



Validation of the Portuguese Version of Hyperacusis Questionnaire and Comparison of Diagnostic Skills with Loudness Discomfort Levels

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Keywords

Hyperacusis, Questionnaire, Validation, Tinnitus

Abbreviations

HQ: Hyperacusis Questionnaire; LDL: Loudness Discomfort Levels; dB SL: deciBel Sensation Level

Introduction

Hyperacusis is a condition entailing increased sensitivity or unusual intolerance to ordinary environmental sounds that would not usually be bothersome, threatening or uncomfortably loud to most individuals, but cause significant distress and impairment in social activities due to physical discomfort, pain or fear of sounds [1-3]. Such negative reaction is related to the physical characteristics of the sounds, and not to the frequency or quality of sounds [4]. Baguley, et al. considered hyperacusis as a central auditory reaction modulated by emotional states such as fear and anxiety [5] and Tyler, et al. discriminates four clinical components of hyperacusis: Loudness, annoyance, fear and pain [6].

A patient's negative reactions may occur in response to any, including low, level of sound. It is postulated that decreased sound tolerance consists of two components: hyperacusis and misophonia, which frequently occur together [4].

Misophonia is defined as an abnormally strong reaction to a sound with a specific pattern and meaning to a given subject. The physical characteristics of the sound are secondary. Reactions to the sound depend on the subject's past history and on non-auditory factors such as the subject's previous evaluation of the sound, his/her belief that the sound is a potential threat or that exposure to it will be harmful, the subject's psychologic profile, and the context in which the sound is presented [7-9].

This symptom has been demanding growing interest because: 1) Its prevalence reaches 8 to 15% of the population [10,11]; 2) It is associated to hearing loss, tinnitus, autism, migraine and other sensory hypersensitivity disorders [12]; 3) It compromises emotional status, sleep, and concentration [13,14], turning familiar, professional and social lives difficult.

There is enough evidence of a positive correlation between hyperacusis and tinnitus [4,14-17]. Thus, having a reliable measurement of hyperacusis would be of interest for managing patients with one or another symptom [18].

The association of a clinical history of discomfort to different daily sounds and altered measurements of Loudness Discomfort Levels (LDL) is the most acceptable diagnosis of hyperacusis, although results lack standardization [4,18,19].

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The thresholds of LDL are decreased in such patients in most frequencies between 500 and 8000 Hz in one or both ears [18-21]. It can be classified as: Mild hyperacusis when LDL is at 80-90 dB in 2 or more frequencies, moderate hyperacusis in presence of LDL at 65-75 dB in 2 or more frequencies and severe hyperacusis in presence of LDL at 60 dB or lower in 2 or more frequencies [19].

Common limitations of measuring LDL are related to the strong dependence on the instruction given. Different instructions may lead to a difference greater than 20 dBHL, therefore the test should be carefully performed [18,22,23] and associated to the clinical history. Moreover, clinical cut-off values of LDL differ among authors (< 90 dBHL [24], < 70 dBHL [18] and between 60-85 dBHL [25]) and test-retest reliability has been questioned [21]. Therefore, the further use of a validated questionnaire might help in standardizing diagnosis, screening the presence of, and establishing severity/impairment levels of hyperacusis.

The Hyperacusis Questionnaire (HQ) was first elaborated by Khalfa, et al. to quantify and characterize hyperacusis [26]. The first part consists of three binary questions that collect information on auditory disorders and noise exposure. The second part consists of 14 questions with a 4-point scale answer each: “no” (0 point), “yes, a little” (1 point), “yes, quite a lot” (2 points), and “yes, a lot” (3 points). Such questions evaluate three major dimensions: Attentional (questions 1-4), social (questions 5-10) and emotional (questions 11-14) with satisfactory internal consistency reliability. The total score ranges from 0 to 42; the higher the score, the greater the hypersensitivity.

The HQ has already been translated into Dutch [27], Italian [28] and Japanese [29] versions that were subsequently validated. Since differences in symptom expression among cultural groups might compromise the validity of the questionnaire [30], we aimed at performing the cross-cultural adaptation and validation of the Portuguese version of HQ, and compared our results to Loudness Discomfort Levels results.

Statistical Analysis

Quantitative personal characteristics and the results of the questionnaires and quantitative scales have been reported in patients using summary measures (mean, standard deviation, median, minimum and maximum) and evaluated qualitative characteristics were described using absolute and relative frequencies [31]. Each issue of hyperacusis questionnaire were described in the two evaluations carried out and evaluated the reproducibility of the questionnaire for each question using the weighted Kappa coefficient with the respective intervals with 95% confidence [32]. The total questionnaire was described

in each evaluation and verified the reproducibility of this method with use of the Intraclass Correlation Coefficient (ICC), with its range with 95% confidence [32]. To check the questionnaire construct validation, it was compared between sexes using Mann-Whitney test [31] and verified the correlation with age using Spearman correlation [31]. Internal consistency was verified the questions of the questionnaire and the total with use of Cronbach's alpha [31], each item removed from the questionnaire and evaluated in the presence of other items. The concurrent validity of the questionnaire was tested using Spearman's total questionnaire correlated with assessments at each frequency of LDL and also compared the total questionnaire between categories of LDL classification using the Kruskal-Wallis test, followed by Dunn's multiple comparisons [33]. The tests were performed at the 5% significance level.

Methods

After receiving approval of the Ethics committee of Hospital Universitário of University of São Paulo, this prospective cross-sectional study was conducted in two phases from August 2015 to March 2016:

- Phase 1: Cross-cultural adaptation and validation of the Portuguese version of HQ: This phase was conducted following the criteria established by Guillemin, et al. [30], which consisted of four steps. First step: Two native Portuguese speakers from Brazil, who were bilingual in English and knew the purpose of the study, were asked to independently translate the original questionnaire from English into Brazilian Portuguese after receiving the permission of the author; Second step: Both original and translated versions were analyzed by a team of professionals (two otolaryngologists and one linguistic professor), which led to a final single version that maintained linguistic consistency while keeping the basic characteristics of the concepts of the original questionnaire; Third step: the final version was translated back into English and compared with the original questionnaire by two other native speakers who did not know the purpose of the study; Fourth step: After a critical analysis of the linguist professor that there were no differences between the original and translated versions, we proceeded to the application and reapplication of the final Portuguese version of HQ in a sample of subjects whose data is displayed below. Data on the clinical application and reapplication of HQ was further analyzed for validation of the questionnaire and establishing a cut-off score for diagnosing hyperacusis. The patients answered questionnaires by themselves. We were available to help if they had any issues understanding the items.

- Phase 2: Comparison between results obtained from HQ and Loudness Discomfort Levels (LDL). The sample

selected to undergo the fourth step of Phase 1 and Phase 2 included all consecutive patients who spontaneously attended a medical appointment in a tinnitus and hyperacusis center, from November 2015 to March 2016, regardless of their main complaints. They were invited to participate and signed the informed consent. The exclusion criteria were patients under 18-years-old and those who refused to undergo any step of the research. The final sample was composed by 136 subjects (100% Caucasians, 64.7% males, 35.3% females). Age varied from 14-89 years (mean = 49.8 years; SD ± 14.3; median = 49).

All patients were asked by the physician assistant to fill out by themselves the Portuguese version of HQ in the waiting room, before medical consultation starts. If they had any doubt, a physician would help to make unclear questions understandable. Among the 136 patients, 101 (74.2%) answered the questionnaire for the second time after the appointment to check for test-retest reliability. Pure-tone audiometry was performed in an acoustic chamber in frequencies from 0.25-16 kHz. Patients who had tinnitus were further submitted to pitch and loudness matching [34]. Loudness Discomfort Levels (LDLs) were obtained for both ears in frequencies from 0.5-4 kHz. The stimuli were presented from 50 dBHL, ascending in steps of 5 dB [22]. Patients were instructed to indicate when the sound delivered through the earphone from the audiometer became uncomfortable, not just loud. The standardized verbal instruction made by the audiologist was: "You will hear sounds that will become louder. Please, press the button when the sound reaches an intensity that you no longer want to hear it and the sound will stop immediately. We want to know which intensity provokes discomfort and not to know if the sound is strong or weak. The sound can be strong and not provoke any hearing discomfort, for example. This test does not offer risk to your hearing even if you hear a sound in the maximum intensity of this equipment" [22]. Two trials were used for each loudness measure to ensure reliability. We adopted the classification of hyperacusis as suggested by Goldstein and Shulman [19] (Table 1).

The quantitative personal characteristics, results of the questionnaires and quantitative scales were statistically reported using summary measures (mean, standard deviation,

Table 1: Classification of hyperacusis according to loudness discomfort levels [14].

Hyperacusis classification	LDL level
None/negative	95 dB or greater in all frequencies
Mild	80-90 dB in 2 or more frequencies
Moderate	65-75 dB in 2 or more frequencies
Severe	60 dB or lower in 2 or more frequencies

median, minimum and maximum). The score of each question and the total score of HQ were described in both test and retest evaluations, and their reproducibility was evaluated through weighted Kappa coefficient and Intraclass Correlation Coefficient (ICC) with 95% confidence intervals. Internal consistency of each question and the total score was verified through Cronbach's alpha, being each question removed from the questionnaire and evaluated in the presence of other questions. The concomitant validity of the questionnaire was tested using Spearman's total questionnaire correlated with assessments at each frequency of

Questionário de Hiperacusia
(tradução para o Português e adaptação cultural de Khalfa et al, 2002)

Nome: _____
 Sexo: () masculino () feminino Idade: _____ Profissão: _____ Endereço: _____
 Contatos: _____

Você está ou tem estado exposto a ruído
 Você tolera menos barulho quando comparado há poucos anos atrás? Você já teve problemas auditivos? Se sim, quais?

		Nio	Sim, pouco	Sim, razoavelmente	Sim, bastante
1	Você já usou tampões por dentro ou por fora dos ouvidos para diminuir a percepção de ruídos (não considere o uso de proteção auditiva durante situações de ruído anormalmente alto)				
2	Você acha difícil ignorar os sons ao seu redor no dia-a-dia?				
3	Você tem dificuldade para ler em locais ruidosos?				
4	Você tem dificuldade para se concentrar em ambientes ruidosos?				
5	Você tem dificuldade para manter conversas em locais ruidosos?				
6	Alguém já lhe disse que você tolera pouco o ruído ou certos tipos de som?				
7	Você é sensível ou particularmente incomodado pelo barulho da rua?				
8	Você acha o ruído desagradável em certas situações sociais (casa noturnas, bares, concertos, coquetéis, fogos de artifício)?				
9	Quando lhe propõem alguma coisa (sair, ir ao cinema, concerto), você já pensa logo no ruído que terá que enfrentar?				
10	Você já recusou convite ou não saiu de casa por causa do barulho a que poderia ser exposto?				
11	Os ruídos ou sons específicos incomodam você mais em um local calmo do que em ambiente com leve som leve de fundo?				
12	O estresse e o cansaço reduzem sua capacidade de se concentrar em ambiente ruidoso?				
13	Você é menos capaz de se concentrar em ambiente ruidoso quando chega no final do dia?				
14	O ambiente ruidoso ou determinados sons lhe causam estresse ou irritação?				

Figure 1: Brazilian Portuguese version of Hyperacusis Questionnaire.

LDL and also compared the total questionnaire between categories of LDL classification using the Kruskal-Wallis test and Dunn's multiple comparisons. The tests were performed at the 5% significance level.

Results and Discussion

Validity and reliability of HQ questionnaire in a Brazilian sample of patients after cross-cultural adaptation

The cross-cultural adaptation of HQ aimed to find equivalence between English and Portuguese versions. Small changes were performed to clarify questions 11 to 14, with no change in the meaning, as soon as we noticed that few subjects repeatedly argued about them (Figure 1).

The quantitative analysis of answers of each item, obtained in the self-application of HQ, is shown in Figure 2. The average total score was 14.2 (SD = 7.9).

Considering the comparisons between test and retest applications (four weeks after first application) of HQ in the 101 subjects who agreed to retest the questionnaire, the results point to:

- a high total reproducibility of HQ (Figure 3), even the reproducibility/agreement of each issue has not been so high, with coefficients close to 0.5, the total of the questionnaire showed reproducibility/higher agreement (ICC = 0.881), very close to 1, indicating high reproducibility of the total questionnaire.

Interclass Coefficient Correlation was > 0.8 (ICC = 0.881), but reproducibility of 5 questions was questioned (concordance coefficient < 0.5).

Two points deserve attention when considering the studies that evaluated HQ: The sample selection and the mean score for diagnosing hyperacusis. Khalifa, et al. selected the sample among the general population, while Meeus, et al., Fioretti, et al., Fackrell, et al., Onishi, et al. [14,26-29] and ourselves studied subjects with primary complaint of tinnitus. Despite a few differences in the target population, the mean score obtained in most studies were quite similar, ranging from 11 to 15. Our mean value was 14.2 (SD ± 7.9), in agreement with the other studies.

Loudness discomfort levels and comparison with HQ

Considering the classification described by Goldstein and Shulman [19], 47.1% (64/136 patients) had normal results (≥ 95 dB), while the 52.9% (72/136 patients) had abnormal LDL, which were distributed in 35.3%, 13.2%

Data	Description (n = 136)	Data	Description (n = 136)
Q1 - n (%)		Q8, n (%)	
No	110 (80,9)	No	27 (19,9)
Yes, a little	15 (11)	Yes, a little	44 (32,4)
Yes, quite a lot	6 (4,4)	Yes, quite a lot	41 (30,1)
Yes, a lot	5 (3,7)	Yes, a lot	24 (17,6)
Q2, n (%)		Q9, n (%)	
No	49 (36)	No	102 (75)
Yes, a little	46 (33,8)	Yes, a little	19 (14)
Yes, quite a lot	26 (19,1)	Yes, quite a lot	5 (3,7)
Yes, a lot	15 (11)	Yes, a lot	10 (7,4)
Q3, n (%)		Q10, n (%)	
No	48 (35,3)	No	110 (80,9)
Yes, a little	49 (36)	Yes, a little	13 (9,6)
Yes, quite a lot	14 (10,3)	Yes, quite a lot	4 (2,9)
Yes, a lot	25 (18,4)	Yes, a lot	9 (6,6)
Q4, n (%)		Q11, n (%)	
No	21 (15,4)	No	30 (22,1)
Yes, a little	59 (43,4)	Yes, a little	48 (35,3)
Yes, quite a lot	26 (19,1)	Yes, quite a lot	28 (20,6)
Yes, a lot	30 (22,1)	Yes, a lot	30 (22,1)
Q5, n (%)		Q12, n (%)	
No	26 (19,1)	No	54 (39,7)
Yes, a little	50 (36,8)	Yes, a little	39 (28,7)
Yes, quite a lot	34 (25)	Yes, quite a lot	25 (18,4)
Yes, a lot	26 (19,1)	Yes, a lot	18 (13,2)
Q6, n (%)		Q13, n (%)	
No	91 (66,9)	No	64 (47,1)
Yes, a little	19 (14)	Yes, a little	40 (29,4)
Yes, quite a lot	19 (14)	Yes, quite a lot	22 (16,2)
Yes, a lot	7 (5,1)	Yes, a lot	10 (7,4)
Q7, n (%)		Q14, n (%)	
No	56 (41,2)	No	18 (13,2)
Yes, a little	39 (28,7)	Yes, a little	47 (34,6)
Yes, quite a lot	26 (19,1)	Yes, quite a lot	34 (25)
Yes, a lot	15 (11)	Yes, a lot	37 (27,2)
TOTAL			
Mean (SD)	14,2 (7,9)		
Median (min;max)	12 (2-37)		

Figure 2: Quantitative analysis of the answers obtained in the self-application of Portuguese version of HQ (n = 136) - a high internal consistency (Figure 3), since the Cronbach's Alpha to all 14 questions was 0.846 (ranging from 0.820 - Q4 - to 0.849 - Q11). Moreover, there was no significant difference in the scores between genders (p = 0.146) or among different ages (r = -0.058, p = 0.499).

Question number/ (SD)	Questionnaire		Concordance coefficient	Question number/ (SD)	Questionnaire		Concordance coefficient
	First application (n = 101)	Second application (n = 101)			First application (n = 101)	Second application (n = 101)	
Q1, n (%)			0,5 (0,300; 0,702)	Q8, n (%)			0,417 (0,289; 0,545)
No	82 (81,2)	80 (79,2)		No	21 (20,8)	17 (16,8)	
Yes, a little	14 (13,9)	12 (11,9)		Yes, a little	35 (34,7)	23 (22,8)	
Yes, quite a lot	3 (3)	6 (5,9)		Yes, quite a lot	29 (28,7)	32 (31,7)	
Yes, a lot	2 (2)	3 (3)		Yes, a lot	16 (15,8)	29 (28,7)	
Q2, n (%)			0,367 (0,226; 0,507)	Q9, n (%)			0,609 (0,442; 0,777)
No	35 (34,7)	52 (51,5)		No	75 (74,3)	76 (75,2)	
Yes, a little	33 (32,7)	22 (21,8)		Yes, a little	15 (14,9)	15 (14,9)	
Yes, quite a lot	21 (20,8)	16 (15,8)		Yes, quite a lot	5 (5)	3 (3)	
Yes, a lot	12 (11,9)	11 (10,9)		Yes, a lot	6 (5,9)	7 (6,9)	
Q3, n (%)			0,564 (0,434; 0,694)	Q10, n (%)			0,554 (0,388; 0,721)
No	35 (34,7)	39 (38,6)		No	83 (82,2)	77 (76,2)	
Yes, a little	33 (32,7)	32 (31,7)		Yes, a little	8 (7,9)	10 (9,9)	
Yes, quite a lot	14 (13,9)	14 (13,9)		Yes, quite a lot	4 (4)	9 (8,9)	
Yes, a lot	19 (18,8)	16 (15,8)		Yes, a lot	6 (5,9)	5 (5)	
Q4, n (%)			0,626 (0,517; 0,735)	Q11, n (%)			0,346 (0,199; 0,493)
No	18 (17,8)	30 (29,7)		No	23 (22,8)	29 (28,7)	
Yes, a little	43 (42,6)	34 (33,7)		Yes, a little	40 (39,6)	30 (29,7)	
Yes, quite a lot	20 (19,8)	16 (15,8)		Yes, quite a lot	18 (17,8)	22 (21,8)	
Yes, a lot	20 (19,8)	21 (20,8)		Yes, a lot	20 (19,8)	20 (19,8)	
Q5, n (%)			0,590 (0,478; 0,701)	Q12, n (%)			0,478 (0,344; 0,613)
No	24 (23,8)	27 (26,7)		No	40 (39,6)	35 (34,7)	
Yes, a little	34 (33,7)	33 (32,7)		Yes, a little	27 (26,7)	28 (27,7)	
Yes, quite a lot	24 (23,8)	20 (19,8)		Yes, quite a lot	18 (17,8)	19 (18,8)	
Yes, a lot	19 (18,8)	21 (20,8)		Yes, a lot	16 (15,8)	19 (18,8)	
Q6, n (%)			0,618 (0,485; 0,752)	Q13, n (%)			0,322 (0,180; 0,463)
No	69 (68,3)	73 (72,3)		No	51 (50,5)	52 (51,5)	
Yes, a little	17 (16,8)	10 (9,9)		Yes, a little	27 (26,7)	16 (15,8)	
Yes, quite a lot	9 (8,9)	9 (8,9)		Yes, quite a lot	18 (17,8)	19 (18,8)	
Yes, a lot	6 (5,9)	9 (8,9)		Yes, a lot	5 (5)	14 (13,9)	
Q7, n (%)			0,557 (0,433; 0,681)	Q14, n (%)			0,613 (0,504; 0,722)
No	42 (41,6)	40 (39,6)		No	14 (13,9)	18 (17,8)	
Yes, a little	32 (31,7)	31 (30,7)		Yes, a little	34 (33,7)	26 (25,7)	
Yes, quite a lot	17 (16,8)	22 (21,8)		Yes, quite a lot	28 (27,7)	24 (23,8)	
Yes, a lot	10 (9,9)	8 (7,9)		Yes, a lot	25 (24,8)	33 (32,7)	

Figure 3: Quantitative comparison between answers obtained in test and retest application of Portuguese version of HQ (n = 101).

and 4.4% of mild, moderate and severe hyperacusis, respectively (Table 1).

The comparison between the total score of HQ and each frequency of LDL ($p < 0.05$) and each classification of LDL ($p = 0.014$) showed a significant and progressive inverse correlation between both tools: The higher the HQ score, the lower the values of LDL, especially for subjects with severe hyperacusis by LDL. However, our sample had only few subjects classified as severe (4.4%) and because of that the correlation between them was relatively low (0.2). We could have had a high correlation in a larger sample.

Studies that have previously compared HQ and LDL have come to different conclusions. Wallén, et al. [35] and Fioretti, et al. [28] also found that higher scores in HQ correlate with lower values in LDL. Wallén, et al. [35] identified the social dimension of HQ as most related to the audiological parameters of hyperacusis. On the other hand, Meeus, et al. [27] and Fackrell, et al. [14] found low correlations between both tools, suggesting that they measure different constructs.

In this regard, having uniform verbal instructions given by the audiologist to the patients when measuring LDL is of utmost importance. This is why we standardized the instructions described by Knobel and Sanchez [22], which were described in Methods. However, individuals with altered LDL may report no clinical complaint of hyperacusis and vice-versa because hypersensitivity to pure tones does not necessarily mean hypersensitivity to everyday sounds. Indeed, although the intrasubject variability is low, the intersubject variability may be rather high, giving LDL a limited diagnostic value [20]. Thus, caution is needed when comparing LDL between studies or when reassessing the same individuals with different types of equipment.

Due to such limitations of both HQ and LDL in quantifying hyperacusis, a further care should be taken when interpreting our results. The relatively strong correlations between both instruments, which were observed in this study only indicate the size of the association, but do not determine how well both methods measure the same construct.

Conclusions

The Portuguese version of Hyperacusis Questionnaire maintained reproducibility and good internal consistency reliability as compared with the original version, and may be an acceptable tool for measuring symptoms of hyperacusis. In addition, the values obtained by such questionnaire showed an inverse correlation with the values obtained from Loudness Discomfort Levels.

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