Development of AIISH Hyperacusis Assessment Toolbox for individuals with tinnitus associated with hyperacusis

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ABSTRACT

Hyperacusis is an auditory hypersensitivity disorder characterized by a heightened perception of the loudness or annoyance level of sounds. Sounds in the moderate to the intense range are perceived as intolerably loud or even painful by patients with hyperacusis. Hypersensitivity to sound and tinnitus (perception of sound or noise in the absence of any external acoustic stimulation) are often co-morbid.

There is no standard protocol for evaluating hyperacusis in individuals with tinnitus. There are few questionnaires to assess hyperacusis related disability and measure of grading hyperacusis which may not be directly applicable to the Indian context. A clear measure of sensitivity to sound is important as there is dearth in standard protocol for evaluating hyperacusis in individuals with tinnitus. Thus, the present study attempts to develop AIISH Hyperacusis Assessment Toolbox for individuals with tinnitus associated with hyperacusis.

As a part of the study, AIISH Hyperacusis Handicap questionnaire (AIISH-HHQ) and AIISH Hyperacusis Assessment Quotient (AIISH-HAQ) for a detailed evaluation of individuals with tinnitus associated with hyperacusis were developed. The developed questionnaire was validated and was further administered on 50 individuals with hyperacusis associated with tinnitus. In addition, it was also attempted to correlate the results of hyperacusis measurements with scores on tinnitus assessment to understand its relationship.

The results of the present study showed that the internal consistency of the questionnaire as determined by Cronbach's Alpha (α) was α = 0.86; and, α = 0.81 for Functional, α = 0.81 for Social, α = 0.69 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. The comparisons between the AIISH Hyperacusis Quotient (HAQ) and the Johnson's Hyperacusis Quotient (JHQ) provided a significant difference among the scores with minimum mean scores for narrow-band noise HAQ (mean = 75.71) and the maximum mean score for JHQ (mean = 78.5). Further, correlation analysis among total AIISH-HHQ scores showed a strong positive correlation with the sub-scale AIISH-HHQ scores and THI scores; and a moderate negative correlation with pure-tone and narrow band noise HAQ scores.

Thus, the study provides an indigenous validated clinical tool for the assessment of hyperacusis for the Indian population. The major implication of the questionnaire is that it helps in the characterization and quantification of the handicap associated with hyperacusis in individuals with tinnitus.

Keywords

Quality of life, Annoyance, Reduced tolerance, Hypersensitivity, Questionnaire

CHAPTER 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 & 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 & Lynn, 1995).

It was stated by Jepsen (1963) that hyperacusis is more often an abnormal sense of discomfort evoked by sounds above the hearing threshold. However, there are other conditions usually confused with hyperacusis like phonophobia (fear of sound) and misophonia (dislike of sound) wherein the major distinction is that the involvement of an emotional response to specific sounds (Jastreboff & Jastreboff, 2003). Jatrebboff and Jastreboff (2015) explained the various related conditions like hyperacusis, misophonia, diplacausis, and polyacousis in terms of nature of problem and the treatment options available for each of these conditions. It was put forth that all these conditions vary with respect to the neurophysiological processes and principles. It is estimated that the prevalence of hypersensitivity to sound in adults is approximately 8 to 15% (Fabijanska, Rogowski, Bartnik, & Skarzynski, 1999). These conditions can influence emotional well-being, concentration, hearing and sleep along with anxiety issues. The major concern for these individuals will be with the interference of speech perception in noise (Tyler et al., 2014). Being affected by many psychological issues it is estimated that about half of patients with hyperacusis also have a psychiatric or anxiety disorder (Jüris, Andersson, Larsen, & Ekselius, 2013).

Among the possible etiologies of hyperacusis are conditions involving the peripheral auditory system (e.g., Bell's palsy, Ramsay Hunt syndrome, noise-induced hearing loss, Ménière's disease), diseases and syndromes of central nervous system (e.g., headaches, depression, head injury, Williams's syndrome, learning disabilities, spinal problems), and hormonal (e.g., Addison's disease) and infectious diseases (e.g., Lyme disease). However, in most cases, hypersensitivity to sound has no known cause (Katzenell & Segal, 2001). However, clinical hyperacusis is a marked intolerance to sounds well below the threshold of hearing (Vernon, 1987). The term has also been used to denote a painful sensitiveness to sounds with no essential relationship between the threshold of discomfort and hearing. 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.

Hypersensitivity to sound and tinnitus (perception of sound or noise in the absence of any external acoustic stimulation (Jastreboff & Hazell, 2004) are often co-morbid. The prevalence of tinnitus among people with hypersensitivity to sound is much higher than in the general population and with estimates of 40% (Jastreboff & Jastreboff, 2000), 79% (Jüris et al., 2013), and 86% (Anari, Axelsson, Eliasson, & Magnusson, 1999a). Similar to tinnitus, several potential pathophysiological mechanisms might lead to hypersensitivity to sound and similar to tinnitus those mechanisms are not mutually exclusive.

Hypersensitivity to sound is described as a pre-tinnitus event as it very often precedes the onset of tinnitus (Jastreboff & Hazell, 1993). They postulated that hypersensitivity to sound is an effect of an increased gain in the central auditory

system and this is also been considered as one possible mechanisms of tinnitus generation. Also, the hypotheses that there is contribution of peripheral mechanisms in the generation of hyperacusis is made through the association between hypersensitivity to sound, tinnitus, and peripheral auditory system damage present in case of stapedectomy, Meniere's disease, and sensorineural hearing loss (Marriage & Barnes, 1995).

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, Halpin, Nam, Levine, & Melcher, 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, Fearnley, Hoare, & Sereda, 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 (Barbara Goldstein & Shulman, 1996).

To craft a tool suitable in both quantification and evaluation of various hyperacusis symptoms, questionnaires screening several aspects of auditory symptomatology are constructed (Khalfa et al., 2002, Bläsing et al, 2010, Dauman and Bouscau-Faure, 2005). Hyperacusis appears to be a subjective phenomenon, which is not easily defined or quantified by objective measurements. Anari and colleagues (Anari, Axelsson, Eliasson, & Magnusson, 1999b) 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 damaged ear with significant hearing loss. Most hyperacusic patients have hearing within normal limits (Johnson, 1999). It is different from recruitment, which is the abnormal growth of loudness in a damaged ear with significant hearing loss. Moreover, hyperacusic patients are often obliged to wear hearing protective devices like earplugs/earmuffs (Preves, Sammeth, Cutting, & Woodruff, 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.

Need for the study

There is no standard protocol for evaluating hypersensitivity to sound. The most common approach includes history taking and measuring uncomfortable loudness levels (ULLs) as a first step in the diagnosis. In people with hypersensitivity to sound ULLs are usually lower than average overall or specific frequencies in both or one ear. There are few measures developed such as Hyperacusis and Loudness discomfort test (Goldstein & Shulman, 1996) and Johnson Hyperacusis Quotient (Johnson, 1999) which can be used for measuring the degree of hyperacusis. However, these measures are not standardized and validated on a large group of population. Thus, there is a need to develop an indigenous assessment tool for grading the degree of hyperacusis in individuals with tinnitus.

Patient-reported outcome measures (questionnaires) are used to measure hypersensitivity to sound specific health-related quality of life and to diagnose hyperacusis. There are few standards and validated hyperacusis questionnaires such as German Questionnaire on Hypersensitivity to Sound (Bläsing, Goebel, Flötzinger, Berthold, & Kröner-Herwig, 2010), Multiple-Activity Scale for Hyperacusis (MASH)

(Dauman & Bouscau-Faure, 2005), and Khalfa Hyperacusis Questionnaire (Khalfa et al., 2002) used to assess hyperacusis related handicap. However, there are no standardized questionnaires developed for the Indian context.

The tolerance to loud sounds can vary across a person, region, and nature of environmental sounds. Thus, there is a need to develop and validate a hyperacusis questionnaire for the Indian population.

The present study aimed to develop a comprehensive AIISH Hyperacusis Assessment Toolbox for individuals with tinnitus associated with hyperacusis. The study developed the AIISH Hyperacusis Handicap questionnaire (AIISH-HHQ) and AIISH Hyperacusis Assessment Quotient (AIISH-HAQ) for a comprehensive assessment of individuals with tinnitus associated with hyperacusis.

Objectives of the study:

- To develop the AIISH Hyperacusis Handicap questionnaire (AIISH-HHQ) for assessment of handicap in individuals with tinnitus associated with hyperacusis.
- To develop AIISH Hyperacusis Assessment Quotient (AIISH-HAQ) for categorizing the degree of hyperacusis.
- To validate AIISH-HHQ and AIISH-HAQ on individuals with tinnitus associated with hyperacusis.
- To check the relationship of AIISH-HHQ and AIISH-HAQ with Tinnitus Handicap Questionnaire and results of tinnitus evaluation (Pitch matching and Intensity matching)

CHAPTER 2

METHODS

The study was carried out in three stages:

Stage 1: Development of the AIISH Hyperacusis handicap questionnaire (AIISH-HHQ)

Stage 2: Development of the AIISH Hyperacusis Assessment Quotient (AIISH-HAQ)

Stage 3: Administration of AIISH-HAT and AIISH-HAQ on individuals with tinnitus associated with hyperacusis.

Stage 1: Development of the AIISH Hyperacusis handicap questionnaire (AIISH-HHQ)

AIISH Hyperacusis handicap questionnaire 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 & 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. As 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) similar pattern was used while developing the questionnaire.

Validation of AIISH-HHQ

The developed questionnaire was provided to five experienced audiologists (with at least 10 years of experience) for validation of the questions. They were rated

based on relevance, grammar, and appropriateness of the questions. Based on their responses obtained from the audiologists, four questions were removed from the final AIISH-HHQ, making it a total of 21 questions. Also, few questions were revised and reframed based on the comments and responses obtained during the validation process. Further, the 21 questions of AIISH-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.'

Participant Selection criteria

The study comprised of 50 individuals with intolerance to sounds as well as tinnitus present for at least 6 months in both the ears. The participants age ranged between 20 to 55 years with mean age being 35.46 (SD = 6.72). The selected participants had hearing sensitivity within normal limits or had hearing loss not more than a mild degree. A total of 28 males (Mean age = 33.23, SD = 5.76) and 22 females (Mean age = 35.49, SD = 7.13) were included in the. The participants selected had a minimum education of matriculation as it was a self-rated 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. Also, 50 individuals without tinnitus with normal hearing sensitivity with the age ranging between 20 to 55 years (mean age = 33.5, SD = 7.21) were considered for the study and served as control group.

The inclusion criteria were being aged 20 years or older, the presence of tinnitus for 6 months or more and constant tinnitus. The exclusion criteria were: active illicit drug use or alcohol dependence, active ear infections, treatable cause of tinnitus, history of psychosis, abnormalities of the ear canal, subjects on medications known to

cause tinnitus (aspirin, ibuprofen, naproxen, etc) which could not be stopped, and currently under another sound or masking therapy for tinnitus.

Stage 2: Development of the AIISH Hyperacusis Assessment Quotient (AIISH-HAQ)

There are no standard measures for assessment of the degree of hyperacusis. There are few measures developed such as Hyperacusis and Loudness discomfort test (Barbara Goldstein & Shulman, 1996), and Johnson Hyperacusis Quotient (Johnson, 1999) which can be used for measuring the degree of hyperacusis. However, these are not standardized and cannot be directly applied to the Indian population. The salient features of these measures were considered to develop AIISH HAQ. The loudness discomfort levels were determined for pure tones and narrowband noise at octaves and mid-octaves from 250 Hz to 8000 Hz. Separate quotients for tones and narrowband noise was calculated and grading of hyperacusis was done as patients with tinnitus and hyperacusis may have more discomfort in the presence of noise apart from pure tone, which can further be used in finding if anyone of them was a better indicator than the other. Unlike JHQ which is calculated by averaging the dynamic range of pure-tones at seven frequencies (250 Hz, 500 Hz, 1000 Hz, 20000 Hz, 4000 Hz, 6000 Hz, and 8000 Hz), the AIISH-HAQ was calculated by averaging the dynamic ranges at 10 different frequencies (250 Hz, 500 Hz, 750 Hz, 1000 Hz, 1500 Hz, 20000 Hz, 3000 Hz, 4000 Hz, 6000 Hz, and 8000 Hz) for both pure-tones and narrow-band noise. As tinnitus and hyperacusis can be present in other frequencies apart from the octaves frequencies, additional frequencies were added in the evaluation and calculation of HAQ.

The main intention in developing AIISH-HAQ was to establish an indigenous assessment quotient by providing more emphasis on the speech spectrum as we come

across these frequencies more frequently. Hence, the frequencies 750 Hz, 1500 Hz, and 3000 Hz were added during averaging. Further, both pure-tone and NBN HAQ were obtained from participants and analyzed along with the JHQ to obtain a better assessment quotient.

A total of 50 individuals with age ranging between 20 to 55 years (mean age = 35.46, SD = 6.27). Individuals with normal hearing with no history of any conductive and neural symptoms were selected as control group (mean age = 33.5, SD = 7.21). The participant selection criteria for the experimental group are the same as mentioned in Stage 1.

Stage 3: Administration of AIISH-HHQ and AIISH-HAQ on individuals with tinnitus associated with hyperacusis.

As the first step of the investigation, a detailed case history regarding the characteristics of tinnitus and intolerance to sound was taken. Individuals with pulsatile tinnitus were excluded from the study. All the participants underwent pure-tone audiometry at octave frequencies between 250 Hz to 8000 Hz and speech audiometry testing. A calibrated dual-channel audiometer (Inventis Piano) was used to determine pure tone thresholds coupled to TDH 39 earphones with MX-41/AR ear cushions for octave frequencies between 250 to 8000 Hz to estimate the air conduction threshold and a bone vibrator (Radio ear B-71) for testing the bone conduction thresholds. The threshold was estimated using the modified Hughson and Westlake procedure (Carhart & Jerger, 1959) in a sound-treated room. After the estimation of pure tone thresholds, the speech recognition threshold (SRT) was determined. The speech identification scores (SIS) were determined at 40 dB SL (ref

SRT). Speech recognition thresholds were obtained using Kannada paired words and Speech Identification Scores (SIS) using Phonemically Balanced (PB) word lists in Kannada language (Yathiraj & Vijayalakshmi, 2005). 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 AIISH-HHQ. The AIISH-HHQ was administered where the client was told to read the questions and provide their responses on a rating scale. 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. AIISH-HHQ.

For AIISH HAQ, the pure tone and narrow band thresholds and uncomfortable levels (UCLs) were determined at octaves and mid-octaves from 250 Hz to 8000 Hz. All UCLs' tests were done giving exact instructions as recommended by the British Society of Audiology (BSA., 2011): I will gradually make the sound louder in your ear, and you must raise your hand as soon as the sound becomes uncomfortably loud. This is not a test to find the loudest sound you can tolerate; it is a test to find what level of sound you find uncomfortable. You should raise your hand only when the sound becomes uncomfortable, but make sure you raise it as soon as the sound reaches that level.

In addition, tinnitus evaluation was done to determine the pitch and intensity of the tinnitus using the procedure recommended by Goldstein and Schulman (1981). Tinnitus handicap inventory (THI) (Newman, Jacobson, & Spitzer, 1996) was administered on all the participants of the experimental group. An attempt was made to determine the correlation between THI score, pitch, and intensity of tinnitus with scores obtained in AIISH-HHQ and HAQ.

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.

CHAPTER 3

RESULTS

The data analysis was carried out using the Statistical Package for Social Sciences [SPSS], software version 17.

Internal consistency and Item-total correlation

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 $\alpha = 0.86$. Further, the internal consistency for the three sub-scales was also determined. It was found that the sub-scales of AIISH-HHQ also showed good internal consistency with $\alpha = 0.81$ for 'Functional', $\alpha = 0.81$ for 'Social', and $\alpha = 0.69$ 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 AIISH-HHQ was found to be good with $\alpha = 0.59$. Also, the average item-total correlation for the three sub-scales was found to be $\alpha = 0.52$, $\alpha = 0.52$, and $\alpha = 0.53$ 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	AIISH-HHQ	AIISH-HHQ sub-scales		
		Functional	Emotional	Social
Cronbach's alpha	0.86	0.81	0.69	0.81
Average-item correlation	0.59	0.52	0.53	0.52

Gender comparisons for AIISH-HHQ sub-scales and total scores

The descriptive statistics for gender comparison is represented in Figure 1(a). To test the hypothesis that males and females differ in the three sub-scales as well as the total AIISH-HHQ score, the Mann-Whitney test was performed. The non-parametric test was done as the two groups were unequal in number [Male (N) = 28 and Female (N) = 22] and also, the Shapiro Wilk's test of normality indicated a non-normal distribution. The results indicated a significant difference, $Z = 2.17$, $p = 0.03$ only for the Social sub-scale. There was no significant difference seen for Functional, Emotional and total AIISH-HHQ scores, $Z = 1.16$, $Z = 1.16$, and $Z = 1.93$ ($p > 0.05$) respectively between males and females.

Duration of hyperacusis comparisons for AIISH-HHQ sub-scale and total scores

The figure representing descriptive statistics (mean & SD) for the comparison of duration of hyperacusis provided in Figure 1(b). To compare the differences seen in the total AIISH-HHQ scores and the subscale scores among individuals with varying years of hyperacusis perception, the Kruskal Wallis H test was conducted. The individuals with the complaint of tinnitus associated with hyperacusis were divided into three groups as 0-6 months (n = 18), 7-14 months (n = 15), and >14 months (n = 17) for the analysis. Although, the mean scores for the three different groups showed difference in the total AIISH-HHQ scores (0-6 months = 31.3, 7-14 months = 33.4,

and >14 months = 29.5, the Kruskal-Wallis test indicated no significant difference for any of the sub-scales and total scores in AIISH-HHQ with, $\chi^2(2) = 1.53$, $p > 0.05$ for 'Functional sub-scale'; $\chi^2(2) = 2.26$, $p > 0.05$ for 'Social sub-scale', $\chi^2(2) = 1.63$, $p > 0.05$ for 'Emotional sub-scale', and $\chi^2(2) = 1.52$, $p > 0.05$ for total AIISH-HHQ scores among any of the groups compared.

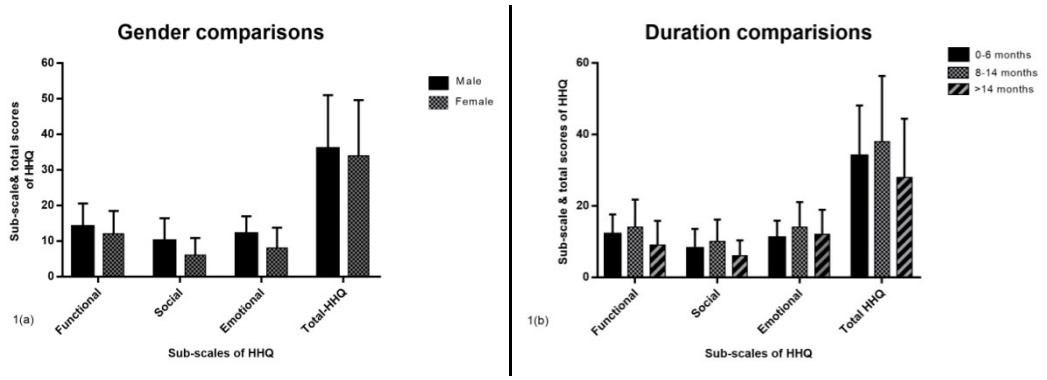

Figure 1. The figure represents the mean and SD values of sub-scale and total AIISH-HHQ scores for gender comparison 1(a) and the comparison of duration of hyperacusis 1(b).

AIISH-HAQ and JHQ comparisons

The descriptive statistics for AIISH-HAQ quotients for both pure-tone and narrow-band noise along with JHQ scores are depicted in Figure 2. The comparison between AIISH-HAQ quotients and JHQ was performed using Friedman's test. A non-parametric Friedman's test of differences among the three scores was conducted and rendered a $\chi^2(2) = 48.81$ which was significant ($p < 0.05$). Further, to test the differences between each pair of scores, the Wilcoxon signed-rank test was conducted. The test indicated that the JHQ score was significantly higher than the HAQ scores for pure-tones with $Z = 3.91$, $p < 0.05$. Similarly, the JHQ score was significantly higher than the AIISH-HAQ scores for narrow-band noise with $Z = 6.09$,

$p < 0.05$; and AIISH-HAQ scores for pure-tone was higher than AIISH-HAQ scores for narrow-band noise with $Z = 3.52$, $p < 0.05$. Thus, the scores of AIISH-HAQ and JHQ were found to be significantly different. The comparison between AIISH-HAQ and JHQ quotients was performed using Friedman's test for control group as well and the results depicted a significant difference, $\chi^2(2) = 69.71$, $p < 0.05$. Further, to test the differences between each pair of scores, the Wilcoxon signed-rank test was conducted. The test indicated a significant difference among all the compared groups, $Z = 6.10$, $Z = 2.39$, and $Z = 5.96$ ($p < 0.05$) for HAQ of pure tone vs HAQ of NBN, HAQ of pure-tone vs JHQ, and HAQ of NBN vs JHQ respectively.

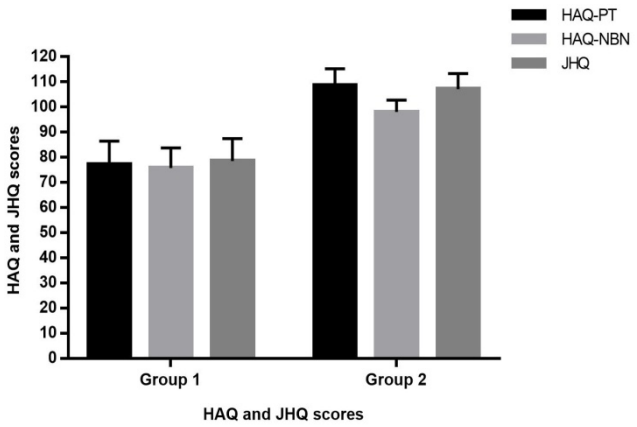

Figure 2. The figure represents the mean and SD values AIISH-HAQ scores for pure-tone and NBN; and JHQ scores. (Group 1 represents individuals with hyperacusis associated with tinnitus, and Group 2 represents individuals without tinnitus and hyperacusis).

In extension, the AIISH-HAQ quotients and the JHQ was compared between the control group that is individuals without tinnitus or hyperacusis and experimental groups that is individuals with hyperacusis associated with hyperacusis. The descriptive statistics depicting the mean and SD for both these groups is provided in

Figure 2. Mann-Whitney tests indicated a significant difference among all the scores compared and the same is represented in Table 2. It was found that the scores for all the quotients assessed indicated a higher mean score for individuals without hyperacusis and tinnitus (Group 2) when compared to individuals with hyperacusis and tinnitus (Group 1). The maximum difference between the two groups was seen using AIISH-HAQ for pure-tones followed by JHQ and HAQ for NBN.

Table 2. Table representing the results of Mann-Whitney test for the comparison of AIISH-HAQ and JHQ scores between groups

	AIISH-HAQ		**JHQ**
	Pure-tone	**NBN**	**Pure-tone**
Z	8.52*	8.92**	8.44**
p	< 0.01	< 0.01	< 0.01

Note. ** depicts significant difference

Gender comparisons for Pure-tone and NBN AIISH-HAQ scores

The descriptive statistics of pure-tones and NBN AIISH-HAQ scores for males and females is represented in Figure 3(a). To compare the gender differences in terms of AIISH-HAQ quotients, the Mann-Whitney test was performed. The results indicated no significant differences for both pure-tone and narrow-band noise AIISH-HAQ with $Z = 1.95$ and $Z = 1.03$ ($p > 0.05$) respectively. Although, the descriptive statistics provided a mean AIISH-HAQ score higher for both pure-tones and NBN in females, there was no statistically significant difference between the two genders.

Duration comparisons for Pure-tone and NBN AIISH-HAQ scores

The descriptive statistics of pure-tones and NBN AIISH-HAQ scores for different durations of hyperacusis is represented in Figure 3(b). To check whether

there is any significant difference between the duration of tinnitus and AIISH-HAQ for pure-tone and narrow band noise; Kruskal Wallis H tests were used. The results indicated no significant difference between duration of hyperacusis and AIISH-HAQ for both pure-tones and narrow-band noise with $\chi^2(2) = 0.58$, $p > 0.05$ and $\chi^2(2) = 1.78$, $p > 0.05$ respectively. Although, the descriptive statistics provided a mean AIISH-HAQ score higher for both pure-tones and NBN in females, there was no statistically significant difference between the two genders.

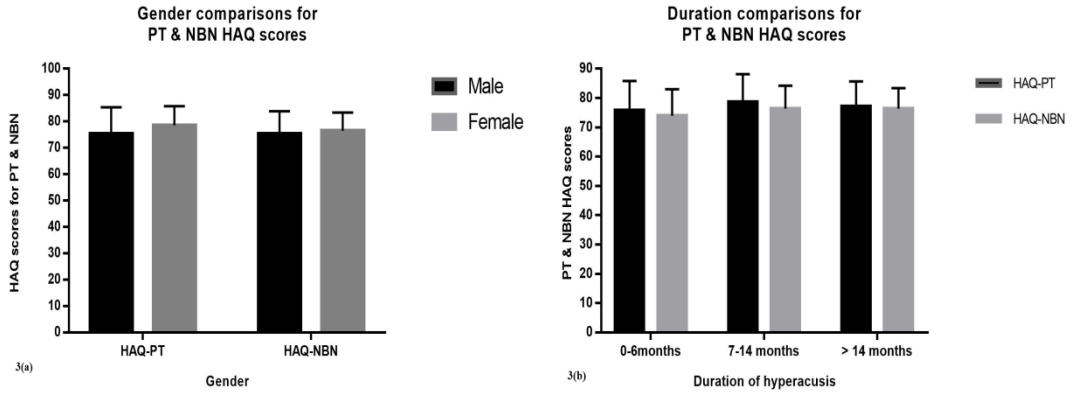

Figure 3. The figure represents the mean and SD values of pure-tone and NBN AIISH-HAQ scores for gender comparison 3(a) and the comparison of duration of hyperacusis 3(b).

Relationship between AIISH-HHQ and THI; and, AIISH-HAQ and HHQ

Further, a correlation analysis was carried out to check the relationship between the total AIISH-HHQ scores along with the sub-scales with that of the THI scores obtained. Spearman's correlation was used to check the relationship between the total and the sub-scales of HHQ, whereas, Kendal's Tau B was used to check the relationship between AIISH-HHQ and THI. The results of Kendal's Tau B correlation suggested a good correlation of THI scores with the total and sub-scale scores of AIISH-HHQ. The results of the same are depicted in Table 3. A strong correlation was seen between AIISH-HHQ scores and its sub-scales

Table 3. Table representing the correlation of THI scores with the total and sub-scale scores of AIISH-HHQ

	Functional	Social	Emotional	AIISH-HHQ	THI
Functional		0.84**	0.60**	0.92**	0.59**
Social	0.84**		0.54**	0.89**	0.66**
Emotional	0.60**	0.54**		0.76**	0.65**
AIISH-HHQ	0.92**	0.89**	0.76**		0.72**
THI	0.59**	0.66**	0.65**	0.72**	

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

To find the correlation between AIISH-HAQ scores for pure-tone and narrow-band noise with that of AIISH-HHQ total and sub-scale scores, Kendal's Tau B correlation test was conducted. The test findings depicted a moderate negative correlation among all the scores correlated and the same is represented in Table 4. Also, there was a negative correlation seen between both the AIISH-HAQ quotients and THI scores. However, the AIISH-HAQ quotients for both pure-tone and NBN were found to be not correlating with pitch and loudness of tinnitus. The results of all the correlation analyses performed are depicted in Table 4. It can be interpreted that there was moderate correlation with AIISH-HAQ scores for pure-tones and NBN with that of the total and subscale scores of AIISH-HHQ and THI. However, there was no correlation of the quotients with the tinnitus pitch and loudness.

Table 4. Table representing the correlation of AIISH-HAQ quotients with the total and sub-scale scores of AIISH-HHQ, THI scores, pitch and loudness of tinnitus

	Pure-tone HAQ	NBN-HAQ
Total AIISH-HHQ	-0.58**	-0.53**
Functional	-0.58**	-0.55**
Social	-0.52**	-0.46**

Emotional	-0.49**	-0.44**
THI	-0.33**	-0.42**
Tinnitus pitch	-0.17	-0.06
Tinnitus loudness	-0.18	-0.09

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

To summarize the obtained results, the AIISH-HHQ questionnaire demonstrated a good internal consistency and average item-total correlation for all the sub-scales. The gender comparisons for AIISH-HHQ indicated no significant difference among the total as well as the sub-scales. Although, the mean scores for the three different groups for duration of hyperacusis showed difference in the total AIISH-HHQ scores, the Kruskal-Wallis test indicated no significant difference for any of the sub-scales and total scores in AIISH-HHQ. Further, the comparison of AIISH-HAQ scores for pure-tone and NBN with JHQ scores indicated a significant difference with higher JHQ scores compared to the AIISH-HAQ scores. Also, the AIISH-HAQ and JHQ quotients provided a significant difference between individuals without and with hyperacusis and tinnitus. The maximum difference was present for pure-tone AIISH-HAQ scores and the minimum for NBN AIISH-HAQ scores. The gender and duration of hyperacusis comparisons for AIISH-HAQ scores indicated no significant difference. The correlation results of AIISH-HHQ total, sub-scale and THI scores indicated a strong correlation between them and a moderate correlation between AIISH-HAQ scores and AIISH-HHQ scores. However, there was no correlation seen between the AIISH-HAQ scores with tinnitus pitch and loudness.

CHAPTER 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 AIISH-HHQ and AIISH-HAQ among the native population.

Internal consistency and Average-item total correlation of AIISH- HHQ

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 AIISH-HHQ score. It indicated a good internal consistency as well as average item-total correlation, suggesting that the AIISH-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 behaviour', $\alpha = 0.83$ for 'Emotional reaction to external Noises' and, $\alpha = 0.81$ for 'Cognitive reactions to Hyperacusis', the HHQ had comparable scores.

Gender comparisons for AIISH-HHQ sub-scale and total scores

The next objective was to check the gender effects seen on different sub-scales of AIISH-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 except for social dimension of hyperacusis, suggesting comparable reactions by both the genders. This difference in social scale could be due to the fact that females express their discomfort more readily when compared to males and also there are more vulnerable to sounds (Khalfa et al., 2002). Besides, the role of hormone-related variability in sensory processing in women is discussed among the possible causes of hyperacusis with unknown etiology (Yilmaz, Taş, & Erdoğan, 2017). Literature has reported gender differences in the individuals with 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 annoyance 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. 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. Also, to check the tolerance level to sounds by males and females using AIISH-HAQ and JHQ scores, Mann-Whitney test was performed and revealed no significant difference between both the genders. The importance of addressing these

differences results in tailor-made therapy approaches. 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.

Duration of hyperacusis comparisons for AIISH- HHQ sub-scale and total scores

The comparison of differences seen in the total AIISH-HHQ scores and the subscale scores among individuals with varying years of hyperacusis perception 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 AIISH-HHQ total scores indicated a slightly higher score for individuals with duration ranging between 7-14 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 AIISH-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 duration of hyperacusis and its effects might answer this hypothesis.

AIISH-HAQ and JHQ comparisons

The comparison results of AIISH-HAQ and JHQ quotients revealed a significant difference for all the three comparisons that is pure-tone AIISH-HAQ scores and JHQ scores, NBN AIISH-HAQ scores and JHQ scores, as well as Pure-tone AIISH-HAQ scores and NBN-HAQ scores. The scores of JHQ were found to be higher followed by pure-tone HAQ and NBN-HAQ. It is known that sounds with same intensity, but with a different frequency will be perceived as having different loudness. The scores could be lowest for NBN as the perception of noise tends to be higher than the tones. Although the JHQ and pure-tone HAQ were assessed using tones, the seven frequency average in case of JHQ may provide a higher quotient than HAQ which is 10 frequency averages. If two sounds have the same intensity and their frequencies lie between about 600 and 2000 Hertz, they will be perceived to be about the same loudness. Outside of this range, that is not the case. For sounds near 3000 to 4000 Hertz, the ear is extra-sensitive; these sounds are perceived as being louder than a 1000 Hertz sound of the same intensity. As the AIISH-HAQ quotients made use of extra frequencies (750 Hz, 1500 Hz, and 3000 Hz) the 3000 Hz sound is perceived to be louder and hence the dynamic range reduces as the threshold is lower due to their extra sensitivity. This could be another possible reason for lower AIISH-HAQ scores compared to JHQ quotients.

Gender comparisons for Pure-tone HAQ and NBN-HAQ scores

Similar to the gender comparisons for AIISH-HHQ questionnaire, there was no significant difference among males and females in the AIISH-HAQ scores for pure-tone and NBN. Both the genders had similar or comparable AIISH-HAQ quotients suggesting the tolerance level dose not vary with gender. Also, the trend of higher AIISH-HAQ scores for pure-tone compared to NBN was followed in both the

genders. Although, a slight difference in terms of increased AIISH-HAQ scores in females was evident, there was no significant difference observed. The higher scores in females could be because females are more vulnerable to sound compared to men (Khalfa et al., 2002). Also, as mentioned earlier the variability in sensory processing due to hormone related changes could make them intolerable to sounds (Yilmaz et al., 2017).

Duration comparisons for Pure-tone HAQ and NBN-HAQ scores

The comparison of pure-tone HAQ and NBN-HAQ scores in terms of duration of hyperacusis perception revealed no significant difference. The mean score was higher for individuals in group with duration ranging between 7-14 months and similar scores for 0-6 months as well as >14 months group. However, the difference was very minimal and no significant difference was obtained. The minimal difference without any significance could be due to effects of habituation at later stages and lack of awareness in the early stages making it less obvious. However, the period intermediate to these two stages may be triggered by somatic senses and cause discomfort. A large scale study tapping the same may answer this hypothesis more clearly.

Correlations

The correlation results revealed a moderate to strong correlation. It is of great importance to analyze this aspect because tinnitus and hyperacusis are co-morbid conditions. There is an apparent association between hyperacusis and tinnitus, wherein 86% of adult patients come with the primary complaint of hyperacusis and secondary complaint of tinnitus (Anari, Axelsson, Eliasson, & Magnusson, 1999) and

40% of patients with a primary complaint of tinnitus experience hyperacusis as well (Jastreboff & 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 AIISH-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. Further correlation analysis of different aspects of hyperacusis and tinnitus performed revealed a moderate negative correlation between AIISH-HAQ score for pure-tone and NBN in comparison with AIISH-HHQ total and sub-scale scores. This analysis was considered to be important as both the expression of an individuals' difficulty using a questionnaire, along with the scores obtained on tests would determine the state of the condition. A moderate negative correlation shows that as the scores on HHQ questionnaire increased, the scores on the subjective tasks like AIISH-HAQ reduced. One of the major concerns was to check whether the patients experience discomfort due to hyperacusis is comparable to or correlating with the outcomes of audiometric data (HAQ). Hence, for individuals whose scores on AIISH-HHQ was low, the HAQ scores for both pure-tone and NBN were high indicating normal functioning without any tolerance problems. A study by Wallén, Hasson, Theorell, and Canlon (2012) reported significant correlations between the Hyperacusis Questionnaire (HQ) and ULLs in both ears for individuals with intermediate (right: −0.328; left: −0.320) and high Emotional Exhaustion (EE) (right: −0.349; left: −0.393), but not with low EE (right: −0.204; left: −0.196). Also, all the correlations were negative, representing that higher HQ scores are correlated with lower ULLs. The strongest correlations were found for the social dimension, indicating that social aspects may correspond best to audiological parameters (ULLs) of hyperacusis. The

different aspects of hyperacusis (Functional, Social, & Emotional) play a major role in describing the type of handicap or difficulty faced by an individual. According to Khalfa et al (2002) the total score of the HQ reflects the phenomenon of hyperacusis, while each subscale provides more clinically relevant information of how different aspects of the disorder affect the individual. Therefore, assessing different dimensions is important during the assessment. In the present study, the strongest correlation was evident for Functional followed by Social and Emotional aspects of hyperacusis. It should be mentioned that significant associations between self-reported hyperacusis and ULLs have been suggested in two previous studies of tinnitus patients (Goebel & Floezinger, 2008; Goldstein & Shulman, 1996). Goebel and Floetzinger (2008) found significant correlations between ULLs and the German questionnaire on hypersensitivity to sound. However the correlation coefficient was rather small ($r = -0.256$). In contrast to the results obtained in the present research, Meeus, Spaepen, Ridder, and Heyning (2010) found that there was no correlation between ULL or dynamic range and Hyperacusis Questionnaire or MASH suggesting incongruence between the complaints of hyperacusis and audiometric measures. A recent study on a similar study population did not find any correlations between ULLs and the German questionnaire on hypersensitivity to sound, implying that the association between this specific hyperacusis questionnaire and ULLs in tinnitus patients is uncertain (Bläsing, Goebel, Flötzinger, Berthold, & Kröner-Herwig, 2010). Goldstein and Shulman (1996) have also suggested association between self-reported hyperacusis and ULLs. However, there are several methodological differences between their study and the present that make it difficult to compare the results.

CHAPTER 6

SUMMARY AND CONCLUSIONS

The study aimed to develop and validate the Hyperacusis Handicap Questionnaire in individuals with hyperacusis associated with tinnitus. The developed AIISH-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 sub-scale scores of AIISH-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. No correlations were found between tinnitus pitch and loudness with that of AIISH-HAQ or HHQ. The results obtained from this study further states that AIISH-HHQ is a validated tool in the assessment of this co-morbid condition.

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 AIISH-HHQ on a larger population for better usage at the clinical set-up. Studies designed to check and validate the AIISH-HHQ in routine audiological evaluations are required to further comment upon its practice.

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 counselling of these individuals. The AIISH-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.

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APPENDIX

AIISH- Hyperacusis Handicap Questionnaire (AIISH- HHQ)

		Never (0)	Sometimes (2)	Always (4)
1.	How often do you prefer covering your ears for certain sounds or to decrease the level of sound?			
2.	How often do you feel uncomfortable in reading or performing tasks in a noisy environment?			
3.	How often do you face problem in concentrating on any task due to the intolerance to sound?			
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 there are certain sounds that bother you more or cause difficulty while conversing?			
10.	How often do you avoid doing certain task or going out due to the fact that 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 (eg. 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 work place 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-