Pilot study on the role of somatic modulation in hyperacusis

Pilot Study on the Role of Somatic Modulation in Hyperacusis

ABSTRACT

Background: Hyperacusis is a reduced tolerance to sounds that often co-occurs with tinnitus. Both symptoms have convergent as well as divergent characteristics. Somatic modulation, changes in pitch or loudness during certain movements, is common in patients with a primary complaint of tinnitus. However, thus far, this is not documented in patients with hyperacusis.

Objective: This study aimed to examine the influence of somatic manoeuvres on the perception of external sounds in patients with a primary complaint of hyperacusis.

Methodology: In this prospective cross-sectional pilot study, 18 patients with a primary complaint of hyperacusis were recruited at the Tinnitus Treatment and Research Center Antwerp (TINTRA). While patients listened to a 1 kHz broadband noise of 30 dB sensation level, six neck manoeuvres (flexion, extension, lateroflexion left/right, traction and compression), three jaw manoeuvres (protrusion, laterotrusion left/right) and one placebo manoeuvre (hand on head) were performed. The primary outcome measure was the change in the perception of the presented sound in terms of loudness and intrusiveness between baseline and each modulation measured by a visual analogue scale (VAS).

Results: No overall significant changes were found, however individual results indicated that five patients presented a clinically relevant change of more than three points out of ten on VAS in terms of hyperacusis after at least one of the executed somatic manoeuvres.

Conclusion: This pilot study did not demonstrate an overall significant change in hyperacusis after somatic manoeuvres but does not rule out the possibility of somatic modulation in some hyperacusis patients.

Trial registration: The protocol of this prospective cross-sectional pilot study was registered on clinicaltrials.gov with registration number NCT04693819.

MEC approval: The Committee for Medical Ethics of the University Hospital Antwerp approved the study on the 2nd of November 2020 (file number: B3002020000192).

Keywords: Hyperacusis; somatic modulation; Tinnitus; Pilot study

1. INTRODUCTION

Hyperacusis can be defined as "a reduced tolerance to sounds that are perceived as normal to the majority of the population or were perceived as normal to the person before their onset of hyperacusis" (1). Due to a lack of consensus on the diagnostic criteria of hyperacusis, the reported prevalence in the general population varies widely from 0.2 to 17.2% (2). The prevalence is higher in women, adolescents, older adults and in people with hearing loss (2, 3). Moreover, women score, in general, higher on the hyperacusis questionnaire (HQ) in comparison to men, suggesting a higher hyperacusis severity (3). Many uncertainties concerning hyperacusis still need to be explored and clarified (4, 5). For example, research priorities are to further examine the prevalence in specific populations, to unravel the heterogeneity in clinical origins, both physical as well as psychological and to look into the existence of meaningful subtypes (4, 5).

The most prominent comorbidity is tinnitus, the perception of a sound without the presence of an external auditory stimulus (11-14). In the general adult population, 10-15% experience tinnitus (11), whereas 86% of hyperacusis patients report tinnitus. Vice versa, 40% of patients

with tinnitus as a primary complaint also experience hyperacusis (15, 16). The co-occurrence of hyperacusis and tinnitus is thought to be the result of a partial overlap in pathophysiological mechanisms due to the involvement of the central nervous system (1, 12, 13, 17). As a consequence, hyperacusis and tinnitus have often been investigated together rather than as distinct phenomena.

Hyperacusis and tinnitus demonstrate many convergent characteristics, but also some divergencies (5). One particular divergence of interest, that up till now has not yet been investigated, is the possibility of somatic modulation. In patients with tinnitus, somatic modulation during certain movements of the neck or jaw, so-called somatic manoeuvres, has been demonstrated in up to 83% (13, 18-20). More specifically, input from the somatosensory and somatomotor system, such as muscle contractions, mechanical pressure on myofascial trigger points or movements of the cervical spine or temporomandibular joint (TMJ), evoke or modulate the auditory percept in certain patients (18-21). The modulation can go both ways. Somatic manoeuvres may reduce as well as worsen the loudness and pitch of the experienced tinnitus. Neither of both is proven to occur more often (18). It is hypothesised that somatic influence on auditory perception may be due to somatosensory-auditory interactions within the central nervous system (19, 22).

Tinnitus management in patients who are responsive to somatic modulation can consist of therapies based upon stimulation of the somatosensory pathways through muscular training, electrostimulation or pharmaceutical approaches (24, 26, 27). Since it is clear that the presence of somatic modulation is partially decisive for the treatment approach for tinnitus, it might be of value to know if this somatosensory input also has an impact on hyperacusis

complaints. Clinical observations by one of the authors (SM) during the treatment of somatic tinnitus patients with hyperacusis revealed that the hyperacusis was the first symptom to disappear after physiotherapy treatment. Furthermore, hyperacusis was more prevalent in somatic tinnitus patients than in patients with other types of tinnitus (28).This raises the question if a form of somatic hyperacusis exists.

Somatic modulation of hyperacusis, although it has never been explored before, might have an essential impact on the treatment approach of these hyperacusis patients that can modulate the perception of the presented sound by use of neck or jaw movements. This prospective cross-sectional pilot study aimed to examine the influence of somatic manoeuvres on the perception of external sounds in patients with a primary complaint of hyperacusis in order to address the current lack of knowledge. The main objective of the somatic experiment was to analyse whether there was a change in hyperacusis after one or more somatic manoeuvres in comparison to the baseline measurement. In addition, secondary objectives were to identify which manoeuvres led to a change between baseline and manoeuvre and what were certain characteristics of patients experiencing a change.

2. METHODS

2.1 Participants

Eighteen adult patients (age 18 years or over) experiencing a primary complaint of hyperacusis were recruited via the Tinnitus Treatment and Research Center Antwerp (TINTRA) of the Ear Nose Throat (ENT) department of Antwerp University Hospital (UZA). Patients were allowed

to have secondary tinnitus, but if they had tinnitus as a primary complaint, they were excluded. Recruitment and data collection were all completed between December 2020 and August 2021.

2.2 Study design

This pilot study was conducted at a single point in time for a sample of eighteen participants. See Figure 1 for the trial course each participant fulfilled. The subsequent components of the study are discussed in further detail in the following paragraphs.

2.3 Baseline measurements

2.3.1 Pure tone audiometry

The audiologist conducted pure-tone audiometry according to the current clinical standards (International Organization for Standardization (ISO) 8253–1:2010) using a two-channel AC-40 audiometer (Interacoustics, Assens, Denmark) in a soundproof audiometric booth. As a transducer, a headphone was used to measure air conduction (AC) thresholds of frequencies ranging from 125 Hz to 8kHz for both the right and left ear. Bone conduction thresholds were determined within a range of 250 Hz to 4 kHz if the AC thresholds within this same range exceeded normal values of 20 dB hearing level (dB HL) in order to determine the type of hearing loss.

2.3.2 Questionnaires

All patients filled out the validated Dutch versions of the following questionnaires using a touch-screen desktop.

The Hyperacusis Questionnaire (HQ) is a valid self-report questionnaire to ascertain hyperacusis complaints. HQ consists of 14 self-rating questions, which were scored on a four-point scale: 0= "no", 1= "a little", 2= "yes, quite a lot" and 3= "yes, a lot"(29, 30). A minimal score of 28 out of a total of 42 is traditionally used as a diagnostic criterion and represents a strong auditory hypersensitivity (29, 30). Higher scores indicate a greater reduction of tolerance to daily sounds (30).

If patients also experienced tinnitus, the Tinnitus Functional Index (TFI) was filled out. The TFI measures the impact and severity of tinnitus. Eight subscores are differentiated, namely intrusiveness, sense of control, cognitive complaints, sleep disturbance, auditory difficulties, relaxation, quality of life and emotional distress. TFI comprises 25 questions, each scored by a ten point-Likert scale. The total score, as well as the subscales, are noted on a scale of 0 to 100, with higher scores representing greater levels of tinnitus-related distress (31, 32).

The Hospital Anxiety and Depression Scale (HADS) is a self-report scale consisting of 14 items, with each item having four answer possibilities. HADS can be divided into two subscales of each seven items that screen for depression and anxiety symptoms. Patients with a minimal score of 8 out of a total of 21 on one or both subscales demonstrate signs of either depression and/or anxiety (33).

142 **2.4 Somatic Experiment**

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144 As well as the pure tone audiometry, the subsequent somatic experiment was performed in a
145 soundproof audiometric booth. The free-field thresholds for a 1 kHz broadband (BB) white
146 noise were measured in order to determine the loudness of the presented noise in the somatic
147 experiment.

148

149 At baseline, participants listened to a 1 kHz BB noise of 30 dB sensation level (dB SL) (i.e. 30
150 dB louder than the hearing threshold) for a duration of 15 seconds. The patients reported the
151 experienced loudness and intrusiveness of the 1kHz BB noise and, if tinnitus was present, the
152 loudness and intrusiveness of the tinnitus perception on visual analogue scales (VAS), which
153 is an instrument to measure intangible quantities (34). In this study, the primary intangible
154 quantities were the loudness and intrusiveness of the presented noise. The VAS comprises a
155 line of 10 cm on which patients had to mark their perception from zero to ten on paper (34).
156 Zero being "silent" or "not intrusive" and ten being "the worst imaginable". This was explained
157 to the patient before the experiment, and above each VAS, the instruction was repeated as a
158 reminder for the patient (34).

159

160 Subsequently, the modulation measurements were conducted. While the subjects listened to
161 the same 1kHz BB noise of 30dB SL for a duration of fifteen seconds, the physiotherapist
162 executed the somatic manoeuvres (see table 1). In random order, ten manoeuvres were
163 performed, of which six cervicogenic manoeuvres, three temporomandibular manoeuvres
164 and one placebo manoeuvre. To check for the presence of a placebo effect, the assessor
165 placed a hand on the patient's forehead while not performing any movement. All manoeuvres

were performed while the patient was in a sitting position. The manoeuvres were executed in the manner they are usually performed in case of the diagnosis of somatic tinnitus, following the protocol of Abel et al. (20). For further specifications on the execution of the manoeuvres, see Supplementary table A. Each manoeuvre was performed for fifteen seconds. Between manoeuvres, there was a break to fill out the VAS again, without auditory stimulation provided, and to receive the instructions for the next manoeuvre.

2.5 Outcome measures

2.5.1 Primary outcome measure: change in hyperacusis on VAS

The primary outcome was the change in the loudness and intrusiveness of the presented noise by means of a VAS with a range from zero to ten. A change in VAS of three between baseline and after modulation was considered to be a clinically relevant change (35).

2.5.2 Secondary outcome measure: change in tinnitus on VAS

The secondary outcome was the change in tinnitus loudness and intrusiveness. Patients who suffered from associated tinnitus did, additionally, need to fill out a VAS for these items. The VAS were constructed and read out in the same manner as the VAS for hyperacusis, discussed above.

2.6 Statistical methods

A non-parametric Wilcoxon ranking test to examine the presence of changes in VAS between baseline and after modulation was performed. The clinical relevance of the results was judged based on the visual graphical representation. All statistical analyses were conducted with IBM® SPSS® Statistics 28.

2.7 Ethics Committee Approval

Participants were informed about the study and were asked to provide informed consent. The Committee for Medical Ethics of the University Hospital Antwerp approved the study on the 2nd of November 2020 (file number: B3002020000192). All participants gave written informed consent prior to the start of the experiment.

2.8 Clinical Trial Registration

The protocol of this prospective cross-sectional pilot study was registered on clinicaltrials.gov with registration number NCT04693819.

3. RESULTS

In total, 18 adult subjects (median age = 47 years ; interquartile range (IQR) = 22.5 years) participated in this prospective, cross-sectional pilot study. The study sample existed of eight male and ten female participants.

3.1 Baseline measurements

All subjects suffered from hyperacusis as a primary complaint (median HQ score = 29; IQR =13.5). Fourteen patients, however, did also demonstrate tinnitus (median TFI-score = 42 ; IQR = 45.4). Nine subjects demonstrated signs of anxiety, of which five also showed signs of depression. One patient failed to fill out the HADS questionnaire due to technical difficulties and was, consequently, not considered for the HADS results. Specifications concerning these questionnaires and the pure tone audiometry can be found in Table 2.

3.2 Somatic experiment measurements

3.2.1 Baseline

With regards to hyperacusis, the median loudness and intrusiveness of the presented sound were respectively 5 and 4.8 at baseline (table 3). Fourteen patients also suffered from comorbid tinnitus as a secondary complaint. These patients did fill in two additional VAS scales concerning tinnitus loudness and tinnitus intrusiveness. The median VAS for tinnitus loudness and tinnitus intrusiveness was 2.3 and 1.8, respectively, at baseline (table 3).

3.2.2 Changes in VAS Hyperacusis

On a group level, no significant changes in loudness or intrusiveness of the presented sound were present during any of the somatic manoeuvres ($p>0.05$). However, five out of 18 participants did demonstrate a clinically relevant change between baseline and at least one of the manoeuvres for either loudness (patient 10), intrusiveness (patient 13) or both (patient 1, 7, 14). These changes were most common after traction (see figures 2A and 2B). For the intrusiveness of the presented sound, a clinically relevant change was also noted for multiple

participants after isometric right lateroflexion and isometric extension (see figures 3 and 4). It has to be noted that one participant (patient 13) demonstrated changes in VAS for the intrusiveness of the presented sound after all manoeuvres except for the placebo manoeuvre (see figure 5).

For information concerning the characteristics of the group of participants that perceived a clinically relevant modulation of their hyperacusis, see Table 4.

3.2.3 Secondary outcomes: tinnitus

No significant changes in tinnitus loudness or tinnitus intrusiveness were present after any somatic manoeuvre ($p>0.05$). However, five out of 14 participants with comorbid tinnitus did demonstrate a clinically relevant change of three points on the VAS for tinnitus loudness or intrusion after at least one of the manoeuvres (patients 16, 18) or both (patients 7, 8, 14). Two of them did also present clinically relevant changes in terms of hyperacusis (patient 7, 14), although this was not for the same manoeuvres except for traction. The changes in tinnitus were most common after traction, isometric left lateroflexion, isometric flexion and TMJ protrusion. It has to be noted that one patient (patient 14) demonstrated changes in tinnitus intrusiveness after all manoeuvres except for the placebo movement.

4. DISCUSSION

This pilot study examined whether hyperacusis can be modulated or evoked by one or more cervical or temporomandibular movements. In general, no statistically significant changes in hyperacusis were present, but five out of eighteen patients did experience a clinically relevant change of at least three points on the VAS for hyperacusis loudness or intrusiveness after at least one manoeuvre. Four patients experienced a clinically relevant change after the traction manoeuvre. A clinically relevant change in intrusiveness of the presented sound was also noted for multiple subjects after isometric right lateroflexion (three patients) and isometric extension (two patients).

The presence of somatosensory influence on the experience of hyperacusis can be explained by central interactions in the brain. Like tinnitus, hyperacusis is hypothesised to be caused by a central mechanism at the level of the central auditory system (36-39). The underlying

mechanism of hyperacusis is proposed to be due to a nonlinear central gain, while tinnitus is due to a linear central gain. The central gain model of hyperacusis suggests that maladaptive neuronal gain in the central auditory system leads to over-amplification of sound-evoked activity and, as a consequence, hyperacusis (36-39). Somatic modulation of tinnitus is caused by altered activity in the connecting fibres between the auditory and somatosensory nervous systems (40-43). This enables the somatosensory system to influence the auditory system (40, 41). Since hyperacusis, like tinnitus, occurs due to alterations at the level of the central auditory system (36-39), the presence of somatosensory influence on the experience of hyperacusis cannot be ruled out.

Hyperacusis also shows similarities with chronic centralisation pain (44). It is hypothesised that the auditory system consists of two parallel pathways: one direct pathway for auditory stimuli from the cochlea to the auditory cortex and one indirect pathway with connections to the dorsomedial nuclei in the thalamus receiving and processing all kinds of sensory input, including somatosensory input, and transmitting it to higher brain regions (44-46). This pathway is also known to play a role in central sensitisation phenomena in chronic pain disorders (44). Chronic pain with central sensitisation is known to be linked to not only an oversensitivity to somatosensory stimuli but also a hypersensitivity to sounds, bright light, tastes, et cetera (44). This central sensitisation mechanism can give rise to hyperacusis in certain pain syndromes by alterations in the sensory convergence pathway in the thalamus and altered activity in brainstem centres, such as the locus coeruleus (44). The amygdala and nucleus accumbens might also play a part in respectively the fear and emotional reactions linked to chronic pain. Likewise, emotional reactions and mental distress are very prevalent in hyperacusis (3, 7, 46-48).

The somatic modulation of loudness and intrusiveness of the presented sound observed in this pilot study were most common after traction. Traction is a passive manoeuvre used for pain relief in patients with cervicalgia, but it can also have a relieving effect on tinnitus complaints (49, 50). This might be due to the theory that traction causes relaxation of the cervical muscles (49). For the intrusiveness of the presented sound, a clinically relevant change was also noted for multiple subjects after isometric right lateroflexion and isometric extension. In this sample of hyperacusis patients, one patient experienced a decrease in intrusiveness of the presented sound and two patients experienced an increase after isometric extension and right lateroflexion.

The changes in tinnitus were additionally analysed to rule out the intermediate influence of tinnitus on hyperacusis during the somatic experiment. Only two of the five participants experiencing a clinically relevant change in hyperacusis had comorbid tinnitus. These two patients did also demonstrate a clinically relevant change in tinnitus, but not after the same manoeuvres except for traction. These results indicate that the presence of comorbid tinnitus cannot be used as a full explanation for the changes in hyperacusis loudness or intrusiveness since three out of five patients presenting somatic hyperacusis modulation solely suffered from hyperacusis. Additionally, this might indicate that the pathophysiological mechanism at the origin of the somatic modulation phenomenon in hyperacusis and in tinnitus may slightly differ.

Three patients indicated no intrusiveness for the presented sound (i.e. score of zero on the VAS). The presented sound was a BB noise of 30 dB SL, which might have been insufficient to elicit the hyperacusis complaints in some participants, but for some patients, this sound was already experienced as pungent. Individualisation of the presented sound, not only in terms of the loudness but also for the type of sound, might be a possible solution. Apart from the dosage of the sound, the dosage of the somatic manoeuvres should be explored further as well. In this pilot study, the manoeuvres were executed in the manner they are usually performed in testing for the presence of somatic tinnitus, following the protocol of Abel et al. (20). However, since tinnitus and hyperacusis aren't completely convergent, it might be an option to alter the dosage of the somatic manoeuvres in order to examine whether the effect would be more or less prevalent if the duration of the isometric contraction were held longer or shorter than 15s or if the contractions are performed submaximally instead of maximally for the patient or if any other parameter is changed. Furthermore, it needs to be explored whether the modulation effect on the hyperacusis complaints is solely present during the execution of the somatic manoeuvres or remains present afterwards and if this is the case , for what duration of time. This might give us valuable information in order to optimise the protocol specifically for hyperacusis patients.

Additionally, it would be recommended in future research to let participants fill out the Neck Bournemouth Questionnaire (NBQ) (51) in order to determine the presence of neck complaints and the Temporomandibular Disorder (TMD) pain Screener (52) to examine possible jaw complaints. Some clinical tests such as palpation of neck and jaw muscles and range of motion testing can be performed to estimate the presence of respectively increased

muscle tension or mobility deficits(53). It must also be noted that not all patients who can somatically modulate their tinnitus have somatic tinnitus according to the diagnostic criteria. Viceversa, not all patients diagnosed with somatic tinnitus are able to somatically modulate their tinnitus (54). Other diagnostic criteria appear to be more decisive in the diagnosis of somatic tinnitus, for example, simultaneous onset or increase/decrease of the tinnitus and neck or jaw complaints (55). Therefore, it might be of crucial importance to also take into account the other diagnostic criteria of somatic tinnitus when looking into the possibility of the existence of somatic hyperacusis.

Even though this exploratory study had a small sample and therefore lacked statistical power, results indicated that a great proportion of this small sample, specifically five out of a total of eighteen subjects, did experience a clinically relevant change in hyperacusis loudness or intrusiveness. The results of the current pilot study demonstrate the need for further research concerning the topic of somatic modulation in hyperacusis. If a somatic modulation phenomenon in hyperacusis can be demonstrated, great research opportunities present themselves. For example, looking into the pathophysiological mechanism behind somatic modulation in hyperacusis and what this tells us about the pathophysiological mechanism of hyperacusis in general. Examining whether there exists something like somatic hyperacusis and if this is indeed the case, what the indications are for hyperacusis treatment. The array of somatosensory-based therapies already proven to be effective in relieving tinnitus distress can then be explored to see if they are also useful in reducing hyperacusis complaints. Future research can also investigate if possible confounding factors such as anxiety, depression, gender, and presence of neck or jaw complaints, which are known to play a role in somatic tinnitus, are also of importance for somatic hyperacusis, if it does exist, and additionally if the

presence of comorbid tinnitus has an influence. Many more research questions are in need of an answer, and a somatosensory influence on hyperacusis might form a whole new research topic to dive into.

5. CONCLUSION

To conclude, this pilot study did not show significant changes in hyperacusis after somatic manoeuvres. However, clinically relevant changes in loudness and intrusiveness after somatic manoeuvres seemed to be present in five out of eighteen hyperacusis patients. Future research on a larger scale should aim to further look into possible clinically relevant changes in hyperacusis due to somatic modulation.

6. AUTHOR STATEMENT:

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