Internal Consistency and Convergent Validity of the Inventory of Hyperacusis Symptoms

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Short title

Validity of the IHS

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Abstract

Objective: The aim was to assess the internal consistency and convergent and discriminant validity of a new questionnaire for hyperacusis, the Inventory of Hyperacusis Symptoms (IHS; Greenberg & Carlos 2018), using a clinical population.

Design: This was a retrospective study. Data were gathered from the records of 100 consecutive patients who sought help for tinnitus and/or hyperacusis from an audiology clinic in the UK. The average age of the patients was 55 years (standard deviation, SD = 13 years). Audiological measures were the Pure Tone Average threshold (PTA) and Uncomfortable Loudness Levels (ULL). Questionnaires administered were: IHS, Tinnitus Handicap Inventory (THI), Hyperacusis Questionnaire (HQ), Insomnia Severity Index (ISI), Generalized Anxiety Disorder (GAD-7), and Patient Health Questionnaire (PHQ-9).

Results: Cronbach's alpha for the 25-item IHS questionnaire was 0.96. Neither the total IHS score nor scores for any of its five subscales were correlated with the PTA of the better or worse ear. This supports the discriminant validity of the IHS, as hyperacusis is thought to be independent of the PTA. There were moderately strong correlations between IHS total scores and scores for the HQ, THI, GAD-7, and PHQ-9, with r = 0.58, 0.58, 0.61, 0.54, respectively. Thus, although IHS scores may reflect hyperacusis itself, they may also reflect the co-existence of tinnitus, anxiety and depression. The total score on the IHS was significantly different between patients with and without hyperacusis (as diagnosed based on ULLs or HQ scores). Using the HQ score as a reference, the area under the receiver operating characteristic for the IHS was 0.80 (95% confidence interval CI: 0.71-0.89) and the cut-off point of the IHS with highest overall accuracy was 56/100. The corresponding sensitivity and specificity were 74% and 82%.

Conclusions: The IHS has good internal consistency and reasonably high convergent validity, as indicated by the relationship of IHS scores to HQ scores and ULLs, but IHS scores may also partly reflect the co-occurrence of tinnitus, anxiety and depression. We propose an IHS cut off score of 56 instead of 69 for diagnosing hyperacusis.

Key words: IHS, hyperacusis, hearing loss, tinnitus, psychometrics, questionnaire, uncomfortable loudness levels

INTRODUCTION

Hyperacusis is intolerance of certain everyday sounds, causing significant distress and impairment in social, occupational, recreational, and other day-to-day activities (Aazh et al. 2016). The sounds may be perceived as uncomfortably loud, unpleasant, frightening, or painful (Tyler et al. 2014). Tinnitus is the perception of sound without any external acoustic sound source. About 55% of patients with tinnitus also report having hyperacusis (Schecklmann et al. 2014). The co-occurrence of tinnitus and hyperacusis was first highlighted by Tyler and Conrad Armes (1983).

Hyperacusis is typically diagnosed using questionnaires. The most commonly used questionnaires are the Hyperacusis Questionnaire (HQ) (Khalfa et al. 2002) and the multiple activity scale for hyperacusis (MASH) (Dauman & Bouscau-Faure 2005). The validity and reliability of the HQ have been questioned by a number of authors (Fackrell et al. 2015; Wallen et al. 2012; Meeus et al. 2010; Fackrell & Hoare 2018). Greenberg and Carlos (2018) pointed out some limitations of both the HQ and MASH. They argued that the reliability and validity of the HQ were unclear and that only 4 of the 14 HQ questions are actually related to hyperacusis. They also pointed out that the MASH has not yet been evaluated for internal consistency, reliability or construct validity. They argued that a reliable and valid questionnaire was needed to provide improved information about hyperacusis severity and differentiation from related categories of heightened auditory sensitivity disorders. Such a questionnaire would also allow clinicians and researchers to assess the efficacy of therapeutic interventions and to evaluate the subjective experience of those suffering from hyperacusis. They introduced a new questionnaire, the Inventory of Hyperacusis Symptoms (IHS; Greenberg & Carlos 2018), that was intended to achieve these goals. It consists of 25 questions and response choices are made on a four-point Likert scale. Hence, the maximum possible score is 100. The authors developed the final questionnaire from a pool of 58 questions, which were created based on qualitative analysis of interviews with five professional audiologists, analysis of patients' posts about sound intolerance to an online support group, a review of the research literature, and current psychometric instruments, e.g. the HQ, the Geräuschüberempfindlichkeit (GÜF) (Nelting et al. 2002), the MASH, and clinical interview forms (Henry et al. 2003; Jastreboff & Jastreboff 2000).

The psychometric properties of the IHS were assessed for 450 participants who were members of online tinnitus and hyperacusis support groups from 37 countries (Greenberg & Carlos 2018). The mean age of the participants was 35 years (SD = 1.6 years). Participants had to complete several questionnaires online, comprising: the IHS, the Patient Health

Questionnaire-4 (PHQ-4) (Lowe et al. 2010), and the World Health Organization WHOQOL-BREF quality of life questionnaire (The WHOQOL Group 1998). Participants were also asked to rate:

(1) The severity of their auditory sensitivity ("Not a problem", A little problem", "Somewhat of a problem", Quite a big problem", "An extreme problem".
(2) Their hearing difficulties ("None", "Mild", "Moderate", "Severe", "Profound")
(3) The severity of their tinnitus ("None", "Mild", "Moderate", "Severe", "Extreme")

The test-retest reliability of the IHS was good, with Cronbach's alpha = 0.93. Mild or greater hearing loss was reported by about 50% of participants and 80% reported tinnitus. Based on PHQ-4 scores, 75% of participants presented with symptoms of anxiety or depression. Greenberg and Carlos (2018) reported that the IHS has five dimensions/subscales (referred to as factors throughout this manuscript): (1) General loudness (items 1, 2, 21), (2) Emotional arousal (items 3-6, 17), (3) Psychosocial (items 12-16, 22-25), (4) Functional impact (items 7-11), (5) Communication (18-20). Construct validity was demonstrated through correlations between IHS total scores and scores for the PHQ-4 ($r = 0.54$) and WHOQOF-BREF ($r = -0.54$). The mean IHS total score was 75 (SD = 15) and it was proposed that a score ≥69 indicates the presence of hyperacusis.

There were some limitations of the study of Greenberg and Carlos (2018), specifically: the study population may not have been typical of those who are treated for hyperacusis in clinical practice, as they were recruited from an online forum. It is important to test the psychometric properties of instruments like the IHS in the population of interest (Mokkink et al. 2010). Additionally, hearing tests were not conducted; no questionnaires assessing the severity of hyperacusis were administered other than the IHS; and the severity of tinnitus was not assessed with a validated questionnaire. Such measures can be used to assess the convergent and discriminant validity of the IHS (Foster & Cone 1995).

The goal of the present study was to assess the reliability and convergent and discriminant validity of the IHS for a population seeking help for tinnitus and/or hyperacusis for whom hearing thresholds had been measured and for whom the results of a wide range of validated questionnaires were available, including questionnaires assessing tinnitus and hyperacusis. Another goal was to assess whether the proposed cut off IHS score of 69 for diagnosing hyperacusis gives outcomes consistent with the cut off scores for other tests used to diagnose hyperacusis, specifically, the HQ and uncomfortable loudness levels (ULLs).

METHODS

Ethical Approval

The study was registered and approved as a clinical audit by the Quality Governance Department at the Royal Surrey County Hospital (RSCH). 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 RSCH.

Study Design and Patients

This was a retrospective cross-sectional study conducted at the Tinnitus and Hyperacusis Therapy Specialist Clinic (THTSC), RSCH, Guildford, UK, which is funded by the UK National Health Service. Data were included for consecutive patients who attended the THTSC within a three-month period for whom the IHS had been completed (*n* = 100). The average age of the patients was 55 years (SD = 13 years, age range 21 to 81 years).

Demographic data for the patients, results of their audiological investigations and the outcomes of their self-report questionnaires 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 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 (BSA 2011a), but with some modifications proposed by Aazh and Moore (2017c) to avoid any discomfort. The starting presentation level at 0.25, 0.5, 2, 3, 4, 6, and 8 kHz was equal to the measured audiometric threshold at the adjacent frequency (e.g., if the threshold at 1 kHz was 20 dB HL, the starting level for measuring the threshold at 2 kHz was 20 dB HL, instead of 50 dB HL as recommended by the BSA). 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 (BSA 2011a): 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 (BSA 2011b), but with the modifications proposed by Aazh and Moore (2017c), to avoid any discomfort. The instructions were "I will gradually make the sound louder in your

ear, and you must press the button (or raise your hand) *as soon as* the sound becomes uncomfortable (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 press the button (or raise your hand) only when the sound becomes uncomfortable; but make sure you press (raise) it as soon as the sound reaches that level." The starting presentation level was equal to the measured audiometric threshold at the test frequency. In addition, levels above 80 dB HL were not used. If the ULL was not reached at 80 dB HL, the ULL at the test frequency was recorded as 85 dB HL. 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, hyperacusis was deemed to be present (Aazh & Moore 2017b).

Inventory of Hyperacusis Symptoms

The IHS has 25 items and the response choices are "not at all" (1 point), "a little" (2 points), "somewhat" (3 points), and "very much so" (4 points). The overall score ranges from 25 to 100. Greenberg and Carlos (2018) proposed that scores between 69 and 79 indicate hyperacusis, scores between 80 and 88 indicate severe hyperacusis and scores ≥89 indicate very severe hyperacusis.

Other Self-Report Questionnaires

The rationale for using the questionnaires listed here is explained under "Data Analyses".

(1) Hyperacusis Questionnaire (HQ; Khalfa et al. 2002)

The HQ comprises 14 items and the response choices are "no" (0 points), "yes, a little" (1 point), "yes, quite a lot" (2 points), and "yes, a lot" (3 points). Cronbach's alpha for the English version of the HQ is 0.88 (Fackrell et al. 2015). The overall score ranges from 0 to 42. Scores of 22 or more were taken as indicating the presence of hyperacusis (Aazh & Moore 2017b).

(2) Tinnitus Handicap Inventory (THI; Newman et al. 1996)

The THI has 25 items, and response choices are "no" (0 points), "sometimes" (2 points) and "yes" (4 points). Cronbach's alpha for the THI is 0.93 (Baguley & Andersson 2003). The overall score ranges from 0 to 100. Scores from 0–16 indicate no handicap, scores from 18–36 indicate mild handicap, scores from 38–56 indicate moderate handicap, and scores from 58–100 indicate severe handicap (Newman et al. 1998).

(3) Insomnia Severity Index (ISI; Bastien et al. 2001)

The ISI comprises seven items that assess the severity of sleep difficulties and their effect on the patient's life. Cronbach's alpha for the ISI is 0.74 (Bastien et al. 2001). Each item is rated on a scale from 0 to 4 and the total score ranges from 0 to 28. Scores from 0-7 indicate no clinically significant insomnia, scores from 8-14 indicate slight insomnia, scores from 15-21 indicate moderate insomnia, and scores from 22-28 indicate severe insomnia (Bastien et al. 2001).

(4) Generalized Anxiety Disorder questionnaire (GAD-7; Spitzer et al. 2006).

This is a 7-item questionnaire for assessment of anxiety symptoms. Patients are asked how often during the last two weeks they had been bothered by each symptom. Response options are not at all (0), several days (1), more than half the days (2), and nearly every day (3). The total score ranges from 0 to 21. The recommended cut-off score for generalized anxiety in the UK mental health system is 8 (IAPT 2011). Cronbach's alpha for the GAD-7 is 0.92 and its test-retest reliability is $r = 0.83$ (Spitzer et al. 2006).

(5) Patient Health Questionnaire (PHQ-9; Kroenke et al. 2001).

This is a 9-item questionnaire for assessment of depression. For each item, a score of 0, 1, 2, or 3 is assigned to the response categories of "not at all", "several days", "more than half the days", and "nearly every day", respectively. The total score therefore ranges from 0 to 27. The recommended cut-off score for caseness for depression in the UK mental health system is 10 (IAPT 2011). Cronbach's alpha for the PHQ-9 is 0.89 and its test-retest reliability is $r= 0.84$ (Kroenke et al. 2001).

Data Analyses

The data were anonymized prior to statistical analysis. Descriptive statistics (means, SDs, and 95% confidence intervals, CI) for the characteristics of the patients and scores for the self-report questionnaires, were calculated. Group differences in IHS scores between patients with and without hyperacusis as diagnosed via HQ scores and ULLmin values were assessed using *t*-tests. A linear regression model was used to assess the variables that significantly predict the IHS score after adjustment for the effect of other variables in the model, including age and gender. The *p* value required for statistical significance was set at $p<0.05$.

Pearson correlation was used to assess convergent and discriminant validity (Lehmann 1988). There are differences of opinion about how to interpret the magnitude of correlation coefficients, *r* (Hemphill 2003). Some authors propose that a correlation is weak if $r < 0.2$, moderate if *r* is between 0.2 and 0.5, and strong if $r > 0.5$ (Hemphill 2003; Cohen 1988). Other authors suggest that $r > 0.7$ corresponds to a strong correlation, *r* in the range 0.5 to 0.7

corresponds to a moderate correlation and *r* < 0.3 corresponds to a weak correlation (Mukaka 2012). We adopted the latter convention. Convergent validity of the IHS would be indicated by moderate or strong correlations with other measures thought to be related to hyperacusis, while discriminant validity would be indicated by weak or zero correlations with measures thought to be unrelated to hyperacusis. The questionnaires that were used in routine practice at the THTSC and were relevant to the construct of hyperacusis were included to assess convergent validity (Mokkink et al. 2010).

Convergent validity was explored by assessing the correlations between the IHS total score and the scores for its five factors with scores for other instruments that measure related constructs. The measures used for convergent validity were:

- The HQ. This is intended to measure hyperacusis, so we expected moderate or strong correlations with IHS scores.
- ULLmin and the average ULL across ears (Average ULLs at 0.25, 0.5, 1, 2, 4 and 8 kHz for both ears). These are measures of loudness tolerance for pure tones presented via headphones in a clinical environment. ULLs do not fully explain the variance of the experienced hyperacusis but they are related to it (Aazh & Moore 2017b; Wallen et al. 2012; Blaesing & Kroener-Herwig 2012; Jastreboff & Jastreboff 2015). Some studies suggest no significant relationship between ULLs and the impact of hyperacusis on the patient's life (Meeus et al. 2010). Therefore, we expected a moderate correlation of the ULL measures with the factor of the IHS related to the general loudness of sounds but lower correlations with other factors.

Discriminant validity was explored by calculating correlation coefficients between the IHS total score and the scores for its five factors with measures of different constructs. The measures used for discriminant validity were:

- The PTA across the frequencies 0.25, 0.5, 1, 2, and 4 kHz for the better ear and for the worse ear. PTA values have been found not to be related to hyperacusis (Aazh et al. 2020b; Aazh et al. 2018b; Aazh & Moore 2017b), so we expected small correlations with IHS scores.
- The THI. This is a measure of the impact of tinnitus on a patient's life. Although the THI score is related to the experience of hyperacusis (Fioretti et al. 2013; Aazh et al. 2017; Cederroth et al. 2020), it measures a different construct, which is the impact of tinnitus, not hyperacusis, on the patients' life. Hence we expected moderate correlations

between THI scores and IHS scores.

- The ISI. This gives a measure of sleep disturbances and is moderately correlated with depressive symptoms related to the experience of tinnitus, hyperacusis and possibly hearing impairment (Aazh et al. 2019a; Aazh & Moore 2019; Clarke et al. 2019). We expected moderate correlations between ISI scores and IHS scores.
- The GAD-7 and PHQ-9. These are measures of state anxiety and depression and scores are strongly correlated with measures of hyperacusis (Hu et al. 2015; Juris et al. 2013; Aazh et al. 2017; Aazh & Moore 2017a). We expected moderate correlations of GAD-7 and PHQ-9 scores with IHS scores.

Cronbach’s alpha was calculated for the IHS and each of its five factors via computing the inter-item correlations or covariance for all pairs of items in the questionnaire and for the scale formed from them (Streiner 2003). A value of Cronbach’s alpha between 0.70 and 0.95 indicates good internal consistency (Terwee et al. 2007).

A Receiver Operating Characteristic (ROC) is a plot of “hits” (correct positive diagnoses, corresponding to sensitivity) against “false alarms” (positive diagnoses when no disease is present, corresponding to 1 – specificity) for different cut off values of a measure (IHS total scores here). In the context of our study, sensitivity refers to the proportion of cases diagnosed as having hyperacusis when hyperacusis was present. Specificity refers to the proportion of cases diagnosed as not having hyperacusis when hyperacusis was not present. Unfortunately, there is no “gold standard” for determining whether or not hyperacusis is present. In the absence of a gold standard, HQ scores were used as a reference for calculating sensitivity and specificity for the IHS, based on different cut off values for the IHS (Mokkink et al. 2012). The area under the ROC curve (AUC) indicates the overall accuracy of the diagnostic tool and is between 0.5 and 1.0. The closer the value is to 1.0, the more accurate is the diagnosis (Hanley & McNeil 1982). The cut off value for the IHS yielding the highest overall accuracy, i.e. the highest percentage of patients classified correctly, was taken as the optimal cut off score indicating hyperacusis for clinical use (Florkowski 2008).

The analyses were restricted to patients with complete data on all variables required for a particular analysis. The number of patients included in each analysis (*n*) is reported. The STATA program (version 13) (StataCorp 2013) was used for statistical analyses.

RESULTS

Characteristic of the Study Population

Fifty two percent of the patients were male. The mean age was 56 years (SD = 11 years) for females and 55 years (SD = 15 years) for males. The difference was not significant (p = 0.85). The means and SDs of the hearing thresholds and ULLs for each ear and each frequency are shown in Table 1. The grand mean PTA across ears was 23 dB HL (SD = 14 dB). The grand mean PTA of the better ear was 18 dB HL (SD = 11 dB). The grand mean PTA of the worse ear was 27 dB HL (SD = 20 dB). Based on the PTA for the better ear, 67% of the patients had no hearing loss, 28% had mild hearing loss, and 5% had moderate hearing loss.

The grand mean of the average ULL across 0.25, 0.5, 1, 2, 4 and 8 kHz was 78 dB HL (SD = 9.9) for both the right and left ears (n = 86). The average value of ULLmin was 77.5 dB HL (SD = 11) (n=90). ULLmin values were 77 dB HL or below, indicating hyperacusis, for 28% (25/90) of patients (Aazh & Moore 2017b). Five patients (5/90) were diagnosed with severe hyperacusis as indicated by a ULL at any frequency of 30 dB HL or less (Aazh & Moore 2018b).

***Tables 1 and 2 ***

The mean scores for the IHS (and its factors), HQ, THI, ISI, GAD-7, and PHQ-9 are shown in Table 2. Based on scores for the HQ, 43% (42/98) of patients had hyperacusis. Based on scores for the THI, 2% of patients (2/99) had no tinnitus handicap, 20% (20/99) had a mild tinnitus handicap, 33% (33/99) had a moderate tinnitus handicap, and 44% (44/99) had a severe tinnitus handicap. Based on scores for the ISI, 18% (18/98) of patients did not have insomnia, 31% (30/98) had mild insomnia, 33% (32/98) had clinically significant insomnia, and 18% (18/98) had severe insomnia. Based on scores for the GAD-7 and PHQ-9, 45% (45/100) of patients had anxiety and 46% (46/100) had depression.

Internal Consistency

Cronbach's alpha for the 25-item IHS was 0.96. Cronbach's alpha was 0.81 for Factor one (General loudness), 0.89 for Factor 2 (Emotional arousal), 0.92 for Factor 3 (Psychosocial), and 0.89 for Factor 4 (Functional impact). Factor 5 (Communication) has only two items, so alpha could not be calculated. Instead, the scale reliability coefficient was calculated, which was 0.89. Overall, the values are high, indicating good internal consistency.

Convergent and Discriminant Validity

Table 3 shows the correlations between IHS total scores and scores for each of its five factors with scores for ULLmin, average ULLs across ears, and PTA for the better and worse ears.

As predicted, the total IHS score and its five factors were weakly correlated with the PTA values. These correlations were not statistically significant. This supports the discriminant validity of the IHS, as hyperacusis is thought to be independent of hearing thresholds (Aazh & Moore 2017b).

The total scores on the IHS were weakly negatively correlated with values of ULLmin and ULL across ears, $r = -0.21$ and -0.22, respectively (lower ULLs are associated with greater hyperacusis). The correlations were somewhat higher in absolute value for Factor 1 (General loudness), $r = -0.26$ and -0.30, respectively.

As shown in Table 4, there was a moderate correlation between IHS total scores and HQ scores, $r = 0.58$. However, there were also moderate correlations between IHS total scores and the scores for the THI, GAD-7, and PHQ-9, with $r = 0.58$, 0.61, 0.54, respectively, suggesting that the IHS scores partly reflect the co-occurrence of tinnitus, anxiety and depression.

Tables 3, 4

Group Differences

Table 5 shows IHS total scores and the scores for its factors for patients who were categorized as having hyperacusis or not having hyperacusis using the ULLmin criterion and the HQ criterion. The difference between categories was significant for the total score for the IHS and the scores for Factors 1 (General loudness), 2 (Emotional arousal) and 4 (Functional impact), for diagnoses based on both ULLmin and HQ scores. The scores for Factors 3 (Psychosocial) and 5 (Communication) were significantly different when the diagnosis was based on HQ scores but not when it was based on ULLmin values.

A regression model showed that variables that significantly predicted the total IHS score were HQ score (regression coefficient, $b = 0.67$, $p = 0.001$), THI score ($b = 0.23$, $p = 0.015$), and GAD-7 score ($b = 0.87$, $p = 0.047$). The regression coefficients were calculated taking into account the effects of other variables in the model that did not significantly predict IHS scores. These comprised: PHQ-9 scores (b = –0.94, p = 0.8), ISI scores (b = 0.3, p = 0.32), ULLmin values (b = –0.1, p = 0.49), PTA of the worse ear (b = 0.06, p = 0.59), PTA of the better ear (b = –0.3, p = 0.17), age (b = 0.04, p = 0.74), and gender (b = –2.14, p = 0.49).

Table 5

Cut Off Scores

The ROC for the IHS, using HQ scores as a reference, is shown in Figure 1. The AUC was 0.80 (95% CI: 0.71-0.89), indicating good accuracy. The cut-off score for the IHS yielding the highest percentage of patients classified correctly (79%) was 56/100. The corresponding sensitivity and specificity were 74% and 82%, respectively. The cut off score of 69 recommended by Greenberg and Carlos (2018) gave a percentage of patients classified correctly of 72.5%, with sensitivity of 48% and specificity of 91%. The sensitivity with this cut off is low, suggesting that a cut off score of 69 for the IHS is too high and a cut off of 56 offers better accuracy and a better balance between sensitivity and specificity.

Figure 1

DISCUSSION

The aim of this study was to assess the internal consistency and convergent and discriminant validity of a questionnaire for hyperacusis, the IHS, using a clinical population rather than using participants who were recruited online, as was done by Greenberg and Carlos (2018). The mean total IHS score for our participants was much lower than found for the participants tested online by Greenberg and Carlos (2018), even for the patients in our sample who were diagnosed with hyperacusis based on ULLmin values or HQ scores. This supports our expectation that the outcomes of the IHS might depend on the population that is studied. The exact characteristics of the population studied by Greenberg and Carlos (2018) are unclear. They stated that “While no specific exclusion criteria prevented any participant from completing the questionnaires, individuals who experience varying levels of auditory sensitivity were sought to gain a sufficiently robust sample size to estimate scoring thresholds between categories of symptom severity”. Our population was based on patients seeking help for their tinnitus, hyperacusis, or both. The IHS is likely to be used most often to assess people seeking help for tinnitus and hyperacusis, so the population used in our study seems more applicable than that tested by Greenberg and Carlos (2018).

Cronbach's alpha for the 25-item IHS was 0.96, which is very high. This high value might indicate a potential for reducing the number of items in the IHS, while maintaining good consistency. The high alpha value might reflect the possibility that more than one construct is measured using the IHS. Consistent with this, our results showed moderate correlations between IHS scores and scores for the THI, GAD-7 and PHQ-9, which are measures of tinnitus severity, anxiety and depression. It is possible that IHS scores partly reflect the distress caused by tinnitus, anxiety, or depression. This is understandable, due to the comorbidity of hyperacusis with tinnitus, depression and anxiety (Valderas et al. 2009; Cederroth et al. 2020). Although the exact mechanisms underlying these comorbidities have not been fully explored, some studies suggest a causal link between hyperacusis and depression (Aazh & Moore 2017a; Aazh et al. 2019b; Assi et al. 2018; Attri & Nagarkar 2010). One could argue that people primarily seek help for their hyperacusis when the anxiety and depression it produces are sufficiently troublesome (Aazh et al. 2014; Aazh et al. 2018a). Hyperacusis is not simply an over-sensitivity to certain sounds. Many people may feel disturbed by certain sounds (e.g., they do not like loud social places and they may dislike the sound of warning sirens) but this does not cause them significant distress or interruption in their daily life. It is only when certain sounds lead to distress and anxiety in everyday life that people seek help for hyperacusis and are diagnosed as having hyperacusis (Aazh et al. 2016). When this happens, symptoms of anxiety and depression often co-occur (Aazh & Moore 2017d). In fact, being sensitive to noise is classified as a personality trait (Weinstein 1978). Noise sensitivity as a personality trait can also be age and sex related, as older people and females tend to be less tolerant of noise (Baliatsas et al. 2016; Stansfeld & Shipley 2015). However, our regression model revealed that IHS scores were not significantly predicted by age or sex.

Tinnitus is also a comorbid construct with hyperacusis and the majority of the patients in our sample had some degree of tinnitus. Untangling the impact of hyperacusis from that of tinnitus in such a population is difficult. For some patients with tinnitus, exposure to certain environmental sounds worsens their tinnitus (Aazh & Salvi 2019). Such patients adopt avoidance behavior or use ear protection (earplugs or muffs), which in turn can lead to development of fear hyperacusis (Tyler et al. 2014). It has also been hypothesized that hyperacusis is a precursor to tinnitus (Jastreboff & Jastreboff 2003; Jastreboff & Jastreboff

2014; Jastreboff & Jastreboff 2015). Finally laboratory studies on animals suggest that hyperacusis and tinnitus share pathophysiological mechanisms (Knipper et al. 2013; Mohrle et al. 2019; Eggermont 2013; Chen et al. 2015). It is not clear whether the IHS can distinguish tinnitus-related distress from hyperacusis-related distress. To assess whether that is the case, IHS scores could be compared for a group of patients with tinnitus but no hyperacusis and a group with tinnitus combined with hyperacusis. Having said this, even in such populations, it may be difficult to disentangle these two constructs. Tinnitus handicap tends to be more severe for patients who also have hyperacusis than for those with tinnitus alone (Schecklmann et al. 2014). More studies are required to explore and explain the interaction between tinnitus, hyperacusis, anxiety, and depression and how these affect the individual (Neale & Kendler 1995). For a review of comorbidity models see Valderas et al. (2009).

The comorbidity of hyperacusis with tinnitus, anxiety and depression and the inability of the questionnaires in this study to distinguish the impact of these conditions on a patient's life can make it difficult for clinicians to plan management strategies. If hyperacusis is the main source of distress, then audiologist-delivered cognitive behavioural therapy (CBT) may be the right course of action (Aazh & Allott 2016; Aazh & Moore 2018a). However, if the main source of distress is an underlying anxiety disorder, then the patient may benefit from more general psychotherapy with mental health professionals (Otte 2011). Another clinical implication is that hyperacusic patients who are very distressed by their tinnitus or exhibit symptoms of anxiety and depression may score highly on the IHS. From a clinical perspective it is important to check for underlying psychological disorders when assessing patients with high IHS or THI scores. Patients whose scores are outside the normal range on psychological questionnaires may be referred to mental health professionals for further investigation and treatment (when needed). A recent study assessing patient's views about the acceptability and relevance of completing certain psychological questionnaires (or screening versions of them) in a tinnitus and hyperacusis clinic showed that patients regard such questionnaires as acceptable and relevant to them (Aazh & Moore 2017d).

Another clinical implication is that clinicians should not rely solely on self-report questionnaires when assessing the impact of hyperacusis or tinnitus on a patient's life. It is very important to perform an in-depth clinical interview to establish whether the distress they experience is related to tinnitus, hyperacusis, mental health disorders, or a combination of these. A recent study showed that there was no statistically significant difference in mean scores for the HQ, THI and audiological measures between patients who after an in-depth clinical interview were deemed not to have any distress linked with their tinnitus or hyperacusis and patients who did present with distress related to tinnitus and/or hyperacusis (Aazh & Moore 2018c). Thus, the in-depth interview provided information that was not provided by the questionnaires or audiological measures.

Previous work has shown that the severity of hyperacusis is not related to PTA values (Aazh & Moore 2017b). Therefore, if the IHS genuinely measures the severity of hyperacusis, IHS scores should not be correlated with PTA values. Our results showed that neither the total IHS score nor scores for its five factors were significantly correlated with PTA values, supporting the discriminant validity of the IHS.

It is believed that low ULLs are associated with hyperacusis (Aazh & Moore 2017b; Aazh & Moore 2017c). Consistent with this, total IHS scores and scores for IHS Factors 1 and 2 were negatively correlated with ULL min values. The correlation for Factor 2 (Emotional arousal) is consistent with the idea that emotional arousal is part of the experience of hyperacusis (Aazh & Allott 2016; Aazh et al. 2019c; Aazh et al. 2014). However, the correlations between IHS factor scores and ULLmin values were all below 0.30. This indicates that, if the IHS provides a valid measure of the severity of hyperacusis, the ULLs measured with pure tones make only a small contribution to the variance of the severity of hyperacusis. This may be the case because hyperacusis often reflects an aversion to specific sounds (not pure tones).

Total IHS scores and factor scores were more highly correlated with HQ scores than with ULLmin values. The correlations with HQ scores ranged from 0.40 for Factor 5 (Communication) to 0.63 for Factor 2 (Emotional arousal), supporting the construct validity of the IHS and consistent with the idea that emotional arousal plays a strong role in the severity of hyperacusis (Aazh & Allott 2016; Aazh et al. 2019c; Aazh et al. 2014; Aazh et al. 2020a; Aazh & Moore 2018c; Aazh & Moore 2018a). A role for emotional arousal is supported by our finding that scores on the GAD-7, which is related to emotional arousal, were moderately correlated with IHS scores.

If the IHS has high construct validity, one might expect the correlation between IHS and HQ scores to be higher than the correlation between IHS and GAD-7 scores. This was not the case: the correlations were 0.58 and 0.61, respectively. This could be related to the shortcomings of the HQ described in the introduction, specifically the fact that some of its questions are not directly related to hyperacusis (Baguley & Andersson 2007). Moreover, the strong relationship between IHS and GAD-7 scores could be due to the fact that the IHS has more items than the HQ assessing the emotional aspects of hyperacusis.

It was found that the cut off total score for the IHS of 69 suggested by Greenberg and Carlos (2018) as indicating hyperacusis would lead to most of the patients in our sample not being diagnosed as having hyperacusis, even though 28% of our patients had ULLmin values ≤77 dB HL and 43% had HQ scores ≥22, both of which have been suggested to indicate hyperacusis (Aazh & Moore 2017b). Also, the cut off score of 69 led to sensitivity of the IHS of only 45%, using diagnosis based on HQ scores as a reference.

The ROC analysis showed that the AUC for the IHS was 0.80. The AUC is a combined measure of sensitivity and specificity that characterizes the validity of diagnostic tests (Hanley & McNeil 1982). The AUC of 0.80 shows that the IHS has a good ability to discriminate patients with hyperacusis from patients without hyperacusis. We propose a cut off value of 56 instead of 69 for diagnosing hyperacusis. The corresponding sensitivity and specificity were 74% and 82%, respectively. We suggest that the Greenberg and Carlos (2018) categories of hyperacusis based on IHS score should be modified to: <56 no hyperacusis; between 56 and 79 mild-moderate hyperacusis; between 80 and 88 severe hyperacusis; ≥89 very severe hyperacusis. More research is needed to assess our proposed categorization of hyperacusis severity.

STUDY LIMITATIONS

This study has several limitations, which are discussed here. First, the analysis was limited to data gathered in a clinic providing treatment to people seeking help for tinnitus and/or hyperacusis. This may not be representative of the entire population of people with hyperacusis. However, the population tested here is representative of those who seek help for hyperacusis and/or tinnitus. A second limitation is that there is no gold standard for diagnosing hyperacusis. Therefore, we were forced to evaluate the sensitivity and specificity of the IHS using an established questionnaire, the HQ, whose validity has been questioned. However, it is reassuring that the results of the IHS were related to the measures of ULLs (higher IHS scores being associated with lower ULLs) and to the scores for other questionnaires assessing tinnitus handicap, insomnia, anxiety, and depression. A third limitation is that we did not have data about the underlying etiologies of the hyperacusis of the participants. Scores for the five factors of the IHS might be different across different etiologies of hyperacusis.

CONCLUSIONS

Cronbach's alpha for the total IHS score was 0.96. Neither the total IHS score nor any of its five factors were correlated with the PTA of the better or worse ear, supporting the discriminant validity of the IHS, since hyperacusis is thought to be independent of the degree of hearing loss. The total IHS score and scores for its five factors were moderately correlated with hyperacusis handicap (as measured via the HQ), tinnitus handicap (as measured via the THI), anxiety (as measured via the GAD-7), and depression (as measured via the PHQ-9). Although this supports the convergent validity of IHS, since all of these constructs are known to be significantly related to hyperacusis, it means that the IHS may not distinguish the distress caused by hyperacusis from the distress caused by tinnitus, anxiety and depression. Therefore, future studies are needed to design questionnaires that can overcome the shortcomings of the current ones as highlighted in this study.

We propose an IHS cut-off value of 56 instead of 69 for diagnosing hyperacusis. With this cut off value, the corresponding sensitivity and specificity of the IHS, using the HQ as a reference, were 74% and 82%, respectively.

From a clinical perspective, it is important to use a wide range of questionnaires to get a better understanding of the symptoms that the patient is experiencing and also to perform an in-depth clinical interview to establish whether the distress they experience is related to hyperacusis, tinnitus, or mental health disorders.

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Figure caption

Figure 1. ROC for the IHS. The area under the ROC was 0.80.

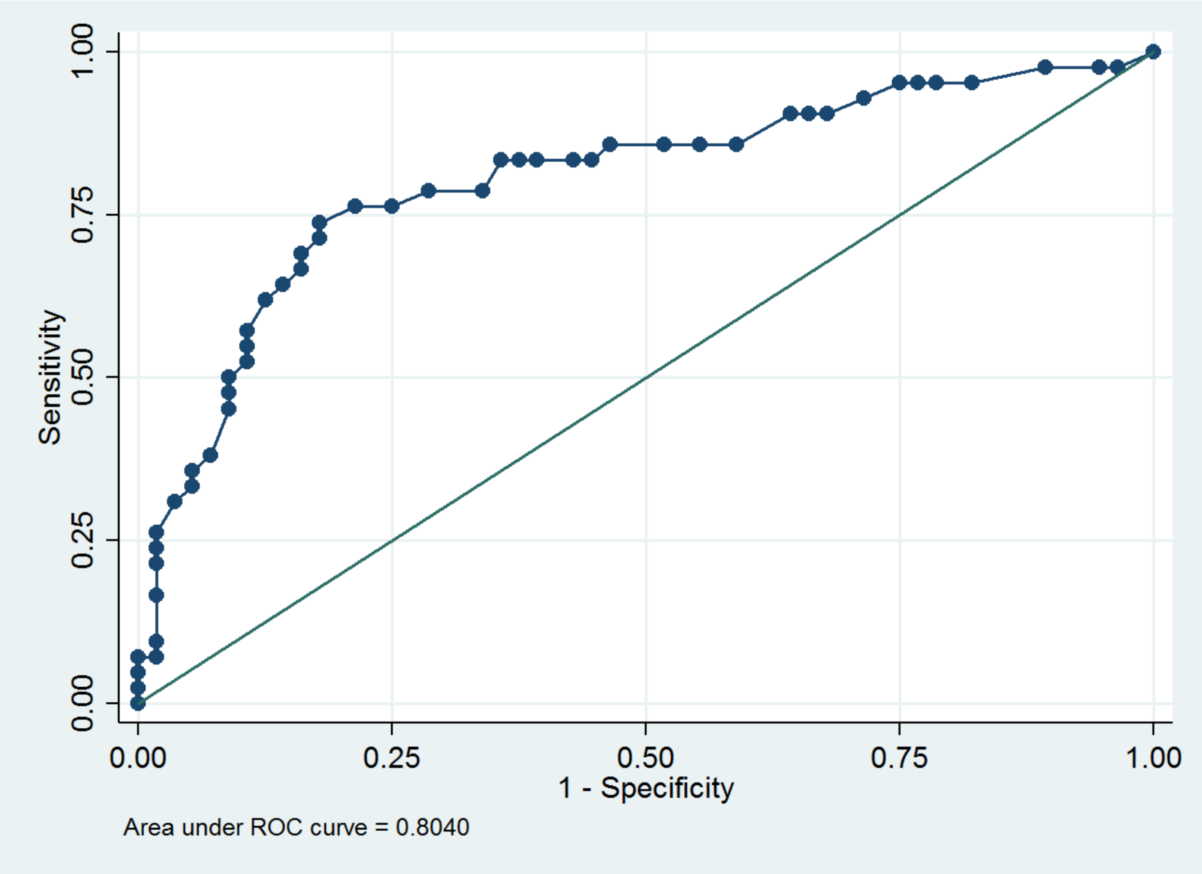

TABLE 1. Means (SD) of HTs in dB HL and ULLs for each ear of the study population. The number of patients included in each analysis is indicated by *n*.

	Frequency, kHz							
	0.25	0.5	1	2	3	4	6	8
HT right	18 (16) *n* = 100	19 (17) *n* = 100	18 (17) *n* = 100	21 (19) *n* = 100	27 (20) *n* = 93	32 (21) *n* = 100	37 (23) *n* = 95	35.5 (26) *n* = 100
HT left	20 (16.5) *n* = 100	20 (17) *n* = 100	19 (19) *n* = 100	23 (20) *n* = 100	30 (22) *n* = 92	35.5 (22) *n* = 100	39 (26) *n* = 93	39.5 (28) *n* = 100
ULL right	78 (11) *n* = 90	79 (11) *n* = 90	80 (9) *n* = 90	79 (11) *n* = 90	79 (11) *n* = 83	79 (12) *n* = 90	79 (12) *n* = 85	76 (14) *n* = 89
ULL left	78 (9) *n* = 90	79 (10) *n* = 90	79 (10) *n* = 90	79 (11) *n* = 90	79 (11) *n* = 82	79 (11) *n* = 89	79 (12) *n* = 83	75 (14) *n* = 87

HT, hearing threshold; ULL, Uncomfortable Loudness Level

TABLE 2. Means and SDs of scores of the study population for the IHS and its factors, and for the HQ, THI, ISI, GAD-7, and PHQ-9. The number of patients is indicated by *n*.

Questionnaire	*n*	Mean	SD
IHS (total)	100	54.4	19
Factor 1 (General loudness)	100	7.6	2.9
Factor 2 (Emotional arousal)	100	12.0	4.5
Factor 3 (Psychosocial)	100	16.0	6.8
Factor 4 (Functional impact)	100	14.7	5.0
Factor 5 (Communication)	100	4.2	2.2
HQ	98	19.4	9
THI	99	55.2	21
ISI	98	14.4	7.1
GAD-7	100	8.2	5.8
PHQ-9	100	10.4	6.7

IHS, Inventory of Hyperacusis Symptoms; THI, Tinnitus Handicap Inventory; HQ, Hyperacusis Questionnaire; ISI, Insomnia Severity Index; GAD-7, Generalized Anxiety Disorder questionnaire; PHQ-9, Patient Health Questionnaire

TABLE 3. Correlations between IHS scores and scores for each of its five factors with audiological measures. The number of patients is indicated by *n*.

	ULLmin	ULL across ears	PTA better ear	PTA worse ear
IHS total	$r = -0.21$	$r = -0.22$	$r = -0.03$	$r = 0.05$
	$p = 0.042$*	$p = 0.04$*	$p = 0.73$	$p = 0.62$
	$n = 90$	$n = 86$	$n = 100$	$n = 100$
Factor 1	$r = -0.26$	$r = -0.3$	$r = -0.04$	$r = 0.07$
(General loudness)	$p = 0.013$*	$p = 0.005$**	$p = 0.69$	$p = 0.45$
	$n = 90$	$n = 86$	$n = 100$	$n = 100$
Factor 2	$r = -0.21$	$r = -0.21$	$r = -0.13$	$r = -0.11$
(Emotional arousal)	$p = 0.043$*	$p = 0.048$*	$p = 0.17$	$p = 0.27$
	$n = 90$	$n = 86$	$n = 100$	$n = 100$
Factor 3	$r = -0.14$	$r = -0.15$	$r = 0.02$	$r = 0.14$
(Psychosocial)	$p = 0.19$	$p = 0.16$	$p = 0.81$	$p = 0.15$
	$n = 90$	$n = 86$	$n = 100$	$n = 100$
Factor 4	$r = -0.21$	$r = -0.2$	$r = -0.09$	$r = -0.03$
(Functional impact)	$p = 0.048$*	$p = 0.064$	$p = 0.34$	$p = 0.75$
	$n = 90$	$n = 86$	$n = 100$	$n = 100$
Factor 5	$r = -0.15$	$r = -0.15$	$r = 0.17$	$r = 0.18$
(Communication)	$p = 0.15$	$p = 0.16$	$p = 0.076$	$p = 0.07$
	$n = 90$	$n = 86$	$n = 100$	$n = 100$

IHS, Inventory of Hyperacusis Symptoms; ULLmin, average ULLs at 0.25, 0.5, 1, 2, 4 and 8 kHz for the ear with the lower average ULL; ULL across ears, average ULLs at 0.25, 0.5, 1, 2, 4 and 8 kHz for both ears; PTA, The pure-tone average across the frequencies 0.25, 0.5, 1, 2, and 4 kHz. * = $p < 0.05$, ** = $p < 0.01$.

TABLE 4. Correlations between total IHS scores and the scores for each of its five factors with scores for the HQ, THI, GAD-7, PHQ-9 and ISI. The number of patients is indicated by *n*.

	HQ	THI	GAD-7	PHQ-9	ISI
IHS total	$r = 0.58$ $p<0.001$*** $n = 98$	$r = 0.58$ $p<0.001$*** $n = 99$	$r = 0.61$ $p<0.001$*** $n = 100$	$r = 0.54$ $p<0.001$*** $n = 100$	$r = 0.43$ $p<0.001$*** $n = 98$
Factor 1 (General loudness)	$r = 0.61$ $p<0.001$*** $n = 98$	$r = 0.37$ $p<0.001$*** $n = 99$	$r = 0.43$ $p<0.001$*** $n = 100$	$r = 0.40$ $p<0.001$*** $n = 100$	$r = 0.32$ $p = 0.0012$** $n = 98$
Factor 2 (Emotional arousal)	$r = 0.63$ $p<0.001$*** $n = 98$	$r = 0.51$ $p<0.001$*** $n = 99$	$r = 0.54$ $p<0.001$*** $n = 100$	$r = 0.45$ $p<0.001$*** $n = 100$	$r = 0.38$ $p<0.001$*** $n = 98$
Factor 3 (Psychosocial)	$r = 0.46$ $p<0.001$*** $n = 98$	$r = 0.53$ $p<0.001$*** $n = 99$	$r = 0.56$ $p<0.001$*** $n = 100$	$r = 0.53$ $p<0.001$*** $n = 100$	$r = 0.36$ $p<0.001$*** $n = 98$
Factor 4 (Functional impact)	$r = 0.49$ $p<0.001$*** $n = 98$	$r = 0.59$ $p<0.001$*** $n = 99$	$r = 0.56$ $p<0.001$*** $n = 100$	$r = 0.49$ $p<0.001$*** $n = 100$	$r = 0.47$ $p<0.001$*** $n = 98$
Factor 5 (Communication)	$r = 0.40$ $p<0.001$*** $n = 98$	$r = 0.45$ $p<0.001$*** $n = 99$	$r = 0.51$ $p<0.001$*** $n = 100$	$r = 0.44$ $p<0.001$*** $n = 100$	$r = 0.29$ $p = 0.004$** $n = 98$

*IHS, Inventory of Hyperacusis Symptoms; THI, Tinnitus Handicap Inventory; HQ, Hyperacusis Questionnaire; GAD-7, Generalized Anxiety Disorder questionnaire; PHQ-9, Patient Health Questionnaire; ISI, Insomnia Severity Index. ** = $p < 0.01$, *** = $p < 0.001$.*

TABLE 5. IHS total scores and scores for its five factors for patients with and without hyperacusis as categorized based on ULLmin values and HQ scores. 95% CIs for the differences and significances of the differences across categories are shown. The number of patients is indicated by *n*.

	Diagnosed based on ULLmin			Diagnosed based on HQ		
	Hyperacusis present Mean (SD)	No hyperacusis Mean (SD)	95% CI and *p* value	Hyperacusis present Mean (SD)	No hyperacusis Mean (SD)	95% CI and *p* value
IHS total	61 (21) *n* = 25	50 (17) *n* = 65	-19.8 to -3.1 *p* = 0.007**	65.8 (18.4) *n* = 42	46 (14.3) *n* = 56	-26.3 to -3.2 *p*<0.001***
Factor 1 (General loudness)	8.8 (2.3) *n* = 25	6.8 (2.9) *n* = 65	-3.3 to -0.72 *p* = 0.003**	9.3 (2.4) *n* = 42	6.3 (2.5) *n* = 56	-3.9 to -1.98 *p*<0.001***
Factor 2 (Emotional arousal)	13.8 (4.7) *n* = 25	10.8 (4.3) *n* = 65	-5.0 to -0.90 *p* = 0.005**	14.6 (4.2) *n* = 42	9.9 (3.6) *n* = 56	-6.3 to -3.12 *p*<0.001***
Factor 3 (Psychosocial)	17.7 (7.7) *n* = 25	14.7 (5.9) *n* = 65	-5.9 to 0.05 *p* = 0.054	19.5 (7) *n* = 42	13.6 (5.5) *n* = 56	-8.3 to -3.3 *p*<0.001***
Factor 4 (Functional impact)	16.5 (5.7) *n* = 25	13.7 (4.4) *n* = 65	-4.9 to -0.49 *p* = 0.017*	17.3 (4.9) *n* = 42	12.7 (3.9) *n* = 56	-6.4 to -2.8 *p*<0.001***
Factor 5 (Communication)	4.5 (2.3) *n* = 25	3.7 (1.9) *n* = 65	-1.78 to 0.13 *p* = 0.089	5.1 (2.2) *n* = 42	3.4 (1.9) *n* = 56	-2.5 to -0.82 *p*<0.001***

*IHS, Inventory of Hyperacusis Symptoms; HQ, Hyperacusis Questionnaire; ULLmin, average ULLs at 0.25, 0.5, 1, 2, 4 and 8 kHz for the ear with the lower average ULL. * = p < 0.05, ** = p < 0.01, *** = p < 0.001.*