EFFICACY OF DIFFERENT CLINICAL ASSESSMENT MEASURES OF HYPERACUSIS – A SYSTEMATIC REVIEW

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CERTIFICATE

This is to certify that this dissertation entitled **'EFFICACY OF DIFFERENT CLINICAL ASSESSMENT MEASURES OF HYPERACUSIS – A SYSTEMATIC REVIEW'** is a bonafide work submitted as a part of the fulfillment for the degree of Master of Science (Audiology) of the student with Registration Number: 20AUD002. This has been carried out under the guidance of the faculty of this institute and has not been submitted earlier to any other University for the award of any other Diploma or Degree.

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CERTIFICATE

This is to certify that this dissertation entitled **'EFFICACY OF DIFFERENT CLINICAL ASSESSMENT MEASURES OF HYPERACUSIS – A SYSTEMATIC REVIEW'** has been prepared under my supervision and guidance. It is also being certified that this dissertation has not been submitted earlier to any other University for the award of any other Diploma or Degree.

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DECLARATION

This is to certify that this dissertation entitled **'EFFICACY OF DIFFERENT CLINICAL ASSESSMENT MEASURES OF HYPERACUSIS – A SYSTEMATIC REVIEW'** is the result of my study under the guidance of Dr. Prashanth Prabhu, Assistant Professor in Audiology, Department of Audiology, All India Institute of Speech and Hearing, Mysore and has not been submitted earlier to any other University for the award of any other Diploma or Degree.

Mysore **Registration No: 20AUD002**

August 2022

Dedicated to

my awesome Maa,

Papa

and

Adi, the best of the best.

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Abstract

Hyperacusis refers to hypersensitivity to auditory stimulus which otherwise has no effect on individuals with hearing sensitivity within normal limits. Hyperacusis prevalence has been found to range from 9 to 15% in the general population. There is no consensus definition on identifying hyperacusis among other decreased sound tolerance disorders. This leads to higher variations in the prevalence of hyperacusis, further affecting the assessment and treatment. This study aims to determine the efficacy of the different clinical assessment measures of Hyperacusis in assessing hyperacusis through a systematic review. This review reports the summary and of the current evidence of clinical assessment measures used for diagnosing hyperacusis. A review of 23 articles was conducted in order to highlight these measures used. Most of the selected studies included retrospective, prospective, survey and experimental in design. Study quality reported overall low risk of bias. Subjective and objective measure in relation to hyperacusis was assessed. This review highlights a definition and protocol that may be used in clinical diagnosis of hyperacusis. It also shows the necessity to standardise and validate assessment measures for the younger population.

Key Words: Hyperacusis, Questionnaire, Assessment, Loudness Discomfort Level (LDL), Uncomfortable loudness level (ULL), systematic review.

Chapter I

INTRODUCTION

Hyperacusis refers to hypersensitivity to hearing. Daily social, professional, recreational, and other activities are severely hindered by a sound sensitivity (Aazh et al., 2016). Vernon (1987) defined hyperacusis as "unusual tolerance to ordinary environmental sounds". (Klein et al., 1990) described hyperacusis as "consistently exaggerated or inappropriate responses to sounds that are neither threatening nor uncomfortably loud to a typical person".

Hyperacusis does not have a single, accepted definition. The American Tinnitus Association describes hyperacusis as a rare condition in which a person's ears cannot tolerate typical environmental sounds. Most of the natural dynamic loudness range of the ears is considered to be lost (American Tinnitus Association, 1989). There is no current universal definition for Hyperacusis nor a universal stand on assessing and interpreting the pathology of hypersensitivity to sounds. It can be thought of as losing the ear's normal dynamic range. Clinically, it is considered an intolerance to everyday environmental sounds otherwise normal to others, whereas their hearing sensitivity is within normal limits. Hyperacusis is an abnormal behavioural response to low-intensity sounds, and everyday sounds are problematic rather than specific. It has many known causes that can be the effectiveness of the medial olivocochlear efferent system, biochemical, neurophysiological, psychological and even social mechanisms which can reinforce the hypersensitivity to everyday sounds (Baguley & Andersson, 2007), as well as associations with a few diseases and syndromes (Williams syndrome, Addison's disease, etc.).

1.1 Prevalence

In the general population, hyperacusis prevalence has been observed to range from 9 to 15% (Andersson et al., 2009). When it is connected to a certain demographic, like Williams syndrome and tinnitus, it is more common (Hall et al., 2015; Rosing et al., 2016). Vernon (1987) reported that just 0.3% of people who suffer from tinnitus show lower tolerance. According to Jastreboff (2004), 25–30% of those with tinnitus also experience hyperacusis. In the general population, hyperacusis affects at least 2% of people in varying degrees. Tinnitus is prevalent in 86 percent of individuals with hyperacusis as their main symptom (Elliasson & Magnusson, 1999). Once a common diagnostic method is devised and put into practise, it will be easier to determine the incidence and prevalence of hypersensitivity to everyday sounds, as opposed to misophonia and phonophobia.

1.2 Etiologiy

Although many instances may have no known cause, there are a number of documented causes and correlations for hyperacusis. Hyperacusis is linked to a few illnesses and disorders, such as depression, fibromyalgia, PTSD (post-traumatic stress disorder), head trauma, Lyme disease, Williams' syndrome, Addison's disease, autism, middle cerebral aneurysm, myasthenia gravis, etc., (D. M. Baguley, 2003; Andersson et al., 2009; Assi et al., 2018; D. (David M.) Baguley & Andersson, n.d.; D. M. Baguley, 2003; Jüris et al., 2013; Klein et al., 1990; Rosing et al., 2016; Viziano et al., 2017).

1.2.1 Hyperacusis and Tinnitus

Hyperacusis and tinnitus are frequently associated (Tyler & Conrad-Armes, 1983). Various authors have stated that the prevalence has been estimated to be between 40 and 86 percent (Anari et al., 1999a; Andersson et al., 2001; Jastreboff & Jastreboff, 2000). Different definitions and criteria for classifying individuals with hyperacusis and tinnitus may have influenced this discrepancy between studies. It is important to note that a significant percentage of the research on tinnitus and hyperacusis comes from tinnitus clinics and may not adequately reflect the general population. It is safe to infer that the reports on hyperacusis are representative of the people that suffer from hearing loss and/or tinnitus. There may be a sizable number of people who have hyperacusis but neither tinnitus or hearing loss as a complaint.

1.2.2 Hyperacusis and Hearing Loss

Hearing and hyperacusis have an ambiguous link. Sensorineural hearing loss has reportedly been experienced by numerous individuals with hyperacusis (Nelson & Chen, 2004; Sood & Coles, 1998). Despite how frequent hearing loss is, it can occasionally be mild or subclinical. Pure tone audiometry (PTA) hearing thresholds of 15-20 dB HL from frequencies 125 to 8000 Hz are regarded as falling within the range of normal hearing sensitivity. This does not imply the absence of hearing loss, merely that there are no current speech or hearing impairments.

1.2.3 Noise Exposure

Another associated risk is occupational noise exposure which has often increased the risk of hyperacusis, usually with tinnitus. Noise exposure can probably be a common cause of hyperacusis, but the data are limited. Additionally, there are findings in the literature linking recreational noise exposure, such as listening to loud music, with hyperacusis (Anari et al., 1999b; Jansen et al., 2009; Kähäri et al., 2003).

1.2.4 Acoustic Trauma/Shock

A relatively recent entity identified some hyperacusis characteristics (McFerran & Baguley, 2007). The symptoms of acoustic shock can be short or long-lasting. There have been many proposals suggesting the mechanism of acoustic shock, for example, the involvement of the tensor tympani muscle (Patuzzi R et al., 2002; Westcott, 2009). Westcott et al. (2009) proposed that an intense impulse can trigger the tonic tensor tympani syndrome, which can cause a frequent spasms. Traditionally it is not linked to the human acoustic reflex. Thus it is unclear to rely on this logic.

1.2.4 The Medial Olivocochlear Efferent System (MOCS)

Anatomically, the MOCS is made up of neurons that emerge from the medial superior olivary (MSO) nucleus and go to the cochlea. The strength of the active amplification of incoming sound is regulated by the efferent fibres, which end on the outer hair cells (OHCs). It shields the cochlea from loud, abrupt sounds (Maison et al., 2002, 2013) and improves its detection of noise signals (Giraud et al., 1995; Micheyl & Collet, 1996). The MOCS's efficacy may differ from individual to individual and may not be enough for some (Lustig, 2006). It can show a decline with age (Kim et al., 2006) and be adversely influenced by head trauma (Attias et al., 2005). Failure of the MOCS and the resulting loss of command over the cochlear amplification gain could lead to a sequel of hyperacusis without any presence of hearing loss, as reported by studies of individuals with brain injury (Attias et al., 2005).

1.3 Assessment / Diagnosis

Most frequently, loudness discomfort levels (LDLs) are utilized to rate hyperacusis. There is disagreement over the frequency and the quantity of repeats for each judgement. The dynamic range (DR) at the particular frequency separates the pure tone thresholds from the LDLs. Again, there is disagreement on the use of the LDL threshold or the DR to determine if hyperacusis is present or absent.

An individual with hearing sensitivity within normal limits can tolerate 100 dB of sound, according to Jastreboff and Jastreboff (2000), who considers LDLs of 100 dB to be normal. "Sensitivity to sound/ hyperacusis," according to Jastreboff & Hazell, 2004, is defined as a decreased dynamic range between threshold and LDL of less than 60 dB. According to Brandy & Lynn, 1995, severe hyperacusis has a dynamic range of 25 to 40 dB . Increased sensitivity to sound is referred to as hyperacusis, a perceptual condition that has varied grades or degrees. The testing procedure used to evaluate hyperacusis varies and is not standardised. The suggested interpretation of test results for identification varies significantly.

1.4 Need for the study

There are very few well-established self-report questionnaires that are targeted for hyperacusis. The most frequently used is the Hyperacusis Questionnaire (HQ) by Khalfa et al. (2002), with 14 items on a 4-point Likert scale. Not all the items included are exclusively appropriate for all cases with hyperacusis, and all aspects of hyperacusis are not covered. This questionnaire has been adapted to other languages like Turkish, Italian, Japanese, etc. (Erinc & Derinsu, 2020; Fioretti et al., 2015; Oishi et al., 2017). Loudness Discomfort Level (LDL) measures used for assessment include the Johnson Hyperacusis Dynamic Range Quotient (JHQ). Rating or severity of hyperacusis is done using this questionnaire from mild to profound based on JHQ scores. Nelting et al., 2002 came up with a questionnaire for the identification of subjective distress levels in relation to hypersensitivity to sound using a broader definition of hyperacusis as "an established collective term for all variations of hypersensitivity to sound". The diagnosis of hyperacusis is a sum of case history, audiological tests, questionnaires and LDL measures. There have not been many studies reported for generalising and validating these measures. Due to this dearth of literature, we aim to determine the efficacy of the different clinical assessment measures to assess the degree and distress due to Hyperacusis.

1.5 Aim of the study

The study aims to determine the efficacy of the different clinical assessment measures of Hyperacusis in assessing hyperacusis through a systematic review.

1.6 Objectives of the study

- Screening and selecting articles relevant to the study that meet the inclusion and exclusion criteria.
- To identify the assessment measures used for diagnosing Hyperacusis.
- Evaluation of the quality of the studies selected.
- Interpret the efficacy of the different clinical assessment measures of Hyperacusis in assessing the severity and distress.

Chapter II

METHOD

A review was done of literature using the following search strategy/keywords, Hyperacusis AND [tests OR Questionnaire OR Assessment OR Loudness Discomfort Level] Limit to humans and English. Inclusion, as well as exclusion criteria, were defined. All possible keyword combinations and related search words and derivatives apply to the research question selected. Databases used for search included PubMed, Google Scholar, Science Direct, AJOL [African Journals Online] and MEDLINE. PRISMA guidelines were used to conduct the review (Parums, 2021).

The eligibility criteria included observational and experimental studies highlighting the efficacy of the questionnaire-based, audiological measures-based assessment tools in assessing individuals with Hyperacusis to derive a standard protocol, including controlled trials. This study did not include case reports, case series, and review articles. A systematic search was carried out for data extraction, using the databases mentioned earlier to obtain articles in English and published in peer-reviewed journals. The search strategy included a comprehensive search terms list to identify relevant articles. This was followed by the title and abstract screening using the inclusion and exclusion criteria mentioned earlier. All eligible articles' full-length texts were procured and reviewed to assess the eligibility as per the criteria. A manual search was also carried out to identify known articles. Disagreements were restored through discussion at the screening stage that was present between the reviewers. As suggested by the PRISMA-P, the reviewers then extracted apposite data from the studies chosen for inclusion. They assessed each study as per the risk of bias assessment (pre-defined) protocols. The extracted information included authors' names and year of publication of the article, research design type, the study population type and their number, methodology of the study, audiological evaluations, questionnaires and other measurement procedures used, the study's outcome, and the merits and demerits of the study.

9770 articles in total were gathered from all databases (with or without the abstracts). 8693 studies were eliminated during title screening, 409 during abstract screening, and 601 were eliminated due to repetition based on a screening of the titles and abstracts of all the studies identified throughout the search in the electronic database. A total of 67 articles were chosen for comprehensive reading. After reading the 67 full-length texts, 44 articles were excluded due to the following reasons: 12 studies had no proper diagnostic criteria for hyperacusis; 10 studies were explicitly about the intervention but had no assessment guidelines; 9 studies were review articles; 8 studies were case reports; and 5 studies were case series.

In the end, 23 articles satisfied the outlined inclusion requirements. Nine of the papers which are included in the current review were retrospective, two were prospective, three were survey studies, and nine were experimental. Based on the assessment method applied to a patient who complained of hypersensitivity to sound, the investigations were further divided. There were six studies utilizing questionnaire-based techniques, six using LDL assessments, and eleven studies combining the two types of data. The PRISMA table in Figure 2.1 provides a summary of the aforementioned.

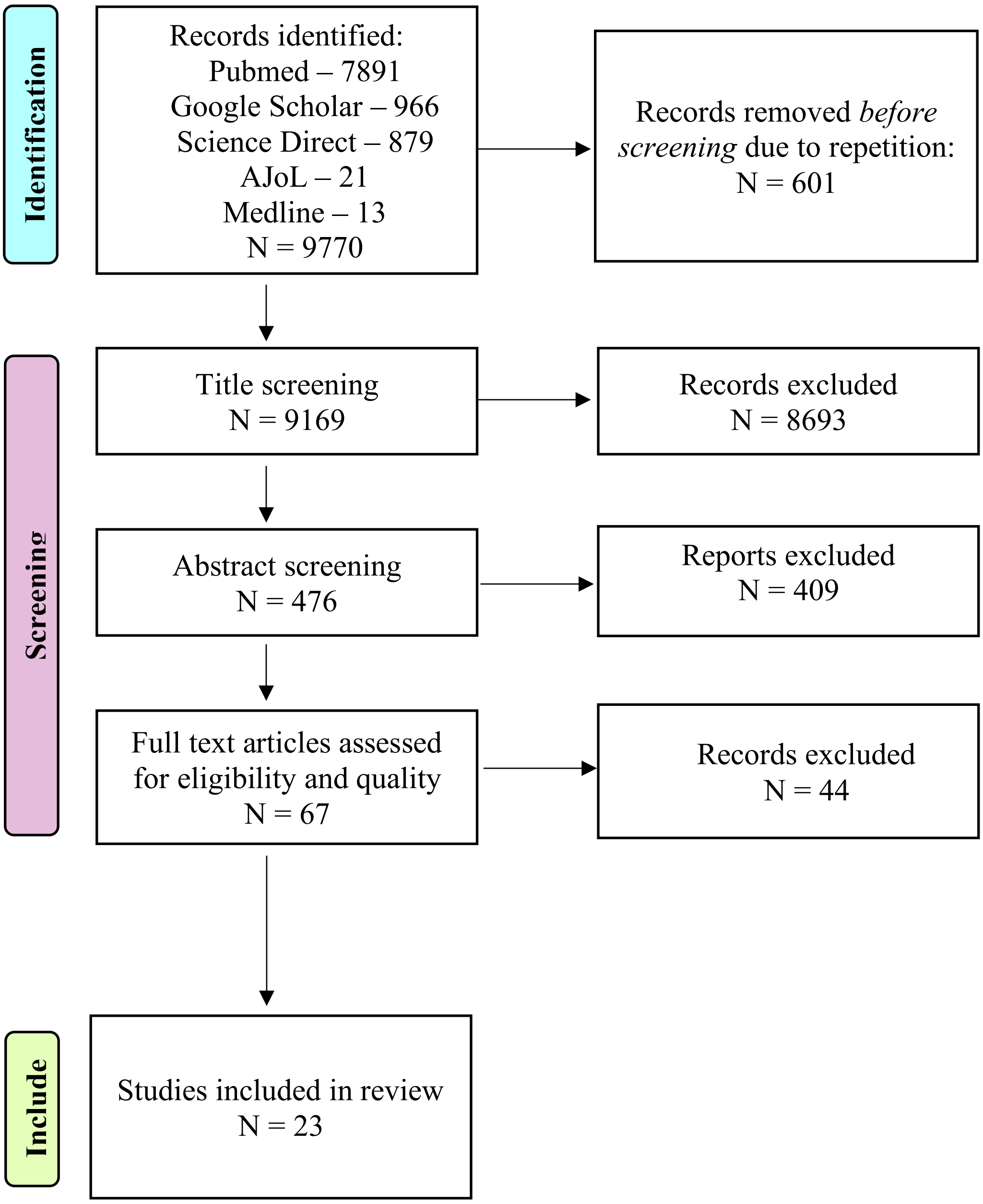


Figure 2.1 *Flow chart depicting the selection process of articles in the systematic review*

Chapter III

RESULTS

3.1 Selection of studies

23 papers were selected for quality assessment and synthesis after the initial search strategy and the inclusion and exclusion criteria were applied. In total, 9770 articles (abstracts included) were found throughout all databases. As part of a pre-selection of these citations, 601 were eliminated owing to repetition, 8693 were eliminated by title screening, and 409 were eliminated by abstract screening after reviewing the titles and abstracts of all the papers located through the manual search from the databases. A total of 67 articles were chosen for thorough reading. After reading all 67 texts, 44 articles were eliminated due to the following reasons: 12 studies had no proper diagnostic criteria for hyperacusis; 10 studies were explicitly about the intervention but had no assessment guidelines; 9 studies were review articles; 8 studies were case reports; and 5 studies were case series. 23 articles ultimately met the requirements for inclusion. Nine of the studies that were reviewed were retrospective, two were prospective, three were survey studies, and nine were experimental. On the basis of the assessment method applied to patients who complained of sound sensitivity, the investigations were further divided. There were six studies adopting questionnaire-based techniques, six using ULL assessments, and eleven studies combining the two types of measurements.

3.2 Summary of data extraction

Data extraction from all the selected articles was carried out and classified using the following criteria-Author and year of publication, research design, research question, type of population, method, outcome, and discussion (merits and drawbacks). The data extraction sheet reveals that studies included were published in the time range of 2002 to 2021. Selected studies mainly consisted of retrospective, prospective, survey and experimental in design. The largest of these studies reported the results of 573 consecutive patients (Aazh & Moore, 2017b). All subjects included in the study attended the Tinnitus and Hyperacusis Therapy Specialist Clinic (THTSC) and their uncomfortable loudness levels (ULLs) were measured. Both the measures to assess hyperacusis was used, questionnaire and ULL measure. Most of the studies preferred using both the measures to identify hyperacusis. There are also new questionnaires that have been developed and some authors have chosen a specific population to administer the hyperacusis measures on (Aazh & Moore, 2018; Silva et al., 2021). Many of the used a similar criterion to indicate the presence of hyperacusis. Many studies used the measures in comorbid conditions as well like tinnitus, WS, anxiety, depression, concussion etc., (Aazh et al., 2019; Aazh & Moore, 2017a, 2017b, 2018; Abouzari et al., 2020; Assi et al., 2018; Blaesing & Kroener-Herwig, 2012; Jüris et al., 2013; Villaume & Hasson, 2017). One universal protocol for the assessment of hyperacusis was not identified, neither a consensus definition used.

Table 3.1 *Data extraction of studies based on Questionnaire and ULL measures*

Study	**Authors and Year**	**Research design**	**Research question**	**Population type (N)**	**Method**	**Outcome**	**Discussion (Merits and drawbacks)**
A1	Aazh et al., 2019	Retrospective study design	To explore the insomnia related factors in patients with tinnitus and/or hyperacusis	444 consecutive patients of tinnitus and/or hyperacusis. Average age of 54 years (Standard deviation of 15 years)	Audiological tests (PTA, ULL) and self-report questionnaires (THI, HQ, VAS, HADS). Multiple-regression analysis to asses relationship between insomnia and other variables.	In patients with tinnitus high prevalence of insomnia. Hyperacusis not significantly associated with insomnia.	Age, gender, ULLmin values, the pure-tone average of the better and worse ears, and the VAS evaluations for tinnitus loudness and influence on life did not significantly predict insomnia. As this study only included a small number of individuals, further research with bigger sample sizes should investigate whether severe hyperacusis is connected to insomnia..
A2	Abouzari et al., 2020	Prospective cohort study design	To assess the effectiveness of a multi-modal migraine	25 patients who reported being abnormally sensitive to	Average LDL levels, VAS-measured hyperacusis discomfort,	According to self-reported and audiometric assessments, the majority of	There are numerous similarities between tinnitus and hyperacusis. With multimodal migraine prophylaxis

			prophylaxis therapy for hyperacusis patients. .	sound and who had symptoms that had persisted for at least six months.	and scores on the modified Khalfa HQ for the severity of hyperacusis were compared between pre- and post- treatment.	hyperacusis patients showed clinical improvement with migraine prophylaxis therapy.	therapy, hyperacusis may be successfully managed and may share a pathophysiologic basis with migraine illness. Limitations: 1. No control group was present. The outcome measurements of hyperacusis are inherently subjective and sensitive to bias. 2. The degree of variation in adherence to the advised regimen was not measured. 3.Unable to evaluate the long-term efficacy of the therapy due to the short research duration. To demonstrate effectiveness, more research is necessary.

A3	Aazh & Moore, 2017	retrospective cross-sectional study	1. to study patterns of ULLs across frequency and their associated factors for individuals with tinnitus and hyperacusis. 2. to review the criteria, based on ULLs and scores for the HQ, for diagnosing hyperacusis	573 consecutive patients with ULLs measurement data included	Age and gender (demographic) information for the patients, as well as the results of their most recent audiological tests (PTA, ULLs), as well as their routine self-report questionnaires (THI, HQ, VAS, HADS, ISI). To predict ULLs, a stepwise linear multiple regression model was developed.	There is significant relationship betwe en ULLmin, HQ score, and age. The impairment caused by hyperacusis was linked to significant across-frequency variability in ULLs.	There is an urgent need for more precise and reliable hyperacusis diagnostic criteria. If the study's cut-off scores are used as a standard for a positive diagnosis, a hyperacusis diagnosis based on HQ scores can be made to be rather consistent with a diagnosis based on ULLs. Low ULLmin values are associated to ageing and high HQ scores. PTAs and ULLmin values are not strongly associated.
A4	Villaume & Hasson, 2017	Survey	To investigate potential associations	A web-based survey included 348 participants,	ULL and HQ are used to evaluate hyperacusis.	Weak correlations with the ULLs and moderate correlations with	The diagnosis and management of hyperacusis should consider the effects

			between hyperacusis and personality factors that are related to health	and 341 of them finished the clinical hearing tests.	The HP5i is used to evaluate personality.	the HQ dimensions were found for the personality trait negative affectivity.	of personality qualities that are relevant to health.
A5	Aazh & Moore, 2017a	Retrospective cross-sectional study	To determine the incidence of discomfort during PTA and the assessment of ULLs in tinnitus and/or hyperacusis patients.	362 consecutive patients with a complaint of tinnitus and/or hyperacusis.	Audiological tests (PTA, ULL) and self-report questionnaires (THI, HQ, VAS, HADS, ISI).	21% or more of patients found the tone presentation levels for PTA and of ULLs to be uncomfortable.	For people who suffer from severe hyperacusis impairment, severe insomnia, or who are female or young, lower sound levels should be used.
A6	Assi et al., 2018	Experimental design	To find relationship between hyperacusis and sport-related concussion.	Participants included 58 college athletes with normal hearing who had either suffered one or more	PTA, LDLs, general questionnaire, HQ, BDI-II, BAI, Loudness growth function was carried out.	Compared to the control group, the concussed group performed better on the HQ and showed more sensitivity to noise. Concussed athletes who self-	Lower LDL levels are associated with higher depressive symptom and hyperacusis scores.

				concussions due to sports (N = 28) or had never encountered a head injury (N = 30).		reported sound sensitivity had lower LDLs, increased Depression and Hyperacusis scores, and different loudness growth functions in comparison to the control group.	
A7	Silverstein et al., 2016	Prospective, longitudinal design.	To determine whether a minimally invasive surgical approach is effective for treating patients with severe hyperacusis .	Two groups were formed from six adult patients (9 ears) having a history of severe hyperacusis (unilateral or bilateral reinforcement procedure).	Transcanal surgical intervention. Pre- and post-operative ULL test, HQ.	The patients reported enhanced quality of life and no difference in hearing postoperatively.	It may be significantly useful to reinforce the round and oval windows with temporalis fascia or tragal perichondrium in individuals with severe hyperacusis who have not responded to conventional therapy.

A8	Jüris et al., 2013	cross-sectional study	To contrast the many methods for measuring hyperacusis that are often used in audiological practise in order to identify the best valid test for assessment.	Hyperacusis was identified in 62 patients between the ages of 18 and 61 after evaluation.	Clinical interview, LDLs test, HQ, HADS was performed.	Significantly negative correlations were detected between nearly all LDL scores and the HQ for the right ear, but none were discovered for the left. Although the HQ and either of the HADS scales did not significantly correlate, LDLs did significantly correlate with the HADS's anxiety subscale.	To identify hyperacusis, clinicians should combine the HQ and HADS with a clinical interview. Made a recommendation to decrease the cut-off for the Swedish edition of the HQ.
A9	Aazh & Moore, 2018	Retrospective cross-sectional study	To determine the proportion of patients who seek treatment for tinnitus and/or	ULLs had been measured for 362 consecutive patients who had attended tinnitus	Criterion: For at least one of the measured frequencies for at least one ear, ULL of 30 dB HL or less is required.	Low ULLs for specific frequencies and no or minimal hearing loss are characteristics of severe hyperacusis. Only	In order to effectively examine individuals with severe hyperacusis, comprehensive audiological, otological, and psychological examinations are advised due to the high incidence

			hyperacusis who have severe hyperacusis, as well as to investigate the factors for severe hyperacusis .	and/or hyperacusis rehabilitation.	Data: PTA, ULLs, THI, VAS, HQ, HADS, ISI, Demographic (age, gender) details.	6 out of 13 people who were given a diagnosis of severe hyperacusis had HQ values greater than 26. This method of calculating the grand mean ULLs indicated a significant correlation with the HQ scores.	of tinnitus, otological abnormalities, and mental health issues.
A10	Aazh et al., 2017	Retrospective cross-sectional study design	To investigate the factors which elderly persons with tinnitus and hyperacusis may find challenging	184 patients with 69 years average age.	Data of patients: PTA, ULLs, THI, VAS, HQ, HADS, ISI,	Significant tinnitus handicap predictors include tinnitus annoyance. The HADS score for depression significantly predicted hyperacusis handicap, the THI score only marginally	Depression foretells the development of hyperacusis disability rather than anxiety actually causing it. It's important to investigate annoyance-related characteristics that can be helpful in developing suitable rehabilitation strategies intended to lessen tinnitus

						predicted it, and the HADS score for anxiety did not. Depression was a significant predictor of insomnia.	impairment in elderly persons. Limitation: Only 30% of the variation in the total HQ scores could be explained by the regression model, suggesting that factors other than those evaluated here are also related to hyperacusis handicap..
A11	Blaesing & Kroener-Herwig, 2012	Experimental study design	To analyse in tinnitus subjects with hyperacusis, the role of sound avoidance and anxiety.	30 controls without tinnitus or hyperacusis and 56 tinnitus patients with or without hyperacusis.	Tinnitus patients with hyperacusis were compared to those with tinnitus without hyperacusis and with healthy controls. The following behavioural indices were measured:	While discomfort was the same for all participants, those with hyperacusis reported significantly higher noise-related avoidance in everyday life and exhibited significantly shorter exposure to a pure tone. Self-reported	Within a hyperacusis treatment plan, confrontation with avoided situations and activities should be considered. This is important with very anxious people because their anxiety is exacerbated by hyperacusis, which can heighten sound sensitivity in the auditory system as a result of sound avoidance. The

					NAQ, STI, TF-12, GÜF, BAI, STAI-T, ULL, and self-exposure to PTs.	avoidance behaviour was significantly correlated with anxiety levels and suffering from hyperacusis.	management of hyperacusis may be significantly aided by regular exposure to sound. Limitations: Only applied to patients with tinnitus and hyperacusis.

Table 3.2 *Data extraction of studies that are based on ULL measures*

Study	Authors and Year	Research design	Research question	Population type (N)	Method	Outcome	Discussion (Merits and drawbacks)
B1	Aazh et al., 2018	Retrospective study	In a tinnitus and hyperacusis clinic, assess the incidence of hyperacusis and the severity of hyperacusis in children and adolescents.	62 patients with 12 years of average age	Young patients' demographic information, as well as the outcomes of their most recent audiological tests (PTA, ULLs).	Children and teenagers who are referred for intervention for their tinnitus and/or hyperacusis share a few traits in common. Based on a ULL of 77 dBHL or less, hyperacusis was diagnosed in more than 85% of patients. There were strong across-frequency fluctuations in ULLs.	Only a small number of kids,out of many, are referred for treatment of their hyperacusis and/or tinnitus. 1. Not all frequencies were used to measure ULLs in order to determine ULLmin. 2. The ULL measurements obtained here lack reproducibility. 3. The results might not be typical of all kids with hyperacusis and tinnitus.

B2	Silva et al., 2021	Experimental study	To evaluate auditory hypersensitivity in WS and assess hyperacusis using standardised protocols to see if it may be connected to WS patients' lack of acoustic reflexes.	17 people with WS, ranging in age from seven to seventeen (10 males and seven females), and 17 people with usual development who are age and gender-matched to the WS patients.	The LDL test responses, as well as ipsilateral and contralateral reflex reactions, were analysed using statistical tests.	In 35.29 percent of WS patients, hyperacusis was present, and in 50% of those cases, it was mild. Hyperacusis was more prevalent in those who lacked the contralateral acoustic reflex, and there was a correlation between hyperacusis and acoustic reflex responses.	However, hyperacusis was less common and may be related to the absence of contralateral acoustic reflexes. Individuals with WS show a high prevalence of auditory hypersensitivity and phonophobia.
B3	Doutoranda em et al., 2005	Experimental design	To establish LDL reference values for those with normal hearing and relating the LDL to the	64 participants with normal hearing between the ages of 18 and 25 (53.1	PTA, LDL test, immittance measurements were carried out.	The LDL and ART did not correlate, and as a result, the ART cannot be utilised to predict the LDL.	There is still no objective measurement that can help with the official diagnosis of intolerance to sound. The LDL should always be carefully

			Acoustic Reflex Threshold (ART).	percent female)			interpreted by the individual and correlated with the anamnesis information.
B4	Urnau & Tochetto, 2012	Cross-sectional, descriptive, non-experimental, quantitative study	To confirm the occurrence and suppression effect of TEOAE, and suppressive effect of TEOAE and laterality, tinnitus, and hyperacusis degrees, association between the degree of hyperacusis and tinnitus in adults with complaints of tinnitus and hyperacusis.	25 normal hearing subjects with hyperacusis and tinnitus complaints	PTA, Immittance measurements, THI for tinnitus degrees classification, and the LDL for hyperacusis classification.	When compared to people with normal hearing who did not have these symptoms, TOAE incidence was reduced. Higher proportion of TOAE suppression in both ears, with the left ear predominating.	There is no correlation between the severity of hyperacusis and tinnitus. There are no correlations between TOAE suppression, handedness, levels of tinnitus, and hyperacusis in patients with normal hearing..

B5	Sheldrake et al., 2015	Experimental study	To find out the audiometric characteristics of hyperacusis patients.	381 patients with primary complaint of hyperacusis	PTA, LDLs test	When compared to a reference group of people with normal hearing, LDLs were found to be much lower, with average values across the frequency range of around 85 dB HL.	In contrast to tinnitus, where hearing loss is a major trigger, LDLs tended to be greater at frequencies when hearing loss was present, indicating that hyperacusis is unlikely to be induced by an increase in hearing threshold. Regardless of the kind or severity of hearing loss, LDLs were reduced over the whole audiometric frequency range, suggesting that hyperacusis may be brought on by a broad increase in auditory gain. This implies that there could not be a shared mechanism between tinnitus and hyperacusis.

B6	Zaugg et al., 2016	Retrospective study design	To investigate the relationship between the SRST and the tonal and speech LDLs .	139 former American soldiers were recruited to offer tinnitus treatment.	Tonal LDLs and SRST were measured	Weak correlations between tone LDLs and SRST as well as between speech LDLs and SRST have been found.	Weak correlations between the measurements imply that LDLs may not adequately reflect a patient's capacity for coping with noise in daily life. By concentrating evaluation efforts on aspects of everyday life affected by a sound tolerance issue, doctors can better understand the aspects of life that each patient feels are significant and vital.

Table 3.3 *Data extraction of studies that are based on Questionnaire measures*

Study	Authors and Year	Research design	Research question	Population type (N)	Method	Outcome	Discussion (Merits and drawbacks)
C1	Viziano et al., 2017	Questionnaire-based survey	To investigate if individuals with complex chemical sensitivities have hyperacusis and noise sensitivity (MCS).	20 healthy participants with matching ages and genders and 18 MCS patients who were chosen using strict diagnostic criteria.	The WNS Questionnaire, the HQ, and the DPOAE were used to find out more information on cochlear function.	WNS, HQ, and qEESI have a strong positive correlation in MCS individuals.	Reduced sound tolerance and noise sensitivity may be potential new features of this condition, which would explain its peculiar phenotype. Indicates a "central" cause for these illnesses in this patient population.
C2	Khalfa et al., 2002	Experimental, Questionnaire study design	A questionnaire assessing many areas of auditory symptomatology was developed in order to	201 participants, aged between 17 and 72, with a mean age of 28.4 (SD = 13.24) years and a	There were 14 self-rating items total. (Attentional, Social, Emotional)	Developed a HQ that was statistically reliable and consistent.	A questionnaire will be valuable in quantifying and characterising the attentional, social, and emotional aspects of the clinical phenomena of hyperacusis.

			establish a tool suited for quantifying and evaluating diverse hyperacusis symptoms.	range of 132 females and 69 males (from the general population, chosen randomly, either hyperacusic or not).			
C3	Fackrell et al., 2015	Experimental study design	To evaluate the validity and reliability of the HQ as a measurement tool	Data from 264 persons who participated in a tinnitus research study.	BDI-II, BDI-FS, BAI, ULLs, HQ, THI, and THQ. Internal consistency, convergent and discriminant validity, floor and ceiling effects, and HQ factor structure were all assessed.	Moderate correlations and high internal consistency were found between the HQ, ULLs, and other health questionnaires.	In a population of individuals with tinnitus, none of the fourteen questions adequately assesses hypersensitivity to sound. Proposed a 2-factor, 10-item version of the HQ. Validation needed using a new sample of individuals with tinnitus and maybe those without tinnitus..

C4	Greenberg & Carlos, 2018	Survey design	To develop a new scale that is valid, concise, and simple to score has to be developed. .	From 469 refined Inventory of Hyperacusis Symptoms (IHS) administrations that were gathered online and represented people from 37 countries with a mean age of 34.8 years, 450 completed survey procedures were evaluated.	Second edition, 25 questions, with a maximum score of 100.	Dimensional structure with five components: communication, functional impact, and general loudness, psychosocial impact, emotional arousal. The degree of hyperacusis was significantly predicted by greater tinnitus symptoms, not by the level of hearing loss, which did not correspond with IHS scores.	These things may need to be restructured in the future, which might lead to clearer loading onto different aspects. Limitations: 1. There isn't a large enough control group without any hyperacusis symptoms recorded, and there are no audiometric measurements like hearing tests and loudness discomfort levels to compare participant self-reports to. 2. It might be difficult to identify possible problems with exaggeration, diminution, and truthfulness of replies collected when utilising an online questionnaire. 3. Analyses and results may not be generalizable to a

							group with a more diversified representation because the sample was approximately 85% Caucasian.
C5	Prabhu & Nagaraj, 2020	Experimental, Questionnaire study design	To develop and evaluate a regional version of the Hyperacusis Handicap Questionnaire (HHQ) for those with tinnitus caused by hyperacusis.	77 people between the ages of 20 and 55 who have hyperacusis and tinnitus.	For validation, 25 questions were taken into account. There are three parts, each with seven questions, that cover "Functional," "Social," and "Emotional" elements.	The questionnaire has good internal consistency. Comparing gender and tinnitus duration, there is no significant difference.	A questionnaire can be used to evaluate the impairment brought on by hyperacusis in tinnitus sufferers. Limitations: 1. The studies' lack of consistency 2. The necessity for a more thorough investigation among various demographics, including children, adults, and senior citizens. 3. More extensive group research is required for generalisation.

C6	Yilmaz et al., 2017	Experimental study design	Using an HQ to investigate the decreased sound tolerance of university students .	536 college students, ages 18 to 25 years, including 300 females and 236 males.	were evaluated using a Khalfa-developed HQ.	Both the overall score and the attentional and affective aspects were significantly higher for females than for males. Subjects who reported exposure to noise or a decline in noise tolerance scored significantly higher than the other participants.	Even among young adults, there is a subset of individuals who show signs of issues that might be caused by lowered tolerance for ordinary sounds. The results demonstrating that the HQ scores of females were higher than those of males indicate the gender gap. In order to establish validity and evaluate the psychometric properties of the Turkish variant of HQ, more research with diverse populations of various age groups is necessary.

3.3 Summary of quality assessment

This systematic review only included studies; case reports and case series were not included. The papers were evaluated using QUADAS-2 (2011), an updated tool for the quality assessment of diagnostic accuracy studies. Patient selection, index test, reference standard, and flow of patients through the study and timing of the index test(s) and reference standard (together referred to as "flow and timing") were the four key domains that made up the QUADAS-2 instrument. This was finished in 4 phases: State the review question in the first phase; develop review-specific instructions in the second phase; assess the main study's published flow diagram in the third phase; construct a flow diagram if none is reported; and assess bias and applicability in the fourth phase. Each domain's bias risk is analysed, and the first three are additionally assessed for concerns about applicability. To assist in assessing the possibility of bias, signalling questions are added. These are connected to the possibility of bias and are designed to help reviewers gauge the probability of prejudice. The risk of bias was categorised using the following criteria: If a study is given a "low" rating on all aspects of bias or applicability, it is permissible to give that study an overall rating of "low risk of bias" or "low concern with application." A study was classified as having "concerns regarding applicability" or being "at risk of bias" if it has a "high" or "unclear" rating on one or more criteria. The risk of bias was lower for all the included research.

Table 3.4 *Quality analysis summary of articles using Questionnaire and ULLs measures*

Study	Risk of Bias				Applicability Concerns		
	Patient Selection	Index Test	Reference Standard	Flow and Timing	Patient Selection	Index Test	Reference Standard
A1	Low risk	Low risk	Low risk	Low risk	Low risk	Low risk	Low risk
A2	Low risk	Low risk	Low risk	Unclear risk	Low risk	Low risk	Low risk
A3	Low risk	Low risk	Low risk	Low risk	Low risk	Low risk	Low risk
A4	Low risk	Low risk	Low risk	Low risk	Low risk	Low risk	Low risk
A5	Low risk	Low risk	Low risk	Low risk	Low risk	Low risk	Low risk
A6	Low risk	Low risk	Low risk	Unclear risk	Low risk	Low risk	Low risk
A7	Low risk	Low risk	Low risk	Low risk	Unclear risk	Low risk	Low risk
A8	Low risk	Low risk	Low risk	Low risk	Low risk	Low risk	Low risk
A9	Low risk	Low risk	Low risk	Low risk	Low risk	Low risk	Low risk
A10	Low risk	Low risk	Low risk	Low risk	Low risk	Low risk	Low risk
A11	Low risk	Low risk	Low risk	Low risk	Low risk	Low risk	Low risk

Low risk High risk Unclear risk

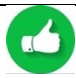
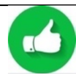
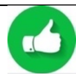
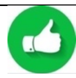
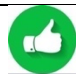
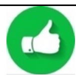
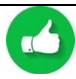
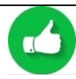
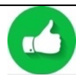
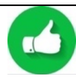
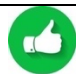
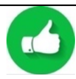
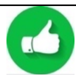
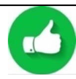
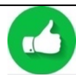
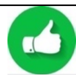
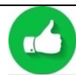
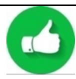
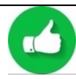
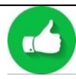
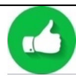
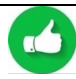
Table 3.5 *Quality analysis summary of articles using ULLs measures*

Study	Risk of Bias				Applicability Concerns		
	Patient Selection	Index Test	Reference Standard	Flow and Timing	Patient Selection	Index Test	Reference Standard
B1	👍	👍	👍	👍	?	👍	👍
B2	👍	👍	👍	👍	👍	👍	👍
B3	?	👍	👍	👍	👍	👍	👍
B4	👍	👍	👍	👍	👍	👍	👍
B5	👍	👍	👍	👍	👍	👍	👍
B6	👍	👍	👍	?	👍	👍	👍

👍 Low risk 👎 High risk ? Unclear risk

Table 3.6 *Quality analysis summary of articles using Questionnaire measures*

Study	Risk of Bias				Applicability Concerns		
	Patient Selection	Index Test	Reference Standard	Flow and Timing	Patient Selection	Index Test	Reference Standard
C1	👍	👍	👍	👍	👍	👍	👍
C2	👍	👍	👍	👍	👍	👍	👍

C3							
C4							
C5							
C6							

 Low risk  High risk  Unclear risk

Chapter IV

DISCUSSION

The present study was carried out to find the efficacy of different clinical assessment measures of Hyperacusis and possibly establish a universal protocol which can be used as an assessment procedure for persons with a complaint of tolerance of everyday sounds. This protocol will also help in the differential diagnosis of patients with various complaints of sound tolerance problems. This will further help discriminate patients with similar complaints of sound tolerance problems which otherwise may be misdiagnosed, thereby having fruitfulness in management by selecting the appropriate patients and finding out the actual efficacy of the management procedures. Twenty-three articles in total were identified and considered for the review.

Many studies mentioned inconsistencies and confusion present in the definition of hyperacusis. The term hyperacusis comes under a general umbrella term which identifies sound tolerance issues, namely, Decreased Sound Tolerance (DST). This includes hyperacusis, misophonia, diplacusis and polyacusis (Jastreboff & Jastreboff, 2012). Hyperacusis has been defined to include all the adverse reactions to any sound concerning its annoyance, loudness, pain and fear (Tyler et al., 2014). Described by the American Tinnitus Association, 1989 as a rare condition in which a person's ears cannot tolerate typical environmental sounds, with most of the natural dynamic loudness range for the ears lost.

There is no standard protocol for evaluating decreased sound sensitivity, specifically hypersensitivity to everyday sounds (Fackrell et al., 2015). Due to this dearth of literature and gap in research, the authors of the current study identified a need to define and propose a standard protocol based on the reported literature. There have been uncertainties regarding the preferred diagnostic or assessment tool. Some of the studies included in this review have introduced in the literature, novel assessment tools, modified the already existing ones, or they have tried to translate and validate the tool in their language (Fackrell et al., 2015; Prabhu & Nagaraj, 2020; Viziano et al., 2017). Some studies have tried to go with a holistic approach by using more than one assessment tool, i.e., a questionnaire-based measure and an LDL measure. In 2017, during the 3rd International Hyperacusis Conference, Khalfa HQ was highlighted as a validated measurement LDLs of 77 dB HL or less, and a score of 22 or more on the Khalfa HQ was mentioned as a measurement for hyperacusis. Although this may seem sufficient initially, the Khalfa HQ was not developed as a tool for diagnosing or used for a younger population.

4.1 Audiometric characteristics

There is rarely any correlation between commonly used audiological tests and hyperacusis (Anari et al., 1999a). However, there have been findings in the literature indicating differences in ULLs across frequencies in children, with ULLs often being lower at 6 and 8 KHz compared to lower frequencies like 0.25 KHz for at least one ear (Aazh et al., 2018). In addition, they noted that 8 kHz was frequently associated to ULLs of 30 dB HL or less, with the mean ULL averaged across hyperacusic ears being 11.2 dB lower at 0.25 than at 8 kHz. Often hyperacusis in children occurs without any evident problems that can be seen in the audiogram and no characteristically strong across frequency variation of hearing thresholds or interaural asymmetry (Aazh et al., 2018).

4.2 Assessment measures

4.2.1 Uncomfortable loudness level based measure

The most common approach to assess DST includes case history taking which can be in the form of personal interview or self-report questionnaire. Along with this, measuring ULLs usually marks the first step in a clinical diagnosis. In the studies included for the current review, many have used ULLs based measures either with other tools such as self-report questionnaires or as the only assessment tool to identify the presence or absence of hyperacusis (Aazh & Moore, 2017b; Blaesing & Kroener-Herwig, 2012). Aazh et al (2018) He observed a higher prevalence of severe hyperacusis in the younger population, specifically in children, who had ULLs of 30 dB HL or less for at least one ear tested and at least one frequency measured. This can be due to less referrals and only children with severe aversions to everyday sounds being referred to a speciality clinic for DST. The above mentioned study also reported higher prevalence in comparison to that of 39% reported by Baguley et al. (2013) for the young population. This may be due to the fact that the study by Aazh et al., 2018 included patients with a complaint of both tinnitus and hyperacusis, whereas the sample for Baguley et al., 2013 included patients with a primary complaint of tinnitus neither did they define the criteria used for diagnosis of hyperacusis.

LDLs are usually measured using pure tone stimuli which might not reflect the natural sounds representing real world sound sensitivity problems the individual is facing. When tested with natural stimuli like a dog's bark or a baby's cry, the LDLs obtained were lower than those obtained by pure tone measurements (Anari et al., 1999a). The LDL measurement values do not serve as a sole hyperacusis diagnostic indicator (Sheldrake et al., 2015). LDLs can depend on many factors like, the instructions given at the time of testing , especially to the younger population. It can also depend on the rapport between the patient and clinician. It is safe to say that we need a wholistic approach for the diagnosis of hyperacusis which may include other symptoms like, aversion, discomfort and annoyance to sound as highlighted by Andersson et al., 2005.

In individuals with WS, most of the studies that have reported a hypersensitivity to sound are based on non-standardised measures, like, interviews with parents, guardian or the individuals with WS themselves (Gallo et al., 2007; Levitin et al., 2005). This can lead to less accuracy in differentiating the DST into different types: Hyperacusis, Misophonia, Phonophobia; as well as algophobia (fear of pain). There is a lot of variance in the report of the prevalence of hypersensitivity to sound where the study included in this article by Silva et al., 2021 mentioned 35.29% whereas studies which used interviews as a measure mentioned a prevalence of 77% and more (Andersson et al., 2005; Levitin et al., 2005). This indicates a need for a standardised protocol for diagnosing hyperacusis. Hyperacusis was mild in 50% of instances. Hyperacusis was more common in patients who lacked the contralateral acoustic reflex, and there was an association between hyperacusis and acoustic reflex responses. More studies needed to generalize this finding.

LDL measure is a highly confident comparison of the individuals own LDLs once there is a good reproducibility achieved, which can be used as a follow up pre- and post- treatment for hypersensitivity to sound, as it was recommended by Gold et al. (2002) and Hazell et al. (2002). Due to its high reproducibility, LDL can be considered a good measure for follow up of patients (Knobel & Ganz Sanchez, 2005).

4.2.2 Questionnaire based measure

Decreased sound tolerance is a very distressing condition and has a detrimental impact on the patients quality of life. The other questionnaires introduced in literature and included in this review include GUF (German questionnaire on hypersensitivity to sound), Multiple Activity Scale for Hyperacusis (MASH), Inventory of Hyperacusis Symptoms (IHS), HHQ, Khalfa (HQ) (Fackrell et al., 2015; Greenberg & Carlos, 2018; Khalfa et al., 2002; Prabhu & Nagaraj, 2020). Some studies included have chosen a specific population i.e., University students, children and adolescents, older individuals, US military veterans, sports related concussed athletes (Aazh et al., 2018; Assi et al., 2018; Yilmaz et al., 2017; Zaugg et al., 2016). There have been attempts at adapting the questionnaire in other languages as well i.e., Turkish, Italian, etc (Viziano et al., 2017; Yilmaz et al., 2017).

The GUF is administered on patients with a complaint of hypersensitivity to sound to subjectively assess the distress associated with it. It is considered to be better suited as an indicator of treatment needs than any audiological tests. This questionnaire is available in both German (15-item self-rating scale) and English (translated version). The German version has good reliability as well as correlation with depression of symptoms. The English version showed unclear statistical properties and factorial structure (Bläsing et al., 2010). An interview-based assessment measure which assesses the level of annoyance caused by hypersensitivity to sound (Dauman & Bouscau-Faure, 2005). This measure has not been assessed for construct validity and internal consistency reliability.

The IHS is a 25 item, 4-point likert scale developed by Greenberg and Carlos (2018). They have reported good statistical properties (internal consistency and convergent validity) as well as usefulness as a clinical and research tool. Due to the inadequate size of the control group without the reported hyperacusis symptoms. They did not have any audiological testing date to confirm the self-reports of the participants. The HHQ is a questionnaire for individuals with a complaint of tinnitus associated with hyperacusis as a comorbid condition. This was developed with the focus of the Indian population in mind, reported noise exposure level varies depending on the different regions (Prabhu & Nagaraj, 2020). The study reports of average item-total correlation and good internal consistency which allow it to be compared to other questionnaires for evaluating hyperacusis.

The most often used questionnaire in the studies included in the current review is the HQ. The validity and reliability of the HQ has also been studied in clinical population with comorbidities like tinnitus, multiple chemical sensitivity (MCS), Williams Syndrome (WS), Insomnia, anxiety, depression (Aazh et al., 2018, 2019; Aazh & Moore, 2017a, 2017b; Blaesing & Kroener-Herwig, 2012; Fackrell et al., 2015; Jüris et al., 2013; Prabhu & Nagaraj, 2020; Silva et al., 2021; Viziano et al., 2017). The original Khalfa HQ was developed for quantifying and characterising the hypersensitivity to sound. It was never indented to be used as an outcome measure for hypersensitivity to sound (Khalfa et al., 2002). The HQ has three subscales (attentional, social and emotional) and uses a cut off score of more than 28 to diagnose an individual as having hyperacusis. This cut off score is too high and underestimates the prevalence of hyperacusis, as the individuals with a complaint of hypersensitivity to sound may not be able to reach a minimum score of 28 to be diagnosed as having hyperacusis (Fackrell et al., 2015; Khalfa et al., 2002; Meeus et al., 2010). There is even a gender difference present where females score more on the HQ than males and are hypothesized to be more sensitive to sounds as well as experience more distress related to hormone and emotional rate changes (Fackrell et al., 2015; Khalfa et al., 2002). There have been contraindicating studies which reports of comparable reaction by both the male and female genders to thw dimensions of hyperacusis (Prabhu & Nagaraj, 2020). Fackrell et al. in 2015 concluded that HQ is not a valid overall measure of sound hypersensitivity seen in UK population with tinnitus and recommended a 10 item questionnaire. As HQ is not an outcome measures questionnaire, there have been other questionnaires developed with improved psychometric properties for monitoring the outcome.

4.2.3 Combined measure

Studies have reported of association of hyperacusis with many other condition like insomnia, depression, anxiety, migraine, tinnitus, sports-related concussed athletes, etc., (Aazh et al., 2019; Aazh & Moore, 2017a, 2017b, 2018; Abouzari et al., 2020; Assi et al., 2018; Blaesing & Kroener-Herwig, 2012; Jüris et al., 2013; Villaume & Hasson, 2017). Aazh et al (2019) reported hyperacusis to have an indirect effect on insomnia which is mediated by anxiety and depression. They also reported significant correlation of ULLmin and HQ scores with Insomnia Severity Index (ISI). All the studies mentioned above has used ULL and self-report questionnaire measure to assess the sensitivity to sound in those comorbid conditions to assess the efficacy of the treatment used. Some have reported ULL measure to be a good measure for follow up patients undergoing treatment for decreased sound tolerance (Silverstein et al., 2016).

There have been evidences in literature that have reported good consistency between the two assessment measures, ULLmin of < 77 dB HL and HQ, where HQ was developed with the intention of being used with adults (Aazh et al., 2018). There have been other studies which have reported of the ULL measures having no correlation between individuals' complaints of hypersensitivity to sound and ULL or DR measurements (Meeus et al., 2010). Aazh and moore (2017b) suggested hyperacusis diagnosis to be based on appropriate cut off values of 77 dB HL or lesser and a score of 22 or more on HQ. They also reported that PTAs are not strongly associated with ULLmin values. Villaume and Hasson (2017) recommended personality trait consideration in diagnosing as well as treatment of hyperacusis. All the studies included in this review under the combined measure (ULL and Questionnaire) should a similar assessment measure used. All the studies used ULL measure with HQ with variations in the adopted criteria for diagnosing hyperacusis.

Due to the high prevalence of tinnitus, otological abnormalities, and mental health issues, comprehensive audiological, otological, and psychological examinations are advised when assessing individuals with severe hyperacusis (Aazh et al., 2019; Aazh & Moore, 2017a, 2017b, 2018; Abouzari et al., 2020; Assi et al., 2018; Blaesing & Kroener-Herwig, 2012; Jüris et al., 2013; Villaume & Hasson, 2017).

4.2.3 Objective measure

Some authors have suggested the use of acoustic reflex thresholds (ART) complementary to the subject test of LDL measures which can be useful in the difficult to test population (younger population, intellectually disabled, etc), making the measure more objective as well as reliable (Kiessling et al., 1996; McLeod & Greenberg, 1979). Some studies tried but did not find a correlation between LDLs and ART (Knobel & Ganz Sanchez, 2005). TEOAE suppression and hyperacusis also do not seem to have an association (Goldstein & Shulman, 1996). Some have tried to connect cochlear neuropathy to hyperacusis which can be present in patients with hearing sensitivity within normal limits with otoacoustic emissions (OAEs) and PTA (Hickox & Liberman, 2014; Plack et al., 2014). We can conclude that there has not been evidences to show correlation between objective measures. There is no evidence pointing towards any solid linkage between the audiological tests done clinically and hyperacusis (Anari et al., 1999a). There is no correlation between DPOAE with HQ in healthy normal subjects as well as patients with MCS (Viziano et al., 2017).

4.2.4 Proposed hyperacusis definition

We saw in the beginning how there is no consensus for a universally accepted definition. As a result, there are several restrictions. The prevalence of hyperacusis as mentioned before varies a lot depending on the referrals made, the assessment measure used, population assessed, etc,. All the definitions suggest that individuals with hyperacusis complain of discomfort when exposed to sounds that are typically acceptable to the general population with hearing within normal limits. It is important to identify the individuals with different types of DST like hyperacusis, phonophobia, misophonia, diplacusis and polyacusis (Jastreboff & Jastreboff, 2012).

We can define hyperacusis as, a discomfort to sounds within range of comfort, impairing daily functioning at the social, emotional and attentional level; which does not show on any objective audiological measure and needs to be assessed by subjective measures such as ULL and self-report questionnaires.

4.2.5 Proposed hyperacusis assessment protocol

When the patient (individual with a complaint of hypersensitivity to sounds) reports to the clinic, the first step is to take a detailed case history which will include all the demographic details, sign and symptoms of the complaints, family history, previous medical reports and presenting complaints. Now, the main question to ask before any further testing will be to ask if the patient has "any hypersensitivity or low tolerance to sounds?”. This will help the clinician with understanding the extent to which the hyperacusis may be present as well as to reduce the risk of higher presentation levels. In this way, the patient is more comfortable with the testing and has more control over the level of presentation of the stimulus. This will minimise the risk of underestimation of the ULLs, increasing the accuracy of diagnosis. The third step is now to carry out audiological tests (PTA in noise and quiet, frequency specific DR, and/or immittance, and/or OAE) to rule out presence of a hearing loss. ULLs calculation should be carried out (Aazh & Moore, 2017b). Along with the ULL values, we would require few more dimensions to be assessed including patient’s attentional, social and emotional aspect with respect to their hypersensitivity to sounds. Specific sounds that causes discomfort to the patient should be noted down ,e.g., dog’s bark, babies cry, etc which will help us better understand frequency specific information spread of the hyperacusis. As mentioned before, ULLs are lesser reported in children compared to adults, there needs to be a standardization of both ULL and Questionnaires for improving assessment of the younger population. Depending on the language preferred by the patient and the availability of the language adapted questionnaire, a self-report questionnaire should be completed. The questionnaire can be 10 item Khalfa’s HQ as proposed by Fackrell et al., 2015. Throughout the testing, proper instructions should be given to the patient. This will be made more clear in the following figure 4.1.

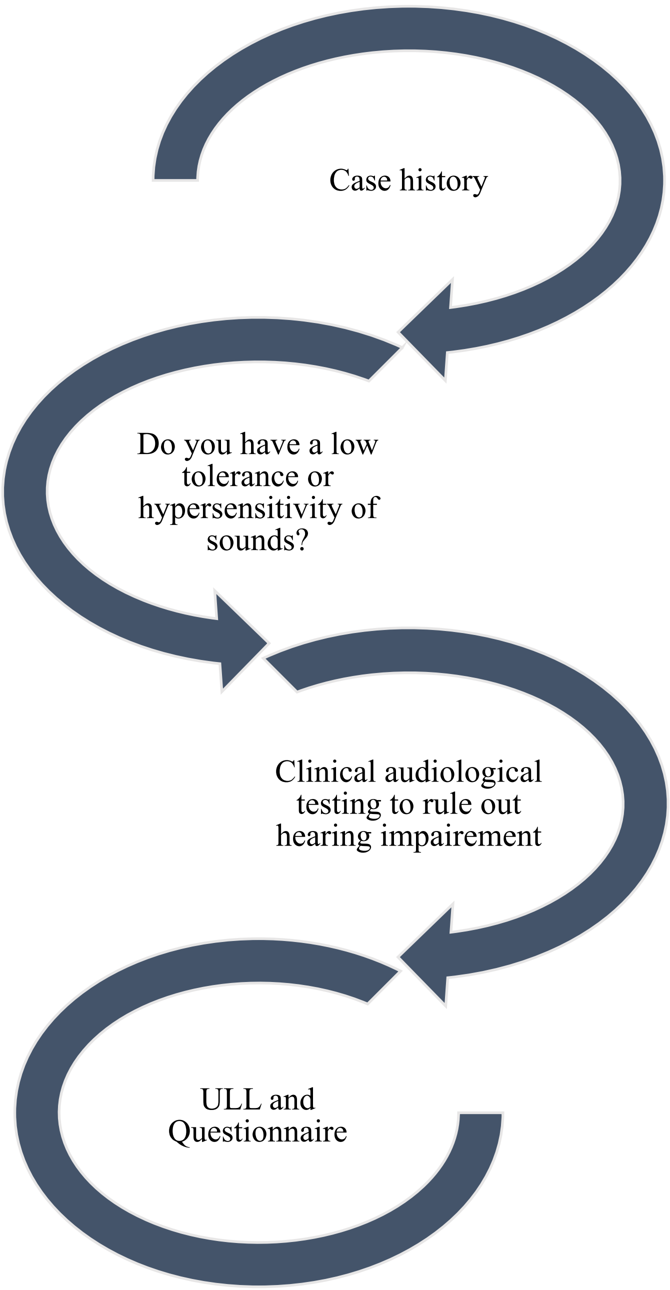


Figure 4.1 *Flow of testing for a patient with a complaints of hypersensitivity to auditory stimulus*

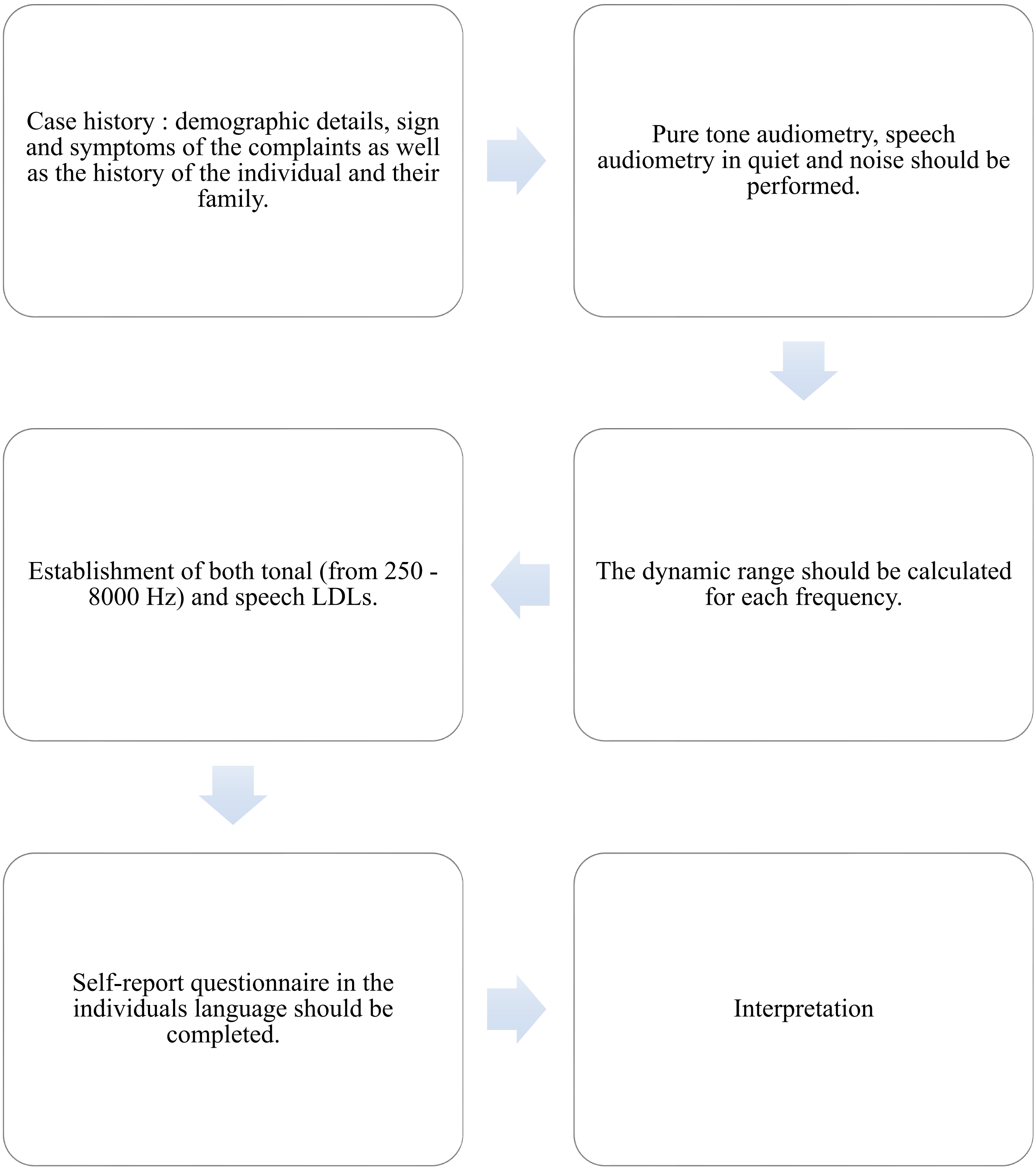


Figure 4.2 *Proposed assessment protocol of Hyperacusis*

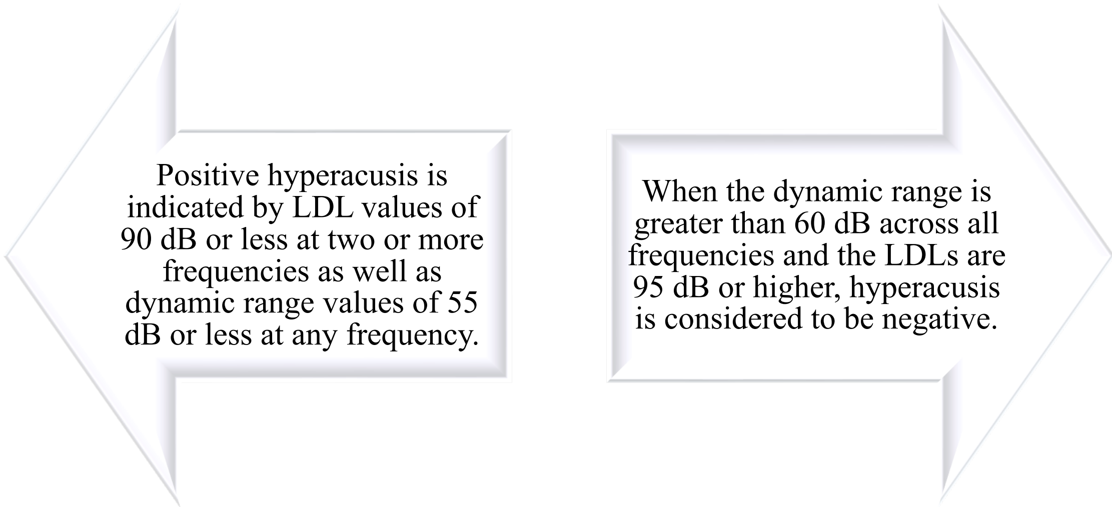


Figure 4.3 *Interpretation of LDLs*

The presence or absence of hyperacusis can be determined when interpreting the LDL scores. According to figure 4.3, hyperacusis is positive if the LDL at two or more frequencies is 90 dB or less, or if the DR at any frequency is 55 dB or less. When LDLs are 95 dB or higher at all frequencies and the dynamic range is 60 dB or higher at all frequencies, hyperacusis is considered to be negative.

Table 4.1 *Classification of severity of hyperacusis based on LDLs*

Severity	DR	LDL
None/Negative	60 dB or greater all frequencies	95 dB or greater all frequencies
Mild	50-55 dB at any frequency	80-90 dB at 2 or more frequencies
Moderate	40-45 dB at any frequency	65-75 dB at 2 or more frequencies
Severe	35 dB or less at any frequency	60 dB or lower at 2 or more frequencies

(Goldstein & Shulman, 1996)

The assignment of severity of hyperacusis can be done using the classification given by Goldstein and Shulman (1996). The self-report questionnaires that can be used to assess hyperacusis should be a standardised questionnaire with good sensitivity and specificity. It should be chosen for the target population (language and age group). All the above mentioned protocol is valid for the adult population. It may be used with children population as well, but we need a standardised and reliable ULL measure and questionnaire developed for the younger population.

Chapter V

SUMMARY AND CONCLUSION

Hyperacusis refers to hypersensitivity of hearing. We define hyperacusis as, a discomfort to sounds within range of comfort, impairing daily functioning at the social, emotional and attentional level; which does not show on any objective audiological measure and needs to be assessed by subjective measures such as ULL and self-report questionnaires.

There is no standard protocol that for evaluating decreased sound sensitivity and specifically hypersensitivity to everyday sounds (Fackrell et al., 2015). Due to this dearth of literature and gap in research, this study was carried out to identify and suggest a standard protocol based on the reported literature. There might be an inaccurate estimation of the incidence of hyperacusis provided by the authors of the articles included. The reason for this inaccuracy lies within the lack of solidarity of the definition used to classify hyperacusis. There is no correlation between ART and LDLs. LDL measure for hyperacusis has high reproducibility making it a good tool for follow up of individuals with hyperacusis undergoing treatment. As of now there are no objective measures that have been used to identify hyperacusis and neither is there a universal protocol used by clinicians which leads to variations in the prevalence as well as the management outcomes of individuals with a complaints of hypersensitivity to sound. We hence proposed a protocol that can be used by clinicians do patients with a complaint of hypersensitivity to sound.

1. Case history : demographic details, sign and symptoms of the complaints as well as the history of the individual and their family.
2. Pure tone audiometry, speech audiometry in quiet and noise should be performed.

3. Each frequency's dynamic range needs to be calculated..
4. Establishment of both tonal (from 250 - 8000 Hz) and speech LDLs.
5. Self-report questionnaire in the individuals language should be completed.
6. Interpretation : diagnosis of presence (with severity) or absence

5.1 Implications of the study

The current study can be used in any clinic to help diagnose an individual reporting with a complaint of hypersensitivity to everyday sounds. The set of protocol can be applied to come as a consensus diagnosis which will not vary in the type and severity of the disorder. This will help in management of hyperacusis as well as increase the accuracy of the prevalence. It will help in differential diagnosis of the individuals with DST. Differential diagnosis is needed in order to select patients for the disorder specific management. This will further increase the efficacy of the better treatment plans. This study has also pointed out the gaps in literature which need to be researched upon.

5.2 Limitations of the study

There might be an inaccurate estimation of the incidence of hyperacusis provided by the authors of the articles included. The reason for this inaccuracy lies in the lack of solidarity in the definition used to classify hyperacusis. Most of the articles included have used different measures, and it cannot be compared with each other. Some studies included retrospective studies where not all the data was available, leading to lesser scope for generalisation of the data on the entire population which the studies have mentioned themselves. Hyperacusis outcome measure are highly subjective, patient reported values and scores. This can make the measurement vulnerable to response bias.

5.3 Future Directions

To make the test and process more appropriate for the younger patient population, a revised set of instructions is required. Further, this will lessen the possibility of discomfort throughout the test, preventing an underestimation of the ULLs The test-retest reliability of ULLs measurements for children is required. A validated hyperacusis questionnaire for children is also required, in addition to the aforementioned.

For the younger age group, it is necessary to investigate various ULL patterns and determine how they relate to the sounds that distress them. It goes without saying that there is a gap in the education of those responsible for making DST referrals to the younger demographic.

Future research is required to use animal models to understand the processes of hyperacusis. Both a universal and standardised technique for diagnosing hyperacusis as well as a screening tool for DST complaints are required. This will aid in the early detection of children with hyperacusis for early intervention who would not be able to undergo the subjective measurements otherwise. The MOC reflex route needs to be validated, especially in children with WS.

The proposed protocol should be validated for individuals with a complaint of hypersensitivity to everyday sounds. The population may be young, adolescents, adults and older adults. Alongside, a standardised objective and subjective combined protocol for diagnosing hyperacusis must be developed. For greater clinical generalisation and population representation, a wider population validation of any of the questionnaires is required.

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