Factors related to Uncomfortable Loudness Levels for patients seen in a tinnitus and hyperacusis clinic

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Short title

Factors related to ULLs for tinnitus and hyperacusis patients

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Abbreviations

BSA	British Society of Audiology
HADS	Hospital Anxiety and Depression Scale
HQ	Hyperacusis questionnaire
HT	Hearing threshold
ISI	Insomnia severity index
NHS	National Health Service
PTA	Pure tone average audiometric threshold
SD	Standard deviation
THI	Tinnitus Handicap Inventory
THTSC	Tinnitus and Hyperacusis Therapy Specialist Clinic
ULL	Uncomfortable Loudness Level
ULLmin	Across-frequency average ULL for the ear with the lower ULL
VAS	Visual Analogue Scale

Abstract

Objectives: The aims were: (1) to explore patterns of uncomfortable loudness levels (ULLs) across frequency and their associated factors for patients with tinnitus and hyperacusis, and (2) to re-evaluate the criteria for diagnosing hyperacusis based on ULLs and scores for the hyperacusis questionnaire (HQ). *Design:* This was a retrospective cross-sectional study. *Study Sample:* 573 consecutive patients for whom ULLs had been measured were included. *Results:* A good correspondence between the diagnosis of hyperacusis based on the across-frequency average ULL for the ear with the lowest ULLs (ULLmin) and hyperacusis handicap based on HQ scores was obtained with cutoff values of ULLmin ≤77 dB HL and HQ score ≥22. A regression model showed significant relationships between ULLmin and the score on the HQ and age. The mean HQ score for patients with a large interaural asymmetry in ULLs was significantly higher than for the remainder. Hyperacusis handicap was associated with strong across-frequency variations in ULLs. *Conclusions:* Appropriate cutoff values for diagnosing hyperacusis are ULLmin ≤77 dB HL and HQ score ≥22. Large interaural asymmetry and large across-frequency variations in ULLs are associated with higher HQ scores.

Key Words: Hyperacusis; uncomfortable loudness levels, psychological assessment; tinnitus

Introduction

There is no universally agreed definition of hyperacusis. Aazh et al (2016) defined hyperacusis as intolerance of everyday sounds that causes significant distress and impairment in social, occupational, recreational, and other day-to-day activities. Several authors and patient groups have given other definitions of hyperacusis (Tyler et al, 2014); Aazh et al, 2014; Jastreboff & Jastreboff, 2014), but all definitions are based on the concept that certain sounds lead to significant distress and/or annoyance for the individual. The sounds may be perceived as uncomfortably loud, unpleasant, frightening, or painful (Tyler et al, 2014).

Audiologists often use Uncomfortable Loudness Levels (ULLs), also called loudness discomfort levels, to determine the lowest sound level at which sounds are perceived to be unpleasantly loud or uncomfortable. The exact instructions are important, and the British Society of Audiology (BSA, 2011) recommends the following instructions: "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."

For normal-hearing people the average ULL across the audiometric frequencies usually lies between 86 and 100 dB HL (Sherlock & Formby, 2005; Knobel & Sanchez, 2006). People with hyperacusis often have lower than normal ULLs in one or both ears (Tyler et al, 2014). ULLs can be used both to diagnose hyperacusis and to assess the severity of hyperacusis. However, the average ULLs reported for patients with hyperacusis vary widely across studies, from 66.3 dB HL (SD =15) (Blaesing & Kroener-Herwig, 2012), to 77 dB HL (Anari et al, 1999), and 83 dB HL (SD = 17) (Sheldrake et al, 2015). This makes the diagnosis of hyperacusis based on ULLs difficult.

The criteria for diagnosis of hyperacusis based on ULLs are not generally agreed and there are wide differences in recommendations across studies. Goldstein and Shulman (1996) and Anari et al (1999) suggested average ULLs across frequency of less than 95 dB HL and 70 dB HL, respectively, as an indication of hyperacusis. Sherlock and Formby (2005) reported that the lowest 5th percentile values of ULLs for 59 normal-hearing adults without sound tolerance problems were 80, 85, 80, and 75 dB HL at 0.5, 1, 2, and 4 kHz, respectively.

Therefore, ULLs of 80 dB HL or above may be considered as within the normal range while ULLs less than 80 dB HL can be considered as abnormally low. The severity of self-reported hyperacusis symptoms for patients diagnosed with hyperacusis using the various proposed ULL criteria has not been evaluated.

There are conflicting reports with regard to the relationship between ULLs and patients' self-reported hyperacusis handicap and psychological aspects of hyperacusis. Zaugg et al (2016) reported a weak negative correlation between across-frequency average ULLs and self-reported sound tolerance problems among 139 patients with tinnitus ($r = -0.23$, $p<0.05$). Meeus et al (2010) did not find a statistically significant relationship between ULLs and scores on the hyperacusis questionnaire (HQ) (Khalfa et al, 2002) ($r = -0.16$, $p = 0.3$) for 46 patients with tinnitus. Contrary to these reports, Blaesing and Kroener-Herwig (2012) reported a moderate and highly significant negative correlation between self reports of hyperacusis and ULLs ($r = -0.49$, $p< 0.001$) for 56 tinnitus patients. In addition, the ULLs were negatively correlated with anxiety levels ($r = -0.352$, $p< 0 .001$).

The criteria for diagnosing hyperacusis handicap based on HQ scores are also not generally agreed. Khalfa et al (2002) suggested a cutoff score of 28 as indicating hyperacusis handicap. Meeus et al (2010) suggested reducing the cutoff score to 26, while Fackrell et al (2015) suggested that the cutoff score of 28 needs to be revaluated but did not propose a definitive value.

Since ULLs and self-report questionnaires (typically the HQ) are routinely used in the audiological assessment of hyperacusis (Tyler et al, 2014; Jastreboff & Jastreboff, 2014; Aazh et al, 2011; Aazh & Moore, 2017), it is important to establish the relationship between the two and to refine the criteria that should be used to diagnose hyperacusis handicap. The aims of this study were: (1) to explore patterns of ULLs across frequency and their associated factors for patients with tinnitus and hyperacusis seen in a National Health Service (NHS) Audiology clinic, and (2) to re-evaluate the criteria for diagnosing hyperacusis based on the outcomes of the measurements of ULLs and scores for the HQ.

Methods

Study design and patients

This was a retrospective cross-sectional study conducted at the Tinnitus and Hyperacusis Therapy Specialist Clinic (THTSC), Royal Surrey County Hospital, Guildford, UK. The data for consecutive patients who attended the THTSC in 2012-14 for whom ULLs had been measured were included (n = 573). The average age of the patients was 55 years (standard deviation, SD = 17 years, range 7 to 95 years). Forty eight percent (273/571) of the patients were male.

Demographic data for the patients and the outcomes of their latest audiological investigations and their routine self-report questionnaires were imported from records held at the Audiology department. These comprised:

(1) Pure tone audiogram measured using the procedure recommended by the British Society of Audiology (BSA, 2004). This involves starting at a clearly audible level for each frequency, decreasing the level in 10-dB steps until the patient no longer responds, and then increasing the level in 5-dB steps. This is repeated to estimate the threshold level, which is the level at which the patient responds on 50% of presentations during an ascending series. All audiometric equipment had been calibrated within the past year.

(2) ULLs measured following the BSA recommended procedure (BSA, 2011). The recommended instructions, described in the introduction, were used.

(3) The following self-report questionnaires: the Tinnitus Handicap Inventory (THI; Newman et al, 1996), the Visual Analogue Scale (VAS; Maxwell, 1978) of tinnitus loudness, the Hyperacusis Questionnaire (HQ; Khalfa et al, 2002), the Hospital Anxiety and Depression Scale (HADS; Zigmond & Snaith, 1983), and the Insomnia Severity index (ISI; Bastien et al, 2001). These questionnaires are routinely given to all patients attending the THTSC for tinnitus or hyperacusis therapy, and they are described briefly below. The clinic receptionists gave the questionnaires in pen and paper format to patients on their arrival and asked them to take a seat in the waiting area and complete the questionnaires prior to being seen by their audiologist.

(4) Age and gender.

Questionnaires

The THI has 25 items, and response choices are "no" (0 points), "sometimes" (2 points) and "yes" (4 points). The overall score ranges from 0 to 100. Scores from 0 to 16 indicate no

handicap, scores from 18 to 36 indicate mild handicap, scores from 38 to 56 indicate moderate handicap, and scores from 58 to 100 indicate severe handicap (Newman et al, 1996).

VAS scores are ratings on a scale from 0 to 10. The VAS score for loudness of tinnitus was assessed by asking the patient to rate the loudness of tinnitus during their waking hours over the last month (It was explained that 0 corresponds to no tinnitus being heard and 10 is the loudest sound that they can imagine).

The HQ comprises 14 items and the response choices are "no" (0 points), "yes, a little" (1 points), "yes, quite a lot" (2 points), and "yes, a lot" (3 points). The overall score ranges from 0 to 42. Scores equal to or above a certain cut-off value indicate strong auditory hypersensitivity. Khalfa et al (2002) suggested a cutoff score of 28, while (Meeus et al, 2010) suggested a cutoff score of 26.

The HADS consists of 14 items each rated from 0 to 3 according to the severity of difficulty experienced. Eight items require reversed scoring, after which anxiety (HADS-A) and depression (HADS-D) subscale totals are calculated. Total scores for each subscale range from 0 to 21. Scores from 0-7 are classified as normal, scores from 8-10 are classified as borderline abnormal, and scores from 11-21 are classified as abnormal (Zigmond & Snaith, 1983).

The ISI comprises seven items that assess the severity of sleep difficulties and their effect on the patient’s life. 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 minimal insomnia, scores from 15-21 indicate moderate insomnia, and scores from 22-28 indicate severe insomnia (Bastien et al, 2001).

Ethical approval

This study was approved by the South West-Cornwall and Plymouth Research Ethics Committee and the Research and Development department at the Royal Surrey County Hospital.

Data analysis

The data were anonymised prior to statistical analysis. Descriptive statistics (means and SDs) for the characteristics of the patients and scores for the self-report questionnaires were calculated. Pearson correlation and t-tests were used to assess the mean differences and relationships between different measures. The *p*-value required for statistical significance was set at $p<0.05$. The variables that had significant correlations with ULLs were included in a regression model to examine whether the variables significantly influenced the ULLs. A stepwise linear multiple regression model was created to predict ULLs, beginning with a full model that included all variables. Then, variables were removed to assess whether their inclusion significantly affected the goodness of fit. Some of the patients did not complete all of the questionnaires or audiological examinations. The analyses were restricted to responders with complete data on all variables required for a particular analysis. The number of patients included in each analysis (*n*) is reported. The STATA programme (version 13) was used for statistical analyses.

Results

Characteristic of the study population

The scores on the THI, VAS, HQ, HADS, and ISI for the study population are shown in Table 1. The means and SDs of the hearing thresholds and ULLs for each ear and each frequency are shown in Table 2. The mean PTA across the frequencies 0.25 to 8 kHz was 22 dB HL (SD = 16) for both the right and left ears ($n = 566$). The mean of the average ULL across 0.25, 0.5, 1, 2, 4 and 8 kHz was 84 dB HL (SD = 14) for the right and left ears ($n = 494$). 126/573 (22%) of the patients had a ULL of 60 dB HL or less for at least one of the measured frequencies, 0.25, 0.5, 1, 2, 3, 4, 6, and 8 kHz, for at least one ear. Table 3 shows the number of individuals with ULLs of 60 dB HL or below for each of the frequencies 0.25, 0.5, 1, 2, 3, 4, 6, and 8 kHz, for the right and left ears. ULLs of 60 dB HL or below occurred most often for the frequency of 8 kHz.

TABLES 1, 2 AND 3 HERE

Diagnostic criteria for hyperacusis based on ULLs and HQ scores

In what follows, the across-frequency average ULL for the ear with lower average ULL is denoted ULLmin. For patients with scores of 26 or above on the HQ, the mean value of ULLmin was 72.7 dB HL (SD =17 dB, $n = 115$, 95% confidence interval, CI= 69.6 to 75.8 dB HL). The mean was significantly ($p < 0.001$) lower than the mean value of ULLmin for

patients with HQ scores below 26, which was 84.6 dB HL (SD = 13 dB, n = 359). If the diagnosis of hyperacusis handicap were based on the value of ULLmin, a cutoff value of 76 dB HL (the upper limit of the 95% CI) would encompass 95% of those with significant hyperacusis handicap, as indicated by an HQ score of 26 or more. The mean HQ score was 16.3 (95% CI: 15.4 to 17.2) for patients with ULLmin >76 dB HL and was 23.6 (95% CI: 22 to 25.2) for patients with ULLmin ≤76 dB HL. This is both interesting and problematic, as it indicates that more than 95% of patients with ULLmin ≤76 dB HL have HQ scores below 26, i.e. not reaching the conventional HQ score used to diagnose hyperacusis handicap.

On these grounds, we believe that the threshold for diagnosing hyperacusis handicap using the HQ needs modification. If the score on the HQ indicating hyperacusis handicap is reduced to 22 (the lower end of the 95% CI for patients with ULLmin ≤76 dB HL), then repeating the above analysis leads to a better-matched outcome between reduced ULLs and abnormal scores on the HQ. For patients with HQ scores of 22 or above, the mean value of ULLmin was 74.4 dB HL (SD =17, n = 163) which was significantly ($p < 0.001$) lower than the mean value of ULLmin for patients with HQ scores below 22 (mean = 85.6 dB HL, SD = 12, n = 311). The boundaries of the 95% CI of the mean ULLmin for patients with HQ scores of 22 or above were 71.8 and 77 dB HL. Given the upper limit of 77 dB HL, it seems reasonable to propose that hyperacusis handicap is indicated by a ULLmin value of 77 dB HL or less. This ULL-based criterion would encompass 95% of those with significant hyperacusis handicap, as indicated by HQ scores of 22 or more. The mean HQ score was 16.1 (95% CI: 15.2 to 17) for patients with average ULLs for the worse ear above 77 dB HL and was 23.5 (95% CI: 22 to 25) for patients with average ULLs of the worse ear ≤77 dB HL. In summary, with appropriate choice of the cutoff values required for the diagnosis of hyperacusis handicap, a good correspondence can be obtained between diagnoses based on ULLs and on HQ scores. Appropriate cutoff values are ULLmin ≤77 dB HL and HQ score ≥22.

Factors related to ULLs

There were significant correlations between ULLmin and scores for: the pure tone average audiometric threshold (PTA) for the ear with better hearing thresholds (r = 0.18, p<0.001, n = 507); the HQ (r = –0.43, p<0.001, n = 474), the anxiety subscale of the HADS (r = –0.21, p<0.001, n = 488), the depression subscale of the HADS (r = –0.20, p<0.001, n = 478), the

THI (r = –0.21, p<0.001, n = 475), the ISI (r = –0.15, p = 0.0015, n = 426), and age (r = 0.37, p<0.001, n = 504). There was no significant correlation of ULLmin values with the VAS score for tinnitus loudness (r = 0.03, p<0.46, n = 451).

Variables that were significantly correlated with ULLmin were included in a linear-regression model. Five variables did not significantly increase the proportion of variance predicted by the regression model. These were the HADS depression score (p = 0.99), the PTA for the ear with better hearing thresholds (p = 0.88), the THI score (p = 0.7), the HADS anxiety score (p = 0.12) and the ISI score (p = 0.23). The remaining two variables in the regression model are shown in Table 4. ULLmin values were significantly associated with age and the HQ score. However, the linear regression model explained only 30% of the variance in the ULLmin values.

Between-ear differences in average ULL

Most of the patients had similar across-frequency average ULLs for the two ears. However, 40/486 patients (8.2%) had a between-ear difference of 10 dB or more. The mean between-ear difference in PTA for these patients was 11.8 dB (SD = 16) which was significantly higher (p<0.001) than the mean for the remainder of the population, which was 5.4 dB (SD = 8). However, among the patients with a between-ear ULL difference of 10 dB or more, 55% had a between-ear difference in their PTA less than 5 dB.

For patients with a between-ear difference in ULL of 10 dB or more, the mean score on the HQ was 22 (SD = 8). This was significantly higher (worse) than the mean HQ score of 17.6 (SD = 9.5) for the remainder of the patients (p = 0.007). Thus, a large interaural asymmetry in ULLs is associated with a higher HQ score. There were no significant differences in anxiety and depression scores as measured via the HADS between patients with interaural asymmetry in ULL ≥10 dB and <10 dB (p = 0.33 and p = 0.2, respectively).

For 6/487 patients (1.2%), the interaural asymmetry in ULL was over 20 dB, the largest value being 27 dB. For these patients, the mean difference in PTA between ears was 4.8 dB (SD = 4), with a minimum of 2 dB and a maximum of 11 dB. Eighty percent of these patients had an across-ear difference in PTA less than 6 dB. Thus, it seems that most cases of large interaural asymmetry in ULLs cannot be explained by interaural differences in PTA.

Variation in ULLs across frequency

The upper part of Table 5 shows the mean differences and the absolute values of the mean differences in ULLs between 8 and 1 kHz (i.e., ULL at 8 kHz minus ULL at 1 kHz), referred to here as 8-1 slope, between 0.25 and 1 kHz, referred to here as 0.25-1 slope, and between 8 and 0.25, referred to here as 8-0.25 slope. The slope values are shown separately for the right and left ears and for the mean across ears (for the slopes preserving the sign). The lower part of Table 5 shows corresponding slope values for the hearing thresholds (HTs).

The mean 8-1 slope across ears was –2.1 dB (SD =11), indicating that for many patients the ULL was lower (worse) at 8 than 1 kHz. However, the mean 8-1 slope in HTs across ears was about 22 dB (SD = 22), indicating that HTs were generally higher (worse) at 8 kHz than at 1 kHz. Thus the trend for lower ULLs at 8 than at 1 kHz occurred despite the fact that HTs were generally higher at 8 than at 1 kHz.

For 16% of patients (80/509) the absolute value of the 8-1 ULL slope for the right ears was ≥20 dB. For 69% of these patients (55/80) the 8-1 ULL slope (including the sign) for the right ears was ≤–20 dB; the ULLs at 8 kHz were 20 to 45 dB lower than the ULLs at 1 kHz. Of these patients, 23% (18/78) had lower HTs at 8 than at 1 kHz and 77% (60/78) had equal or higher HTs at 8 kHz than at 1 kHz. Similarly, for 14% of patients (72/504) the absolute value of the 8-1 ULL slope for the left ears was ≥20 dB. For 65% of these (47/72), the 8-1 ULL slope was ≤–20 dB for the left ears; the ULLs at 8 kHz were 20 to 45 dB lower than the ULLs at 1 kHz. Of these patients, 19.5% (14/72) had lower HTs at 8 than at 1 kHz and 80.5% (58/72) had equal or higher HTs at 8 kHz than at 1 kHz. Overall, these results confirm that ULLs are often markedly lower at 8 than at 1 kHz, whether or not the HT is lower at 8 than at 1 kHz.

TABLES 4 AND 5 HERE

For the slopes averaged across ears, correlation analyses showed significant relationships between the 8-1, 0.25-1, and 8-0.25 ULL slopes and the 8-1, 0.25-1, and 8-0.25 HT slopes (Table 6). The correlations were strongest, and were positive, for cases where the HT slopes were mostly positive, i.e. for the 8-1 and 8-0.25 slopes.

The mean score on the HQ was 22 (SD = 8.7) for patients with absolute ULL 8-1 slope across ears ≥20 dB (n = 51). This HQ score was significantly higher (worse) than the HQ score of 17.5 (SD = 9.5) for patients with absolute ULL 8-1 slope across ears <20 dB (n = 409) (p=0.0012). Thus, large across-frequency changes in ULL are associated with poorer HQ scores.

There were no significant differences between the groups with absolute ULL 8-1 slopes across ears above or below 20 dB in anxiety or depression scores as measured via the HADS (p = 0.19 and p = 0.27, respectively).

Discussion

Diagnostic criteria

There is an urgent need for more precise and more consistent diagnostic criteria for hyperacusis, both to guide treatment in clinical settings and for research studies investigating basic mechanisms of hyperacusis and evaluating different treatment options. Previous researchers have suggested that there is a need to modify the cutoff score for the HQ required to diagnose hyperacusis handicap (Fackrell et al, 2015; Meeus et al, 2010). Our results showed that a diagnosis of hyperacusis handicap based on HQ scores can be made reasonably consistent with a diagnosis based on ULLs if the following cutoff scores are adopted for a positive diagnosis: the average ULL at 0.25, 0.5, 1, 2, 4 and 8 kHz for the ear with the lower average ULL, ULLmin, should be ≤77 dB HL and the HQ score should be ≥22. With these cutoff values, 95% of patients with HQ scores meeting the criterion will also meet the criterion based on ULLs, and vice versa. Hence, we recommend that the cutoff score for diagnosing hyperacusis handicap based on the HQ score should be reduced from 26 to 22. However, these cutoff values for ULLs and HQ scores lead only to a binary decision; hyperacusis handicap is either present or absent. Further work needs to be conducted to determine the relationship between the severity of hyperacusis and ULLs and HQ scores.

Factors related to ULLs

The regression model showed that ULLmin values increased significantly with increasing age. The origin of this effect is not clear. It may partly reflect the fact that audiometric thresholds tended to increase with increasing age. However, in the regression model, the effect of PTA for the better ear was not significant, while the effect of age was significant, so

the association of ULLmin with age probably cannot be fully attributed to changes in PTA with age. It is possible that the effects of age are related to degeneration of auditory neurons; loss of the neurons that typically respond at high sound levels could lead to reduced loudness of high-level sounds, and hence higher ULLs, while having little or no effect on audiometric thresholds (Makary et al, 2011; Sergeyenko et al, 2013).

The regression analysis also showed that ULLmin values tended to decrease with increasing HQ score. Consistent with the latter effect, Zaugg et al (2016) reported a weak negative correlation between across-frequency average ULLs and self-reported sound tolerance problems among 139 patients with tinnitus ($r = –0.23$, $p<0.05$). However, Meeus et al (2010) assessed ULLs for 46 tinnitus patients with or without hyperacusis, most of whom had mild high-frequency hearing loss, and, in contrast to our results, they did not find any significant relationship between ULLs and scores on the HQ ($r = –0.16$, $p = 0.3$). The discrepancy probably occurred because only a few of their patients had sound tolerance problems: the mean ULL for their population was over 100 dB HL, which is higher than the mean ULL for our sample, and even among patients reported to have low tolerance to sounds, the mean HQ score was only 20.

Our data clearly show that high HQ scores are associated with low ULLmin values. However, the linear regression model explained only 30% of the variance in the ULLmin values, indicating that a large amount of the variance in the ULLmin values is not accounted for. There is a need for more research exploring the factors related to ULLs in patients with hyperacusis.

Relationship between ULLs and HTs

One theory of tinnitus and hyperacusis is based on the idea that peripheral hearing damage (hearing loss) leads to increased central gain to compensate for the reduced input from the periphery (Salvi et al, 1990). In the case of hyperacusis, this increased gain might be applied (inappropriately) to sounds of all levels, resulting in the perception of medium and high-level sounds as being too loud. If this theory is applicable to most cases of hyperacusis, then ULLs should be inversely related to HTs. On the other hand, one might argue that hearing loss would lead to reduced loudness for most sounds, thereby offsetting the effect of increased central gain. Thus it is difficult to make firm predictions based on this theory.

Several authors have reported that ULLs become lower for patients with unaided hearing loss or after a period of reduced auditory input produced by the use of ear plugs (Formby et al, 2003; Hamilton & Munro, 2010; Munro & J., 2009). However, it has also been reported that there is no relationship between ULLs and PTAs for patients with hyperacusis (Anari et al, 1999; Formby et al, 2007). Sheldrake et al (2015) reported that across-frequency average ULLs were positively correlated with PTAs for 381 patients with hyperacusis ($r = 0.36$, $p<0.01$). However, the correlation was mainly driven by the fact that the ULL at a given frequency cannot be lower than the HT at that frequency, so if the HT is high, for example 100 dB HL, the ULL must also be high. For patients with no or mild hearing loss, there was a very wide range of ULL values. Our patients mostly had no or mild to moderate hearing loss and our results showed a small but significant correlation between ULLmin and the PTA for the ear with better hearing thresholds ($r = 0.18$, $p<0.001$, $n = 507$). Overall, these results do not support the simple prediction, based on the theory of increased central gain, that ULLs should decrease as HTs increase. However, as noted above, it is difficult to make firm predictions from this theory, since increasing hearing loss will generally lead to reduced loudness at any given frequency (Moore & Glasberg, 2004), offsetting any possible effects of increased central gain.

Interaural asymmetry

There was a between-ear difference in across-frequency average ULLs of 10 dB or more for 40/486 patients (8.2%). For 6/487 patients (1.2%) the between-ear difference was over 20 dB, with a maximum of 27 dB. For patients with a between-ear difference in ULL of 10 dB or more, the mean score on the HQ of 22 was significantly higher than the mean HQ score of 17.6 for the remainder of the patients. Thus, a large interaural asymmetry in ULLs is associated with a higher HQ score.

There is little information in the literature about between-ear differences in ULLs for patients with hyperacusis. Anari et al (1999) reported that 27% of their patients had self-reported hypersensitivity to sound in one ear only. However, they did not report ULL values and did not analyse differences between patients with unilateral and bilateral complaints. Formby et al (2007) reported that less than one percent (1/68) of their patients had unilaterally reduced ULLs. More recently Juris et al (2013) used ULLs below 90 dB HL in one or both ears (averaged across 0.5, 1, 2 and 3 kHz) as one of their criteria for including patients in a study

of hyperacusis. They reported that 95% (59/62) of the included participants met the criteria for both ears. No further details were given, so the magnitude of any interaural asymmetry is unknown.

A large between-ear difference in ULLs might indicate some specific abnormality in monaural pathways. Further studies of people with large between-ear differences in ULL could give some insight into whether the intolerance to sound is mainly due to increased loudness perception in the ear with lower ULLs (which could be studied by comparing the loudness of sounds presented alternately to the two ears), to a dislike of specific sounds when presented to that ear, or to a dislike of a class of sounds with certain spectral characteristics.

If a global psychological or neurological component is predominant in producing hyperacusis, then it seems unlikely that it would affect one ear more than the other. Consistent with this, most of our patients and those in the studies reviewed above had similar ULLs for the two ears. However, our results showed no significant differences in anxiety and depression scores as measured via the HADS between patients with between-ear differences in ULL of 10 dB or more and those with differences less than 10 dB.

Differences across frequencies

The 8-1 ULL slope was ≥20 dB for the right ears of 47/504 patients and for the left ears of 55/509 patients. Past studies have typically shown that ULLs averaged across participants did not change markedly across the frequency range (Formby et al, 2007; Meeus et al, 2010; Sheldrake et al, 2015), but the authors did not report the ULL variations across frequency for individual patients. In our study the mean score on the HQ was 22 (SD = 8.7) for patients with absolute 8-1 ULL slopes across ears ≥20 dB (n = 51), which was significantly higher (worse) (p<0.001) than the HQ score of 17.5 (SD = 9.5) for patients with absolute 8-1 ULL slopes across ears <20 dB (n = 409). Hyperacusis handicap may typically be characterized by strong across-frequency variations in ULL.

The most common frequency associated with ULLs of 60 dB HL or below was 8 kHz; this occurred for 76 patients. This is consistent with the finding of Sheldrake et al (2015) that the mean ULL at 8 kHz for patients with hyperacusis was about 7 dB lower than the average ULL at 0.25, 5, 1, 2, 3, 4, and 6 kHz. Our finding is also consistent with the results of de

Klaver et al (2007), who assessed ULLs for 15 patients with regional pain syndrome and hyperacusis; the mean ULLs across patients were 45, 55, 55, 50, 55, and 45 dB HL at 0.25, 0.5, 1, 2, 4, and 8 kHz, respectively. Thus, for these patients, the (remarkably low) mean ULLs were lower at 0.25 and 8 kHz than at other frequencies. The ULLs for our study population showed a similar trend, but the variation across frequency was less pronounced, possibly because our study population included people whose primary complaint was tinnitus rather than hyperacusis.

Formby et al (2007) did report ULLs separately for tinnitus patients with hyperacusis as their main complaint (135 ears, 68 patients) and patients with tinnitus only (140 ears, 70 patients). For the hyperacusis group, the mean ULLs were approximately 90 dB HL at 1, 2, and 4 kHz and 95 dB HL at 8 kHz; ULLs at 8 kHz were not lower than for other frequencies. This may have been a consequence of the fact that the audiometric thresholds of their patients at high frequencies were higher (worse) than those of our study population.

The strong across-frequency variations in ULLs for our patients classified as having hyperacusis might be an indication of adverse reactions only to specific sounds, which is consistent with the definitions of annoyance and fear hyperacusis (Tyler et al, 2014) and misophonia (Cavanna & Seri, 2015; Kumar et al, 2017; Jastreboff & Jastreboff, 2014). Future studies should explore the pattern of ULLs for individual patients and their relationship to the everyday sounds that are found to be aversive by the patients.

Conclusions

We have shown that a diagnosis of hyperacusis handicap based on ULLs can be made consistent with a diagnosis based on HQ scores by appropriate choice of cutoff values for the two measures. Recommended cutoff values are ULLmin ≤77 dB HL and HQ score ≥22. With these cutoff values, 95% of patients with ULLmin values meeting the criterion will also meet the criterion based on HQ scores, and vice versa. Values of ULLmin tend to decrease with increasing HQ score and to increase with increasing age.

Hyperacusis handicap is often associated with strong across-frequency variation in ULLs. Strong between-ear differences in ULL occur only infrequently, but such differences are associated with higher HQ scores. ULLmin values are not strongly associated with PTAs.

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TABLE 1. Means and SDs of scores of the study population on the tinnitus handicap inventory (THI), the hyperacusis questionnaire (HQ), the visual analogue scale (VAS) of tinnitus loudness, the hospital anxiety and depression scale (HADS), and the insomnia severity index (ISI). The number of patients is indicated by *n*.

Questionnaire	*n*	Mean	SD
THI	530	44.7	24
HQ	527	18	9.5
VAS (Tinnitus loudness)	504	6	2
HADS (Anxiety)	546	8.6	4.6
HADS (Depression)	545	6	4.4
ISI	477	12	7.2

TABLE 2. Means (SD) of hearing thresholds (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	17.8 (15) *n*=567	18.7 (15.3) *n*=571	19.3 (16.9) *n*= 571	21.7 (19) *n*=571	27.5 (19.9) *n*= 502	32.3 (22.2) *n*=570	38.7 (24.4) *n*= 513	40.5 (27.9) *n*=566
HT Left	17.6 (16.3) *n*=568	18.7 (16.8) *n*=569	18.6 (16.6) *n*=569	22 (19.1) *n*=569	29.1 (20.9) *n*= 505	34 (22.8) *n*=569	40 (24.1) *n*= 511	41.2 (27.2) *n*= 567
ULL Right	83.1 (14.4) *n*=517	84.7 (14.6) *n*=567	85.2 (14) *n*=573	84.6 (15) *n*=571	86.3 (14.7) *n*=344	85.7 (15.5) *n*=562	86.5 (16.4) *n*=376	82.3 (18.4) *n*=509
ULL Left	83 (14.5) *n*=509	84.4 (14.7) *n*=559	84.6 (14.3) *n*=565	84.38 (15.2) *n*=564	86.1 (15) *n*=345	86 (16) *n*=555	86.3 (16) *n*=376	82.4 (18.1) *n*=503

TABLE 3. The number of patients who had a ULL of 60 dB HL or less at each frequency for the right and left ears (n = 573).

	Frequency, kHz							
	0.25	0.5	1	2	3	4	6	8
Right ear	49 (8.6%)	49 (8.6%)	39 (6.8%)	49 (8.6%)	28 (4.9%)	58 (10.1%)	33 (5.8%)	76 (13.3%)
Left ear	54 (9.4%)	51 (8.9%)	44 (7.7%)	54 (9.4%)	31 (5.4%)	54 (9.4%)	37 (6.5%)	69 (12%)

TABLE 4. Variables included in the final version of the stepwise linear regression model for predicting ULLmin (n = 398), together with regression coefficients, p values, and 95% CI values. Variables are listed according to the value of the regression coefficient (highest first).

	Regression coefficient	p value	95% CI	
HQ score	–0.6	<0.001	–0.73	–0.47
Age	0.3	< 0.001	0.21	0.38

TABLE 5. The mean differences and the means of the absolute values of the differences (with SDs) in ULLs and HTs between 1 and 8 kHz, 0.25 and 1 kHz, and 0.25 and 8 kHz for the right and left ears. The number of patients included in each analysis is indicated by *n*.

	8-1 kHz	Maximum difference	0.25-1 kHz	Maximum difference	8-0.25 kHz	Maximum difference
ULL Right	–1.6 (12) *n*=504	45	–1.25 (8) *n*=511	35	–0.4 (13) *n*=499	60
ULL Right (abs)	8.2 (8)		5.2 (6)		8.8 (9)	
ULL Left	–2.3 (12) *n*=509	50	–1.7 (8) *n*=517	50	–0.7 (14) *n*=501	65
ULL Left (abs)	8.5 (9)		5.1 (6)		9.2 (10)	
ULL across ears	–2.1 (11) *n*=495		–1.4 (7) *n*=508		–0.75 (12) *n*=490	
HT Right	22.7 (23) *n*=567	85	–0.93 (11) *n*=568	65	23.7 (25) *n*=567	90
HT Right (abs)	25.2 (20)		7.2 (8.4)		26.8 (22)	
HT Left	21.6 (23) *n*=566	90	–1.4 (10) *n*=567	45	22.8 (25) *n*=563	95
HT Left (abs)	24.5 (20)		7 (8)		26 (22)	
HT across ears	22.1 (22) *n*=562		–1.2 (9) *n*=564		23.1 (24) *n*=559	

TABLE 6. Correlations (and *p* values) between the slopes of the ULLs and slopes of the HTs, calculated over different frequency ranges.

	8-1 slope HT	0.25-1 slope HT	8-0.25 slope HT
8-1 slope ULL	0.48 ($p < 0.001$) $n = 487$		
0.25-1 slope ULL		0.2 ($p<0.001$) $n = 502$	
8-0.25 slope ULL			0.54 ($p<0.001$) $n = 480$