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Development and validation of Hyperacusis Handicap Questionnaire in individuals with tinnitus associated with hyperacusis

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ABSTRACT

Introduction: Hypersensitivity to sound and tinnitus are often co-morbid and can influence emotional well-being, hearing, sleep, concentration, cause anxiety, and interfere with speech perception in noise. *Purpose:* A clear measure of sensitivity to sound is important as there is dearth in standard protocol for evaluating hyperacusis in individuals with tinnitus. Although there are a few questionnaires to assess hyperacusis, a direct application of these questionnaires in the Indian context would be unfavorable. *Objectives:* The study attempts to develop and validate an indigenous Hyperacusis Handicap Questionnaire (HHQ) for individuals with tinnitus associated with hyperacusis.

Method: A total of 25 questions were considered for validation. Further, 21 questions were subdivided into three sections of seven questions each, tapping, 'Functional,' 'Social,' and 'Emotional' aspects of the condition. It was administered on 77 individuals with tinnitus associated with hyperacusis in the age range of 20–55 years for further validation. A total score was obtained by adding all the three sub-scales. *Results:* The internal consistency of the questionnaire was determined by Cronbach's Alpha (α) was $\alpha = 0.85$; and, $\alpha = 0.83$ for Functional, $\alpha = 0.81$ for Social, $\alpha = 0.7$ for Emotional subscales suggesting that the questionnaire can be used for the assessment of handicap associated with hyperacusis in individuals with tinnitus. Also, no significant difference in terms of gender and duration of tinnitus comparisons were seen.

Conclusions: The obtained results suggest that HHQ will aid in the characterization and quantification of the handicap associated with hyperacusis in individuals with tinnitus.

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1. Introduction

Tinnitus is the perception of ringing, buzzing, or whistling sounds in the absence of external auditory stimulation. If tinnitus lasts for more than six months, it is regarded as more or less chronic (Davis and El Refaie, 2000). Also, another common condition that causes pathological auditory hypersensitivity is termed as hyperacusis. Similar to tinnitus, hyperacusis also varies across individuals making it difficult to have a universal harmony of hyperacusis test data for the meaning, assessment, and interpretation (Brandy and Lynn, 1995). It was reported by Vernon (1987) that hyperacusis is

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an unusual tolerance to ordinary environmental sounds and, more critically, as 'consistently exaggerated or inappropriate responses to sounds that are neither threatening nor uncomfortably loud to a typical person' (Klein, Armstrong, Greer, & Brown III, 1990). Finally, the most current definition of hyperacusis given by Khalfa et al. (2002) refers to discomfort for sounds that would be acceptable to most normally hearing people. As hyperacusis appears to be a subjective phenomenon, it is not easily defined or quantified by objective measurements.

To craft a tool suitable in both quantification and evaluation of various hyperacusis symptoms, a questionnaire screening several aspects of auditory symptomatology has been constructed (Khalfa et al., 2002). Hyperacusis appears to be a subjective phenomenon, which is not easily defined or quantified by objective measurements. Anari and colleagues (Anari et al., 1999) suggested that uncomfortable loudness levels (ULLs) of 70 dB HL or less be used as a criterion for diagnosing loudness hyperacusis. It is different from recruitment, which is the abnormal growth of loudness in a

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damaged ear with significant hearing loss. Most hyperacusic patients have hearing within normal limits (Johnson, 1999). Moreover, hyperacusic patients are often obliged to wear hearing protective devices like earplugs/earmuffs (Preves et al., 1995), to avoid noise or other sounds causing discomfort. The advantage is that the loudness discomfort level is increased, but the threshold of hearing is also likely to be elevated.

Despite receiving identical sound stimulation levels, subjects with diminished sound-level tolerance (i.e., hyperacusis) show elevated activation in the auditory midbrain, thalamus, and primary auditory cortex compared with subjects with normal tolerance (Gu et al., 2019). The results directly link hyperacusis and tinnitus to hyperactivity within the central auditory system. Hypersensitivity to external sounds is often co-morbid with tinnitus and may be significant for adherence to certain types of tinnitus management (Fackrell et al., 2015). Based on the clinical experience of 4000 patients having subjective idiopathic tinnitus as their primary complaint, not every patient with tinnitus reported of hyperacusis. Similarly, not every patient with hyperacusis had tinnitus. Due to the lack of consistency between the two symptoms being presented together, it is important to explore whether or not there is a correlation between the two phenomena (Goldstein and Shulman, 1996). Therefore, a clear measure of sensitivity to sound is important as there is dearth in standard protocol for evaluating hyperacusis in individuals with tinnitus. Although there are a few questionnaires to assess hyperacusis, a direct application of these questionnaires in the Indian context would be unfavorable. The study attempts to develop and validate an indigenous Hyperacusis Handicap Ouestionnaire (HHO) for individuals with tinnitus associated with hyperacusis. The main objectives of the study includes developing a questionnaire mainly focusing on the India population considering the cultural diversity, lifestyle, and the levels of noise exposure (as individuals residing in different regions have different levels of noise exposure in turn affecting the tolerance levels, finding the internal consistency and average item-total correlation of the subscales and total HHQ scores, comparing the results of HHQ questionnaire in terms of gender and duration of tinnitus, and analysing the correlation between HHQ subscale scores with HHQ total scores as well as Tinnitus Handicap Inventory scores (THI).

2. Method

The HHQ was developed considering the salient features of different questionnaires developed for the assessment of Hyperacusis. The standard and validated hyperacusis questionnaires such as German Questionnaire on Hypersensitivity to Sound (Bläsing et al., 2010), Multiple-Activity Scale for Hyperacusis (MASH) (Dauman and Bouscau-Faure, 2005) and Khalfa Hyperacusis Questionnaire (Khalfa et al., 2002) were used as a reference. Questions were framed considering if it is appropriate for an Indian context. Most of the questionnaires use a 4-point rating scale ranging from no (scoring 0 points), yes, a little (scoring 1 point), yes, quite a lot (scoring 2 points) to yes, a lot (scoring 3 points). A similar pattern was used while developing the questionnaire.

Validation: The developed questionnaire was provided to five experienced audiologists (at least ten years of experience) for validation of the questions. They were asked to rate the questions based on three parameters, being: relevance, grammar, and appropriateness of the questions. Based on their responses obtained from the audiologists, four questions were removed from the final HHQ, making it a total of 21 questions. Also, a few questions were revised and reframed based on the comments and responses obtained during the validation process. Further, the 21 questions of HHQ were divided into three sub-scales tapping, 'Functional,' 'Social' and 'Emotional' domains of the complaint. A 3

point rating scale was used wherein, 0 indicated 'Never,' 2 indicated 'Sometimes,' and four indicated 'Always.' For validation of a clinical population, the questionnaire was administered on 50 individuals with tinnitus associated with hyperacusis.

Participant Selection Criteria: Individuals with tinnitus present for at least six months in both the ears ranging from 20 to 55 years were considered for the study. The selected participants had a primary complaint of intolerance to sounds with hearing sensitivity within normal limits. Individuals were also investigated if they had any co-existing conditions resembling psychological issues associated with intolerance to sounds like fear or dislike to sounds to eliminate subjects with phonophobia and misophonia, respectively. The participants were excluded if they were subjected to recreational noise exposure as there are chances of having a highfrequency hearing loss, if they were under any medications for other ailments or were using any hearing protective devices, and if they had pulsatile tinnitus. An equal number of males and females were included in the study. The participants had completed at least their matriculation so that there were no variables as it was a selfrated questionnaire. They were proficient in reading English and understanding the questions. Those individuals with a history of conductive symptoms and neural pathology were excluded from the study.

Procedure: As the first step of the investigation, a detailed case history regarding the characteristics of tinnitus and intolerance to sound was taken. All the participants underwent pure-tone audiometry at octave frequencies between 250 Hz and 8000 Hz and speech audiometry testing. Further, immittance evaluation and otoacoustic emissions were carried out to rule out any conductive pathology and to check the functioning of outer hair cells. Individuals who met the inclusion criteria were further selected in the self-rating task for HHQ. The participants were provided with HHQ and were instructed regarding the rating scales. The scoring was done by adding the total from each sub-scale. A final score was obtained by summing the 'Functional, 'Social,' and 'Emotional' sub-scale scores. Also, THI was administered at the end of the session for the correlation of HHQ and THI scores.

Ethical consideration: In the present study, all the testing procedures were carried out using non-invasive techniques, adhering to the guidelines of the Ethics Approval Committee of the institute. All the procedures were explained to the patients, and informed consent was taken from all the participants of the study.

3. Results

The data analysis was carried out using the Statistical Package for Social Sciences [SPSS], software version 17. The internal consistency and average item-total correlation of the questionnaire were determined. The first analysis that checked the Cronbach's Alpha (α) for the internal consistency revealed a very good internal consistency with $\alpha = 0.85$. Further, the internal consistency for the three sub-scales was also determined. It was found that the subscales of HHQ also showed good internal consistency with $\alpha = 0.83$ for 'Functional', $\alpha = 0.81$ for 'Social', and $\alpha = 0.7$ for 'Emotional' domains.

The second analysis was to check the average item-total correlation for all the 21 questions taken together and also for each of the sub-scales. The average item-total correlation for the HHQ was found to be good with $\alpha = 0.6$. Also, the average item-total correlation for the three sub-scales was found to be $\alpha = 0.55$, $\alpha = 0.53$, and $\alpha = 0.52$ for 'Functional', 'Social', and 'Emotional' domains respectively. The result of Cronbach's alpha and item-total correlation is shown in Table 1. Overall, the results suggest that the questionnaire can be used for the assessment of handicap associated with hyperacusis in individuals with tinnitus.

Table 1

Cronbach's alpha and average item correlation values for the questionnaires and their subscales.

Measures	HHQ	HHQ sub-scales		
		Functional	Emotional	Social
Cronbach's alpha Average-item correlation	0.85 0.6	0.83 0.55	0.70 0.52	0.81 0.53

To test the hypothesis that males and females differ in the three sub-scales as well as the total HHQ score, the Mann-Whitney test was performed. The non-parametric test was done as the two groups were unequal in number [Male (N) = 41 and Female (N) = 36] and also, the Shapiro Wilk's test of normality indicated a non-normal distribution. The results indicated a significant difference Z = -2.22, p < 0.05 only for the Social sub-scale. There was no significant difference seen for Functional, Emotional and total HHQ scores Z = -1.19, Z = -1.21, and Z = -1.83 (p > 0.05) respectively between males and females. The figure representing the median and standard deviation for gender comparison is provided in Fig. 1(a).

To compare the differences seen in the total HHQ scores and the subscale scores among individuals with varying years of tinnitus perception, the Kruskal Wallis H test was conducted. The individuals with the complaint of tinnitus were divided into three groups as 0–6 months, 7–14 months, and >14 months for the analysis. Although, the mean scores for the three different groups showed difference in the total HHQ scores (0–6 months = 31.9, 7–14 months = 33.9, and >14 months = 28.75), the Kruskal-Wallis test indicated no significant difference for any of the sub-scales and total scores in HHQ with, $\chi^2(2) = 1.63$, p > 0.05 for 'Functional sub-scale'; $\chi^2(2) = 2.22$, p > 0.05 for 'Social sub-scale', $\chi^2(2) = 1.53$, p > 0.05 for total HHQ scores among any of the groups compared. The figure representing the median and standard deviation for the comparison of duration of tinnitus is provided in Fig. 1(b).

Further, a correlation analysis was carried out to check the relationship between the total HHQ scores along with the subscales with that of the THI scores obtained. The results of Spearman's correlation suggested a good correlation of THI scores with the total and sub-scale scores of HHQ. The results of the same are depicted in Table 2. A strong correlation was seen between HHQ scores and its sub-scales.

Table 2

Table representing the correlation of THI scores with the total and sub-scale scores of HHQ.

	Functional	Social	Emotional	HHQ	THI
Functional		0.84**	0.61**	0.92**	0.64**
Social	0.83**		0.56**	0.89**	0.72**
Emotional	0.60**	0.56**		0.77**	0.75**
HHQ	0.91**	0.87**	0.75**		0.78**
THI	0.65**	0.74**	0.72**	0.79**	

Note. ** indicates significant correlation at p < 0.01.

4. Discussion

Hyperacusis and tinnitus are a highly subjective phenomenon, and hence, it is essential to develop measures native to the region so that a considerably larger population can be assessed. Also, it is important to develop questionnaires appropriate to the culture and social background. Considering all the factors mentioned above, the present study attempted to develop and validate the HHQ among the native population.

The first objective of the study was to characterize the internal consistency and the average item-total correlation, which after the analysis, showed a good consistency for the sub-scales as well as the total HHQ score. It indicated a good internal consistency as well as average item-total correlation, suggesting that the HHQ is a useful tool in the assessment procedure of hyperacusis in individuals with tinnitus. Also in comparison with other questionnaires like the Khalfa Hyperacusis questionnaire which had an internal consistency of $\alpha = 0.66$ for 'Attention', $\alpha = 0.68$ for 'Social', and V = 0.67 for 'Emotional' dimensions; and, G Ü F having $\alpha = 0.92$ with no large differences for the subscales; $\alpha = 0.82$ for 'Actional/ somatic behavior', $\alpha = 0.83$ for 'Emotional reaction to external Noises' and, $\alpha = 0.81$ for 'Cognitive reactions to Hyperacusis', the HHQ had comparable scores.

The second objective was to check the gender effects seen on different sub-scales of HHQ. This analysis was taken up to screen and check whether different aspects of hyperacusis that are functional, emotional, and social have different reactions by males and females. It was found from the analysis that there was no significant difference among males and females in any of the dimensions of hyperacusis, suggesting comparable reactions by both the genders. However, previous studies have reported



Fig. 1. The figure represents the median and SD value subscale and total HHQ scores for gender comparison 1(a) and the comparison of duration of hyperacusis 1(b).

gender differences in the case of tinnitus. One such study was done by Seydel et al. (2013), wherein the analysis for gender differences was carried out using pre-therapeutic scores for different aspects like annoyance due to tinnitus, stress, and proactive coping strategies and so on. Besides, the effects of these aspects on age and duration of tinnitus were also analyzed. It was found that irrespective of the duration of tinnitus and age, the annovance due to tinnitus and perceived stress was prominent in women when compared to men. Also, a lower score was secured for women in proactive coping strategies, sense of coherence, and personal resources. Although there was a gender difference in tinnitus-related distress, these could vary in terms of age and duration of tinnitus. The importance of addressing these differences results in tailor-made therapy approaches. For instance, stress management might be a better option for younger patients, and physical exercise or relaxation techniques would be of help in older women. However, in the present study, there was no significant difference in terms of gender. As both tinnitus and hyperacusis are a highly variable phenomenon, it is possible to arrive at results that vary from one population to another.

The comparison of differences seen in the total HHQ scores and the subscale scores among individuals with varying years of tinnitus perception being the third objective revealed no significant difference. Most of the studies done in individuals with tinnitus have not mentioned the possible reason for differences in scores for the hyperacusis questionnaires. However, in the present study, although there was no significant difference observed, the mean HHQ total scores indicated a slightly higher score for individuals with duration ranging between 7 and 14 months. One possible reason for this could be that individuals with hyperacusis and tinnitus might be unaware of the condition being present and might try to overcome it. Nevertheless, the condition might trigger their somatic senses and hinder the routine functioning later on (7-14 months), leading to higher scores in HHQ. During the later stages (>14 months), the person might get habituated with the condition. However, this is suggested as one of the reasons and may vary among individuals. The participants of the study also provided deviant responses, and hence, further studies dealing with a duration of hyperacusis and its effects might answer this hypothesis.

The last objective was to correlate the scores obtained in THI with the total score and subscale scores of HHQ. The results revealed a moderate to strong correlation. It is of great importance to analyze this aspect because tinnitus and hyperacusis are comorbid conditions. There is an apparent association between hyperacusis and tinnitus, wherein 86% of hyperacusis patients are also presented with tinnitus (Anari et al., 1999) and 40% of patients with a primary complaint of tinnitus experience hyperacusis as well (Jastreboff and Jastreboff, 2000). Hence, a good correlation between hyperacusis and tinnitus measures aids in a better understanding of the everyday problems faced by individuals who have a co-morbid condition. Overall, the HHQ having good internal consistency and strong correlation with the THI is a serves as a tool in assessing hyperacusis associated with tinnitus in different dimensions.

5. Summary and conclusion

The study aimed to develop and validate the Hyperacusis Handicap Questionnaire in individuals with hyperacusis associated with tinnitus. The developed HHQ exhibited a good internal consistency and average item-total correlation, which was comparable with the other hyperacusis questionnaires. Also, a moderate to strong correlation was seen between the THI scores and the subscale scores of HHQ along with its total score. However, there was no significant difference seen when the analysis was done in terms of gender and duration of hyperacusis. The results obtained from this study further states that HHQ is a validated tool in the assessment of this co-morbid condition.

5.1. Limitations of the study and future directions

The lack of consistency among studies indicates the necessity of examining the epidemiology of tinnitus and hyperacusis in children and adolescents with a set of standardized criteria. Also, a detailed study among different populations, including children, adults, and older adults, will serve beneficial in arriving at normative ranges for different age groups is necessary. Further research is required for the validation of HHQ on a larger population for better usage at the clinical set-up. Studies designed to check and validate the HHQ in routine audiological evaluations are required to further comment upon its practice.

5.2. Implications of the study

The major implication of having a questionnaire is that it helps in the characterization and quantification of the handicap associated with hyperacusis in individuals with tinnitus. This plays a major role in the management and counseling of these individuals. The HHQ helps in assessing the post-therapy outcome measures. Along with the outcome measures, the patients can also be detailed about the improvement from the treatment for these conditions.

Declaration of competing interest

There is no conflict of interest to disclose.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.joto.2019.12.004.

Appendix

Hyperacusis Handicap Questionnaire (HHQ).

N	lever	Sometimes	Always
	0)	(2)	(4)

How often do you prefer covering your ears for certain sounds or to decrease the level of sound? 1. How often do you feel uncomfortable reading or performing tasks in a noisy environment? 2.

3. How often do you face problem in concentrating on any task due to the intolerance to sound? (continued)

•				
		Never (0)	Sometimes (2)	Always (4)
4.	How often do you think that your routine, as well as work related performance, has decreased due to intolerance to sound?			
5.	How often have you felt that you cannot enjoy music because of intolerance to sound?			
6.	How often do you find it difficult to listen for a longer duration when surrounded by many sounds?			

- 7. How often do you feel difficulty in listening to music using earphones or headphones?
- 8. How often do you face tolerance problems while conversing in a noisy situation?
- 9. How often do you feel that certain sounds bother you more or cause difficulty while conversing?
- 10. How often do you avoid doing a certain task or going out because you have to be in a noisy place/situation?
- 11. Have you ever felt isolated among a group of people due to intolerance to sound (e.g. Party/other functions?
- 12. How often people tell you that you cannot tolerate sounds or your tolerance level for certain types of sound are very less?
- 13. How often do you prefer staying in-door because you feel that you might have to face loud sounds outside?
- 14. How often have you felt like changing your workplace because of excessive sound?
- 15. How often do you feel sad that you cannot tolerate certain sounds like traffic noise?
- 16. How often do you feel that a noisy place brings more stress and irritation?
- 17. How often do you get angry when you are surrounded by sounds?
- 18. How often are you scared of any particular sound?
- 19. How often have you faced emotional problems due to intolerance to sound?
- 20. How often do you feel disappointed due to the face that intolerance to sound is affecting your relationship with family and friends?
- 21. How often do you feel irritated because of sounds?

Total Score

*Questions 1 to 7 tap 'Functional handicap'; 8 to 14 tap 'Social handicap'; and 15 to 21 tap 'Emotional handicap' of the client. Sub-scores for each domain can also be obtained by adding the scores of respective questions falling in each category. **Sub-scores**:

Functional-	Social-	Emotional-

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