

A Cross-Sectional Study of the Prevalence and Factors Associated With Tinnitus and/or Hyperacusis in Children

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Objectives: The aim of this study was to determine the prevalence of tinnitus and/or hyperacusis in Danish children aged 10 to 16 years, and to assess associations between tinnitus or hyperacusis and other relevant factors.

Design: A cross-sectional study based on a previously established child cohort. A total of 501 children were enrolled in the project. The study was performed in eight mainstream schools and data were collected during an 8-week period from October 27, 2014 to December 16, 2014.

Results: Using broad tinnitus research questions, the prevalence of any tinnitus was 66.9%; of noise-induced tinnitus (NIT) was 35.7%; and of spontaneous tinnitus (ST) was 53.7%. Bothersome tinnitus was reported by 34.6% of the children with any tinnitus, 23.2% of the whole population. Few children were severely bothered (2.4%, 1.6%, respectively). It was significantly more common for children with NIT to report tinnitus episodes lasting for minutes or longer than for children with ST ($p = 0.01$). Girls were more likely than boys to be bothered by tinnitus [Odds ratio (OR) = 2.96; 95% confidence interval (CI) 1.34 to 6.51; $p = 0.01$]. 14.6% of the children reported hyperacusis, and 72.6% of those reporting hyperacusis were bothered by it, 10.6% of the whole population. The odds of having hyperacusis were 4.73 (1.57, 14.21) times higher among those with ST compared with those without ST. Furthermore, hyperacusis was associated with sound avoidance behaviors such as experience of sound-induced pain in the ear (OR = 2.95, 95% CI 1.65 to 5.27; $p < 0.001$), withdrawal from places or activities (OR = 3.33; 95% CI 1.44 to 7.69; $p = 0.01$), or concerns about sound could damage the hearing (OR = 1.85, 95% CI 1.06 to 3.31; $p = 0.03$).

Conclusions: Tinnitus and hyperacusis are common in children but prevalence is dependent on tinnitus definitions. Only a few children are severely bothered by tinnitus. In the case of hyperacusis, children may exhibit sound avoidance behavior.

Key words: Childhood, Hyperacusis, Prevalence, Tinnitus.

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INTRODUCTION

Tinnitus is a symptom defined as the experience of sound in the head or ears not related to any external source (Baguley et al. 2013). There is no standard diagnostic criterion for tinnitus and various types of questions have been used in epidemiological studies. The inconsistency in defining and reporting tinnitus and the lack of standard questions for tinnitus makes comparison across studies challenging (McCormack et al. 2016). The most common type of question used in adult tinnitus studies is some version of “tinnitus lasting for more than five minutes at a time” (Eggermont 2012; McCormack et al. 2016).

A recent systematic review showed that tinnitus prevalence in children is also population, context, and question dependent (Rosing et al. 2016). In the published literature, different age ranges have been used, different populations and settings have been examined, and the boundaries among childhood, adolescence, youth, and adulthood are unclear (Baguley et al. 2013). With this taken into account, published estimates of the prevalence of tinnitus in children vary between 23.5% (Savastano 2007) and 62.2% (Graham 1981) of children with hearing loss and between 4.7% (Mahboubi et al. 2013) and 54.7% (Sanchez et al. 2016) of children with normal hearing or population-based samples.

Noise-induced hearing loss is often associated with tinnitus, and hearing loss from exposure to noise can either be temporary or permanent, depending on the level or duration of the exposure (Axelsson & Barrenas 1992; Henderson et al. 2011). Three prevalence studies among children have differentiated between noise-induced tinnitus (NIT) and spontaneous tinnitus (ST) (Holgers 2003; Holgers & Juul 2006; Juul et al. 2012). NIT was defined as sound present after listening to loud music or other loud sounds and ST were defined as sound present without having listened to loud music or other loud sound. The same prevalence questions were used in the three studies involving various populations and age groups, identifying a prevalence of NIT within the range of 2.5–53% and of ST within the range of 12–46% (Holgers 2003; Holgers & Juul 2006; Juul et al. 2012). However, only the study by Holgers and Juul (2006) included a measure of tinnitus severity. Recently, the classification “clinically significant tinnitus” has been suggested, defined as noises lasting more than seconds and being bothersome (either slightly or severely) (Humphriss et al. 2016). Humphriss et al. (2016) found ST in 28.1% of 7092 children aged 11 years, and a point prevalence of clinically significant tinnitus to be 3.1%.

For bothersome tinnitus, similar measurement challenges are found with a prevalence variation between 0.6% (Park et al. 2014) and 49.2% (Holgers & Juul 2006) depending on the questions used, response options, population, and age ranges studied (Rosing et al. 2016). Some studies calculate bothersome tinnitus as a proportion of the whole population of children, whereas others calculate from the group of children with

tinnitus (Rosing et al. 2016). It is unknown how many children are troubled to such a degree that they need help to cope with it. Very few children are seen in established tinnitus clinics (Baguley et al. 2013).

As with tinnitus, there is no standard diagnostic criterion for hyperacusis and epidemiological studies of children have been undertaken with various questions used including aspects such as oversensitivity to noise, pain in the ears, and if the child has been bothered by any kind of loud noise (Rosing et al. 2016). Hyperacusis in children has been less studied than tinnitus, and there is no consistency among definitions of how hyperacusis should be measured (Widen & Erlandsson 2004; Coelho et al. 2007a; Landalv et al. 2013). Reports of the prevalence of hyperacusis in children vary between 3.2% (Coelho et al. 2007a) and 17.1% (Widen & Erlandsson 2004).

Baguley defines hyperacusis as “abnormal, lowered tolerance to sound” (Baguley 2003), a definition that others have supported (Moller 2007). Hyperacusis is used as a general term for decreased sound tolerance, regardless of the emotional impact or source of sound (Baguley 2014). An associated term is misophonia (McFerran 2016), which is applied to persons with a strong dislike of sounds, most commonly sounds produced by another person, for example, eating and respiration sounds. Phonophobia (McFerran 2016) is applied to persons with a fear of certain sounds, but the term is also used within the neurological literature to describe intolerance to sounds in migraine headaches (Asha’ari et al. 2010).

Considering adults, Tyler and colleagues (2014) suggested focusing on four categories of hyperacusis to capture the general perception and associated reactions: loudness, annoyance, fear, and pain. Loudness hyperacusis occurs when moderately intense sounds are judged to be very loud compared to what a person with normal hearing would perceive. Annoyance hyperacusis is a negative emotional reaction to sounds that are often, but not always, reported as being loud. Fear hyperacusis is defined as an aversive response to sounds that results in an anticipatory response and avoidance behavior. Annoyance and fear could be considered as self-report emotional reactions, whereas pain hyperacusis might be a physical response or emotional reactions, or both. This typology of hyperacusis has not yet been applied to children.

Hyperacusis can lead to changes in behavior such as avoiding loud situations, social interactions, and public transport (Sheldrake et al. 2015). A distinct pattern of avoidance behaviors can be present with hyperacusis, including avoidance of sound due to actual pain in the ears, irritation and annoyance, and a fear of sound injuring hearing and lead to worse hyperacusis (and tinnitus) (Baguley et al. 2013). Therefore, a three-component model for understanding hyperacusis involving consideration of sensitivity, annoyance and fear of injury has been introduced, with the recommendation that both the classical auditory system and also systems of emotion and behavior must be involved when addressing hyperacusis. As such, hyperacusis is both physiological and psychological at the same time (Baguley & Andersson 2007).

Given the lack of standardized assessment methods for tinnitus and hyperacusis, one way to analyze and compare data is to use assessment methods from previously published studies.

Three studies on NIT and ST are useful in separating NIT from other tinnitus types and are designed specifically to address a childhood population (Holgers 2003; Holgers & Juul

2006; Juul et al. 2012). Concerns have been raised (Viani 1989; Savastano 2007) that there is a risk of both over and underestimates of childhood tinnitus prevalence. Although efforts have been made to focus on study design, questioning techniques and ensuring that research questions are designed and asked in ways that children understand, this is an area in which further knowledge would be valuable, particularly when it comes to the prevalence of hyperacusis in various patient groups.

The aim of the present study is to estimate the prevalence of NIT, ST, bothersome tinnitus, clinically significant tinnitus, and hyperacusis in a group of Danish children from 10 to 16 years. A secondary aim is to explore associations between NIT, ST, bothersome tinnitus, or hyperacusis and other relevant factors.

MATERIALS AND METHODS

Setting

The present study was a substudy of the Childhood Health, Activity, and Motor Performance School Study, Denmark (CHAMPS-study, DK), a longitudinal school-based cohort study (Wedderkopp et al. 2012). The county council of the Municipality of Svendborg, Denmark, created six Sport Schools with increased levels of suitable physical activities, which made it possible to study the health outcomes in these children while comparing them to children who attended the “normal” schools of the region using the design of a “natural experiment” (Wedderkopp et al. 2012). Children and parents from preschool to fourth grade were initially invited to participate in the research program from both sport schools and control schools and the study was kept open, so that new children could enter and leave the study at any time. Therefore, the number of children in the cohort and the specific analyses will differ according to time and the research question asked. At baseline $n = 1218$ children agreed to participate (Klakk 2013). The present study was performed as a cross-sectional study and $n = 1296$ children were enrolled in the CHAMPS-study and were invited to participate.

Children were invited through their parents, from whom written informed consent was obtained along with information on age, gender, and otological history. The study was performed in eight mainstream schools and data were collected during an 8-week period from October 27, 2014 to December 16, 2014. Testing and interviews were conducted in school settings.

Ethical committee approval was obtained from The Regional Committees on Health Research Ethics for Southern Denmark before the start of the project; ID S20140043 and registration with the Danish Data Protection Agency was undertaken.

Population

In total, 1296 children and adolescents from the sixth to ninth grade, from eight public schools, were invited to participate in this study. The heterogeneity of the population reflects that of the general Danish school population (Wedderkopp et al. 2012).

A total of 518 children signed up for participation (40%). Thirty-seven children (7.14%) failed otoscopy and were referred for earwax removal. Seventeen did not get the earwax removed during the 8 weeks data collection window, so a total of 501 children participated in the project. The response rate was 54% for children in the sixth grade, 47% for the seventh grade, 35% for the eighth grade, and 23% for the ninth grade. Figure 1 shows the flowchart of the participation process.

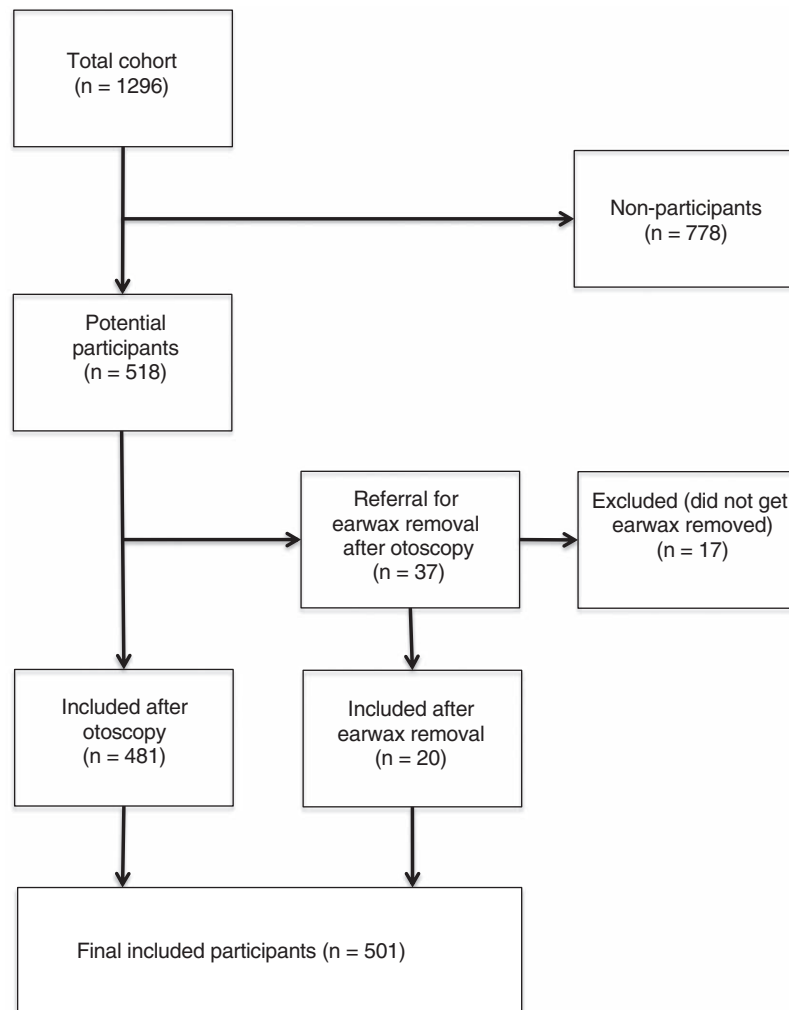


Fig. 1. Flowchart of participants.

To assess the representative nature of the participants, the education level of the mothers of the participants were compared with the whole CHAMPS population. No difference was found among the 1141 available questionnaires.

Study Protocol and Pilot Study

The study protocol, questionnaires on tinnitus and hyperacusis characteristics, and data collection sheet derived from a prevalence study by Humphriss et al. (2016) were used in a modified version translated into Danish with permission. The English wording of the questions is demonstrated in Table 1. Regarding the outcome definition for NIT and ST, the present study used the same questions used in three earlier studies on NIT and ST (Holgers 2003; Holgers & Juul 2006; Juul et al. 2012). Thus, NIT was defined as subjects answering “yes” to question No. 1 and ST was defined as subjects answering “yes” to question No. 2 (Table 1). Any tinnitus was defined as subjects reporting either NIT, ST, or both types of tinnitus. The question for bothersome tinnitus was the same as in the study by Humphriss et al. (2016) with the same response options (question No. 4, Table 1). Also, the definition of clinically significant tinnitus as used by Humphriss et al (2016) was used. Clinically significant tinnitus was therefore defined as having

a tinnitus sound more than seconds in duration, and being bothered by it.

A pilot study obtained in September 2014 tested the questionnaire in 10 children and identified that the children did not understand the Danish version of the sentence: Do you ever experience over-sensitivity to particular sounds? when their comprehension was assessed with a direct question. The hyperacusis question was changed to: Are everyday sounds ever too loud for you? and questions on sound tolerance were added to gain additional information. The reviewed questionnaire was successfully tested on further five children.

The framework of the study was developed by close and detailed discussion by two of the researchers (S.N.E. and D.M.B.) with their clinical experience balancing the need to be simple and accessible to the children, with the need to capture the problem.

Otological History

Before the session, parents had provided information on history of ear infections, otitis media, hearing loss and if the child had had a tympanic membrane operation, ventilation tube insertion, removal of tonsils or adenoids, or other middle ear surgery.

TABLE 1. Questionnaire

Questions or Themes	Answer Choices
Tinnitus measures	
(Q1) After listening to loud music or other loud sounds or noise, have you heard any sort of sounds in your head or ear even after that the loud music or noise has been turned off?	Yes/no
(Q2) Do you ever get noise in your head or ears without first having listened to loud music or other sounds?	Yes/no
(Q3) Do you have it today?	Yes/no
(Q4) Do the noises bother you?	Not bothered/slightly bothered/severely bothered
Tinnitus characteristics	
(Q5) Which ear is it in?	Left/right/both/head/don't know
(Q6) Can you describe the noise?	Buzzing/whistling/other
(Q7) Is it low or high pitch?	Low/high/don't know
(Q8) Is the noise loud or soft?	Loud/soft/don't know
(Q9) Can the noise describes as:	Clicks/pulsed/no/don't know
(Q10) How often do you hear the noises?	Each day/every few days/each week/each month/every few months/ each year/don't know
(Q11) Do you hear the noises	Sometimes (intermittently)/all the time (continuously)/don't know
(Q12) How long do the noises last?	Seconds/minutes/hours/don't know
(Q13) How long had you had the noises?	Days/1 week/1 month/several months/1 year/several years/don't know
Hyperacusis measures	
(Q16) Are everyday sounds ever too loud for you?	Yes/no
(Q17) Are they too loud today?	Yes/no
(Q18) Does it bother you when everyday sounds are too loud for you?	Not bothered/slightly bothered/severely bothered
(Q19) How often are everyday sounds too loud for you?	Each day/every few days/each week/each month/every few months/ each year/don't know
(Q20) How long have everyday sounds been too loud for you?	Days/1 week/1 month/several months/1 year/several years/don't know
(Q21) Does it happen all the time or does it come and go?	All the time/comes and go/don't know
Sound-avoidance behaviors	
(Q14) Do you sometimes experience sounds that you do not like?	Yes/no
(Q15) How often do you experience sounds that you don't like?	Each day/every few days/each week/each month/every few months/ each year/don't know
(Q22) Have you ever experienced sound like pain in the ear?	Yes/no
(Q23) Do you stay away from places or activities because you think the sounds will be too loud for you?	Yes/no
(Q24) Do you ever use ear protection?	Yes/no
(Q25) How worried are you that sound could damage your hearing?	Not worried/slightly worried/severely worried
Other sensory sensitivities	
(Q26) Are you ever bothered by any of the following	Light/colors/touch/pin/smell/taste

Data Collection

All enrolled children had passed the otoscopy to eliminate the risk of cerumen causing tinnitus and/or hearing loss. They were interviewed before or after the hearing measurement session. A hearing therapist specifically trained for this purpose performed a structured interview and ensured that the child understood the questions. If the child hesitated, the question was repeated. If the child still looked doubtful, the question was rephrased with as few changes as possible.

Questionnaires

The children were asked to complete a questionnaire in a structured interview by verbally responding to questions given to them by the hearing therapist. The questions are listed in Table 1. The presence of tinnitus was investigated in three different ways. Q1 in Table 1 explored any experience of NIT. ST was explored with Q2, and point prevalence of any kind of tinnitus was explored with Q3. Finally, troublesome tinnitus was

studied with Q4. Children who answered yes to Q1 or Q2 were further questioned regarding tinnitus characteristics (Q5 to Q13 in Table 1).

As a measure of hyperacusis, children were asked about everyday sounds ever being too loud (Q16) and if this was present at the time of interview (Q17). The children were asked if their hyperacusis was bothersome (Q18). Sound avoidance behaviors (Q14 to Q15 and Q22 to Q26) and other sensory sensitivities (Q26) were investigated among all 501 children.

Before the session, parents had provided information on history of ear surgery or intervention. All tests were conducted according to the ISO 8253 recommended procedure for audiometry, using the shortened ascending technique for determining threshold in 5 dB steps. The test procedures included otoscopy and tympanometry using Otoflex 100 from Otometrics. Tympanometry results were interpreted within the modified Jerger classification system (Fiellau-Nikolajsen 1983). Audiometry tests were made by final year student audiologists within an elective course under supervision from a trained audiologist. Pure-tone

audiometry was conducted using an Astra 2 audiometer with OtoInsert phones. Headphone ME70 (transducer TDH39) were used, if insert-measurement could not be performed. The hearing thresholds were measured at 500 to 8000 kHz and bone conduction threshold at 500 to 2000 kHz. High-frequency pure tone average (HF-PTA) was calculated on the left and right ear as the mean hearing thresholds of 3, 4, 6, and 8 kHz. Low-frequency pure tone average (LF-PTA) was equally calculated as the mean hearing thresholds of 250 Hz, 500 Hz, 1 and 2 kHz. The first ear tested was randomly selected to eliminate systematic bias as learning effect (getting better as the test progresses) or getting tired (and performing worse over time). Audiometry was performed in the school setting, in sound-treated or quiet rooms. The audiometry results were used to refer children to ENT or hospital for further tests, if the hearing thresholds were higher than 20 dB HL at any frequency.

Data Entry, Data Cleaning, and Reduction of Data

Data were entered separately by two persons independently using EpiData double entry and compared afterwards by the first author. Errors were corrected based on original paper forms. The questions of bothersome tinnitus were recoded to yes/no answers used for association analysis, but otherwise kept divided into the response options as presented in Table 1. This was done to distinguish, when needed, between bothered or not bothered or between degrees of being bothered. The answer to duration of noise was recoded into three groups: duration of seconds, more than seconds (e.g., minutes or hours), and “don’t know” answers.

Analyses

STATA version 14.1 for Mac (Stata Corporation, College Station, TX, USA) was used for statistical analysis. Logistic regression was used to test the hypothesis that the outcomes of interest (NIT, ST, bothersome tinnitus, and hyperacusis) were significantly associated with the explanatory variables (tinnitus characteristics, otological history, auditory measures, presence of hyperacusis, sound avoidance behaviors, and other sensory sensitivities). All explanatory variables including variables regarded as confounding factors (age, gender, ENT diseases, previous treatment with ventilation tubes, removal of tonsils or adenoids, middle ear operation as describes in the section

“otological history”) were initially included in all the models and stepwise eliminated again one by one from the regression models using $p > 0.1$ as elimination criteria. If $p < 0.1$ the variable was associated to the outcome of interest and was retained in the final model. The level of Statistical significance was set at $p < 0.05$. Age and gender retained in all models despite nonsignificant influence on the models analyzing tinnitus outcomes.

RESULTS

A total of 501 children with an age range from 10.9 to 16.6 years (mean 13.7, SD 1.1) were tested, 55% (n = 275) were female.

Tinnitus Prevalence

Tinnitus of any kind was reported by 66.9% (n = 335). The prevalence of NIT and ST were 35.7% (n = 179) and 53.7% (n = 269), respectively. Both NIT and ST were experienced by 22.6% (n = 113). The relation between children with NIT, ST, any tinnitus, and no tinnitus is illustrated in Figure 2. Tinnitus point prevalence (tinnitus being present at the time of interview) was found to be 5.8% (n = 29). Table 2 indicates the age distribution of children with tinnitus. Prevalence estimates are presented in Table 3.

Tinnitus Characteristics • As shown in Table 4, 40.0% (n = 134) of the children with any tinnitus reported that their tinnitus was “seconds” in duration. Fifty-one percent (n = 171) of the children with tinnitus reported that their tinnitus was minutes in duration and 7.5% (n = 25) reported that their tinnitus was hours in duration. Tinnitus was experienced each month in 29.0% (n = 97) of the children, each week in 24.2% (n = 81) of the children and every few days in 18.2% (n = 61) of the children. A total of 9.3% (n = 31) of the children experienced their tinnitus each day. Furthermore, tinnitus was experienced for several years in 55.5% (n = 186) of the children. Intermittent tinnitus was reported by 84.8% (n = 284), and continuous tinnitus by 14.3% (n = 48) of the children.

Bothersome Tinnitus • Bothersome tinnitus was reported in 34.6% (n = 116) of children with any tinnitus, 23.2% of the whole population. Bothersome tinnitus was experienced in 40.2% (n = 72) within the NIT group, and in 37.6% (n = 101) within the ST group. In the group with both NIT and ST, 50.4% (n = 57) were bothered. Bothersome tinnitus on the day of the interview

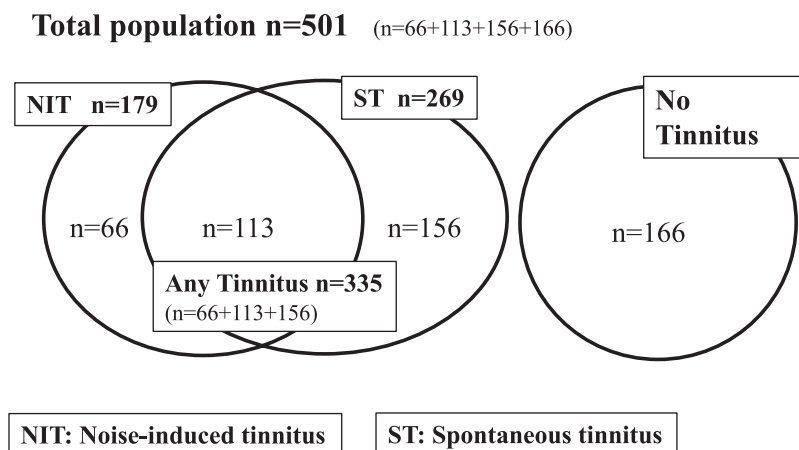


Fig. 2. Relation between children with noise-induced tinnitus (NIT), spontaneous tinnitus (ST), any tinnitus, and no tinnitus.

TABLE 2. Number of Danish children at different ages with different types of tinnitus and hyperacusis

Age (years)	Number (N =)	Noise-Induced Tinnitus	Spontaneous Tinnitus	Any Tinnitus	Hyperacusis
10	1	1	1	1	1
11	12	5	8	9	4
12	148	55	79	95	29
13	162	61	90	115	19
14	94	31	49	60	11
15	78	23	39	49	9
16	6	3	3	6	0
Mean age (SD)	13.7 (1.1)	13.6 (1.1)	13.6 (1.1)	13.6 (1.1)	13.4 (1.1)
Total (N=)	501	179	269	335	73

N = Total study population.

was reported as 44.8% ($n = 13$) of children with any tinnitus, 2.6% of the whole population. 2.4% ($n = 8$) of the children with any tinnitus were severely bothered by the sound. Of those, five reported a duration of minutes, while the remaining three children reported a duration of seconds. There were no children with severely bothersome tinnitus reporting a duration of hours.

Clinically Significant Tinnitus • Clinically significant tinnitus showed a prevalence of 23.3% ($n = 78$) of children with any tinnitus, 15.6% of the whole population.

Hyperacusis Prevalence

Hyperacusis was found in 14.6% ($n = 73$) of the children. As shown in Figure 3, hyperacusis was more common in ST. In total, 72.6% ($n = 53$) of children with hyperacusis, 10.6% of the whole population were bothered by it; 9.6% of children with hyperacusis, 1.4% ($n = 7$) of the whole population were severely bothered, and 63.0% ($n = 46$) of children with hyperacusis, 9.2% of the whole population were slightly bothered. Hyperacusis point prevalence was reported by 2.2% ($n = 11$). All the children with hyperacusis on the day of the interview were bothered by it. Experience of the dislike of sounds was reported in 56.9% ($n = 285$). Experience of sound-induced pain in the ear was reported by 23.8% ($n = 119$), and 7.2% ($n = 36$) of all the children stayed away from places or activities because they thought the sounds would be too loud. The distribution of children with hyperacusis, hyperacusis and tinnitus, their combinations, and no tinnitus or hyperacusis is illustrated in Figure 3.

Otological History

Overall 98.8% ($n = 495$) of the parents provided information regarding their child's otological history. Of those, 30.9% ($n = 153$) reported that the child had at least one ventilation tube insertion. Tonsils had been removed in 5.3% ($n = 26$) of the cases and adenoids were removed in 12.1% ($n = 60$) of the children. Among the 499 children for whom information on hearing aid use was obtained, 1.0% ($n = 5$) used hearing aids. Information regarding concerns about their child's hearing were provided by 99.4% ($n = 498$) of the parents, and showed that 8.0% ($n = 40$) of the parents were concerned.

Factors Associated With the Reporting of Tinnitus

The associations between tinnitus characteristics and other factors influencing the reporting of tinnitus are given in Table 5 for ST and NIT, respectively. In summary, children with ST had 4.7 times higher odds of having hyperacusis than children without ST. It was more common for children with NIT to report tinnitus lasting for minutes or longer than for children with ST [χ^2 (1 d.f.) = 6.6, $p = 0.01$, $N = 219$]. Only a few children ($n = 20$) reported tinnitus lasting for hours, and tinnitus lasting for hours were not demonstrated to be significantly related neither to NIT nor ST. Children reporting both ST and NIT were more bothered than children reporting ST or NIT [χ^2 (1 d.f.) = 19.5, $p < 0.001$, $N = 335$]. Children with NIT were more likely to be bothered by pain due to sound or reporting bothersome tinnitus.

TABLE 3. Prevalence of different types of tinnitus in Danish children aged 10–16 years

	%	n/N	Male		Female	
			n	%	n	%
Any tinnitus (NIT and/or ST)	66.9	335/501	146	29.1	189	37.7
NIT	35.7	179/501	75	15.0	104	20.8
ST	53.7	269/501	118	23.6	151	30.1
NIT and ST	22.6	113/501	47	9.4	66	13.2
Any tinnitus at the time of interview	5.8	29/501	13	2.6	16	3.2
Tinnitus population						
Bothersome any tinnitus	34.6	116/335	40	11.9	76	22.7
Bothersome NIT	40.2	72/179	23	12.8	49	27.4
Bothersome ST	37.6	101/269	34	12.6	67	24.9
Bothersome NIT and ST	50.4	57/113	17	15.0	40	35.4
Bothersome any tinnitus at the time of interview	44.8	13/29	4	13.8	9	31.0
Clinically significant tinnitus	23.3	78/335				

NIT, noise-induced tinnitus; *ST*, spontaneous tinnitus; *n*, number of cases with tinnitus; *N*, total study population.

TABLE 4. Characteristics of tinnitus in Danish children aged 10–16 years

Characteristic	N	Descriptor	n	%		
Duration of noises (any tinnitus)	335	Seconds	134	40.0		
		Minutes	171	51.0		
		Hours	25	7.5		
		Do not know	5	1.5		
		Each day	31	9.3		
Frequency of noises (any tinnitus)	335	Every few days	61	18.2		
		Each week	81	24.2		
		Each month	97	29.0		
		Every few months	43	12.8		
		Each year	14	4.2		
		Do not know	8	2.4		
		Length of history (any tinnitus)	335	One week	4	1.2
				One month	13	3.9
Several months	38			11.3		
One year	48			14.3		
Several years	186			55.5		
Do not know	46			13.7		
Intermittency (any tinnitus)	335	Sometimes	284	84.8		
		All the time	48	14.3		
		Do not know	3	0.9		

N, study population size; *n*, number of cases with the specific tinnitus characteristics.

Audiometry showed that HF hearing thresholds were significantly elevated in the right ear but with a very small difference (mean 3.96 dBHL, SD 7.3) compared to the left ear (mean 3.69 dBHL, SD 7.1) only in children reporting ST.

Factors Associated With the Reporting of Bothersome Tinnitus

Factors related to the reporting of bothersome tinnitus are presented in Table 6. Girls were significantly more likely to be bothered by tinnitus ($p = 0.007$). Children, who described their tinnitus as “loud” and that they heard the sound all the time, were more bothered, as were younger children.

Factors Associated With the Reporting of Hyperacusis

The presence of hyperacusis was significantly higher in children with ST. Sound avoidance behaviors explored as experience of sound-induced pain in the ear, withdrawal from places

or activities, or concerns about sound could damage the hearing were significantly associated with reporting of hyperacusis, as shown in Table 7.

DISCUSSION

Main Findings

This study shows that tinnitus in children is frequent and the prevalence can vary considerably depending on whether severity of tinnitus and the duration and frequency of tinnitus episodes are included in the tinnitus definition. Many studies use broad questions addressing tinnitus without eliminating cases with a short duration of sound or related to external sources, which has been proposed by the National Study of Hearing in the UK (Davis 1989). This definition of prolonged ST includes a sound lasting for 5 min or longer, and not occurring only immediately after exposure to loud noise. As a way to differentiate between experience of “historical tinnitus” and “present

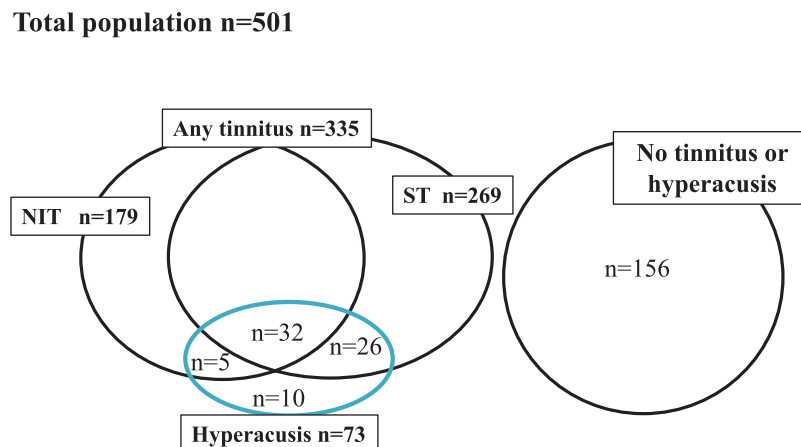


Fig. 3. Relation between children with noise-induced tinnitus (NIT), spontaneous tinnitus (ST), hyperacusis and no tinnitus or hyperacusis.

TABLE 5. Multivariate logistic regression of factors associated with the outcome of having spontaneous tinnitus or noise-induced tinnitus in Danish children aged 10–16 years

	Spontaneous (n/N = 263/323)			Noise-Induced (n/N = 179/335)		
	n	OR [95% CI]	p	n	OR [95% CI]	p
Age (years)		0.85 [0.66–1.11]	0.24		0.94 [0.77–1.15]	0.55
Female gender		0.94 [0.52–1.72]	0.85		0.91 [0.57–1.44]	0.67
Laterality						
Both	118	1 (ref)		*		
Left	39	1.31[0.46–3.75]	0.62	*		
Right	66	0.77[0.35–1.69]	0.51		*	
Head	54	0.45 [0.20–1.03]	0.06		*	
Don't know	46	1.01 [0.40–2.58]	0.98		*	
Duration of noises Seconds	132	1 (ref)		134	1 (ref)	
Minutes/hours	191	0.52 [0.28–0.98]	0.04	196	1.84 [1.15–2.92]	0.01
Hyperacusis	61	4.73 [1.57–14.21]	0.01		*	
Bothersome tinnitus		*		116	1.63 [1.01–2.63]	0.05
Worries that sound can damage hearing	156	0.58 [0.32–1.04]	0.07	163	1.49 [0.95–2.33]	0.08
Bothered by pain		*		105	1.88 [1.14–3.09]	0.01
High frequency PTA left		0.91 [0.83–0.99]	0.03		*	
High frequency PTA right		1.19 [1.07–1.31]	0.001		*	

Variables that were kept in the final model following logistic regression and backward elimination of nonsignificant variables are shown. Numbers in bold represent significant findings. The total population N = 335 are subjects with both spontaneous tinnitus and noise-induced tinnitus (any tinnitus). The total number of subjects with spontaneous tinnitus or noise-induced tinnitus is given as a fraction of the total population in the two models. The size of subgroups is shown in the column n.

The size of the population with spontaneous tinnitus is N = 323 because 5 observations answering: "Don't know" to the "Duration of noises" question were omitted from the analyses. Additionally, 7 subjects not completing all the high pure-tone audiometry frequencies were not included in the model. Thus, 323 subjects were included in the final model investigating the outcome of spontaneous tinnitus.

ref, reference; OR, odds ratio; HF-PTA, High-frequency pure tone average, which was calculated on the left and right ear as the mean hearing thresholds of 3, 4, 6, and 8 kHz.

All variables included in the two models are shown and no additional adjustments were done.

*Indicates variables that were excluded from the particular model except age and gender, which was kept in the models despite nonsignificant influence. All associated factors including variables regarded as confounding factors were initially included in both models and stepwise backward eliminated again one by one from the regression models using $p > 0.1$ as elimination criteria.

TABLE 6. Multivariate logistic regression of factors associated with the outcome of having bothersome tinnitus among Danish children aged 10–16 years (N = 192)

	n	OR [95% CI]	p
Age (in years)		0.70 [0.51–0.97]	0.03
Female gender		2.96 [1.34–6.51]	0.01
Fluid in the ear	33	0.33 [0.09–1.20]	0.09
Hearing loss	9	0.19 [0.03–1.10]	0.06
Ventilation tube insertion	41	3.55 [0.95–13.19]	0.06
Adenoids removed	60	0.27 [0.07–1.12]	0.07
Loudness			
Soft	71	1 (ref)	
Loud	43	3.83 [1.51–9.69]	0.01
Do not know	2	0.43 [0.20–9.02]	0.59
Intermittency			
Intermittent	94	1 (ref)	
Continuous	20	4.34 [1.48–12.73]	0.01
Don't know	2	Missing	
Frequency of experience of dislike of sounds			
Each day	17	0.77 [0.28–2.07]	0.60
At least once a week	32	1 (ref)	
At least once every few months	24	0.27 [0.11–0.62]	0.01
Each year	1	0.24 [0.23–2.53]	0.24
Don't know	1	0.04 [0.00–1.17]	0.06
Concerns that sound can damage hearing	57	2.01[0.97–4.16]	0.06
Bothered by light/colors	82	2.13 [1.08–4.19]	0.03
Bothered by smell	97	0.46 [0.23–0.92]	0.03
High frequency PTA right		1.07 [1.01–1.13]	0.02

Variables that were kept in the final model following logistic regression and backward elimination of nonsignificant variables are shown. All variables included in the model are shown and no additional adjustments were done.

Numbers in bold represent significant findings.

ref, reference; n, number of cases; HF-PTA, high-frequency pure tone average, which was calculated on the left and right ear as the mean hearing thresholds of 3, 4, 6 and 8 kHz. Missing indicates that no subjects had the outcome of bothersome tinnitus within the specific category.

TABLE 7. Multivariate logistic regression of factors associated with hyperacusis among Danish children aged 10–16 years (N = 495)

	n	OR [95% CI]	p
Cases	73		
Age		0.74 [0.57–0.96]	0.02
Female gender	46	1.36 [0.76–2.43]	0.30
Adenoids removed	14	2.09 [1.00–4.37]	0.05
Spontaneous tinnitus	58	3.70 [1.95–7.03]	<0.001
Experience of sound-induced pain in the ear	34	2.95 [1.65–5.27]	<0.001
Withdrawal from places or activities	13	3.33 [1.44–7.69]	0.01
Concerned that sound can damage hearing	45	1.87 [1.06–3.31]	0.03
Bothered by lights/colors	24	1.90 [1.00–3.64]	0.05

Variables that were kept in the final model following logistic regression and backward elimination of nonsignificant variables are shown. Numbers in bold represent significant findings. n, number of cases; OR, odds ratio.

tinnitus”, a measure of point prevalence was included in the present study.

Prevalence figures and severity ratings are therefore difficult to compare across studies due to a number of substantial methodological differences related to tinnitus definitions and survey methodology, which is a key challenge in tinnitus research (McCormack et al. 2016).

In this study, 35.7% (n = 179) of the children indicating NIT reported that tinnitus symptoms lasted for minutes or longer. NIT was significantly associated with the sensation of bothersome tinnitus and the sensation of bothersome pain in the ears. Coelho and colleagues found that history of noise exposure among children aged 5 to 12 years was a risk factor for tinnitus [an odds ratio (OR) of 1.8] and for troublesome tinnitus (an OR of 2.8) (Coelho et al. 2007b). However, it is unclear exactly how history of noise exposure was defined in their study, since only sparse details were given: “children who related exposure to noise and were able to describe the source were classified as having a positive history of noise exposure.” (Coelho et al. 2007b, p.182). Holgers and Pettersson (2005) found that adolescents who attended concerts or clubs or discos and experienced TTS from noise exposure had an odds ratio (OR) of 1.4 to present ST and of 2.0 to NIT. These numbers increased to an OR of 2.8 to present ST and 8.4 to NIT when comparing participants who sometimes experienced TTS to participants who did not have TTS. The wording of the research question used to identify TTS is not reported in the article, which limits interpretation of the data.

NIT has been the subject of studies that indicate a range in prevalence from 2.5% to 74.9% (Holgers 2003; Widen & Erlandsson 2004; Holgers & Juul 2006; Coelho et al. 2007b; Juul et al. 2012; Gilles et al. 2013; Landalv et al. 2013), and concerns have been raised that young people could be at risk of hearing loss due to unsafe listening practices. The prevalence of NIT in the existing study confirms previously published results. The prevalence of NIT indicates an important role for education regarding hearing protection and safe levels of noise exposure, and this is an area for further research.

One might assume that the prevalence of NIT increases with age as a consequence of loud music exposure in teens and adolescents compared with younger children. However, in this study investigating children aged 10 to 16 years, no association between age and NIT was found. This could be due to the fact that the age range was too small, and with few individuals identified in each age group. Only one child was between 10 and 11 years, and only 6 children between 16 and 17 years.

In the study by Holgers and Juul (2006), 22.6% reported tinnitus to be annoying sometimes or more often. As in the present study, they found that the girls were more annoyed by their tinnitus, and they found a weak correlation to the experience of NIT. However, it is unclear if the numbers in the study by Holgers and Juul (2006) derives from the total population or the population of children with tinnitus.

This study supports the view that the high prevalence of tinnitus in childhood does not necessarily indicate that a large number of children are in need of pediatric tinnitus-specific interventions. Indeed, Park et al. (2014) found that although 17.7% of their population-based sample of 12- to 19-year-olds reported tinnitus, only 0.6% of those with tinnitus, 0.1% of their whole population, reported severe discomfort. Recently, Humphriss et al. (2016) found that 84% of their population-based study of 11-year-old children reporting tinnitus were not bothered by their symptom and would not need referral for tinnitus-specific interventions. A recent study on referral patterns and interventions used for Danish children showed that only a small proportion of children with tinnitus underwent referral for tinnitus interventions (Rosing et al. 2016). Similarly, Savastano (2007) found that, although 34% of 6- to 16-year-old children seen at an ENT-department reported tinnitus, only 4.8% of those with tinnitus, 1.6% of their whole population, were significantly worried.

Tinnitus that met the definition used in this study of “clinically significant tinnitus” was reported by 15.6% (n = 78) of the whole population. This is higher than the 3.1% of the total population as shown by Humphriss et al. (2016). They used the same measurement for duration of tinnitus and definition of bothersome tinnitus to calculate the number of children with clinically significant tinnitus, but they only looked at children age 11 with ST and utilized a different tinnitus question. The younger children studied in the Humphriss et al. study and the different format of the tinnitus question may explain the differences between this study and the Humphriss et al. study.

We found a strong association between ST and hyperacusis. Associations between hyperacusis and tinnitus are well documented (Coelho et al. 2007a; Baguley et al. 2013; Hall et al. 2016), although this may not imply causality. Hyperacusis prevalence defined as are everyday sounds ever too loud for you? was estimated as 14.6% (n = 73), which is within the range of the findings from Widen and Erlandsson (2004), who reported that 17.1% of 1285 children aged 13 to 19 years considered themselves to be oversensitive to noise. However, a lower prevalence was found by Coelho et al. (2007a), who found 3.2%

of 499 children aged 5 to 12 years to be bothered by any kind of sound or noise, and also Hall et al. (2016), who found that 3.68% of 7097 children who are 11-year-old ever experience over-sensitivity or distress to particular sounds. These studies, like the present study, were performed among the general population and in school settings. The question in the present study was broad and might have resulted in overestimation the number of children with hyperacusis. Furthermore, the present study as with Widen and Erlandsson (2004) studied relatively older children compared to the studies by Coelho et al. (2007a) and Hall et al. (2016), and these age differences may be involved in the prevalence differences among studies.

Widen and Erlandsson (2004) found that those who reported both tinnitus and hyperacusis were more worried before noise exposure, and that 59.2% experienced pain in the ear associated with loud noise. Hall et al. (2016) found that 42.9% of the children with hyperacusis reported behavioral sound avoidance of places or activities because of hyperacusis. Thus, results from Hall et al. and from Widen and Erlandsson are comparable with the present study, even though a statistical analysis of the significance of these associations only was carried out in the present study.

A recent classification framework for decreased sound-tolerance (Tyler et al. 2014) indicated a subcategory of pain-hyperacusis. That work only considered the adult population and our data indicates that this experience occurs to children also.

Study Strengths and Weaknesses

Low Response Rate • The main limitation of this study is the 40% response rate, which may have biased the results regarding tinnitus prevalence especially if the responders report tinnitus more frequently than nonresponders. The response rate can partly be explained by the fact that the researchers were only allowed to communicate indirectly with parents, in writing via the school. The education level of the mothers of the participants were compared with the whole CHAMPs population and showed no difference between those who participated from those who did not, so it is therefore unlikely that educational levels of the families have influenced the participation rate. However, we found varying response rates from the sixth to ninth grade, where the response rates were highest for the sixth-grade children and lowest for the ninth grade. This may reflect a lack of motivation to respond with increasing age in adolescence. It is therefore possible that motivation of participation in a study can bias the prevalence of tinnitus and hyperacusis in any direction, if knowledge of these conditions before the study can motivate or demotivate participation in the study. It is not possible to analyses in greater detail how nonresponders of the questionnaire differ from responders in order to estimate the importance of the bias related to lack of participation in the study.

Representativeness • It is possible that only those people with an interest in or experience of tinnitus or hyperacusis (either parental or child) agreed to be enrolled. It could also be that families with children with existing or history of hearing problems were more willing to take part in this study, both would cause a risk of overestimate of experiences of tinnitus and/or hyperacusis. In our study, 30.9% of the children had had at least one ventilation tube insertion. This is similar to the findings of Djurhuus et al. (2014) demonstrating at least one middle ear

ventilation tube insertion for 3 in 10 Danish children. According to parental report, 1% of the children in our study used hearing aids. It is assumed that all children with hearing aids have hearing loss, and so the number of children with hearing loss is higher than those with hearing aids. This is comparable to the work by Sudan et al. (2013), who showed a prevalence of hearing loss at age 7 of 1.7% of 54,680 Danish children. Moreover, this study was not based on a weighted sample, thus these findings cannot be extrapolated to the population at large. Therefore, the ability to make any inferences regarding prevalence is limited.

Lack of Validation of the Outcome Measurement • In the present study, there was no test for the consistency of the answers over time, and the lack of test-retest data is a limiting factor and affects the validity of the questions and responses. This underscores the need for future studies to more thoroughly consider issues related to validity, which would likely improve the quality and reliability of the questions, so studies can be directly compared.

Clinical Relevance

According to the findings above, approximately one child out of 40 would be expected to be severely bothered by tinnitus. This could have an important educational impact on the children affected. A recent study (Rosing et al. 2016) found that only a small number of Danish children with tinnitus or hyperacusis are identified by ENTs or the Educational-Psychological Advisory services in each municipality. Since the present study along with earlier published articles indicates that children are not likely to self-report their tinnitus (Mills et al. 1986; Shetye et al. 2010), it seems that children with bothersome tinnitus are overlooked using the current approach. A step forward could be looking for some “soft signs” (like reports of listening difficulties in noise and quiet, dislike or distrust of one ear or sleep difficulties) as recommended by the British Society of Audiology guidance for the management of tinnitus in childhood (Kentish 2015).

The present study also finds that most children who experience hyperacusis are bothered by it. Explicitly, teachers could look for the sound avoidance behavior as a “soft sign” of hyperacusis. The burden that hyperacusis places upon the child is important to acknowledge so that appropriate rehabilitative strategies can be offered, and appropriate commissioning of services be undertaken.

Further studies designed to address experiences of tinnitus and hyperacusis in children could be well served by qualitative research and narrative research (Kentish et al. 2000; Emond & Kentish 2013).

There is currently neither an internationally recognized standard definition for tinnitus/hyperacusis nor a unique internationally recognized classification of these diagnoses. This raises additional difficulties in comparing results across studies. Depending on the questions used in each individual study, the reported data may represent the prevalence of anything from brief to lifetime experiences of tinnitus, with or without considerations about duration or severity. Study design and the study population need to be closely linked and the tinnitus question asked must reflect the age of the child. Recall bias and interview bias should be carefully reflected upon in future studies, as well as potential confounders such as gender, socioeconomic

situation, geographical location, and other health issues like hearing status.

There is an opportunity for researchers within the field of pediatric tinnitus and hyperacusis to find agreement on a standard way of assessing, defining, and reporting prevalence and severity of tinnitus and/or hyperacusis. It would be valuable to obtain standardized information on prevalence, with well-defined questions and tight age ranges, or sampling at a specific age. The most important aspects of designing a questionnaire about tinnitus and hyperacusis in children will lie in phrasing the question so it is age-relevant, making sure the child understand the words used and employing sound-related topics and experiences the child can remember.

CONCLUSION

Tinnitus is prevalent among children but reports of prevalence are dependent on the exact tinnitus question and thereby the definition of tinnitus in the study. Furthermore, duration of tinnitus in each tinnitus episode and the frequency of tinnitus episodes can influence the prevalence as well. Children with NIT were more likely to find the tinnitus bothersome, which could indicate an important role for education around hearing protection and safe levels of noise exposure. Additionally, children with hyperacusis show sound avoidance behaviors, and rehabilitative efforts should be directed toward children where hyperacusis and bothersome tinnitus impact their lives.

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