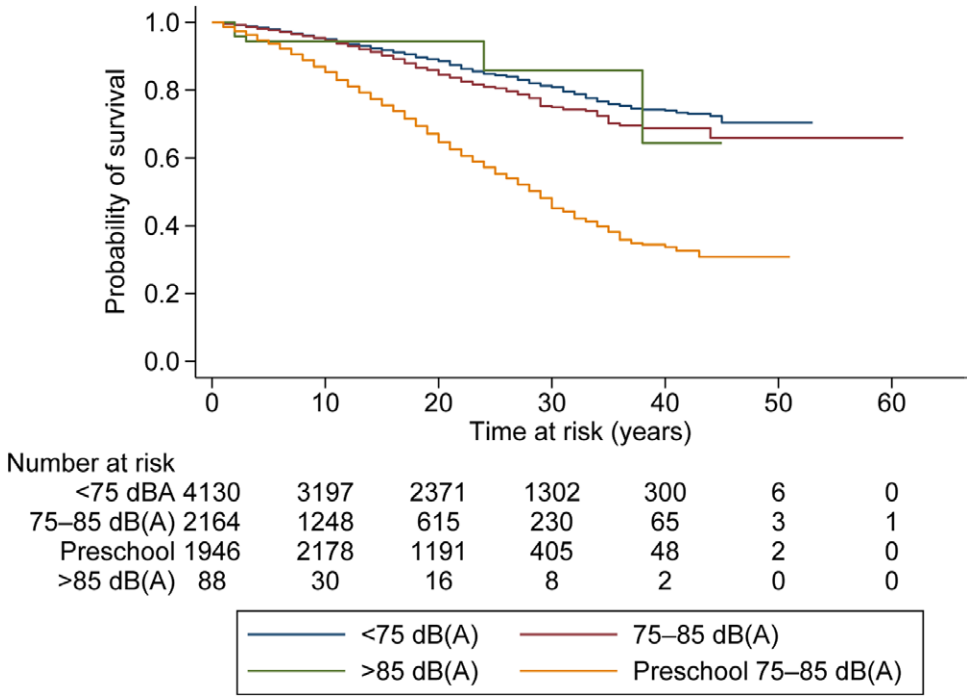

Fig. 2. Kaplan-Meier survival estimates showing the crude probability of hyperacusis (at least a few times each week) onset in relation to person-time at risk in the four occupational noise exposure groups as analyzed in model 2, assigned by the Swedish Job-Exposure Matrix for noise, with time at risk including all years in occupational noise exposure from first reported occupation until end of study

task exposes the personnel to high sound levels directly into the ear, which are difficult to prevent with other means than HPDs. There may be similarities between this exposure and acoustic incidents with sudden intense sounds described in call centers, where exposed operators report hyperacusis and otalgia (McFerran & Baguley 2007; Parker et al. 2014).

The finding of increased risk of hyperacusis both below and above the 8-hour permissible noise exposure limit 85 dB(A) may be explained by the fact that the regulation focuses primarily on reducing the risk of noise-induced permanent hearing threshold shifts as measured by pure-tone audiometry. Thus, the risk of other hearing-related symptoms such as hyperacusis, which are not readily detected by pure-tone audiometry, may be underestimated. Additionally, the regulation does not fully account for intermittency and repeated instances with loud sound levels. This could provide an additional explanation for the substantially higher HR found in the preschool exposure group. A study by Sjödin et al. (2012) showed that the sound environment in preschools is highly intermittent, with more than 100 one-second loggings per hour exceeding 85 dB(A) equivalent levels. In the same study, the prevalence of hyperacusis was found not to correlate with sound level measurements (Sjödin et al. 2012). Importantly though, that analysis assessed neither the exposure at the time of hyperacusis onset, nor the accumulation of exposure preceding onset. The hypothesis of additional risk of hyperacusis in intermittent exposure levels may be supported by studies showing that hyperacusis is common among musicians (Halevi-Katz et al. 2015; Kähäri et al. 2003), and a study showing an association between hyperacusis and self-reported noise exposure among young healthy military recruits (Muhr & Rosenhall 2010). Both musicians and military personnel are exposed to intermittent or even impulse sound, though generally with higher sound levels than that found in preschools.

Contrary to our hypothesis, the main analysis did not show a significantly increased risk among women working in occupations with sound levels >85 dB(A). Although the point estimate was above one, it was uncertain, with wide CI. The sensitivity analysis using the most strict outcome definition was significant with even higher HR, but also wider CI. This indicates that a type-II error due to low power is a less likely explanation for the non-significant results in the main analysis. Most of the events in the highest exposure group remained in the sensitivity analysis, indicating that most of these events were more severe (more frequently occurring). The result from the main analysis could have been interpreted as indicating that hearing conservation programs and noise abatement measures, which are mandatory according to Swedish law whenever workers are exposed to these sound levels, have a protective effect. However, the sensitivity analysis indicate that that is not the case for a severe outcome. One should bear in mind that the regulated noise exposure limits are not set to prevent every case of hearing loss. Our results suggest this may be true also for daily hyperacusis symptoms.

Although noise exposure has been suggested as the most common cause of hyperacusis (Aazh et al. 2014; Anari et al. 1999; Axelsson & Hamernik 1987; Tyler et al. 2014), evidence of causal relationships are so far mainly based on experimental research (Knipper et al. 2013; Pienkowski et al. 2014). In contrast, evidence of noise exposure causing hearing loss is well established, including occupational exposure (Engdahl & Tambs 2010; Nelson et al. 2005). This is important to consider, since hearing loss is one of the main hypothesized initiating mechanism for hyperacusis (Auerbach et al. 2014; Eggermont 2018; Knipper et al. 2013; Pienkowski et al. 2014). Preschool personnel have been reported to have significantly worse pure-tone hearing thresholds compared to unexposed controls (Sjödin et al. 2012).

TABLE 3. Additional outcome sensitivity analysis using more strict definitions of the outcome hyperacusis as: (a) at least several times each week and (b) every day

Occupational Noise Exposure	Hyperacusis Events, n	Person-Years At Risk‡	Incidence Rate per 1000 Person-Years IR	(95% CI)	Crude Model HR	(95% CI)	Adjusted Model HR	95% CI
(a) Outcome defined as at least several times each week								
Model 1†								
<75 dB(A)	335	93,547	3.6	(3.2–4.0)	(reference)		(reference)	
75–85 dB(A)	809	81,712	9.9	(9.2–10.6)	3.6*	(3.2–4.1)	2.9*	(2.5–3.3)
>85 dB(A)	5	949	5.3	(2.2–12.7)	2.2	(0.9–5.5)	2.2	(0.9–5.4)
Model 2, separating preschool teachers§								
<75 dB(A)	335	93,547	3.6	(3.2–4.0)	(reference)		(reference)	
75–85 dB(A)								
Preschool teachers (036.10)	675	50,158	13.5	(12.5–14.5)	4.0*	(3.5–4.6)	3.1*	(2.7–3.7)
All other occupations within 75–85 dB(A)	134	31,554	4.2	(3.6–5.0)	1.3*	(1.1–1.6)	1.3*	(1.1–1.6)
>85 dB(A)	5	949	5.3	(2.2–12.7)	1.6	(0.7–3.9)	1.7	(0.7–4.2)
(b) Outcome defined as every day								
Model 1†								
<75 dB(A)	152	95,096	1.6	(1.4–1.9)	(reference)		(reference)	
75–85 dB(A)	366	84,911	4.3	(3.9–4.8)	3.6*	(2.9–4.4)	2.8*	(2.3–3.5)
>85 dB(A)	4	964	4.1	(1.6–11.1)	4.1*	(1.5–11.7)	3.8*	(1.4–10.3)
Model 2, separating preschool teachers§								
<75 dB(A)	152	95,096	1.6	(1.4–1.9)	(reference)		(reference)	
75–85 dB(A)								
Preschool teachers (036.10)	308	52,785	5.8	(5.2–6.5)	4.7*	(3.8–5.8)	3.7*	(3.0–4.6)
All other occupations within 75–85 dB(A)	58	32,126	1.8	(1.4–2.3)	1.5*	(1.1–2.0)	1.5*	(1.1–2.0)
>85 dB(A)	4	964	4.1	(1.6–11.1)	3.7*	(1.4–10.3)	3.6*	(1.3–9.8)

The models show hazard ratio (HR) of hyperacusis (event) among women in relation to occupational noise exposure assigned by the Swedish Job-Exposure Matrix (JEM) for noise based on the occupation held at year of onset of hyperacusis or at the end of the study if no event, with frailty regression modeling accounting for individual variation in survival times, reflecting for example exposure during years prior to hyperacusis onset.

**Statistical significance ($p < 0.05$).*

†Adjusted for the same confounders as in the main analysis model 1: age, the highest education attained, household income, and family history of hearing loss.

‡Person-years at risk was defined as years exposed to occupational noise until onset of hyperacusis (event) or time of survey if no event occurred, including overlapping years in different occupations for n = 67 subjects.

§Adjusted for the same confounders as in the main analysis model 2: age, the highest education attained, household income, family history of hearing loss, and ever changed jobs because of noise at work.

CI, confidence interval; HR, hazard ratio; IR, incidence rate.

Hearing thresholds were, however, generally within the normal range (≤20 dB HL), consistent with a study that did not find a significantly increased risk of disabling hearing loss (pure-tone average ≥35 dB HL) among preschool teachers (Engdahl & Tambs 2010). It is important though to note that "clinically normal" hearing thresholds (≤20 dB HL) within the standard 0.25 to 8 kHz range does not rule out peripheral auditory disorders, as discussed in a review by Pienkowski (2017a). For example, despite often having normal or near normal pure-tone hearing thresholds, preschool personnel have shown forward masking results similar to that of noise-exposed industry workers, possibly suggesting inner hair cell dysfunction (Lindblad et al. 2014). As mentioned, studies have found hyperacusis in subjects without apparent hearing loss (Anari et al. 1999; Tyler et al. 2014). Findings in experimental studies suggest that cochlear synaptopathy (hidden hearing loss) with its loss of synapses between inner hair cells and type I auditory nerve fibers, resulting from noise exposure, may explain hyperacusis in participants without pure-tone hearing loss (Hickox & Liberman 2014; Liberman et al. 2016). Cochlear synaptopathy has often been elicited with higher sound levels than those occurring in preschool, and over shorter time periods. However, it has recently been suggested to occur as a consequence of long-term noise exposure even at 75 dB SPL (sound pressure level), albeit then failing to find support for hyperacusis when assessed as acoustic startle reflexes in animals (Pienkowski 2017b). Thus, although synaptopathy could be a possible explanation of hyperacusis in participants without apparent hearing loss, strong evidence is still lacking for cochlear synaptopathy and succeeding hyperacusis at noise levels around 75 to 85 dB(A), as found in this study.

Research also suggests that stress may be a risk factor for auditory disorder and that stress may increase the susceptibility of detrimental noise effects on the auditory system (Canlon et al. 2007; Canlon et al. 2013). A study by Hasson et al. (2013) found that women with emotional exhaustion who were exposed to acute stress had more decreased uncomfortable loudness levels

(ULLs) compared with women with low emotional exhaustion. The psychoacoustic measurement of ULLs (sometimes referred to as measurements of loudness discomfort levels, LDLs) show on average discomfort at lower sound levels in patients with hyperacusis complaints compared to normative data (Sheldrake et al. 2015). Thus, Hasson et al. (2013) argued that their results could indicate that stress causes hyperacusis. In a previous explorative study, we found that both stressful working conditions and symptoms of long-lasting stress acted as a modifiers in the association between self-reported noise exposure and hyperacusis, increasing the risk (Fredriksson et al. 2017). If stress is a risk factor for hyperacusis, the substantial HR found among preschool workers compared with other occupations within the same noise exposure group, could be, at least partly, explained by more stressful working conditions and more pronounced risk of stress-related disorder among preschool workers as compared to other occupations (Fredriksson et al. 2019; Wieclaw et al. 2006). Causal relationships between stress and hyperacusis are yet to be determined.

Study Strengths and Weaknesses

A major strength of our study is the sample size, which gave good power in the statistical analysis. The response rate of 38% and 51% may pose some restrictions on the generalization of our results. Bias may have been introduced if exposed participants with the outcome responded to a greater extent. However, in an earlier non-response analysis of these data, we found a higher relative risk of self-reported hearing loss in non-responders than in responders, both among preschool teachers and among women in general, and similarly for the outcome tinnitus among preschool teachers (Fredriksson et al. 2019). We have not been able to assess potential response bias of reported occupations underlying the noise exposure assessment. However, by using the JEM, we avoid common method bias from self-report of both outcome and exposure, which is another strength.

A strength in our exposure assessment was that the occupational history underlying the exposure assignment by the JEM was coded blinded to the outcome. Furthermore, we were able to capture the full occupational history, or nearly so, for each individual, and thus also account for exposure preceding hyperacusis onset. The impact of exposure prior to onset is difficult to discern in relation to possible mechanisms causing hyperacusis, as there is still limited evidence. For hearing loss, the accumulation of exposure is commonly regarded as a risk. The statistical analysis clearly indicated that the data were heterogeneous, such that individuals within the same exposure group had different survival times, possibly as an effect of preceding exposure. The random frailty component used in the regression model takes this into account.

Although the advantages of using the JEM for objective exposure assessment outweigh many of the disadvantages, the limitations of the pre-defined exposure intervals should be considered. First, the wide exposure intervals prohibit the assessment of detailed dose–response relationships. Second, the reference group, <75 dB(A), could be argued not to be completely unexposed. About a third of all the hyperacusis events in the main analysis, with the outcome defined as at least a few times each week, occurred in the reference group. A large number of these events occurred among elementary school teachers. Further analysis is needed, but the results are consistent with a previous study showing a high prevalence of hyperacusis among schoolteachers (Meuer & Hiller 2015). Third, the interval 75 to 85 dB(A) does not differentiate the lower action value 80 dB $L_{Aeq,8h}$ regulated by the Swedish Work Environment Authorities, hindering assessment of potential preventive effects of the regulation. Fourth, assigning noise exposure based on job families (3-digit-level NYK codes), as is done in the current JEM, may introduce exposure misclassification due to variation in exposure between different work tasks and job titles within the same job family. A compensating strength was that our data were coded on a more detailed 5-digit level, which gave us the opportunity to separate preschool workers from non-preschool workers. Finally, the use of the JEM as objective noise exposure assessment has the limitation that individual or situational factors influencing noise exposure cannot be accounted for. We were also not able to control for use of HPDs, although previous studies have shown that self-report of hearing protection devices use may actually overestimate the exposure reduction effect (e.g. Neitzel & Seixas 2005), and thus potentially underestimate the exposure effect. Still, compared with self-report, the JEM assessed noise exposure is a major strength and an important contribution to the research in this area.

The outcome hyperacusis was self-reported using a single question with a defining statement aimed to assess loudness and pain hyperacusis. A strength is that the question has been verified using cognitive interviews, which allows for understanding of how the respondents react to the question and the response alternatives, and of their recall strategies (Willis 2005). In total, ten participants, including five preschool personnel not included in this study, in the ages 21 to 58 years, were interviewed (Fredriksson et al. 2019). As a result of the interviews, the authors introduced the clarifying definition “feel discomfort or pain of everyday sounds” to capture the typical characteristics of loudness and pain hyperacusis, as the supposed mechanisms relating to noise exposure have been described in relation to these characteristics, rather than to conditions such as misophonia or phonophobia. Although a recently published study shows promise in terms of reaching consensus on a definition and description of hyperacusis, at least within an audiological clinical setting (Adams et al. 2020), there is as of yet no standard assessment of hyperacusis and there is generally limited evidence on validity of questionnaires (Fagelson & Baguley 2018). A variety of single-item questions have been used in epidemiological studies to assess the prevalence of hyperacusis, mostly among adults (Andersson et al. 2002; Hannula et al. 2011; Paulin et al. 2016), but also among children and teenagers (Nemholt et al. 2020; Widén & Erlandsson 2004). Large epidemiological surveys often demand single-item assessments for practical reasons. Hence, the use of a more comprehensive instrument such as the 14-item Hyperacusis Questionnaire (Khalfa et al. 2002), which quantifies hyperacusis symptomatology, including avoidance behavior and cognitive and emotional consequences, was not feasible in this study. Another strength is that the clarifying statement attempts to distinguish hyperacusis from general noise sensitivity, as discussed as relevant by Anari et al. (1999). General noise sensitivity, often studied in relation to environmental noise exposure, can be defined as an internal state that increases the reactivity to noise (Job 1999). It has an attitudinal component, but a weaker correlation to psychoacoustic assessments of auditory function (Ellermeier et al. 2001; Miedema & Vos 2003). Another important consideration is the choice of definition of the binary

outcome used in the statistical analysis. We initially used a less strict definition in the main analysis, which we considered relevant in relation to working life, rather than a strict definition, which would perhaps correspond better to a clinically relevant outcome. However, as a standard definition of hyperacusis is lacking, we included additional sensitivity analyzes in which we assessed more strict definitions of hyperacusis, reflecting more frequently occurring symptoms. This is a major strength, which could hopefully also further the discussion of outcome definitions. As is well known, symptom occurrence increase when a less strict definition is used. This was reflected as higher incidence rates in the main analysis, compared with the sensitivity analyses where there were fewer events in relation to a longer total time at risk. Importantly though, our results also indicated that a less strict definition could underestimate the risk (HR) of exposure, particularly among highly exposed subjects. Finally, although the long follow-up time made possible by the retrospective study design is a major strength of our study, we cannot exclude recall bias in reported year of hyperacusis onset. There was a low occurrence of missing data for the reported year, but the previously mentioned cognitive interview found that participants generally felt uncertain about the exact year of onset, unless they recalled a distinctive incident in relation to the onset. Thus, a further analysis regarding the effect of recall bias is suggested.

Clinical Relevance and Further Studies

The results of increased risk of hyperacusis in relation to occupational noise exposure below the permissible exposure limit should have implications for occupational health care, occupational medicine, and audiological management of occupational disability claims relating to hyperacusis cases, particularly among women working in preschool, where a substantial risk was found. In this aspect, validated diagnostic criteria are needed, either psychoacoustic or self-report methods, or a combination. For ULLs, there is a need to find sensitive and specific diagnostic cut-offs. Preliminary cut-offs for ULLs from <95 dBHL (Goldstein & Shulman 1996), to <70 dB HL (Anari et al. 1999), or ≤77 dB HL in the worse ear (Aazh & Moore 2017), have shown varying diagnostic capabilities. A new test using natural sounds has recently been found to have similar or even slightly higher accuracy compared to tests using pure tones, even without having to present uncomfortably loud stimuli (Enzler et al. 2021). For self-report questionnaires, there is a particular need for validated scales and items. Recent studies have shown promising results of the psychometric properties and validity of a new inventory of hyperacusis symptoms (Greenberg & Carlos 2018; Aazh et al. 2020). Validity of single-items should also be addressed as well as definitions and operationalization of the outcome. Ideally, evidence will determine whether different subtypes of hyperacusis are related to different mechanisms and etiologies, and if so, the diagnostic assessment should be able to distinguish between them, considering both perceived symptoms, degree of bother or distress as well as functional impairments.

The results in this study indicate a need for improved risk assessment, hearing prevention measures, and improved routines for including noise and hyperacusis in the systematic work environment assessment in non-industrial occupations, particularly that of preschool worker, but also in other occupations exposed to noise above 75 dB(A), such as worker in a kitchen or restaurant, bartender, waitress, dentist and cleaner. Further studies should also address the fact that we found a third of the hyperacusis events within the reference group, <75 dB(A), of which a large number were related to working as elementary schoolteachers. The educational sector, including both preschool and elementary school, has been specifically pointed out by the European Agency for Safety and Health at Work, regarding noise exposure as a risk deserving much more attention (EU-OSHA 2013).

CONCLUSIONS

This study indicates that occupational noise exposure below the currently regulated 8-hour exposure limit of 85 dB $L_{Aeq,8h}$ increases the risk of hyperacusis among female workers in general, and in particular among women working in preschool. A more than three times increased risk was found among women working in preschool, while an increased risk of at least 30% was found in other occupations with noise exposure 75 to 85 dB(A). These results were confirmed in a sensitivity analysis with a strict division of preschool workers and non-preschool workers as well as in analyses using more strict definitions of the outcome reflecting more frequently occurring symptoms. The substantial risk among preschool personnel is argued, based on earlier studies, to be explained by infrequent use of hearing protection, frequent exposure to screaming in the ears, and possibly also simultaneous exposure to stressful working conditions. Prospective studies are needed to confirm a causal effect of noise on hyperacusis onset, and more narrow exposure intervals are needed to establish a dose–response relationship. However, this study already suggests a need for risk assessments, including improved hearing prevention measures, and noise abatement measures, in occupations with noise levels below the exposure limit. The results could also have implications for management of occupational disability claims relating to hyperacusis, particularly among preschool workers.

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