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Psychometric Evaluation of the Hyperacusis Impact Questionnaire (HIQ) and Sound Sensitivity Symptoms Questionnaire (SSSQ) Using a Clinical Population of Adult Patients with Tinnitus Alone or Combined with Hyperacusis

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Abstract:

Background: Hyperacusis can be defined as an intolerance of certain everyday sounds, which are perceived as too loud or uncomfortable and which cause significant distress and impairment in the individual's day-to-day activities. It is important to assess symptoms of sound intolerance and their impact on the patient's life, so as to evaluate the need for treatment and to assess the effectiveness of treatments.

Purpose: To evaluate the psychometric properties of the Hyperacusis Impact Questionnaire (HIQ), and the Sound Sensitivity Symptoms Questionnaire (SSSQ). The 8-item HIQ focuses on assessing the impact of hyperacusis on the patient, while the 5-item SSSQ is designed to assess the type and severity of sound intolerance symptoms.

Research Design: This was a retrospective cross-sectional study.

Study Sample: 266 consecutive patients who attended a Tinnitus and Hyperacusis Therapy Clinic in the UK within a six-month period. 55% were female. The average age was 54 years (standard deviation = 16 years).

Data Collection and Analysis: Data were collected retrospectively from the records of patients held at the audiology department. Audiological measures were pure-tone audiometry and Uncomfortable Loudness Levels (ULLs). Questionnaires administered in addition to the HIQ and SSSQ were: Tinnitus Handicap Inventory, Hyperacusis Questionnaire (HQ), and Screening for Anxiety and Depression in Tinnitus.

Results: Exploratory factor analysis suggested one-factor solutions for both the HIQ and SSSQ. Multiple-causes multiple-indicators (MIMIC) models showed some small influences of gender but negligible effects of age for both the HIQ and SSSQ. ROC analysis showed no significant effects of covariates on the ROC curves. Cronbach's alpha was 0.93 for the HIQ, and 0.87 for the SSSQ, indicating high internal consistency. Convergent validity was supported by moderate correlations between HQ and HIQ scores and between SSSQ scores and ULLs.

Conclusions: The HIQ and SSSQ are internally consistent questionnaires that can be used in clinical and research settings.

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Abstract

Background: Hyperacusis can be defined as an intolerance of certain everyday sounds, which are perceived as too loud or uncomfortable and which cause significant distress and impairment in the individual's day-to-day activities. It is important to assess symptoms of sound intolerance and their impact on the patient's life, so as to evaluate the need for treatment and to assess the effectiveness of treatments.

Purpose: To evaluate the psychometric properties of the Hyperacusis Impact Questionnaire (HIQ), and the Sound Sensitivity Symptoms Questionnaire (SSSQ). The 8-item HIQ focuses

on assessing the impact of hyperacusis on the patient, while the 5-item SSSQ is designed to assess the type and severity of sound intolerance symptoms.

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Results: Exploratory factor analysis suggested one-factor solutions for both the HIQ and SSSQ. Multiple-causes multiple-indicators (MIMIC) models showed some small influences of gender but negligible effects of age for both the HIQ and SSSQ. ROC analysis showed no significant effects of covariates on the ROC curves. Cronbach's alpha was 0.93 for the HIQ, and 0.87 for the SSSQ, indicating high internal consistency. Convergent validity was supported by moderate correlations between HQ and HIQ scores and between SSSQ scores and ULLs.

Conclusions: The HIQ and SSSQ are internally consistent questionnaires that can be used in clinical and research settings.

Key words

Psychometrics

Questionnaire

Hyperacusis

tinnitus

Introduction

Hyperacusis can be defined as an intolerance of certain everyday sounds, which are perceived as too loud or uncomfortable and which cause significant distress and impairment in the individual's day-to-day activities. There are other definitions of hyperacusis but most of them are largely in agreement with the definition proposed here, with some differences in details (1, 2). Based on the definition proposed here, hyperacusis may be diagnosed if: (1) the patient's sound intolerance is related to the level of certain everyday sounds; (2) the patient experiences significant distress as a result of their sound intolerance; (3) sound intolerance significantly affects their day-to-day activities; (4) the patient's sound intolerance is not better explained by another disorder (e.g., hearing loss, post-traumatic stress disorder, depression, social phobia, or psychosis). This does not mean that hyperacusis cannot be present in combination with other disorders. However, it is important to determine whether the main source of sound-induced distress is hyperacusis, other disorders, or a combination of them.

It is important to assess symptoms of sound intolerance and their impact on the patient's life in order to assess the need for treatment and assess the improvement produced by a treatment. According to World Health Organization's International Classification of Functioning, Disability and Health, the severity of symptoms of a condition is different from the impact of the condition on the patient's life (3). Although the severity of symptoms is often correlated with the impact of the condition on the patient's life, they are not the same construct. For example, two individuals with the same moderate degree of hearing loss may experience different degrees of difficulty in their day-to-day life. There are various personal and environmental factors that can influence on the impact of an impairment on the patient's

life (4, 5). Therefore, it is important to be able to measure the severity of symptoms and impact separately, in order to guide the rehabilitation process.

Hyperacusis is often assessed using questionnaires. Validated questionnaires in English are the Hyperacusis Questionnaire (HQ) (6) and the Inventory of Hyperacusis Symptoms (IHS) (7, 8). The HQ has 14 items covering: the symptoms of hyperacusis and hearing loss, the types of sounds that can trigger hyperacusis, and the impact of hyperacusis on the patient's life. The IHS has 25 items that mainly focus on assessing the impact of hyperacusis on the patient's life, but some of the items assess the severity of symptoms of hyperacusis. Thus, total scores for both the HQ and the IHS reflect two different constructs, making it difficult to distinguish the severity and type of symptoms of hyperacusis from the impact of the hyperacusis.

The aim of this study was to evaluate the psychometric properties of two new short questionnaires that were developed for clinical use at the Tinnitus and Hyperacusis Therapy Specialist Clinic (THTSC), Royal Surrey Foundation Trust (RSFT), UK, namely the Hyperacusis Impact Questionnaire (HIQ) and the Sound Sensitivity Symptoms Questionnaire (SSSQ). The items in the HIQ were chosen to be largely consistent with some of items in the HQ and IHS that solely focus on assessing the impact of hyperacusis on the patient. In contrast, the SSSQ was designed to assess the type and severity of sound intolerance symptoms, based on the categories of hyperacusis described by Tyler et al (9).

Materials and methods

Ethical approval

The study was registered and approved as a clinical audit by the Quality Governance Department at the RSFT. Further analysis of the data was approved by the South West-

Cornwall and Plymouth Research Ethics Committee and the Research and Development department at the RSFT.

Study design and patients

This was a retrospective cross-sectional study conducted at the RSFT. Data were included for 226 consecutive patients who attended the THTSC within a six-month period and who had completed the HIQ and SSSQ. Demographic data for the patients, the results of their audiological investigations and their responses for other self-report questionnaires (described below) were imported from their records held at the Audiology Department. All questionnaires were completed prior to the start of any treatment, at each patient's first visit to the clinic. Patients completed the questionnaires in the clinic waiting area in pen and paper format without involvement of their audiologist.

Audiological measures

Audiological measures were:

(1) Pure-tone audiogram measured using the procedure recommended by the British Society of Audiology (10), but with some modifications proposed by Aazh and Moore (11) to avoid any discomfort. The severity of hearing loss was categorized based on the values of the pure-tone average (PTA) across the frequencies 0.25, 0.5, 1, 2, and 4 kHz, as recommended by the British Society of Audiology (10): Mild (20 – 40 dB HL), Moderate (41 – 70 dB HL), Severe (71 – 95 dB HL) and Profound (over 95 dB HL).

(2) Uncomfortable Loudness Levels (ULLs) measured following the BSA recommended procedure (12), but with the modifications proposed by Aazh and Moore (11), to avoid any discomfort. The across-frequency average (0.25, 0.5, 1, 2, 3, 4, 6, and 8 kHz) ULL for the ear

with lower average ULL is denoted ULLmin. When ULLmin was ≤ 77 dB HL, sound intolerance was deemed to be present (13).

Instruments and procedures

In addition to the new questionnaires, the HIQ and SSSQ (Tables 1 and 2), participants completed several validated questionnaires, namely the Tinnitus Handicap Inventory (THI) (14), the Hyperacusis Questionnaire (HQ) (15), and the Screening for Anxiety and Depression in Tinnitus (SAD-T) (16). As this was a retrospective study based on routine clinical data, not all of the patients completed all of the measures. The number of patients with complete data for each analysis is reported when appropriate. All questionnaires used are described below.

Hyperacusis Impact Questionnaire

The HIQ has eight items that ask respondents to rate how often (in number of days) over the past two weeks each of several situations occurred because of certain environmental sounds that seemed too loud to the respondent, but that other people could tolerate well. The items were developed based on common complaints of hyperacusis patients in the clinic and the items are broadly consistent with certain items from the HQ and IHS that exclusively focus on the impact of hyperacusis on the patient. Scores of 0, 1, 2, and 3 are assigned for frequencies of 0-1 days, 2-6 days, 7-10 days and 11-14 days, respectively. The response choices were chosen to be in keeping with response choices for commonly used psychological questionnaires in the UK's National Health Service, namely the Patient Health Questionnaire (PHQ-9)(17) and the Generalised Anxiety Disorder (GAD-7)(18). Those questionnaires also ask "over the last 2 weeks, how often have you been bothered by the following problems?". The response choices in the PHQ-9 and GAD-7 are "Not at all",

“Several Days”, “More than half the days”, and “Nearly every day”. We used 0-1 days, 2-6 days, 7-10 days and 11-14 days instead, in order to make the response choices more clear cut.

Sound Sensitivity Symptoms Questionnaire

The five-item SSSQ requires respondents to rate the number of days during the past two weeks that they experienced each of a number of symptoms, including loudness hyperacusis, pain or discomfort hyperacusis, annoyance hyperacusis/misophonia, and fear hyperacusis.

The method of scoring is the same as for the HIQ.

Hyperacusis Questionnaire

The HQ has 14-items, each rated on a 4-point Likert scale from “no” to “yes, a lot” (15). Cronbach’s alpha for the English version of the HQ is 0.88 (19). The overall score ranges from 0 to 42. Scores of 22 or more were taken as indicating the presence of hyperacusis (13).

Tinnitus Handicap Inventory

The THI has 25 items, each rated for its impact using a three-point scale: “yes” (4 points), “sometimes” (2 points) and “no” (0 points). Cronbach’s alpha for the THI is 0.93 (20). The overall score ranges from 0 to 100.

Screening for Anxiety and Depression in Tinnitus

The SAD-T contains four items that match those for the physical health questionnaire (PHQ-4; 21). Each item is rated on a four-point Likert scale. Two items relate to experiences of anxiety and worry and two items relate to the experience of anhedonia and feeling down, depressed or hopeless. Cronbach’s alpha for the SAD-T, based on responses from a tinnitus and hyperacusis clinical population, is 0.85. This was calculated but not reported during a study on the acceptability and relevance of psychological questionnaires in the assessment of

patients with tinnitus and/or hyperacusis (16). The overall score ranges from 0 to 12. Scores of 4 or more indicate symptoms of anxiety and/or depression.

Data analysis

Psychometric analysis

Latent variables for the HIQ and SSSQ were evaluated separately by exploratory factor analysis (EFA) for categorical items, using the weighted least squares mean and variance adjusted estimator (WLSMV) (22). Eigenvalues above 1 were used to identify the most suitable model fit (Guttman-Kaiser criterion) (23, 24).

To further test the number of factors to be retained for the HIQ and SSSQ, parallel analysis, based on minimum rank factor analysis (PA-MRFA), was conducted (25). To first check if it was appropriate to use polychoric correlation, the skewness and kurtosis for each item of the scale were calculated; values above one were considered to indicate that it was appropriate for polychoric correlation to be used (26, 27). The PA-MRFA method compares the real-data percentage of common variance and the 95th percentile of the random percentage of variance based on 500 PA-MRFA random datasets. For a dimension to be retained, the real-data percentage of common variance must be greater than the 95th percentile of the random percentage.

The identified factor structures were evaluated by measures of absolute and relative fit. Absolute fit measures were the relative chi-square (χ^2/df : values close to 2 indicate a close fit; 28), Root Mean Square Error of Approximation (RMSEA, values less than 0.05 are required for a close fit; 29), and the Standardized Root Mean Residual (SRMR: values below 0.05 suggest a close fit; 30). Two measures of relative fit were used, the Tucker-Lewis Index (TLI, values higher than 0.9 are required for a close fit; 31) and the Comparative Fit Index

(CFI, values higher than 0.95 are required for a close fit; 29). The range of communalities, i.e. the proportion of each variable's values that is explained by the latent factor, was calculated for all items; high communalities are required and low communalities (0.0-0.4) are indicative of items that may require removal (32).

Potential effects of gender and age were assessed using multiple indicator multiple cause (MIMIC) models (33, 34). MIMIC is a method for assessing if an item in a questionnaire is biased in the sense that responses to that item differ for individuals with different demographic characteristics (e.g., age or gender), irrespective of the differences in the severity of their hyperacusis symptoms or its impact on their lives (35). The standardized model coefficients for direct effects were considered; values below 0.36 are considered small in magnitude (36).

Logistic regression and Receiver Operating Characteristic (ROC) curves

Logistic regression models were used to assess the odds of hyperacusis having a significant impact (for the HIQ) and of experiencing sound sensitivity symptoms (for the SSSQ) based on a number of predictive measures. The analyses were based on Receiver Operating Characteristic (ROC) curves, which are plots of sensitivity (the proportion of positive cases that are classified as positive) against (1 - specificity) (where specificity is the proportion of negative cases that are classified as negative). Unfortunately, there are no widely accepted "gold standards" for assessing the impact of hyperacusis or for assessing the severity of systems of hyperacusis. Hence, the ROC analyses were based on imperfect standards, which we refer to in this paper as "bronze standards". For the HIQ, the bronze standard for classifying a case as positive (hyperacusis having a significant impact versus non-significant impact) was a score ≥ 22 on the HQ (13). For the SSSQ, the bronze standard for classifying a

case as positive (experience of abnormal sound sensitivity versus no abnormal sound sensitivity) was $ULL_{min} \leq 77$ dB HL (13).

Area under the ROC curve (AUC) values were assessed as excellent for values from 0.9 to 1, good for values from 0.8 to 0.9, and fair for values from 0.7 to 0.8. The Youden J (37) criterion was used to choose a cutoff point on each ROC curve giving an appropriate balance between sensitivity and specificity, where $J = \text{sensitivity} - (1 - \text{specificity})$. The Kappa value of Cohen (38) was used to assess the degree of agreement in classification based on scores for the new questionnaire compared with those for previously validated measures (39).

Before the optimal cut-off points for the HIQ and SSSQ were established, the Stata command *rocreg* (40, 41) was used to test for the presence of significant covariates affecting the ROC curve. The ROC curve can be influenced by covariates in two ways. First, a significant covariate can affect the discriminatory accuracy of a measure, i.e., the ability to discriminate between cases and controls at each level of a covariate. The presence of such covariates was assessed through testing of the null hypothesis that the covariate coefficient was equal to 0 (with probability $p \leq 0.05$), by bootstrapping to produce a 95% confidence interval of the covariate coefficient. Covariate-specific ROC curves are produced for each level of a significant covariate. For example, if gender was a significant covariate, then a male-specific ROC curve and a female-specific ROC curve would be produced. Second, the questionnaire score can be affected by a covariate, such as gender, as well as by the latent variable. In this case, significant covariates would be identified by a significant t-test result ($p \leq 0.05$). When significant covariates are identified, an ROC regression model with the covariates is used to provide a covariate-adjusted ROC curve.

Reliability and validity

The reliability of each scale was assessed using Cronbach's alpha (42), the value of alpha if an item is deleted (AID), and the item total correlation values (ITC; 43). To conclude that a measure is reliable, alpha is required to be greater than 0.7 and the AID lower than the alpha value.

Concurrent validity was established by calculating the correlations between HIQ/SSSQ scores and score for previously validated measure of hyperacusis, and also by comparing HIQ and SSSQ scores with ULLs. Discriminant validity was explored by calculating correlations between HIQ and SSSQ total scores and PTA values for the better ears. PTA values have been found not to be related to hyperacusis (13, 44, 45), so we expected small correlations of PTA values with HIQ and SSSQ scores. The non-parametric Spearman's correlation coefficient (ρ) was used to assess validity, due to the skewness of the data. A value of $\rho \geq 0.7$ is considered as strong, $0.4 \leq \rho \leq 0.6$ is considered as moderate, and $\rho \leq 0.3$ is considered as weak (46).

The Mplus software (47) was used for the EFA and MIMIC analysis, parallel analysis was carried out using a freeware program called FACTOR (48), and the statistical software SPSS version 25.0 (IBM 49) and STATA (version 13) (50) were used for the rest of the analysis.

Results

Sample characteristics

Of the 266 participants, 120 (45%) were male and 146 (55%) were female. Their ages ranged from 17 to 97 years with a mean of 54 years (standard deviation, SD = 16 years). The grand mean PTA across ears was 22.4 dB HL (SD = 15 dB) (n = 253). The PTA did not significantly differ between genders ($p = 0.5$). The grand mean PTA was 19 dB HL (SD = 13 dB) for the better ear and 26 dB HL (SD = 18.9 dB) for the worse ear. Based on the PTA for

the better ear, 64% of the participants had no hearing loss, 29% had mild hearing loss, and 7% had moderate hearing loss.

All patients had tinnitus. ULLmin values were 77 dB HL or below, indicating sound intolerance, for 30% of patients (60 out of 198 patients with available ULLmin values). The average value of ULLmin was 80.2 dB HL (SD = 6.9) for the men ($n = 84$) and 75.8 dB HL (SD = 10.4) for the women ($n = 114$), and this difference was significant ($t = 3.33$, $df = 196$, $p = 0.001$). The mean SAD-T score was 3.6 (SD = 3.8) for the men ($n = 117$) and 4.6 (SD = 4.2) for the women ($n = 142$) and the difference approached but did not reach significance ($p = 0.06$). Of 248 participants who answered the questions about their mental health history, 116 (47%) reported a past mental illness and 98 (40%) were under mental health care.

Psychometric analysis

Exploratory factor analysis

HIQ

EFA for the 8-item HIQ was based on scores for 232 participants and was carried out using Oblimin rotation. One eigenvalue above 1 (6.092) was produced by the sample correlation matrix, which according to the Guttman-Kaiser criterion supports a one-factor model for the data. The parallel analysis based on MRFA, using appropriate polychoric correlations, revealed one variable with real-data common variance greater than the 95th percentile variance, indicating the number of dimensions in the HIQ to be one. The goodness of fit indices for the one-factor solution indicated an adequate fit (relative $\chi^2 = 3.421$, RMSEA = 0.099 ($p = 0.001$), CFI = 0.99, TFI = 0.99 and SRMR = 0.05). Therefore, the one-factor solution was accepted. Table 3 shows the factor loadings for the one-factor solution. The

numbers refer to the specific items in the HIQ. The factor loadings were all greater than 0.77. The range of communalities for the HIQ was 0.59 to 0.81.

SSSQ

One eigenvalue above one (3.632) was produced by the sample correlation matrix. Parallel analysis based on MRFA, with polychoric correlations, indicated one dimension for the SSSQ. The goodness of fit indices for the one-factor model indicated an adequate but not excellent fit (relative $\chi^2 = 4.314$, RMSEA = 0.114 ($p = 0.014$), CFI = 0.99, TFI = 0.99 and SRMR = 0.05). However, no stable two-factor solution was found, so the one-factor solution was preferred. Table 3 shows the factor loadings for the one-factor solution. The factor loadings were all greater than 0.70. The range of communalities for the SSSQ was 0.49 to 0.85.

MIMIC

MIMIC models were fitted to assess possible effects of gender and age on the responses to individual items of the HIQ and SSSQ.

HIQ

Adjusted for age, and for a given level of the impact of hyperacusis, gender affected the expected scores for items HIQ3 (lack of concentration), HIQ4 (unable to relax), and HIQ8 (tire quickly) by 0.197, 0.238, and 0.282 points, respectively, on a scale from 0 to 3, with women giving higher scores. These effects are considered to be of small magnitude.

Adjusted for gender, significant effects of age were found for two items. The expected scores for HIQ3 and HIQ4 decreased with increasing age, but only by -0.008 and -0.009 units per year, respectively, which can be considered negligible.

In summary, there was a small effect of gender and a negligible effect of age.

SSSQ

Adjusted for age, for a given sound sensitivity score, the expected score for item SS1 (sounds “too loud”) was higher for women than for men by 0.306 units on the 0-3 scale. There were no effects of gender or age for the four remaining items. As there was an effect of gender for only one item, and the effect had a small magnitude, the overall score for the SSSQ can be considered as unaffected by gender or age.

Logistic regression

The odds of receiving a diagnosis of an impact of hyperacusis based on an HQ score ≥ 22 increased significantly with increasing HIQ score, each one-unit increase in HIQ score giving a 15% increase in the odds of a positive diagnosis of hyperacusis impact based on the HQ score (Table 4).

The odds of receiving a diagnosis of having abnormal sound sensitivity based on ULLmin ≤ 77 dB HL increased significantly with increasing SSSQ score, each one-unit increase in SSSQ score giving a 12% increase in the odds of a positive diagnosis of abnormal sound sensitivity based on ULLmin (Table 4).

ROC curves

Given that small effects of gender and age were found in the way that participants responded to certain items on the HIQ and SSSQ, ROC regression analysis was performed to determine whether covariate-adjusted or covariate-specific ROC curves should be created. The analysis showed that age and gender did not have significant covariate effects upon the ROC curves, on either their performance or its discriminatory ability, for the HIQ or SSSQ ($p > 0.05$ in all

cases). Thus, it was not necessary for covariate-adjusted or covariate-specific ROC curves to be created.

HIQ

Using HQ scores as a reference, the AUC for the HIQ was 0.77, which is considered as fair (Figure 1, Table 5). The highest value of J was 0.54 (Appendix 1, Table A1), and this occurred for a score of 11.5 on the HIQ, for which sensitivity was 0.74 and specificity was 0.80. Cohen's Kappa was moderate at 0.49 for agreement between diagnosis using the HIQ and HQ when a cut-off point of 11.5 was used (Table 6).

SSSQ

Using ULLmin as a reference, the AUC for the SSSQ was 0.67, which is considered as poor (Figure 2, Table 5). This probably partly reflects the fact that ULLmin scores are themselves imperfect indicators of sound sensitivity. The highest value of J [sensitivity - (1- specificity)] was only 0.268 (Appendix 1, Table A2), and this occurred for a score of 8 on the HIQ, for which sensitivity was 0.52 and specificity was 0.75. Cohen's Kappa was 0.27 for agreement between diagnosis using the SSSQ and using ULLmin when a cut-off point of 8 was used, which is only fair (Table 6). However, as can be seen in Figure 2, the ROC curve for the SSSQ is somewhat jagged. Assuming that the underlying distributions are normal, an AUC of 0.67 corresponds to a discriminability index, d' , of 0.511. For this value of d' , a better balance between sensitivity and specificity is obtained using a cutoff score of about 4.7, for which sensitivity is 0.60 and specificity is also 0.6. Since scores can only take integer values, in practice scores above 4 would be taken as indicating the presence of sound sensitivity. Cohen's Kappa for an SSSQ cut-off score of 4 was 0.22.

Reliability

Cronbach's alpha was 0.93 for the HIQ and 0.87 for the SSSQ, indicating good internal consistency for both questionnaires. No problematic items were found, since alpha did not increase when any single item was deleted and the ITC values were neither too high nor too low (range: HIQ, 0.64-0.82; SSSQ, 0.57-0.77).

The internal consistency of the SAD-T was replicated in this sample, with a satisfactory Cronbach's alpha of 0.91. Cronbach's alpha was not increased by the deletion of any item and the ITC values were between 0.76 and 0.84, which is satisfactory.

Validity

Age was not significantly correlated with HIQ score but it was significantly negatively correlated with SSSQ score (Table 7). Scores for the SAD-T and THI were moderately correlated with scores for the HIQ and the SSSQ, providing evidence for construct validity. Evidence for concurrent convergent validity was provided by moderate correlations between HIQ and SSSQ scores and HQ scores, as well as a moderate correlation between SSSQ scores and ULLmin values. The correlations between HIQ scores and ULLmin and PTA values of the better ear and between SSSQ scores and PTA values of the better ear were weak, indicating discriminant validity of the HIQ and SSSQ.

Discussion

In this study we evaluated the psychometric properties of the HIQ, which is designed to assess the impact of hyperacusis on a patient's life, and the SSSQ, which is designed to assess symptoms of abnormal sound sensitivity, in a clinical sample of patients with tinnitus of whom about 30% also had hyperacusis. Both questionnaires were found to be internally consistent, with Cronbach's alpha of 0.93 for the HIQ and 0.87 for the SSSQ.

EFA showed that both the HIQ and the SSSQ are one-factor questionnaires. The one-factor nature for the HIQ may indicate that the psychological and functional components of hyperacusis are not separable. This is consistent with the definition of hyperacusis, which is deemed to be present only if the sound intolerance causes significant distress in a patient's life, affecting their day-to-day activities and/or mood.

To the authors' knowledge the MIMIC model has not previously been used to evaluate any of the questionnaires available for assessment of hyperacusis. MIMIC is a method for assessing differential item functioning (DIF). When a significant DIF occurs for a specific questionnaire item, this indicates a bias for that item, in that responses to that item differ for individuals with different demographic characteristics (e.g., age or gender), irrespective of the differences on the variable under study (35). For example, DIF is present if men and women, or young and old people, or black and white people, with a given severity of the impact of hyperacusis respond differently to an item on a hyperacusis impact questionnaire. It is important to minimize such biases to ensure that a questionnaire is valid for a range of demographic characteristics.

In this study, a MIMIC model for the HIQ showed that for a given level of the impact of hyperacusis, after adjusting for age, gender affected the scores for items HIQ3 (lack of concentration), HIQ4 (unable to relax), and HIQ8 (tire quickly) by 0.197, 0.238, and 0.282 units, respectively, with women scoring higher. Similarly, for the SSSQ, for a given sound sensitivity, the score for item SS1 (sounds "too loud") was higher for women than for men by 0.306 units. These effects of gender may be related to the fact that on average women had significantly lower ULLmin values than men. Items similar to HIQ3, HIQ4 and HIQ8 are used in the IHS and the HQ, so it is important for future studies to explore if similar effects of gender occur for those questionnaires and how they can be minimized. Future studies using larger samples should also use the MIMIC model or other methods for testing DIF (35) to

assess whether there are biases for items in the HIQ and SSSQ linked to demographic factors that were not included in this study.

To sum up, there were minor effects of gender on scores for a few items of the HIQ and SSSQ. However, the ROC regression analyses showed that age and gender did not have significant covariate effects upon the ROC outcome, on either its performance or its discriminatory ability, for the HIQ or SSSQ. Thus, the scores are largely invariant with respect to age and gender. It is important that future studies explore the impact of measurement bias both in clinical practice and research.

The total scores for the HIQ and SSSQ were moderately correlated with total HQ score. This indicates that the HIQ and SSSQ are measuring related things to the HQ but are not measuring an identical construct. A strong correlation was not expected, because these new questionnaires were designed specifically to assess separately the impact of hyperacusis and severity of its symptoms, while the HQ assesses the severity of symptoms of hyperacusis combined with its impact. In addition some of the items of the HQ do not appear to assess hyperacusis-related constructs (19). For example, item 5 of the HQ is about “Difficulty listening to conversations in noise”, which is probably more related to hearing loss than to hyperacusis. In addition, scores for item 1 of the HQ “Use earplugs or earmuffs to reduce noise” are only weakly correlated with scores for other items of the HQ, indicating that scores for this item may not be related to hyperacusis or its impact (19). Scores for both the HIQ and the SSSQ were moderately correlated with scores for the SAD-T and THI. This shows that they are measuring different but related constructs. The close relationship between hyperacusis, tinnitus and mental health is well known (8, 51).

SSSQ scores were moderately correlated with ULLmin values but HIQ scores were only weakly correlated with ULLmin values. This is as expected, since the HIQ is not

intended to measure severity of the symptoms of hyperacusis, while ULLmin values do partly reflect the severity of hyperacusis. As expected, the correlations between HIQ and SSSQ scores and PTA values of the better ear were very small. This is consistent with past studies that suggest that hyperacusis is not related to audiometric thresholds (13, 15, 52).

As noted earlier, there is no widely accepted gold standard for assessment of the impact of hyperacusis. The HIQ was intended to remedy this gap by solely assessing the impact of hyperacusis, without dilution or contamination by other constructs such as hearing difficulty and severity of sound sensitivity symptoms. Therefore, we did not expect to obtain high sensitivity and specificity when using the HQ as a reference for determining a cut off value for the HIQ. Future studies should further examine how the severity of the impact of hyperacusis should be classified based on HIQ total score.

The SSSQ included items intended to assess different types of sound sensitivity, ranging from various forms of hyperacusis (items 1, 2, 3, and 5) to misophonia (item 4). Given that there is no gold standard for assessing the type or severity of the symptoms of hyperacusis, we used ULLmin values as reference for the ROC analysis of the SSSQ. There are several studies that suggest that ULLmin values ≤ 77 dB HL are associated with the experience of hyperacusis (13, 53, 54). ULLs may also be reduced among patients with misophonia, and Jastreboff and Jastreboff (55) reported that misophonia is almost always present in cases of severe hyperacusis. Severe hyperacusis is also characterized by reduced ULLs mainly at higher frequencies (56). So, it seemed reasonable here to use ULLmin values as a reference, even though they are an imperfect measure of the severity of sound sensitivity symptoms (57). Future studies should further explore how to classify sound sensitivity symptoms into different categories of severity based on the SSSQ total score.

Conclusions

In this paper we introduced two new brief questionnaires: the HIQ, which assesses the impact of hyperacusis on a patient's life, and the SSSQ, which assesses the types and severity of symptoms of sound intolerance. Both questionnaires were found to have high internal consistency. The HIQ and SSSQ can be used to assist audiologists and other clinicians in making a diagnosis of hyperacusis. A cutoff point of 11/24 for the HIQ gave a sensitivity of 0.74 and a specificity of 0.8 in the diagnosis of hyperacusis, using diagnosis based on HQ scores as a reference. For the SSSQ, a cut-off point of 4.7 gave a sensitivity of 0.60 and a specificity of 0.60 in diagnosing sound tolerance problems, using ULLmin values as a reference.

MIMIC models revealed biases for certain items in the HIQ and SSSQ, although the biases were small. These items have also been used in some other hyperacusis questionnaires. Therefore, future research should explore this further, not only for the questionnaires tested here but also for other commonly used hyperacusis questionnaires.

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Conflict of Interest

None declared.

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Figure 1 Receiver operating characteristic (ROC) curve for the hyperacusis impact questionnaire (HIQ). The area under the curve value was 0.77.

Figure 2 Receiver operating characteristic (ROC) curve for the sound sensitivities questionnaire (SSSQ). The area under the curve value was 0.67.

Table 1. Hyperacusis Impact Questionnaire (HIQ)

Please answer each item to the best of your ability as close to your experience as possible.				
Over the last 2 weeks, how often would you say each of the following has occurred because of certain environmental sounds that seemed too loud to you but that people around you could tolerate well?				
	0-1 day	2-6 days	7-10 days	11-14 days
1. Feeling anxious when hearing loud noises	0-1 day	2-6 days	7-10 days	11-14 days
2. Avoiding certain places because it is too noisy	0-1 day	2-6 days	7-10 days	11-14 days
3. Lack of concentration in noisy places	0-1 day	2-6 days	7-10 days	11-14 days
4. Unable to relax in noisy places	0-1 day	2-6 days	7-10 days	11-14 days
5. Difficulty in carrying out certain day-to-day activities/tasks in noisy places	0-1 day	2-6 days	7-10 days	11-14 days
6. Lack of enjoyment from leisure activities in noisy places	0-1 day	2-6 days	7-10 days	11-14 days
7. Experiencing low mood because of your intolerance to sound	0-1 day	2-6 days	7-10 days	11-14 days

8. Getting tired quickly in noisy places	0-1 day	2-6 days	7-10 days	11-14 days
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Table 2 Sound Sensitivity Symptoms Questionnaire (SSSQ)

Over the last 2 weeks, how often have you been bothered by any of the following problems?				
1. Having a problem tolerating sounds because they often seem “too loud” to you?	0-1 day	2-6 days	7-10 days	11-14 days
2. Pain in your ears when hearing certain loud sounds? Examples: loud music, sirens, motorcycles, building work, lawn mower, train stations.	0-1 day	2-6 days	7-10 days	11-14 days
3. Discomfort (physical sensations other than ear pain) in your ears when hearing certain loud sounds?	0-1 day	2-6 days	7-10 days	11-14 days
4. Feeling angry or anxious when hearing certain sounds related to eating noises, lip smacking, sniffing, breathing, clicking sounds, tapping?	0-1 day	2-6 days	7-10 days	11-14 days
5. Fear that certain sounds may make your hearing and/or tinnitus worse?	0-1 day	2-6 days	7-10 days	11-14 days

Table 3. Standardised factor loadings from the exploratory factor analysis of the HIQ and SSSQ

	Item	Factor 1
HIQ	HIQ4	0.901
	HIQ3	0.890
	HIQ5	0.890
	HIQ7	0.888
	HIQ6	0.886
	HIQ1	0.863
	HIQ2	0.772
	HIQ8	0.771
SSSQ	SS3 – discomfort	0.922
	SS2 – pain	0.881
	SS1 – tolerate	0.803
	SS5 – fear	0.770
	SS4 – misophonia	0.703

HIQ, Hyperacusis Impact Questionnaire, SSSQ, Sound Sensitivity Symptoms Questionnaire

Table 4. Odds ratios (OR) for the HIQ diagnosing an impact of hyperacusis (based on HQ score ≥ 22) and the SSSQ diagnosing abnormal sound sensitivity (based on ULLmin ≤ 77 dB HL) with standard errors in parentheses, together with *p* values and 95% confidence intervals (CI)

predictor	OR (SE)	p value	95% CI
HIQ	1.15 (0.05)	0.001	1.06 - 1.25
SSSQ	1.12 (0.04)	0.001	1.05 - 1.20

HIQ, Hyperacusis Impact Questionnaire, SSSQ, Sound Sensitivity Symptoms Questionnaire

Table 5. AUC values for the HIQ and SSSQ, with standard errors in parentheses, together with 95% confidence intervals (CI).

Measure	AUC (SE)	95% CI
Hyperacusis		
HIQ	0.77 (0.07)	0.64 - 0.91
SSSQ	0.67 (0.04)	0.59 - 0.75

HIQ, Hyperacusis Impact Questionnaire, SSSQ, Sound Sensitivity Symptoms Questionnaire

Table 6. Cohen's Kappa (with *p* values in parentheses) for diagnosis agreement using a cutoff score of 11.5 for the HIQ and cutoff scores of 4 and 8 for the SSSQ.

	Cohen's Kappa (<i>p</i>)	
	HQ	ULL category
HIQ	0.49 (<0.001)	
SSSQ (4)		0.22 (0.001)
SSSQ (8)		0.26 (<0.001)

HIQ, Hyperacusis Impact Questionnaire, SSSQ, Sound Sensitivity Symptoms Questionnaire

Table 7. Spearman's correlation coefficients (with *p* values in parentheses) between scores for the HIQ and SSSQ and other variables, specifically age, PTA of the better ear, scores for the SAD, HQ, and THI, ULLmin values and hyperacusis category based on ULLmin.

	HIQ		SSSQ	
Measure	n	ρ (<i>p</i>)	n	ρ (<i>p</i>)
Age	233	-0.07 (0.233)	245	-0.20 (0.001)
PTA of better ear	225	0.18 (0.008)	237	0.06 (0.33)
SAD-T	229	0.64 (<0.001)	243	0.47 (<0.001)
HQ	67	0.59 (<0.001)	71	0.68 (<0.001)
THI	66	0.54 (<0.001)	70	0.56 (<0.001)

Hyperacusis category based on ULLmin	175	0.19 (0.011)	184	0.27 (<0.001)
ULLmin	175	-0.22 (0.003)	184	-0.33 (<0.001)

Note. PTA, pure tone average across the frequencies 0.25, 0.5, 1, 2, and 4 kHz; SAD-T, Screening for Anxiety and Depression Questionnaire; HQ, Hyperacusis Questionnaire; THI: Tinnitus Handicap Inventory; SSSQ: Sound Sensitivity Symptoms questionnaire; HIQ: Hyperacusis Impact Questionnaire; HQ: Hyperacusis Questionnaire; ULLmin, Uncomfortable Loudness Level (ULL) averaged across the frequencies 0.25, 0.5, 1, 2, 3, 4, 6, and 8 kHz for the ear with lower average ULL; n, number of participants; p, probability value

Appendix.

Table A1. Co-ordinates of the ROC curve for the Hyperacusis Impact Questionnaire. The right-most column shows the values of the Youden J.

Cut-off point (\geq)	sensitivity	1 - specificity	J = sensitivity - (1-specificity)
-1.00	1.000	1.000	0.000
0.50	0.947	0.756	0.192
1.50	0.947	0.711	0.236
2.50	0.895	0.644	0.250
3.50	0.895	0.600	0.295
4.50	0.789	0.533	0.256
5.50	0.789	0.489	0.301
6.50	0.789	0.444	0.345
7.50	0.789	0.400	0.389
8.50	0.789	0.333	0.456
9.50	0.737	0.289	0.448
10.50	0.737	0.267	0.470
11.50	0.737	0.200	0.537
12.50	0.632	0.200	0.432
13.50	0.579	0.178	0.401
14.50	0.526	0.178	0.349
15.50	0.526	0.156	0.371
16.50	0.474	0.133	0.340
17.50	0.421	0.133	0.288
18.50	0.421	0.089	0.332
19.50	0.316	0.044	0.271
20.50	0.263	0.022	0.241
21.50	0.211	0.022	0.188
22.50	0.211	0.000	0.211
23.50	0.158	0.000	0.158
25.00	0.000	0.000	0.000

Table A2.

Coordinates of the ROC curve for the Sound Sensitivity Symptoms Questionnaire. The right-most column shows the values of the Youden J.

Cut-off point (\geq)	sensitivity	1 - specificity	J = sensitivity - (1-specificity)
≥ 0	1.000	1.000	0.000
≥ 1	0.946	0.742	0.204
≥ 2	0.875	0.656	0.219
≥ 3	0.786	0.539	0.247
≥ 4	0.714	0.453	0.261
≥ 5	0.589	0.398	0.191
≥ 6	0.571	0.328	0.243
≥ 7	0.518	0.273	0.245
≥ 8	0.518	0.250	0.268
≥ 9	0.429	0.219	0.210
≥ 10	0.268	0.156	0.112
≥ 11	0.268	0.133	0.135
≥ 12	0.196	0.117	0.079
≥ 13	0.179	0.078	0.101
≥ 14	0.161	0.070	0.090
≥ 15	0.089	0.047	0.042
> 15	0.000	0.000	0.000



