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Demographic data, referral patterns and interventions used for children and adolescents with tinnitus and hyperacusis in Denmark

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ABSTRACT

Objectives: To investigate whether children and adolescents with tinnitus and/or hyperacusis are seen in Ear-Nose-Throat (ENT) clinics and to report the clinical data, treatment and referral patterns of these children. To describe the population of children and adolescents with tinnitus and/or hyperacusis found in Educational- Psychological Advisory services (EPAs) and Centres for special Education for Adults (CEAs) and to identify the referral patterns and interventions used for the children in each of these settings.

Methods: A prospective study within 15 ENT clinics was conducted from June 2014 to February 2015. All children with a primary complaint of tinnitus and/or hyperacusis was reported. No changes in daily practice regarding diagnostics, treatment or referral were made. A retrospective case review was undertaken during a five-year period from 01/01/2009 to 31/12/2013 in each Danish municipality and region.

Results: In the prospective ENT study, 12 children were identified and in the retrospective CEA/EPA study 69 children were identified. The 12 children seen by ENT (8 females and 4 male) had an age range from 5.7 to 14 years. The majority of the 69 children seen by CEA/EPA ($n = 50$, 72.5%) had been diagnosed with tinnitus as a primary complaint. Hyperacusis was the primary complaint in 9 cases (12.8%), and both tinnitus and hyperacusis were reported in 11 cases (15.7%). The findings of this study indicate that a majority of children with tinnitus and/or hyperacusis are seen in settings designed for adult audiological rehabilitation. Counselling, including explanations and discussion of coping strategies was the most commonly reported intervention. Intervention methods corresponding with guidelines for tinnitus management in adults were used in 11 cases (15.9%).

Conclusion: Overall only a small number of children with tinnitus and/or hyperacusis were identified in this systems, suggesting that either the children are seen at general practitioner level or not being referred at all. It may also be the case that the incidence of troublesome tinnitus in childhood is lower than the epidemiological data proposes. Referral pathways indicate a general uncertainty about which services provide acquire sufficient intervention. This study indicates that clinicians working at tinnitus services for adults (CEAs) collectively have the skills to help older children, and that a service development focus should be on the younger children as this point.

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1. Introduction

Tinnitus is a symptom defined as a whistling or buzzing sound in the head or ears not related to any external source [1].

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Hyperacusis is the experience of reduced tolerance of sound of moderate or low intensity [2] and is often present in association with tinnitus, but also without. Tinnitus and/or hyperacusis can be experienced in both adults and children, and there has been an increase in studies on tinnitus in children and adolescents over the last 20 years.

The prevalence of tinnitus perception in adults is 10–15%, but the prevalence of troublesome tinnitus in adults is lower (0.5–2%) [1]. The prevalence of childhood tinnitus is broadly equivalent to that in adults, although differences in tinnitus definitions, the tinnitus question used to establish prevalence and whether or not it has been tested if the child understands the question make it difficult to compare across studies [3]. Childhood tinnitus prevalence estimates of tinnitus vary between 4.7% [4] to 54.7% [5] in normally hearing or population-based samples, and from 23.5% [6] to 62.2% [7] of the population of children with hearing loss. Reports of the prevalence of hyperacusis in young people vary from 3.2% [8] to 17.1% [9]. Humphriss et al. [10] found that 28.1% of 7092 11-year-old children had any spontaneous tinnitus (not associated with noise exposure). The prevalence of clinically significant tinnitus (defined as lasting minutes or hours and being bothersome), was reported to be 3.1%. Results of hyperacusis from the same study reported by Hall et al. [11] found that 3.7% of the 11-year-old children reported hyperacusis.

The prevalence of troublesome tinnitus in children ranges from 0.6% [12] to 49.2% [13], with different words used for research question [3], but it is unknown how many children need help coping with it. Only a few children are seen in established tinnitus clinics [14]. Studies have found that children rarely complain of tinnitus spontaneously [15–17], and when they do, there is probably a significant pathological cause [17]. Kentish [18] found that the child developed coping strategies to deal with tinnitus sound itself. Parents and general practitioners (GPs) may think that nothing can be accomplished by seeking clinical support [14]. Furthermore, a survey on UK GPs regarding their practice in adults with tinnitus showed that GPs with an interest in or experience of ENT were less likely to refer patients onwards [19]. In Denmark, treatment of children with tinnitus and/or hyperacusis can be provided in both medical and non-medical settings. Medical based settings for assessment and treatment of childhood tinnitus and/or hyperacusis include ENT, located in clinics or hospitals. Non-medical settings includes Centres for special Education for Adults (CEA), who provide intervention and support for adults with speech, hearing, reading and sight disorders, or Educational- Psychological Advisory services (EPA) for children.

The Danish Guidelines on Assessment and Treatment of tinnitus specifically aimed for medical staff, particular ENTs, recommend that adult patients with tinnitus should be referred to services that specialize in tinnitus management. These include Audiology Departments or CEA [20]. These guidelines state that there is currently no specific knowledge about tinnitus prevalence or annoyance in children. A Good Practice Guideline on Assessment of Tinnitus and Hyperacusis in adults, developed for hearing therapists [21], recommend that tinnitus assessment and intervention should be based on the neurophysiological model of tinnitus [22] and that the Danish version of Tinnitus Handicap Inventory, THI-DK, be used in assessment and as an outcome measure [21,23]. The UK British Society of Audiology (BSA) Guidelines for Good Practice in Childhood Tinnitus recommend that health care professionals involved should have appropriate paediatric skills, and knowledge of care pathways and law-making relevant to the paediatric population instead of being an add-on to tinnitus clinics for adults [24,25].

Currently no similar clinical guidelines exist for managing childhood tinnitus in Denmark or other northern European countries.

Currently only one study [26] has looked specifically at a simplified version of Tinnitus Retraining Therapy [22] outcomes for children with tinnitus. Outcome after six months of treatment were reported to be significant improvement in 81.4% of the cases ($n = 48$), defined as the decrease of at least three of the measured parameters by and minimum of 20%.

As a way of exploring the discrepancy between high tinnitus prevalence numbers and low clinical referral numbers, an eight-month prospective study has been carried out in fifteen Danish ENTs and any cases seen at EPAs or CEAs within a five-year period has been collected retrospectively. The objectives of the study were as follows:

- To investigate whether children and adolescents with tinnitus and/or hyperacusis are seen in ENT clinics and to report the clinical data, treatment and referral patterns of these children.
- To describe the population of children and adolescents with tinnitus and/or hyperacusis found in EPAs and CEAs and to identify the referral patterns and interventions used for the children in each of these settings.

2. Methods

2.1. Organizational structure of the Danish health system

The Danish health system is a tax-based, decentralized health system where hospital care, GP services and public health services are largely free of charge at the point of service [27]. Five regions main responsibility is to deliver hospital care and agree service level with medical doctors through primary healthcare contracts (including GP and ENT specialists in private practice) and 98 municipalities are responsible for providing services such as rehabilitation, public health, school health services and some aspects of prevention. Both regions and municipalities play a critical role in planning and providing public services [28].

In Denmark, patients can self-refer to GPs and directly to ENTs (primary care). Access to hospital care (secondary care) is free, provided there is a referral from a GP [27] or an ENT. These services are paid by the regions. Primary care also includes non-medical services provided and paid by the municipality polity [29]. Each municipality has an EPA service, that covers special needs education for children between the age of 0–18 years within the education system. The commissioning for these is covered by the local municipality according to its internal political and financial structure.

Primary care non-medical services are also provided in CEA, run by the region, but paid by the municipalities. CEAs provide services for. All cost of assessment and treatment in CEAs is covered by the municipal system and *not* the regional system. Within the structure of the Danish health care system, GPs and ENTs cannot refer a child to a CEA without approval from the local EPA.

As stated by the previously mentioned guidelines for medical doctors [20], adult patients with tinnitus should be referred to CEAs for treatment. Since no such guidelines have been established for children, it could be assumed that similar referral pathways would be used for children with tinnitus. This would mean that children with tinnitus and/or hyperacusis found in the medical-setting system (Hospitals or ENTs) would be referred to CEAs for treatment, without first recurring a commissioning approval from the municipal system.

Ethical committee approval was obtained before the start of the project; ID S20140043 and registration in the Danish Data Protection Agency was made, as stipulated by the law J.nr. 2008-58-0035.

2.2. ENT study (medical settings)

As the Danish Health System does not require ENTs to report what diagnoses they use for their patients into a central register, it was not possible to investigate contact with childhood tinnitus and/or hyperacusis in ENT-practice by a registry study. Instead a prospective study was conducted and all cases were therefore enrolled prospectively. The study was conducted among all fifteen Danish ENTs working on the island of Funen. 8.6% (80.689/943.476) of all Danish children in the age range of 5–18 years were living in this area at September 1, 2014 [30]. The ENTs were asked to report all children with a primary complaint of tinnitus and/or hyperacusis seen during an eight-month period.

ENTs were asked not to change their daily practice, neither regarding diagnostics, treatment or referral. Each child's medical record, including results of audiometry and tympanometry were sent to Odense University Hospital in order to get the child registered in the project. All data was collected over the period June 2014 to February 2015, and data were confirmed by email in July 2015.

Children that exceeded the age of 5–18 years old, were excluded from the study. A lower age limit of 5 years was chosen because of the high incidence of otitis media in children below that age [31], and because of difficulties in reliable reporting of tinnitus [3]. In all, fifteen children were found. For further analysis, three cases (20%) were excluded as their age was below five years, resulting in 12 cases.

2.3. EPA and CEA study (non-medical settings)

A retrospective case review was undertaken using a structured email questionnaire to identify and gather information about any cases of children and adolescents (aged 5 to 18) with tinnitus and/or hyperacusis referred to EPAs and CEAs across Denmark. 98 EPAs and 16 CEAs spread over the five Danish counties were contacted by email. Data was collected in the period 01/01/2009 to 31/12/2013, and reminder emails were sent twice. The wording of the questionnaire is detailed in [Appendix 1](#).

Data were collected in different ways due to different patient management systems in the services. Some cases were found by entering a diagnosis code or search word 'tinnitus'/hyperacusis' in a digital database. In other services, the hearing therapist recalled having seen a child with tinnitus, and then went through records manually, or by entering the child's name in the local digital database.

3. Results

3.1. ENT study (medical settings)

From the fifteen ENTs who participated in this study, nine reported no cases. The remaining ENTs reported one, one, one, two, three and four cases, respectively. [Table 1](#) provides an overview of clinical data, referral and treatment of the included cases.

There were a total of 12 cases with an age range from 5.7 to 14 years (mean age 9.2 years, SD 2.24). Eight cases were female, and four cases were male. Half of the cases ($n = 6$) were diagnosed with tinnitus, four cases with hyperacusis (33.3%) and two cases with both tinnitus and hyperacusis (16.7%). Half of the children had grommets inserted in one or both ears. One child had a conductive hearing loss and in another child the audiogram indicated a hereditary hearing loss.

The most common action by the ENT clinicians was referring the children to hospital based secondary care. Nine children were directly referred to the hospital (75%), and one child was referred to

the hospital after initial intervention by the ENT. In one case, the ENT was performing a follow-up on a previous middle ear disease. Only one case received treatment at the ENT, however no follow-up appointment was deemed necessary. After being referred to the hospital, nine of ten cases were given intervention. The type of intervention used at the hospital were counselling including explanation ($n = 7$), sound therapy ($n = 5$) and coping strategies ($n = 4$). Other topics of intervention included sleep, hearing protection and therapy with Bedside Noise Generator, as detailed in [Table 1](#).

3.2. EPA and CEA study (non-medical settings)

The response rate was 75.7% ($n = 75$) from the EPAs and 71.7% ($n = 11$) from the CEAs. Eighty-nine cases were identified of which 20 cases were excluded and 69 cases remained. [Fig. 1](#) shows the exclusion process as well as the distribution of cases. Demographic data regarding age, gender and diagnosis was recorded, as were referral patterns and intervention techniques used.

3.2.1. Population

There was a total of 69 cases (mean age 14.5 years, SD = 3.3). Thirty-eight cases were male (55.1%) and 31 cases were female (44.9%). Seven cases (10%) were aged less than 10. Referral year was reported in 67 cases. The mean number of cases in each of the responding centre per year was 0.16 overall; 0.87 case per year was seen in CEAs and 0.056 case per year was seen in EPAs ([Figs. 2 and 3](#)).

The majority of the cases ($n = 50$) had been diagnosed with a primary complaint of tinnitus (72.5%), 8 cases (11.6%) were given the diagnosis of hyperacusis and the remaining 11 cases (15.9%) were reported having both tinnitus and hyperacusis. An additional auditory or developmental diagnosis was reported in 26 of the cases distributed as follows: in 17/50 cases of tinnitus (34%), in 5/8 cases of hyperacusis (62.5%) and in 4/11 cases with both tinnitus and hyperacusis (36.4%).

In children with tinnitus, hearing loss was found in 10/50 cases (20%). Three of the cases had linguistic difficulties, possibly related to their hearing loss. For children with hyperacusis, 2/8 (25%) suffered from hearing loss. Other additional diagnoses included: head trauma, hearing loss, ADHD, behavioural and learning difficulties, reduced eyesight, tunnel vision and "massively sensitive senses in general". The additional diagnoses for each of the four children with both tinnitus and hyperacusis were respectively: unilateral hearing loss, phonophobia, Auditory Processing Disorder (APD) and headaches and blurred vision.

3.2.2. Referral patterns

We found 48 cases in CEAs (69.6%), and 21 in EPAs (30.4%). There was a significant difference in age, ($p < 0.0001$, t -test) with children referred to CEAs being older (mean age = 15.7 year, SD 2.4) than children referred to EPAs (mean age = 11.6 year, SD 3.3). The referral pathways to and from EPAs and CEAs are shown in [Fig. 4](#).

Group 1 represents children referred from the audiology departments to EPAs or CEAs. 61.8% of these cases were referred directly to CEAs without approval for commissioning from EPAs, with a mean age of 15.5 years (age range 10.11–18 years, SD = 2). The remaining 38.2% were referred from audiology departments to EPAs, with a mean age of 11.4 years (age range 6–17 years, SD = 3.4).

A similar pattern is seen in Group 2, referrals from ENTs, where 93% were referred directly to CEAs and the remaining 7% to EPAs. The mean age for children referred from ENTs to CEAs was 15.7 years (age range 10–18.8 years, SD = 2.8). One child was referred from ENT to EPA (13 years).

Table 1
Clinical data, referral and intervention.

Case	Age	Gender	Diagnoses	Audiometric comments	ENT	Hospital	Further referral
1	7,9	Female	Tinnitus Hyperacusis	Right ear: grommet insertion Left ear: Type C tympanometry	Referral to hospital	Counselling incl. explanation Sound therapy	–
2	5,7	Female	Hyperacusis	Bilateral grommet insertions	Referral to hospital	Counselling incl. explanation Coping strategies Avoid hearing protection	To EPA (inform school)
3	11,5	Female	Tinnitus Auditory processing disorder Mucoid otitis media Chronic nonsuppurative otitis media	Bilateral grommet insertions Bilateral hearing loss 30 dB HL at 250 Hz	Referral to hospital	Counselling incl. explanation Stress	–
4	9,4	Male	Tinnitus Cerebral palsy Auditory processing disorder Suppurative otitis media, unspecified Acute tonsillitis, unspecified Acute sinusitis, unspecified	Bilateral grommet insertions	Sound therapy Referral to hospital	Counselling incl. explanation Counselling about APD Sound therapy Sleep	To EPA (test bedside noise generator)
5	7,0	Male	Hyperacusis	Bilateral grommet insertions	Referral to hospital	Hyperacusis is not bothersome No need for treatment	–
6	7,1	Female	Hyperacusis	Bilateral Type C tympanometry	Referral to hospital	Incomplete assessment due to lack of translator (the child's parent did not speak Danish)	–
7	9,4	Female	Tinnitus	No comments	Referral to hospital Cancelled (tinnitus complaint stopped)	–	–
8	10,4	Male	Tinnitus	No comments	Referral to hospital	Counselling incl. explanation Stress Sound therapy Treatment with masker Sleep Coping strategies	–
9	14,0	Female	Tinnitus	Bilateral scars after previous grommet insertion Left ear: Tympanometry with high peak pressure	Referral to hospital	Counselling incl. explanation Coping strategies Sound therapy	–
10	7,4	Male	Hyperacusis	Obstruction of Eustachian tube Bilateral grommet insertions Right ear: Type C2 tympanometry	Follow up on previous middle-ear disease Assessment	–	–
11	11,2	Female	Tinnitus Hyperacusis	No comments	Counselling incl. explanation Referral to hospital	–	–
12	9,5	Female	Tinnitus	Audiogram indicate hereditary hearing loss. Bilateral detritus	Referral to hospital	Counselling incl. explanation Coping strategies Sound therapy Sleep Hearing protection in music class	–

EPA: Educational-Psychological Advisory service.

APD: Auditory processing disorder.

Group 3 represents children referred to CEA from other places than the audiology departments or ENT. This group had the highest mean age of the population (mean age 16.1, SD = 2.4, age range 9–18). The majority of cases in this group were over the age of 16 years and in some way contacted the centres on their own initiative (self-report, private clinic, GP and CEA). We found no self-reported cases less than 16 years old.

Respectively, group 4 represents children referred to EPAs from other places than the audiology departments or ENT and had the lowest mean age of the population (mean age 11.7, SD = 3.1, age range 8–17.9). Most of these cases were between the age of nine to fifteen and were mostly referred from or already in the school/EPA system due to other previous difficulties.

Among the cases referred to CEAs, 20 were onward referred to other service providers (41.7%). The majority (85%) were referred from CEA to EPA for approval of the child to be treated at CEA and funding authorization. Ten of the cases referred to EPAs were onward referred to another service provider (47.6%). For 80% of these cases, the EPAs stated that they had referred the child to respectively a CEA (n = 5) or an Audiology Department (n = 3), because they believed the knowledge and experience needed to help the children was better in those services.

There is a difference in referral patterns when looking at age.

21.7% of the children were in the age range 5–10 year (n = 15), while 78.3% were in the age range of 11–18 year (n = 54). Thirteen of the children under 11 years were initially referred to EPAs (13/15). Two children under 11 years were referred to CEAs (2/15). Forty-five of the children in the age range 11–18 were initially referred to CEAs (45/54), while nine were referred to EPAs (9/54). Children referred to EPAs had a mean age of 11.5 years (range 6–17.9 years), while children referred to CEAs had a mean age of 15.7 years (range 9–18.8 years).

One specific CEA reported more than 30% of all cases (n = 22) with a mean of 4.4 cases each year. This could reflect local well established referral arrangements.

3.2.3. Intervention strategies

Of the total sample, eight families did not attend further appointments. In four of those cases the parents initially had to get a financial approval from the local EPA. Four families received unspecified counselling from another service provider. Three cases involved children with special needs and were given specific counselling. In eighteen cases, management intervention techniques were not reported. Table 2 shows the management intervention techniques specified in the remaining 36 cases. Most cases received intervention methods from multiple categories in

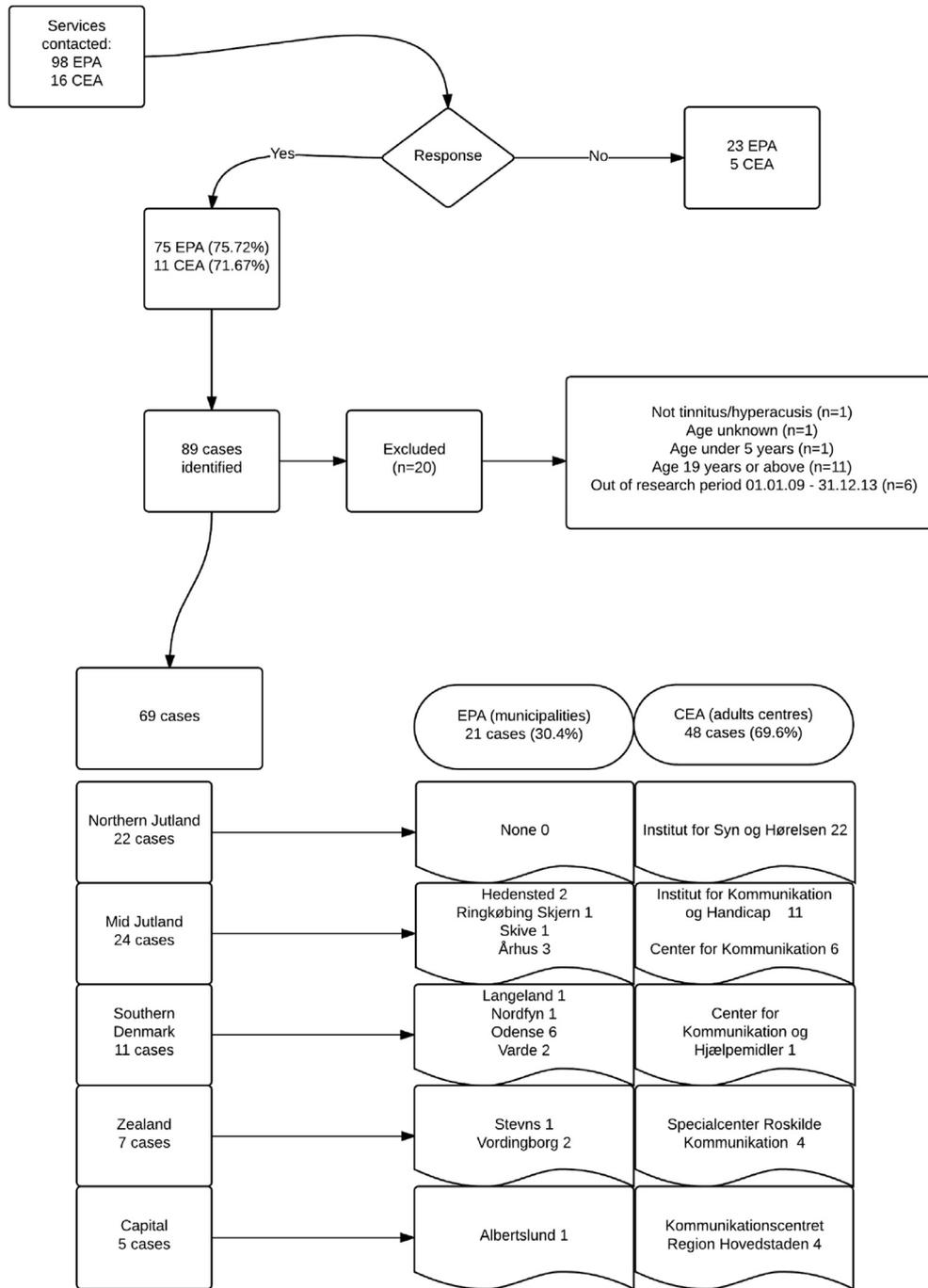


Fig. 1. Flowchart non-medical settings.

accordance to personal need.

Counselling including explanation for the main diagnosis was the most commonly used intervention method followed by coping strategies, which were used more at CEAs (n = 9) than EPAs (n = 2). Counselling based on the neurophysiological model (n = 8), the use of cognitive therapy (n = 3) and THI-DK (n = 3) were all types of intervention only used at CEAs. Issues related to sleep were addressed at both CEAs (n = 7) and EPAs (n = 3) and a bedside sound generator were used in seven of the ten cases.

Apps for mobiles were more commonly used at EPAs. Counselling about hearing protection was given more at EPAs (n = 4) than CEAs (n = 1). Guidance on how the family can support the child and

knowledge about tinnitus specifically addressed to the parent was provided (n = 6), mostly at CEA, while tinnitus impact at school (n = 6) was mostly addressed at EPA. Four case were given counselling about anxiety and depression and in one case suggestions for exercises to minimize tensions in neck and jaw were given.

In twelve cases intervention was offered to the child but did not involve parents. In eleven of them, this could be explained by the high age of the children involved (age range 15–18, mean = 17.1, SD = 1.16). The remaining case was a child aged nine, who reportedly were not significantly troubled by the tinnitus, which can explain why little/no intervention was given.

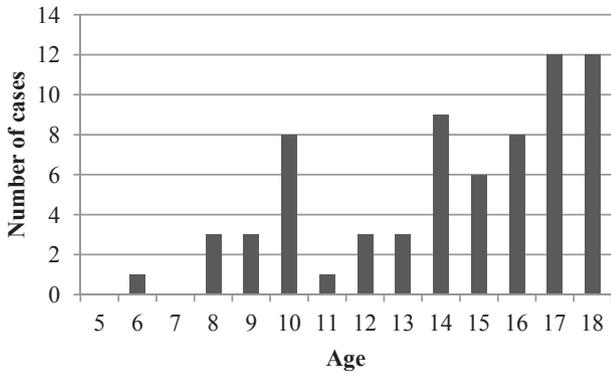


Fig. 2. Age distribution of cases.

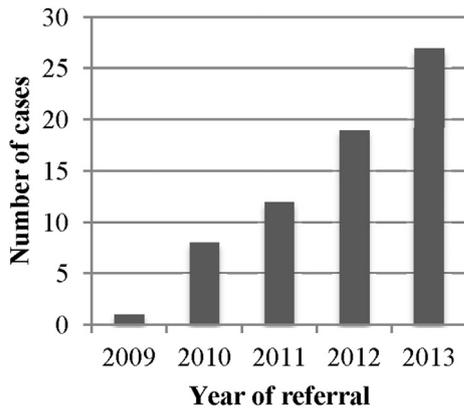


Fig. 3. Year of referral.

4. Discussion

4.1. Main findings in medical settings

Only 6 out of 15 ENTs reported having seen any children with tinnitus and/or hyperacusis. In the geographically defined area Funen, inhabited by 8.6% of all Danish children between 5 and 18 years old, 12 children were found during an 8-month period. This corresponds to an incidence of 22.3 children per 100,000 per year, which indicate that only a small number of children with tinnitus and/or hyperacusis are found and treated at ENT clinics.

A mean age of 9.2 years, and the fact that more than half of them had or had had grommets, could indicate that the majority of these children were initially seen at the ENTs because of middle ear problems. Almost all children were onward referred to an audiology department at Hospital, which could suggest that the knowledge for treating childhood tinnitus and/or hyperacusis is not sufficient at ENT practices. Topics of intervention included at the audiology department are similar to the ones suggested in The UK BSA Guidelines [24]. Appropriate paediatric settings also recommended by these guidelines are provided both by ENTs and hospital. Nevertheless, it could be assumed that the hospital has a wider established paediatric audiological service and have a critical mass, which supports the argument that intervention for children with tinnitus and/or hyperacusis could be provided at the hospitals.

4.2. Main findings in non-medical settings

According to a recent study by Humphriss et al., approximately one child per class of 30 would be expected to have clinically significant tinnitus at age 11 years [10]. Detailed statistical information on the Danish society is provided by StatBank Denmark [30]. Table 3 shows the number of 11-year-old Danish children in the

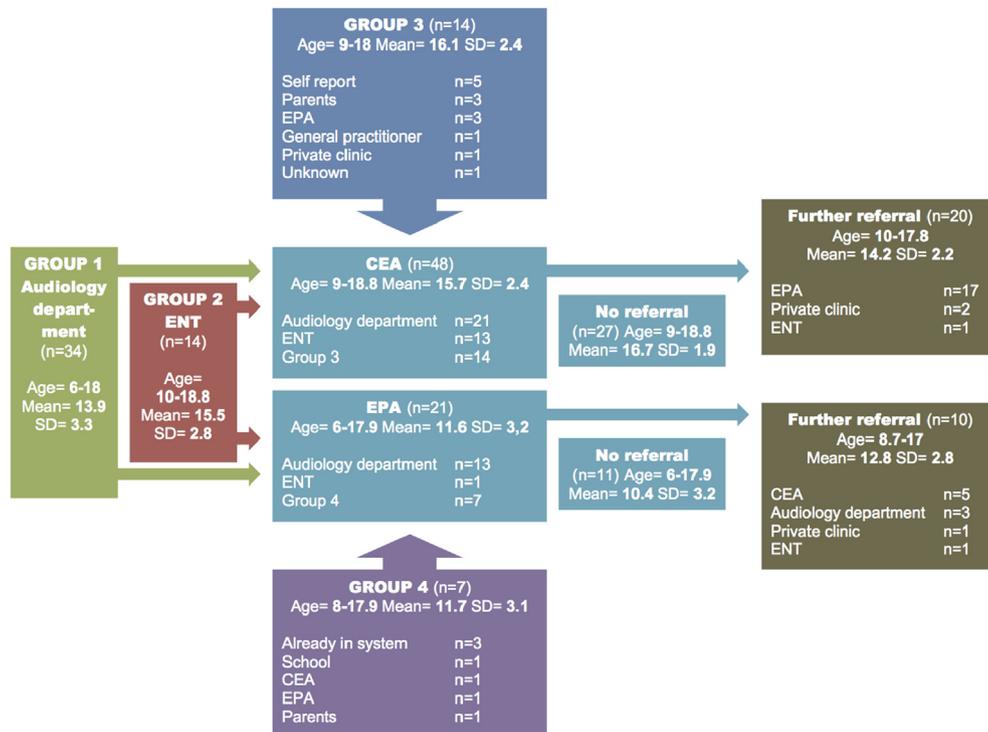


Fig. 4. Referral patterns. Abbreviations: ENT: Ear, Nose and Throat Specialists, CEA: Centre for special Education for Adults, EPA: Educational-Psychological Advisory service.

Table 2
Management intervention techniques.

	Total (n = 36)	CEA (n = 24)	EPA (n = 12)
Counselling incl. explanation	28	18	10
Coping strategies	11	9	2
Sleep	10	7	3
Neurophysiological model	8	8	0
Bedside	7	6	1
Sound therapy (apps)	6	2	4
Parents	6	4	2
School	6	2	4
Hearing protection	5	1	4
Hearing loss and other diagnoses	4	0	4
Anxiety	4	2	2
THI-DK	3	3	0
Cognitive therapy	3	3	0
Exercises (jaw and neck)	1	1	0

studied period. The table also provides an estimated numbers of children expected to have clinically significant tinnitus according to the study by Humphriss et al. The centres examined in the present study only saw very few children in total, and only one child at age 11 during the five years. However, the increase of referral from 2009 to 2013 may indicate a raising awareness of children having tinnitus and/or hyperacusis by health care professionals working in the field of audiology.

Ninety percent of the children were aged 10 years or older (n = 62). This number is very similar to the study by Baguley et al. [14], who found 7% aged under 10 referred to tinnitus clinics. They found a higher number of children having both tinnitus and hyperacusis (39%) than the present study (15.7%). The mean age of the children in this study (14.4 years) is the same as the mean age reported by Bartnik et al. [26] (age range 7–17).

Two patterns of onward referral were identified in the non-medical settings. Referrals from EPAs to CEAs occurred because healthcare professionals at the EPA felt that managing tinnitus in a child was beyond their scope of practice. Referrals from CEAs to EPAs was related to funding approval. The use of this pathway potentially prolongs the time period before the child can receive accurate information and help. Furthermore, there seems to be a lack of understanding about where appropriate services are located. This could lead to a lack of accurate information and counselling about tinnitus for the child and its family and could prevent the child from learning practical strategies for tinnitus management.

The main intervention techniques found in this study have also been reported by other studies [14,18]. Aside from these techniques this survey additionally identified use of the neurophysiological model, THI-DK and coping strategies. This is most likely due to the influence of the practice of adult tinnitus treatment [21].

Despite recommendation from the BSA Guidelines [24], the majority of the children in this survey were seen in tinnitus clinics for adults. In eleven of these cases, intervention techniques were used according to guidelines for adult tinnitus management. This is an indication that healthcare professionals, transfer tinnitus management intervention techniques for adults to children. If this is the case, there is a risk of not getting age appropriate help, if referred to CEA.

4.3. Study strengths and weaknesses

This study relied upon reliable retrospective responses from health care professionals. In general, there is a risk of underestimates due to lack of time to find and report cases. No information on tinnitus or hyperacusis definition, tinnitus severity or question used by each ENT to examine the child are provided in this study. This could be a potential bias, but since the children only were seen if they had a complaint of tinnitus and/or hyperacusis, the risk over over-reporting must be low. Some of the EPA and CEA centres that did not respond to the survey could potentially have provided additional data. The manual search option was the only options for some centres to locate cases, and there is a risk of recall bias, which could lead to missing data. Since the referral patterns shows a flow from and to CEAs/EPAs, there could be a risk that the same child has been reported twice (both from a CEA and a EPA). To avoid this, all cases were reviewed for similarities that could indicate duplication: none were found.

4.4. Clinical relevance

4.4.1. Low number of children

The finding in the present study adds on to the discrepancy between prevalence studies and clinical findings, and the need to distinguish between tinnitus sensation and tinnitus severity. Also the lack of methodological and population consistency among studies indicates that the prevalence data should be interpreted with caution [3]. Giving the epidemiological data for clinically significant tinnitus, large numbers of children would then either be seen at medical settings as GPs, ENTs, hospital or would be undiagnosed. There could be a risk that the primary care system as a whole not is aware of childhood tinnitus, and the role of Danish GPs in assessment and treatment on childhood tinnitus and/or hyperacusis are still to be explored. In an international comparison of health systems for tinnitus, Hall and colleagues [32] observed that GPs often are the 'first port of call' for a patient seeking help with their tinnitus, while most ENTs see referred patients, typically from GPs. However, all Danish residents have free direct access to both GPs and ENTs.

If only a small number of children and adolescents have tinnitus

Table 3
Number of 11-year old children in Denmark 2009–2013.

Year	2009	2010	2011	2012	2013	Total
Number of 11 years children in Denmark	68,983	67,534	67,323	68,090	66,273	338,203
Expected tinnitus cases (one child out of 30)	2299	2251	2244	2270	2209	11,273

and/or hyperacusis to an extent that requires intervention from a special service with critical mass, only few specialist services for children with tinnitus and/or hyperacusis are needed.

4.4.2. Referral patterns

This system review provides a description of the Danish commissioning system for assessment and intervention for childhood tinnitus and hyperacusis, and can therefore contribute to service development. Comparing referral pathways in the medical and non-medical settings, showed two main reasons for further referral.

The majority of cases found in the ENT study were referred solely to the hospital and 20.3% of the cases referred to the non-medical settings came from the ENT which suggests that ENT practices do not have enough knowledge to treat childhood tinnitus and/or hyperacusis. The non-medical study also showed a strong referral pathway from audiology departments to non-medical settings, suggesting that the competence does not lie there either, or that the guidelines for assessment of adult tinnitus for medical staff [20] also are followed regarding children. Lastly, professionals at EPAs reported referring children to CEAs or hospital service, because they believed the knowledge and experience needed to help the children was better in those services. These referral patterns show a general insecurity about which services have sufficient knowledge about childhood tinnitus and/or hyperacusis, and provide assessment and intervention for these symptoms. Good practice guide for (adult) tinnitus care published by the English Department of Health [33] emphasised the importance of clear guidelines and criteria for referral.

The second main driver of further referrals regarded concerns about commissioning due to the structure of the Danish health care system. A strong referral pattern from CEAs to EPAs for approval of commissioning for their services were identified. Bogason [34] found in a report on the Danish CEAs that optimal rehabilitation was challenged by administrative and organizational problems and there is established unnecessarily burdensome and a costly decision-making system. Along with our findings, this emphasizes the need for improvement of this part of the Danish health-care system. An OECD review of health care quality in Denmark [28] concluded that even though municipalities have been asked to take on additional health care responsibilities, they report having little capacity to take on new roles in primary care. This study indicates that clinicians working at adults' tinnitus services (CEAs) collectively have the skills to help older children, and that the focus for service development as this point should be on the younger children.

4.4.3. Intervention methods

Presently, most minors seen in this system with tinnitus and/or hyperacusis are adolescents, which clinicians collectively have the skills to help. In the non-medical study, we found that many services actually remodel services used for adults, to fit these adolescents, but believe that their approach could be more systematic and better organized.

The majority (67%) of the children and young people found in the non-medical study were aged 15 or over. Whilst having specialist services for children with tinnitus is a counsel of excellence, in resource limited health care systems, these young people could have a slightly modified version of adult care without harm being likely. The need for specialist paediatric tinnitus and/or hyperacusis services could then focus upon the younger children.

It will be difficult to develop and maintain accurate knowledge and intervention skills for childhood tinnitus in EPAs, if only sparse referrals are made for tinnitus and/or hyperacusis in each of the 98 municipalities. The structure of the Danish healthcare system,

along with the lack of clear guidelines for treatment of children with tinnitus, could indicate diverse referral pathways across the country. However, only children with troublesome tinnitus need multidisciplinary-based interventions. Improvements could potentially be achieved through development of specialist paediatric tinnitus services in Denmark, at the maximum one in each of the five counties. Children with troublesome tinnitus and/or hyperacusis could be referred to centres with specialist expertise and critical mass, and local health care professionals could receive further education on the topic and supervision.

Declaration of interest

The authors report no declaration of interest.

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Ethics approval

Ethical committee approval was obtained before the start of the THACAD project; ID S20140043 and registration in the Danish Data Protection Agency was made, as stipulated by the law J.nr. 2008-58-0035.

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Appendix 1. Questionnaire

Dear xxx

PhD fellow, Master of Arts in Audiologopedics, Susanne Nemholt is currently doing a PhD thesis studying children and young people aged 5 to 19 with tinnitus and/or hyperacusis. In Denmark, we do not have a comprehensive registry that collects information on children/young people with this diagnosis, and we therefore know very little about the group.

I am contacting you in order to gain more knowledge about these children and young people.

Hyperacusis is:

A dislike of certain everyday sounds such as cars, trains, hairdryer, is unusual. However, in some children, over-sensitivity to sound can become more of a problem causing upset, annoyance and distress to the child. The child may become very frightened of the noise and avoid situations where it is likely to occur e.g. birthday parties, cinemas, school playgrounds and travel by train or bus.

Tinnitus can be described as a sound, such as buzzing or ringing, that is heard in the ears or inside the head.

I would like you to answer the following question: *Have your EPA/CEA been in contact with a child/young person with tinnitus and/or hyperacusis within the last 5 years?*

If "no": please return this mail to me.

You can either write an answer to me, or simply send a blank email back to me so I know you have received the email and have reported back that you have had no contact with a child/young

person with tinnitus and/or hyperacusis.

If “yes”: please answer the following questions for each child as well:

Age of the child (first meeting):

Diagnosis:

Who referred the child to EPA/CEA?

What kind of counselling did the parents receive?

What kind of treatment did the child receive?

How many appointments did the child received?

How many appointments did the parents received?

What kind of professionals did the child/parents see?

What is your profession?

Do you have any information regarding the case you think would be useful for me to know about, please note them here.

Please answer by returning this mail to me.

Thank you for your cooperation. It is very appreciated that you took the time for helping gaining more knowledge about children and young people with tinnitus and/or hyperacusis.

If you have any questions, please do not hesitate to contact me at: (phone) or (email).

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