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study protocol for a systematic review**

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BMJ Open Prevalence of tinnitus and/or hyperacusis in children and adolescents: study protocol for a systematic review

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ABSTRACT

Introduction: There is some debate as to what extent epidemiological data for the prevalence of childhood tinnitus can be relied on. While indications are that the prevalence is relatively high, referral numbers for children with tinnitus are reported to be low and many of the studies have a number of methodological difficulties. We describe the protocol of a systematic review aimed at assessing the prevalence of tinnitus and/or hyperacusis in children and young people.

Methods and analysis: We will include studies of any design (except case reports or case series) comparing the prevalence of tinnitus and/or hyperacusis in children and young people with and without hearing loss, any known external exposure and psychological disorders. We will search the following databases: PubMed, EMBASE and Scopus. No restrictions of language will be applied in the search strategy but during the article selection language is limited to English, German and Scandinavian languages. Primary and additional outcomes will be the prevalence of tinnitus/hyperacusis and the severity, respectively.

Ethics and dissemination: No ethical issues are foreseen. The results will be published in a peer-reviewed journal and presented at national and international conferences of audiology and paediatrics.

Trail registration number: This review protocol is registered in the PROSPERO International Prospective Register of Systematic Reviews, registration number CRD42014013456.

INTRODUCTION

Tinnitus is a symptom defined as the experience of perceiving sounds without any known audible external sound,¹ which is the definition used in this article. Tinnitus can be perceived in one ear or both ears or inside the head. In order to eliminate cases with short duration ringing in the ear, the most commonly used question in epidemiological studies is some version of ‘tinnitus lasting for more than 5 min at a time’.² For the adult population the prevalence of tinnitus falls in

Strengths and limitations of this study

- Clearly established purpose, as well as a systematic and transparent approach.
- Comprehensive search strategy with search and data extraction conducted independently by two authors.
- During the article selection, language is limited to English, German and Scandinavian languages.
- This study will gather well-known published studies to determine the prevalence of tinnitus and/or hyperacusis across studies. This knowledge is important to know the extent of the problem.
- We expect some heterogeneity between studies, which makes it difficult to compare across studies.

the range 10–15%. Unfortunately, there has been no consensus regarding definition of tinnitus used for phrasing appropriate questions and the populations studied vary widely.^{1–3} Factors like age, hearing status and previous noise induction influences the prevalence outcome. An yet unpublished attempt to calculate a worldwide-pooled prevalence estimate of tinnitus in adults has not been possible due to the wide variability in tinnitus definitions among studies.² The number of people with troublesome tinnitus is lower than the overall prevalence,¹ as many people have tinnitus that is not bothersome. It has been noted that the association between tinnitus annoyance and perceived intensity is weak.³

The prevalence of tinnitus in children has been reported and estimates range from 3% to 58%. The available studies have a wide variability in the population studied, some reports considering children with hearing loss, psychological conditions or children who have been noise exposed in comparison with children where these conditions are not known.⁴

Hyperacusis is often present in association with tinnitus. Several definitions of the term hyperacusis are in general use. Jastrebroff and Hazel define hyperacusis as an abnormal sound sensitivity arising from within the



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auditory system, either peripheral or central.⁵ They suggest that decreased sound tolerance consists not only of hyperacusis, but it also consists of a fear of sound (which they refer to as *phonophobia*) or a strong dislike of sound (which they call *misophonia*). In their description, patients with misophonia or phonophobia will have abnormally strong reactions of the limbic and autonomic nervous system without involvement of the auditory system, as defined in hyperacusis. The term phonophobia is also used within the neurological literature to describe intolerance to migraine headaches, adding to further confusion in its own real definition.⁶ Jastrebroff and Hazel use the factor of fear to disambiguate hyperacusis from phonophobia, which can be troublesome in the sense that it lacks the possible influence of context. Environmental factors include both the perceived content of sound (eg, sound intensity and quality) and support and relationships. Another attempt to define hyperacusis is from Coelho *et al*⁷ defining hyperacusis as lowered loudness discomfort levels associated with an abnormal annoyance to sounds. Baguley has described hyperacusis as an abnormal, lowered tolerance to sound,⁸ a definition which others have supported.⁹ This definition gives the possibility of including factors like mood and context to understand the variation of decreased sound tolerances between and within individuals, and will be used in this article.

The prevalence studies in this area are, given the complexity and inadequacy of terminology and definitions, diverse and frugal. The scarce statistics show an approximate prevalence of about 10–15%.¹⁰ Approximately 40% of people with tinnitus also experience hyperacusis.¹¹ The reported range of co-occurrence of hyperacusis and tinnitus is very variable and can vary from 7.3% to 79%.¹²

Prevalence studies of childhood hyperacusis are even sparser. Coelho *et al*⁷ found a prevalence of 3.2% using their previous introduced definition on a population of 506 children, age 5–12 years. An unpublished study on 7,093 11 years old children living in Bristol finds the prevalence of hyperacusis to be 3.68% using the question: “Do you ever experience oversensitivity or distress to particular sounds?” (D Baguley, personal communication, 2014).

There is some debate as to what extent epidemiological data for the prevalence of childhood tinnitus and hyperacusis can be relied on. Although some studies indicate a high prevalence, there is a low rate of spontaneous complaints and a recent study shows that the number of children seen with a primary complaint of tinnitus represents just a small fraction of the total number of patients seen for tinnitus in four European clinics with an established and internationally known tinnitus programme.¹³

This calls for caution when the epidemiological data for the prevalence is interpreted and indicates that it is important to attempt to understand what underpins the variation. Accurate estimates of the true prevalence are of value in planning diagnostic and intervention services.

It has been stated that the prevalence figures vary widely depending on the populations studied, methodologies used and the definition of tinnitus. The majority

of the studies have not ascertained tinnitus severity and/or complaint behaviour. The age range of children studied varies and there is a big degree of difference between definitions (tinnitus, hyperacusis) and measures (severity, perception, annoyance). It has been suggested that difficulties in interviewing children may lead to different answers.¹⁴ It is possible that study design, the definition of tinnitus and differences related to the included participants explain the inconsistencies in currently available studies.

OBJECTIVES

The aims are to assess the degree of variation among prevalence studies of tinnitus and hyperacusis in children, and to provide an overall summary of prevalence diversity.

We will conduct a systematic review of published studies to address the following objectives:

- ▶ To systematically review studies of the epidemiology of tinnitus and hyperacusis in children and young people in order to establish the reported prevalence estimates;
- ▶ To determine factors implicated in the variability of estimates, including those deriving from definitions;
- ▶ To investigate which methodological factors may determine differences in prevalence estimates.

We want to find possible explanations for the high degree of variation between different epidemiological studies to address the main study question: ‘Is the prevalence of tinnitus and/or hyperacusis in children/young people (aged 5–19 years) higher in individuals, who have either hearing loss, psychological conditions or have been noise exposed in relation to children with tinnitus and/or hyperacusis where these conditions are not known?’ Since the variable ‘severity’ is generally less reported than ‘perception’, we will explore the prevalence of chronic bothersome tinnitus whenever possible.

METHODS

Methods for this systematic review have been developed according to recommendations from the PRISMA checklists and the PRISMA Flow Diagram will be used to describe the flow of information through the different phases of the systematic review.¹⁵ This paper uses systematic and quantitative methods to examine reasons for variation in prevalence estimates.

Selection criteria

Study type

Studies will be selected and screened according to the research question and PICOS criteria. They will be included if they are original articles from peer-reviewed scientific journals published in English, German, Swedish, Norwegian or Danish.

Population

Children and young people aged 5–19 years will be included. If the age included in the studies falls outside

this range, the study will be included if the prevalence is rated for different ages.

Comparisons: All studies, excluding case series and case studies, will be considered.

Outcome

The primary outcome will be the point prevalence of tinnitus and/or hyperacusis in individuals aged 5–19 years. The secondary outcome will be the consequences of tinnitus and/or hyperacusis (severity, annoyance, bothersome).

Exclusion criteria

Case study: children under the age of 5 and over the age of 19 years.

Search methods for identification of studies

Electronic searches

The strategy for the electronic search has been developed with the assistance of librarians from the University of Southern Denmark. The first author will perform electronic searches in the databases PubMed, EMBASE and SCOPUS. No language period of publication limitations will be applied in the initial search.

The search period will be from 1960 to 2014. Our search has no restriction in the search period, as we want to include all studies in the review.

PubMed search

The matrix includes the relevant diagnoses (first category) and the topic of the studies we want to review (second category). As PubMed uses specific terms for various age groups, filters will be used to get the right population. Table 1 lists the search words used in the PubMed search.

The following MeSH terms and/or free text syntax will be used for PubMed

((“hyperacusis”[MeSH Terms] OR “hyperacusis”[All Fields]) OR (“tinnitus”[MeSH Terms] OR “tinnitus”[All

Fields]) OR misophonia[All Fields] OR (“hyperacusis”[MeSH Terms] OR “hyperacusis”[All Fields]) OR “phonophobia”[All Fields])) AND ((“epidemiology”[Subheading] OR “epidemiology”[All Fields] OR “epidemiology”[MeSH Terms]) OR (“epidemiology”[MeSH Terms] OR “epidemiology”[All Fields] OR “epidemiologic”[All Fields]) OR (“epidemiology”[Subheading] OR “epidemiology”[All Fields] OR “prevalence”[All Fields] OR “prevalence”[MeSH Terms]) OR (“epidemiology”[Subheading] OR “epidemiology”[All Fields] OR “morbidity”[All Fields] OR “morbidity”[MeSH Terms]) OR (“epidemiology”[Subheading] OR “epidemiology”[All Fields] OR “occurrence”[All Fields] OR “epidemiology”[MeSH Terms] OR “occurrence”[All Fields]) OR (“epidemiology”[Subheading] OR “epidemiology”[All Fields] OR “incidence”[All Fields] OR “incidence”[MeSH Terms])) AND ((“infant”[MeSH Terms] OR “child”[MeSH Terms] OR “children”[All Fields] OR “adolescent”[MeSH Terms]) OR “adolescence”[All Fields] OR “young adult”[MeSH Terms])

EMBASE and SCOPUS search

The matrix includes the relevant diagnoses (first category), the topic of the studies we want to review (second category), and the relevant population groups (third category). Table 2 lists the search words used for EMBASE and SCOPUS search.

The search terms and syntax for EMBASE and SCOPUS will be as follow:

(Tinnitus or Hyperacusis* OR misophonia OR phonophobia) AND (epidemiology OR epidemiologic OR prevalence or morbidity OR occurrence OR incidence) AND (infant OR child OR children OR adolescent OR adolescence OR young adult)

Searching other resources

Manual searches will include scanning of reference lists of relevant papers.

Table 1 Matrix for PubMed search

First category			Second category		
Tinnitus/hyperacusis	N=		Epidemiology	N=	Filter*
Tinnitus	(n=)	AND	Epidemiology	(n=)	Preschool child
OR			OR		Child
Hyperacusis	(n=)		Prevalence	(n=)	Adolescents
OR			OR		Young Adults
Hyperacusis	(n=)		Morbidity	(n=)	
OR			OR		
Misophonia	(n=)		Occurrence	(n=)	
OR			OR		
Phonophobia			Incidence	(n=)	

*“Preschool child” retrieves citations about persons 2–6 year old, “Child” about persons 6–12 years, “Adolescents” about persons 12–18 years, and explodes to broaden the age group (child-preschool, infant (+6), etc.). The filters “Preschool Child: birth-6 years”, “child: 6–12 years”, “adolescents: 12–18 years” and “Young Adults: 19–24 years” will be used.

**Table 2** Matrix for EMBASE and SCOPUS search

First category	N=		Second category	N=		Third category	N=
Tinnitus/hyperacusis			Epidemiology			Population	
Tinnitus	(n=)	AND	Epidemiolog*	(n=)	AND	Infant	(n=)
OR			OR			OR	
Hyperacus*	(n=)		Prevalence	(n=)		Child	(n=1)
OR			OR			OR	
Hyperacous*	(n=)		Morbidity	(n=)		Children	(n=)
OR			OR			OR	
Misophonia	(n=)		Occurrence	(n=)		Adolescent	(n=)
OR			OR			OR	
Phonophobia			Incidence	(n=)		Adolescence	
						OR	
						Young adult	

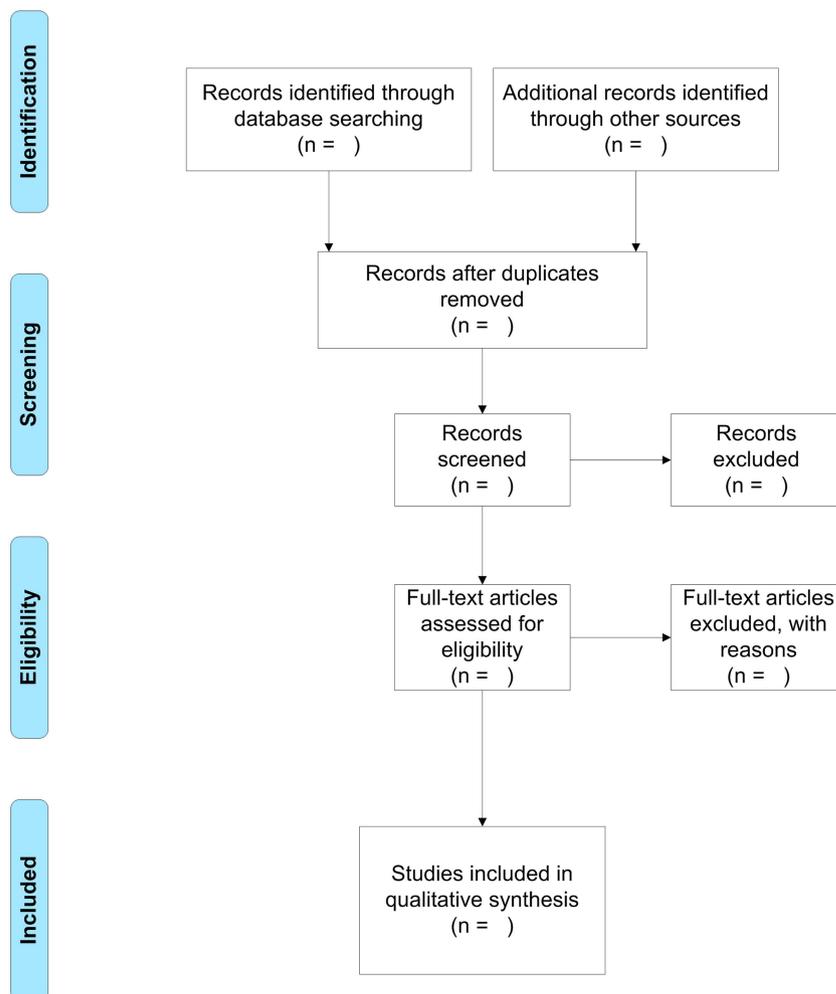
Identification and selection of studies

Studies identified with electronic and manual searches will be listed with citation, titles and abstracts in Endnote. Duplicates will be found using the Endnote function 'find duplicates' to compare each set of duplicate references. [Figure 1](#) provides an overview of the search flow chart.

The eligibility process will be conducted in two separate stages.

- Two authors (SN and JS) will independently screen title and abstracts of all non-duplicated papers and will exclude those not pertinent. A final list will be agreed with discrepancies resolved by consensus between the two authors. When consensus is not reached, a third author (DB or NW) will act as arbitrator. If any doubt about inclusion exists, the article will proceed to the next stage.

Figure 1 Search flow chart (PRISMA flow diagram).



- Agreement between the two authors will be assessed and reported using κ -statistics and overall agreement.
- The full-text version of the articles passing stage 1 screening will be downloaded and assessed for eligibility by two authors (SN and JS), independently. Discrepancies will be resolved by consensus between the two authors and if needed, a third author (DB or NW) will act as arbitrator.

DATA EXTRACTION

Two researchers (SN and JS) will independently perform data extraction; any discrepancies will be resolved by consensus between the two authors. If this is not possible, another author (DB or NW) will make a judgement on the data entered and act as an arbitrator.

Definition of checklist items

The principles from Grading of Recommendations Assessment, Development and Evaluation (GRADE) are useful in order to evaluate the overall quality of evidence

Box 1 Descriptive items

Descriptive checklist

- First author, year of publication
- Design (type of study)
 - ▶ Correlational study
 - ▶ Cross-sectional
 - ▶ Case-control
 - ▶ Cohort
- Study temporality
 - ▶ Prospective
 - ▶ Retrospective
- Setting
 - ▶ Clinical
 - ▶ Epidemiological population study
- Study population
 - ▶ Number
 - ▶ Gender
 - ▶ Age
 - ▶ Method to establish the hearing status (self-reported, audiometric)
- Psychiatric comorbidities
- Data collection
 - ▶ Questionnaire
 - ▶ Interview
 - ▶ Test
- Tinnitus
 - ▶ Definition (selected articles will be grouped by their definition:
 1. No clear definition of tinnitus
 2. Limited to either a sound lasting for more than 5 min or exclusion of noise induced tinnitus
 3. Limited to a sound lasting for more than 5 min and exclusion of noise induced tinnitus.)
 - ▶ Description
 - ▶ Consequences (severity, duration, etc)
- Hyperacusis
 - ▶ Definition (according to Baguley *et al*, noise sensitivity, annoyance/irritation, fear of sound and injury)
 - ▶ Description
 - ▶ Consequences (severity)

Table 3 Quality items

Quality checklist	Yes	No
Clear tinnitus/hyperacusis definition		
Tinnitus/hyperacusis well defined to participants		
Tested for question understood by child		
Clear separation between tinnitus/hyperacusis perception and tinnitus/hyperacusis severity		
Clear separation of aspects of tinnitus		

for the studies¹⁶ and will be used if possible. It is most likely, however, that the quality of the studies is unsuited to evaluate using GRADE. In that case, relevant articles will be reviewed according to the following checklist, in which the authors have defined a set of criteria consisting of descriptors, quality items and study results that is considered essential for this review. **Box 1** shows the descriptive items that will be extracted. **Table 3** provides an overview of the quality items and **table 4** shows the results checklist. Assessment of study quality and bias in included studies is shown in online supplementary file 1.

Data will be extracted and inserted in an Excel sheet.

- Descriptive items
- Quality items

No fixed set of generally accepted quality criteria has been found that suits this type of literature review and therefore quality criteria has been chosen in consideration of factors important for a systematic review of prevalence. We want to look at the following:

- ▶ Which definition of tinnitus/hyperacusis is used and how is it diagnosed?
- ▶ Does the study provide a clear separation between tinnitus/hyperacusis perception and severity?
- ▶ Another possible reason for the prevalent differences could be found in the variations of studied groups. Therefore, we will compare the prevalence findings of the studies in relation to the selected participants: normal hearing, hearing loss, noise exposure and psychological problems.

- Results

Table 4 Results

Prevalence	Tinnitus	Annoyance	Hyperacusis
All			
Boys			
Girls			
<13 years			
>13 years			
Normal hearing			
Hearing impaired			
Noise induced			
Data collection			
▶ Questionnaire			
▶ Interview			
▶ Performed hearing test			

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Contributors SSN, JHS were involved in study design, implementation, analysis and writing. NW and DMB were involved in study design, writing and editing.

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Competing interests None.

Provenance and peer review Not commissioned; externally peer reviewed.

Data sharing statement All recorded data from the data extraction process will be available on request to the extent that it is not included in the systematic review article.

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