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Hyperacusis : demographic, audiological, and clinical characteristics of patients at the ENT department

**Reference:**

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## 1 Abstract

2 **Purpose:** To document whether patients with and without hyperacusis differ from each other on  
3 demographic, audiological and clinical characteristics.

4 **Methods:** Based on the Hyperacusis Questionnaire's (HQ) cut-off ( $HQ > 28$ ), a total of 2301 participants  
5 were divided into patients with and without hyperacusis. Demographic data, scores on self-reported  
6 questionnaires (Tinnitus Functional Index (TFI), Visual Analogue Scale of tinnitus loudness ( $VAS_{loudness}$ ),  
7 Hospital Anxiety Depression Scale (HADS)) and audiological parameters were retrospectively analysed  
8 to determine differential factors between the two groups.

9 **Results:** In total, 10,9 % of the patients was classified as hyperacusis patients ( $n = 251$ ). They reported  
10 a significant, higher tinnitus severity (mean difference of 19 points on TFI) and mental distress (mean  
11 difference of 4 points on the HADS subscales) ( $p < .001$ ) than patients without hyperacusis. Moreover,  
12 this group consisted of more women (45% % in hyperacusis group vs. 35 % in non-hyperacusis group)  
13 and women scored significantly higher on the HQ ( $p < .001$ ) and TFI ( $p < .01$ ).

14 **Conclusion:** Patients with hyperacusis have distinctive characteristics. The presence of hyperacusis in  
15 combination with tinnitus can indicate a higher need for psychoeducation. Patients that present  
16 themselves with hyperacusis without tinnitus complaints remain a minority, yet might be  
17 underdiagnosed. Hence, future studies should disentangle tinnitus from hyperacusis. In clinical  
18 practice, greater efforts are required to increase knowledge about hyperacusis as a primary or  
19 secondary complaint and to provide individualized treatment for these patients.

## 1. Introduction

Hyperacusis is defined as a reduced tolerance to sound(s) perceived as normal to the majority of the population or perceived as normal to the person before the onset of hyperacusis [1]. It is a self-reported auditory symptom which is often associated with tinnitus (i.e. the perception of a sound in absence of an auditory source). Common underlying mechanisms have been proposed for both symptoms. The central gain model states that reduced cochlear output can cause a maladaptive over-amplification of peripheral input, giving rise to higher spontaneous activity (tinnitus) and/or higher stimulus-evoked neural activity (hyperacusis) [2]. Moreover, similar treatments have been proposed for both conditions, such as Tinnitus Retraining Therapy (TRT) and Cognitive Behavioural Therapy (CBT). However, recent literature suggests that tinnitus and hyperacusis should be seen as distinct phenomena [3]. The field of hyperacusis is young and many research questions are still to tackle with regard to its epidemiology, pathophysiology, assessment and treatment [for the full overview see 3]. The current study aims to systematically evaluate the demographic, audiological and clinical characteristics of patients with and without hyperacusis in order to provide insights in the similarities and differences of the two clinical populations.

The large overlap between tinnitus and hyperacusis has already been identified by epidemiological research. The prevalence of hyperacusis in a tinnitus population (i.e. 7-79%) has been estimated to be much higher when compared to the prevalence in the general population (i.e. 8-15%) [4-10]. Similarly, the prevalence of tinnitus in a hyperacusis population (i.e. 40-86%) is reported to be much higher in comparison with the general population (i.e. 10-15%) [6, 11-15]. Hyperacusis has not been studied as a standalone problem for many years. Nevertheless, prevalence rates have demonstrated that there are hyperacusis patients without tinnitus.

Hyperacusis and tinnitus are associated with hearing loss and show altered neural activities in auditory and non-auditory brain areas [16]. Yet, a recent study by Hofmeier, et al. [17] demonstrated functional biomarkers that distinguish between tinnitus with and without hyperacusis. More specifically, the comorbidity was associated with a more widespread signal amplification process through overactive thalamo-cortical activity. Furthermore, divergent characteristics of tinnitus and hyperacusis were highlighted by Baguley & Hoare (2018). For example, tinnitus is often lateralized and intermittent, while hyperacusis is mostly bilateral and constant. As such, it has been suggested that these two conditions should follow a separate diagnostical approach in order to provide effective treatment [3, 17]. For example, hyperacusis can be a symptom of superior semi-circular canal dehiscence syndrome, in which case surgery can be offered [18].

The interplay between tinnitus and hyperacusis formed the central focus of earlier studies [e.g. 8, 19]. Gilles et al. (2014) found that patients with severe tinnitus more often also perceive hyperacusis compared to patients with a lower degree of tinnitus severity. Similarly, Schecklmann and colleagues proposed the comorbidity of hyperacusis as a useful criterium for defining a sub-type of tinnitus with a greater need for treatment [8]. In their study, a total of 55% of the tinnitus patients were characterized as hyperacusis patients. The comorbidity was associated with younger age, reduced quality of life, increased tinnitus-related distress and mental burden. Furthermore, these patients rated their subjective hearing level as worse and their tinnitus as louder and higher pitched, whereas the audiologic measurements were unable to confirm this patient-reported outcome. With regard to the tinnitus-related characteristics (e.g. tinnitus duration, laterality, character), the authors did not find a relationship with the presence of hyperacusis. The main limitation of this study was the definition used to identify hyperacusis patients. Tinnitus patients who answered 'yes' to the question "Do sounds cause you pain and physical discomfort?" were considered as hyperacusis patients in the analysis. However, hyperacusis can also exist without these aspects being present [1]. Moreover, the authors acknowledged that this question might be interpreted with relation to the tinnitus and therefore reflecting tinnitus burden.

The present study aims to build on these pioneering studies by identifying patients with hyperacusis based on the cut-off score of the Hyperacusis Questionnaire (HQ) [20].

## Methods

The intake data of 2301 patients who were registered at the Tinnitus Treatment and Research centre Antwerp (TINTRA) at the ENT department of the Antwerp University Hospital (UZA) (i.e. a tertiary referral centre with expertise in evaluation and treatment of tinnitus) were analysed for the current study. The patients consulted the department for tinnitus and/or hyperacusis complaints between 26-Nov-2014 and 13-Mar-2020. A more extensive dataset with regards to tinnitus characteristics and hearing status was available in the research database of the ENT department (OpenClinica LLC, an electronic CRF application), including 622 patients who were enrolled in a clinical study [e.g. 21, 22-26].

### *1.1. Ethical Committee*

The Committee for Medical Ethics of the Antwerp University Hospital / University of Antwerp approved the study (19/43/485). The study was conducted according to the Helsinki Declarations.

## 1.2. Questionnaires

All 2301 patients filled out the validated Dutch versions of the following questionnaires on a touch-screen desktop. The scoring of the questionnaires was computerized.

### 2.2.1. Hyperacusis Questionnaire

Sound tolerance was questioned with the Hyperacusis Questionnaire (HQ), consisting of 14 self-rating items with a four-point answer scale [20, 27]. The total score ranges from 0 to 42 with higher scores indicating higher levels of sound tolerance complaints. A score higher than 28 is used as a cut-off score for hyperacusis [20].

### 2.2.2. Tinnitus Functional Index

The effect of tinnitus on eight domains (intrusiveness, sense of control, cognitive interference, sleep, auditory difficulties, relaxation, quality of life and emotional distress) was assessed by the Tinnitus Functional Index (TFI). This questionnaire assesses tinnitus-related burden and is favoured over the Tinnitus Questionnaire (TQ) by Jacquemin, et al. [28] as it is much shorter and has a similar convergent validity and even slightly higher agreement with the self-reported perceived effect. It consists of 25 questions with Likert scale responses from 0 to 10. The total score ranges from 0 to 100, with a higher score representing a higher tinnitus severity and accompanying burden. More specifically, a score above 25 is considered as a clinically significant complaint of tinnitus that requires treatment [29, 30].

### 2.2.3. Visual Analogue Scale of tinnitus Loudness

The mean tinnitus loudness in the last four weeks was scored on a Visual Analogue Scale for tinnitus loudness (VAS<sub>loudness</sub>) from 0 (absence of tinnitus) to 100 (as loud as possible) [31].

### 2.2.4. Hospital Anxiety Depression Scale (HADS)

In order to screen for symptoms of anxiety and/or depression, the Hospital Anxiety Depression Scale (HADS) was filled out by the patients. This questionnaire consists of 14 items with four-point (0-3) response categories [32, 33]. For each subscale, a score between 8 and 10 indicates a 'borderline' psychological morbidity and a score higher than 10 indicates 'case' of anxiety or depression.

### 1.3. Audiological examination

For the subsample of 622 patients, further analyses were performed on a more extensive dataset, including audiological data and tinnitus characteristics, of the research database of the ENT department. The following audiological tests were part of at least one of the research protocols in this database.

#### 2.3.1 Pure tone Audiometry

Air conduction thresholds between 125 Hz and 8 kHz were determined during a pure tone audiometry according to the clinical standards (ISO 8253-1,2010) using a two-channel Interacoustics AC-40 audiometer in a soundproof audiometric booth. Pure tone averages of the low frequencies (PTA<sub>low</sub>: 0.5, 1 and 2 kHz) and high frequencies (PTA<sub>high</sub>: 1, 2 and 4 kHz) were calculated. This data was missing for 10 patients.

#### 2.3.2 Speech audiometry in quiet

Speech reception in quiet (SPIQ) was conducted in a soundproof booth by use of the Dutch open-set NVA lists [34], with each list consisting of 12 monosyllabic words. The speech reception threshold (SRT) was determined by calculating the sound level of the speech stimuli at which the patient could recognize 50% of the phonemes correctly. This test was not part of all research protocols and was available for 76 patients.

#### 2.3.3 Speech audiometry in noise

Speech audiometry in noise (SPIN) was conducted in a soundproof booth by use of the Leuven Intelligibility Sentence Test (LIST) [35]. This Dutch sentence test consists of 35 lists of 10 sentences with an equivalent difficulty. The SRT, which is the 50% correct identification point, was calculated by using an adaptive procedure (fixed noise level of 65 dB SPL, 2 dB down - 2 dB up procedure) and expressed in dB SNR. This test was not part of all research protocols and was available for 256 patients.

#### 2.3.4. Tinnitus information

Tinnitus characteristics were available from the anamnesis, such as side of tinnitus (unilateral left, unilateral right, bilateral, central), tinnitus type (pure tone, noise, polyphonic) and aetiology (idiopathic, otologic, non-otologic, psychological). Furthermore, psychoacoustic analysis with regards to tinnitus pitch and loudness was part of the dataset. Tinnitus pitch (expressed in Hz) and tinnitus loudness (expressed in dB Sensation Level (dB SL)) were determined. A forced-choice technique was applied for frequency matching. The patient had to choose between two pure tones or small band noises (depending on the type of tinnitus) of different frequencies presented in the contralateral ear

until the frequency was found that was most similar to his/her tinnitus. This frequency was used to determine the tinnitus loudness in the ipsilateral ear. The patient could finetune the intensity with 1 dB SL accuracy. The test was performed in a soundproof booth. This test was not part of all research protocols and was available for 197 patients.

#### *1.4. Statistical analysis*

In order to identify patients with a clinically significant complaint of tinnitus or hyperacusis respectively, the TFI and HQ were dichotomized based on the cut-off of these questionnaires (i.e. TFI > 25 and HQ > 28). For statistical analyses, we compared data of patients with and without hyperacusis using chi-square-tests for categorical variables and two-sided independent t-tests for continuous variables. Pearson correlation coefficients were calculated when interested in the relationship between two continuous variables. A p-value less than .05 was considered as statistically significant. Adjustment for multiple testing was performed by controlling the false discovery rate. Data were analysed using SPSS statistical software version 27 (SPSS Inc., Chicago, IL, USA).

## **2. Results**

In the sample of 2301 patients who were registered at TINTRA, 10.9 % have hyperacusis (i.e. 251 patients) and 80.3 % have clinically significant complaints of tinnitus (i.e. 1848 patients) according to the self-report questionnaires. There is a considerable overlap between the two groups, with 233 patients reporting clinically significant complaints of tinnitus as well as hyperacusis. In other words, 13 % of patients with clinically significant complaints of tinnitus also have hyperacusis and 93 % of patients with hyperacusis have clinically significant complaints of tinnitus. For 18.9 % of the patients, no clinically significant complaints of either tinnitus nor hyperacusis were present. Figure 1 provides more insight in the tinnitus severity and hyperacusis severity of the patients in this sample.

### *2.1. Self-reported questionnaires*

Patients with hyperacusis show significantly higher scores on the TFI (and its subscales), HADS and VAS<sub>loudness</sub> compared to patients without hyperacusis ( $p < .001$ ) (see table 1 for mean differences, t-values and p-values of the two-sided independent t-tests). Figure 2 visualises the scores on these questionnaires for both groups.

## 2.2. Demographics

The gender distribution shows 55 % men and 45 % women for the patients with hyperacusis and 65 % men and 35 % women for patients without hyperacusis (see table 1 for the demographic data of patients with and without hyperacusis). This gender ratio significantly differs between patients with or without hyperacusis complaints ( $p < .01$ ). Moreover, a gender difference is apparent in terms of tinnitus and hyperacusis severity, with women scoring significantly higher on the HQ ( $p < .001$ ) and TFI ( $p < .01$ ) (Figure 3).

The mean age of patients was not significantly different between patients with hyperacusis and without hyperacusis (table 1). The total score of the TFI is significantly, positively correlated with age ( $r = .05$ ,  $p < .05$ ). However, the correlation between the HQ score and age is not significant ( $r = .03$ ,  $p > .05$ ).

## 2.3. Audiological and tinnitus characteristics

In the subsample of 622 patients, for whom audiological data and tinnitus characteristics are available, 15.9 % have hyperacusis (i.e. 99 patients) and 90.7 % have clinically significant complaints of tinnitus (i.e. 564 patients). There is also a considerable overlap between the two groups, with 98 patients reporting clinically significant complaints of tinnitus as well as hyperacusis. For 9.2 % of the patients, no clinically significant complaints of either tinnitus nor hyperacusis were present.

There are no clear differences in terms of audiological (PTA, SPIQ, SPIN) or tinnitus characteristics (pitch, loudness, side, type, aetiology) in the patients with hyperacusis compared to patients without hyperacusis (table 2). Yet, a small, positive correlation between duration of tinnitus and hyperacusis severity is found ( $r = .11$ ,  $p < .01$ ). However, this correlation does not reach significance after correction for multiple testing.

## Discussion

The current study systematically evaluated different demographic, audiological and clinical characteristics of patients with and without hyperacusis from a large sample of patients who registered at the ENT department, providing insights in the similarities and differences of the two clinical populations. The major differences between the hyperacusis group compared to the non-hyperacusis group found were in terms of gender (i.e. 45% women in hyperacusis group vs. 35 % women in non-hyperacusis group), tinnitus distress (i.e. higher tinnitus distress in hyperacusis group with a mean difference of 19 points on TFI) and mental distress (i.e. higher mental distress with a mean difference of 4 points on the HADS subscales) and these results are in line with previous studies [8, 11, 12, 19]. Hence, the presence of hyperacusis in tinnitus can indicate a higher need for psychoeducation. On the



other hand, patients with hyperacusis were not significantly older in the current study, whereas Schecklmann, et al. [8] demonstrated that these patients were younger. This can be due to differences in defining hyperacusis complaints, namely a score of >28 points on the HQ versus response to a single question.

A higher tinnitus distress and more anxiety and depression symptoms were demonstrated in patients with hyperacusis complaints, suggesting common or cooperating mechanisms. Hence, the presence of hyperacusis indicates that the need for treatment might be higher. The risk for comorbid anxiety and depression disorder being higher in patients with hyperacusis is an important red flag for clinicians to bear in mind [10, 12].

The associations with audiological and tinnitus characteristics were analysed to shed light on the pathophysiology of hyperacusis. No significant associations were found, similarly to Schecklmann, et al. [8]. Future studies with a prospective design are needed to confirm this finding, as the current data was limited to a smaller sample size. The small correlation between duration of tinnitus and hyperacusis severity, which was insignificant after multiple testing correction, raises the question whether hyperacusis is a predisposition for tinnitus rather than a consequence. Hence, it challenges the idea that hyperacusis may be present prior to the development of tinnitus [8]. Under the framework of the fear-avoidance model (i.e. presence of tinnitus or hyperacusis leading to catastrophizing thoughts, fear, and avoidance behaviours), both scenarios could be true. Independently of tinnitus or hyperacusis being present first, these auditory symptoms might reinforce each other through increased fear and attention in a dynamic relationship [36].

While more men present themselves at our department with complaints of tinnitus and/or hyperacusis, it was interesting that there were relatively more women in the hyperacusis group and that women showed more severe complaints. This result is in agreement with gender differences previously reported in the field of tinnitus and hyperacusis [10, 11, 37], and can be explained by maladaptive coping strategies being more likely in women [38]. Other proposed theories are an intermediate influence of emotional exhaustion and stress on auditory sensitivity in women [39] and/or differences in stress coping, as there is a well-established relationship between stress and hearing problems [39]. Furthermore, a higher prevalence of women in the somatic tinnitus population has been reported [37], which raises questions regarding the somatic component in the hyperacusis population. A recent article by Michiels, et al. [40] showed a higher prevalence of hyperacusis in a group of patients with a higher degree of somatic tinnitus, which is in line with the earlier study by Schecklmann, et al. [8]. However, other studies did not demonstrate this finding [41]. Hence, future studies on this topic are needed to shed light on this possible association. In the field of chronic pain,

similar findings are apparent. (Pain-) hyperacusis (i.e. a condition where moderate intensity levels evoke pain) is also a significant symptom in this field. Suhnan, et al. [42] indicated that central sensitisation, in which nociceptive circuits become hypersensitive and spread from the periphery to higher centres in the brain, can explain this comorbidity.

The major strength of the present study is that all patients filled out both the TFI and HQ. While these data are continuous, dichotomisation is possible because of their cut-off scores. Hence, analyses looked into the effects of the presence of hyperacusis and/or tinnitus complaints, as well as into the severity of these complaints. Even though some authors have discussed whether the original cut-off score of the HQ is too high [43], this original cut-off was used in the current study in order to ensure that only the patients with clinically significant hyperacusis were identified as having hyperacusis.

The current findings cannot be extrapolated to the general population, as the data of patients with hyperacusis and/or tinnitus complaints were gathered in a tertiary referral centre with expertise in evaluation and treatment of tinnitus. Patients that present themselves with hyperacusis without tinnitus complaints remain a minority, yet might be underdiagnosed. The recent definition of hyperacusis: "A reduced tolerance to sound(s) perceived as normal to the majority of the population or perceived as normal to the person before the onset of hyperacusis" [1] might support defining the group of patients we are investigating, leading to optimization of clinical guidelines for diagnosing hyperacusis and further treatment.

To develop a full picture of hyperacusis and its treatment options, future studies should disentangle tinnitus from hyperacusis. For many years, pathophysiology studies did not focus on hyperacusis as a standalone problem and treatments were focused on hyperacusis as a comorbidity of tinnitus. As such, tinnitus treatments, such as CBT for tinnitus and TRT, were often offered. Evidence on this approach is limited as most studies focused on hyperacusis as a secondary complaint. However, positive effects on hyperacusis are shown, while the treatments were focused on the hearing loss [e.g. 44] or the tinnitus [e.g. 45].

In conclusion, 11,1 % of the patients who consult the ENT department with tinnitus and/or hyperacusis complaints were classified as hyperacusis patients. At presentation in the clinic, this group consisted of more women and hyperacusis patients reported a higher tinnitus and mental distress. While there is a considerable comorbidity between hyperacusis and tinnitus, the two symptoms should be further disentangled. An important implication for clinical practice is to pay special attention to patients with a primary complaint of hyperacusis, who may not find their way to the ENT department.

#### Statements and Declarations

No conflict of interest for any of the authors is applicable.

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396 Figure legends

397 **Fig. 1** Density plot with density contours around the individual data points of the total scores of the  
398 Tinnitus Functional Index (TFI) and Hyperacusis Questionnaire (HQ) scores in the sample ( $r = .40$ ,  $p <$   
399  $.001$ )

400 **Fig. 2** Boxplots representing the self-report questionnaires for patients with and without hyperacusis.  
401 The scores on the Tinnitus Functional Index (TFI), Visual Analogue Scale (VAS) for tinnitus loudness and  
402 Hospital Anxiety Depression Scale (HADS) subscales) were significantly different between the two  
403 groups ( $p < .001$ )

404 **Fig. 3** Boxplots representing the scores on the Hyperacusis Questionnaire (HQ) and Tinnitus Functional  
405 Index (TFI) in male and female patients. The HQ and total TFI score is higher in female patients  
406 compared to male patients ( $p < .01$ )