

CERTIFICATE

This is to certify that this dissertation entitled '**Efficacy of Tinnitus Retraining Therapy in treating Hyperacusis – A Systematic Review**' is a bonafide work submitted as a part for the fulfilment for the degree of Master of Science (Audiology) of the student Registration Number: 20AUD003. This has been carried out under my guidance and has not been submitted earlier to any other University for the award of any other Diploma or Degree.

Mysuru

August 2022

Dr. M. Pushpavathi

Director

All India Institute of Speech and Hearing
Manasagangothri, Mysuru 570 006

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Mysuru
August 2022

Dr.Ajith Kumar U
Guide
Professor in Audiology
Department of Audiology,
All India Institute of Speech and Hearing
Manasagangothri, Mysuru 570 006

DECLARATION

This is to certify that this dissertation entitled '**Efficacy of Tinnitus Retraining Therapy in treating Hyperacusis – A Systematic Review**' is the result of my own study under the guidance of **Dr.Ajith Kumar U**, 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.

Mysuru

Registration Number: 20AUD003

August 2022

**This dissertation is dedicated to
My Mother**

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TABLE OF CONTENTS

	Contents	Page Number
	List of Tables	i
	List of Figures	ii
	Abstract	
Chapter 1	Introduction	1
Chapter 2	Methods	6
Chapter 3	Results	9
Chapter 4	Discussion	31
Chapter 5	Summary and Conclusion	42
	References	43

LIST OF TABLES

<u>Table number</u>	Caption	<u>Page Number</u>
3.1	Study Characteristics of the selected articles	12
3.2	Results of the quality assessment for the selected randomised control trial study	23
3.3	Results of the quality assessment for all the selected cohort studies	24
3.4	Results of the quality assessment for all the selected literature review studies	26
3.5	Results of the quality assessment for all the selected diagnostic studies	27

LIST OF FIGURES

<u>Table number</u>	Caption	<u>Page Number</u>
3.1	PRISMA flowchart of the selection process of articles that were included in the review	10

ABSTRACT

Tinnitus Retraining Therapy (TRT) is a well-accepted treatment modality for tinnitus. It is developed as a practical application of Jastreboff's neurophysiologic model. Tinnitus is observed to be frequently accompanied by hyperacusis. Both these disorders have a common pathophysiology. Hence, they are expected to respond to TRT, which addresses the processes that contribute also to their etiology. This is a systematic review which proposes to elucidate the efficacy of TRT in the treatment of hyperacusis.

CHAPTER I

INTRODUCTION

Tinnitus retraining therapy (TRT) is a treatment modality which uses the habituation technique to improve the quality of life of the tinnitus patient. TRT functions by manipulating the limbic, autonomic and auditory systems. Thus, these systems respond less to abnormal stimuli. The same treatment technique is advocated for hyperacusis. This study is a systematic review of the efficacy of TRT in the treatment of hyperacusis.

Tinnitus is an annoying symptom with a moderately high prevalence encountered by otologists. It can interfere with a person's day-to-day activities. Tinnitus is described by patients as ringing, hissing, buzzing, cricket-like, or escaping steam (Jastreboff, 2011). Jastreboff describes tinnitus as 'as a phantom auditory perception, namely perception of sound without corresponding vibratory, mechanical activity in the cochlea' (Jastreboff, 2011).

Decreased sound tolerance, which includes two components, hyperacusis and misophonia, is seen concomitantly with tinnitus (Jastreboff, 2011). A patient can be diagnosed as having decreased sound tolerance when an exaggerated negative reaction is displayed to a sound which is tolerable to normal individuals (Jastreboff & Jastreboff, 2015).

Hyperacusis has been defined by Baguley as 'unusual tolerance to ordinary environmental sounds' and, more pejoratively, as 'consistently exaggerated or inappropriate responses to sounds that are neither threatening nor uncomfortably loud to a typical person (Baguley, 2003).

According to Baguley et al, 'hyperacusis' is used to describe the experience of everyday sounds being perceived as intense and overwhelming. Other terms used to express the same phenomenon are 'decreased' or 'reduced sound tolerance' (Baguley & Hoare, 2018). Stedman's Medical Dictionary defines hyperacusis as 'Abnormal acuteness of hearing due to increased irritability of the sensory neural mechanism'.

An increased gain within the auditory pathways leads to the condition known as hyperacusis. The intensity and spectrum of sound are the only determinants of hyperacusis. Enhanced functional connections between the auditory and the limbic and autonomic nervous systems are the reason for the occurrence of misophonia. Here the specific patterns of sound cause the reaction. The measure of total spectral energy is inconsequential (Jastreboff, 2011).

Tinnitus patients have co-existent hyperacusis. This is seen in 25–30% of tinnitus patients, according to studies by authors across the globe (Jastreboff, 2011). In the TRT based on the neurophysiologic model of Jastreboff, categorization of TRT patients is done first as the treatment protocol has been designed differently for each category. This categorization is based on the presence or absence of hyperacusis (Jastreboff & Jastreboff, 2015). Studies done independently by various authors have univocally acknowledged the positive effect of TRT in treating hyperacusis (Jastreboff & Jastreboff, 2015). Hyperacusis is amenable to the desensitization approach, which is the mainstay of TRT. Statistically significant results and clinical improvement have been noted in hyperacusis with TRT. This is a finding put forth by various researchers (Jastreboff & Jastreboff, 2015). Jastreboff found the presence of misophonia in about 60% of the patients. Hence, for the success of tinnitus treatment, first, decreased sound tolerance and its components must be diagnosed correctly and treated (Jastreboff, 2011).

Unlike misophonia, hyperacusis is amenable to the protocol of desensitization. Two months are usually sufficient for attenuating or curing hyperacusis (Jastreboff, 2011). Misophonia entails a longer duration of treatment which is more complex and involves specific protocols to be adhered to (Jastreboff, 2011).

Jastreboff introduced the neurophysiological model of tinnitus based on which the TRT was developed (Jastreboff, 2011). The underlying concept of the neurophysiological model of tinnitus is that many systems in the brain need to be considered when studying tinnitus and providing therapy (Jastreboff, 2011). TRT is done in clinical settings (Jastreboff & Jastreboff, 2015). The limbic and autonomic nervous systems are implicated in producing tinnitus. This is explained by the symptoms described by the patients. These include anxiety, annoyance, and strong emotional reactions. Tinnitus is diagnosed clinically by the activation of these systems and the symptoms produced thereby (Jastreboff & Jastreboff, 2002).

The auditory pathways bear signals which activate the autonomic nervous system (Jastreboff & Jastreboff, 2002). The method used in TRT is habituating this activation, thereby increasing the patient's tolerance to it (Jastreboff & Jastreboff, 2002). This is the primary objective of TRT (Jastreboff, 2011). The strength of these signals is decreased through sound therapy (Jastreboff & Jastreboff, 2002). In TRT, the patient is first habituated to the reactions evoked by tinnitus, following which the patient is habituated to the tinnitus perception (Jastreboff, 2011). TRT aims to retrain the brain so that it gets habituated to tinnitus. In the absence of TRT, tinnitus can activate the sympathetic nervous and limbic systems as a result of habituation of reaction and involves the cerebral cortex due to habituation of perception (Jastreboff, 2007). There are various other methods to achieve habituation of tinnitus. TRT differs from these in that it essentially incorporates two features which are based on the principles of the

neurophysiological model of tinnitus. They are (1) counselling, wherein tinnitus is reclassified into another category of neutral signals instead of negative ones and (2) sound therapy, put forth by Jastreboff and Hazell, which attempts to weaken neuronal activity related to tinnitus (Jastreboff & Jastreboff, 2006).

Tinnitus and hyperacusis utilize the exact neurophysiologic mechanisms and involve the same systems in the brain (Jastreboff & Jastreboff, 2002). Neuronal activity occurs in relation to tinnitus and also in hyperacusis. In the latter, external sound stimuli induce abnormal enhancement of this activity (Jastreboff & Jastreboff, 2002). A common treatment modality is thus plausible as both conditions have the common goal of total extinction of connections linking the neurophysiologic systems (Jastreboff & Jastreboff, 2002). The causative internal pathologic mechanism being similar, hyperacusis patients are predisposed to developing tinnitus, and the two are likely to co-exist (Jastreboff, 2011). The tinnitus signal is also enhanced by the increased gain in the auditory pathways that causes hyperacusis (Jastreboff & Jastreboff, 2002).

Hyperacusis is found in 0.2-17.2% of the population. It increases to 3.8-67% in the special population and reaches 4.7-95% in patients having special diseases. It is found to be increased in adolescents and older adults. Women have a significantly higher prevalence of hyperacusis than men. The prevalence in those with hearing impairment shows a significant increase compared to those who possess hearing within normal limits. Several diseases and conditions with greater exposure to sound have been identified as high-risk factors for hyperacusis (Ren et al., 2021).

Hyperacusis is diagnosed based primarily on the patient complaint of inability to tolerate various types of sounds along with decreased threshold values in the Loudness Discomfort Levels (LDL) or Uncomfortable Loudness Levels (ULL)

(Sanchez & Pereira, 2019). Hyperacusis has not been proved to be amenable to medical management. In a report of successful treatment of hyperacusis in two male children, Sanchez et al has used Ginkgo biloba leaf extract along with sound enrichment⁹. There is no clear causal relationship between anxiety and hyperacusis though present evidence suggests a possible link between them (Aazh & Allott, 2016). Hence, cognitive behavioural therapy has its limitations in the treatment of hyperacusis (Aazh & Allott, 2016). Thus, TRT is a treatment modality to be strongly considered in the treatment of hyperacusis.

1.1 Justification of the study

Several researchers have reported their views and experiences on this treatment method for hyperacusis. However, the efficacy of tinnitus retraining in the management of hyperacusis has not been systematically reviewed so far. Therefore, the current study was taken up to systematically document the efficacy of TRT in the management of hyperacusis. This study is proposed to be an aid to the audiologist in planning the treatment of hyperacusis effectively.

1.2 Aim

The present study aims to a systematic review on Efficacy of Tinnitus Retraining Therapy in the treatment of individuals with hyperacusis.

1.3 Objectives

To systematically review the efficacy of TRT in treating hyperacusis.

CHAPTER II

METHODS

The systemic review was conducted based on the Preferred Reporting Items for Systematic Review and Meta-analyses statement (PRISMA statement) (Page et al., 2021). A systematic literature search was carried out for peer-reviewed articles published from 2000 to 2022.

2.1 Information sources

The following databases were extensively searched for studies on the efficacy of Tinnitus Retraining Therapy in treating hyperacusis: PubMed/Medline, Google Scholar, Science Direct, and Scopus. Lists of references and citations were searched manually for further relevant studies.

2.2 Search strategy

The search was carried out using the following key terms, related search phrases, derivatives, and MeSH words relevant to the study combined with Boolean operators such as 'AND,' 'OR,' 'NOT. "Tinnitus" OR "Tinnitus retraining therapy" AND "hyperacusis" OR "loudness intolerance" OR "loudness discomfort level" OR "decreased sound tolerance" were used as the key terms for searching studies.

2.3 Study selection

The specific inclusion and exclusion criteria for the selection of studies were as follows:

2.3.1 Inclusion Criteria

- All parameters for measuring the efficacy of TRT will be included.
- Original and review articles published over the past 20 years
- Observational Analytical Studies, Experimental studies, Cross-sectional studies, and Review articles shall be included

2.3.2 Exclusion Criteria:

- Articles with low methodological quality and language apart from English.
- Case reports, animal studies, studies with insufficient data, and studies with duplicated data will be excluded

2.4 Data extraction

The search results were combined using the Rayyan QCRI (Qatar Computing Research Institute) and Mendeley desktop reference manager system, and the duplicate studies were eliminated. The studies that met the inclusion criteria were identified by screening the titles and abstracts retrieved from the search strategies. Thereafter, the full text of the potential studies was retrieved and matched to see if they were eligible. The extracted data included: article title, author details with their affiliation, year of publication, research design, study population, sample size, age group, comparison group, method of outcome measures and keywords specific to finding Efficacy of Tinnitus Retraining Therapy in treating Hyperacusis.

2.5 Quality assessment:

The Critical Appraisals Skills Programme (CASP) was used to conduct a methodological quality assessment of the included studies. The finding has been shown in the result section in detail.

CHAPTER III

RESULTS

A total of 11,264 articles were identified using database searches, with 10 duplicates eliminated. A total of 11,251 articles were included in the title/abstract screening. Following titles and abstracts review, 44 articles were selected for the full-length article screening. Fifteen articles matched the inclusion criteria in the study. The remaining 26 articles were excluded mainly because of the study design (pilot study, letter to the editor, case reports) and irrelevant study population (study population had no complaint of hyperacusis.). A detailed Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flow chart for the selection of the study is shown in Figure 3.1

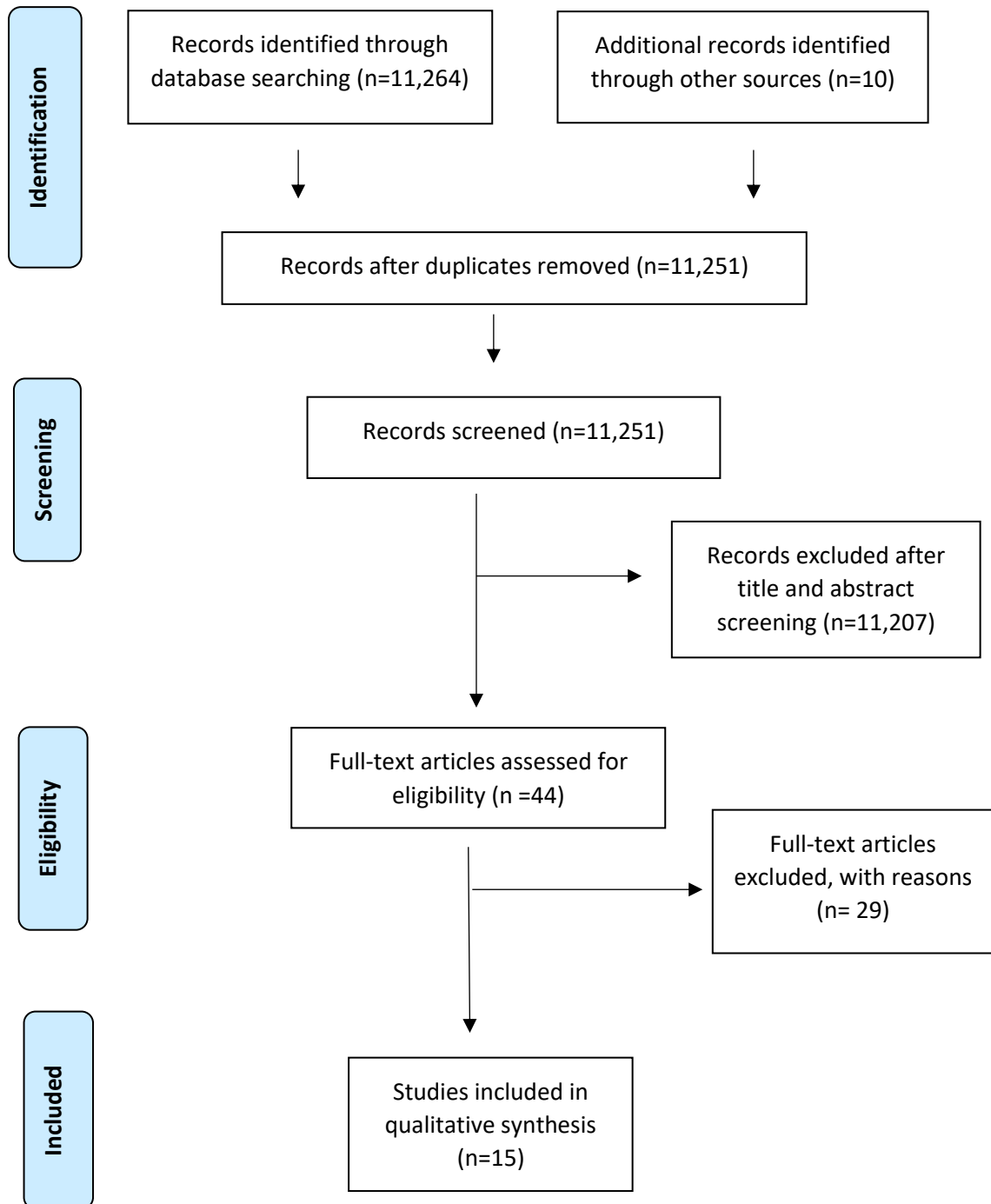


Figure 3.1: PRISMA flowchart for the selection process of articles included in the review

3.1 Study Characteristics

Out of the 15 articles finalized for review, five focused on the population with hyperacusis as the primary complaint, nine focused on the population with tinnitus as a primary complaint, and one focused on the population with tinnitus and hyperacusis both as a primary complaint. Amongst the 5 articles which focused on Hyperacusis as primary complaint, three studies state the use of TRT, and two studies state the use of other treatment approaches. Furthermore, nine studies focused on population with tinnitus as primary complaint and one study on both tinnitus and hyperacusis as primary complaint. To describe the efficacy of treatment of hyperacusis, studies used various outcome measures. Table 3.1 summarizes the type of treatment evaluated, type of outcome measured, study design, study population details, and study outcomes.

Table 3.1 *Study Characteristics of the selected articles*

STUDY	RESEARCH DESIGN	RESEARCH QUESTION	POPULATION	PARAMETERS USED	OUTCOME
Aazh et.al (2016)	Observational Cross sectional Analytical Survey	Is there significant difference in the therapies for tinnitus and hyperacusis as judged by the patients?	Attendees of a clinic for tinnitus and hyperacusis	Questionnaire for the service evaluation survey Scale from 1 to 5	Counseling had the greatest score.
Smit et.al (2022)	Observational Cross sectional Analytical	What are the ways tinnitus treatment is used, what are their outcomes and what are the factors that influence tinnitus?	Online attendees of support community for tinnitus patients	Surveys were done on tinnitus treatment and its results and the influencing factors	According to usage the treatment modalities were as follows: tinnitus therapies, self-administered sound therapy and natural supplements. Tinnitus treatment was significant in in females, those with high impact of

					tinnitus, hearing loss and hyperacusis.
Zhangh et.al (2011)	Observational Cross sectional Analytical	What are the different factors resulting in recovery, and how are they related to tinnitus symptoms?	Tinnitus treatment records of 253 patients were scrutinized and 126 attributes were applied and the association between treatment factors and recovery were investigated	Clustering and new Temporal Features TRT Data Collection, Information Retrieval, Mining Text Data, Temporal Feature Design for Continuous and Categorical Data, System Overview	On applying TRT, the association between treatment factors and recovery was assessed in the first of 3 experiments done. According to Rule 13 of the results, the presence of tinnitus, hyperacusis, and hearing loss leads to chance of improvement

Alam et.al (2017)	Observational Longitudinal Analytical Cohort study	What is the success rate of treating tinnitus patients with TRT when noise from mobile phones is used for sound therapy?	30 male adult patients	<p>(1) Awareness of tinnitus when awake during the last week. Expressed in percentage</p> <p>(2) Time for which symptoms were present expressed as percentage</p> <p>(3) Life factors affected</p>	In persons with hyperacusis with or without tinnitus, hearing loss irrelevant, and having no kindling effect 50% showed improvement and in persons with hyperacusis (intolerance towards loud sounds) with or without tinnitus and on symptoms after noise exposure 30% showed significant improvement.
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Turhan et.al (2020)	Observational Longitudinal Analytical Cohort study	What are the outcomes of masking, counseling, and a new sound therapy protocol in subjective tinnitus patients?	60 subjective tinnitus patients	Tinnitus handicap questionnaire	Median tinnitus handicap questionnaire (THQ) score differences in the beginning of the study to the second month and to the fourth month were significantly prominent in hyperacusis negative patients.
Grewal et.al (2014)	Systematic Review	What are the outcomes of TRT and CBT?	Medline (using Ovid) and the Web of Science, up to and including February 2013	Critical appraisal of 4 TRT and 10 CBT studies	Results from Wide Band Noise generators are dependent on the level of tinnitus (Jastreboff scale), the use of hearing aids, and hyperacusis

Hazell et.al (2002)	Observational Longitudinal Analytical Cohort study	What are the predisposing factors, triggers and outcomes after TRT?	187 consecutive patients from the Tinnitus and Hyperacusis Centre, London. with a primary diagnosis of decreased sound tolerance (DST)	Number of life factors affected and loudness discomfort levels (LDL)	Following treatment in a tinnitus retraining therapy programme loudness discomfort levels reached normal levels as follows: 44.8% by 6 months, 51.4% by 15 months and 60.4% by 25months
Bartnik et.al (2012)	Observational Longitudinal Analytical Cohort study	What is the epidemiology, audiological profile and preliminary effects of the therapy in children with troublesome tinnitus?	143 children consulted in the Tinnitus Clinic, Institute of Physiology and Pathology of Hearing, AK Kampinos 1, 01-943 Warsaw, Poland in 2009	Proper categorization of Tinnitus Retraining Therapy (TRT)	Hyperacusis is present in category III and may be present in category IV of 'TRT mixing point-level' close to the place where the sound from the instrument blends with the tinnitus.

Scherer et.al (2019)	Randomized Clinical Trial	Does treatment with tinnitus retraining therapy with conventional sound generators result in better outcomes than tinnitus retraining therapy with placebo sound generators or standard of care when used in subjective debilitating tinnitus and audiometric normal to mild hearing loss?	Eighty-three individuals	One-year	The positive and significant effects on anxiety and depression, which were achieved immediately after tinnitus habituation therapy, were maintained.
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Potgieter et.al (2020)	Scoping Review	What is the literature in major databases regarding hyperacusis in children?	The underling factors on which the research topic is based	(1)The aim and research questions, (2) selection of relevant studies (3) team based selection of the study (5) collation of results, summarisation and reporting, (6) review by clinical experts who were not part of stages 1–5. treatment program and Survey	Psychological therapy, sound therapy, tinnitus retraining therapy, medication and neuro-rehabilitation are used for treating children showing hyperacusis.
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Silverstein (2016)	Prospective, longitudinal cohort	Is there a decrease in hypersensitivity to sound before and after surgical procedure for the treatment of hyperacusis?	Severe hyperacusis patients who are not children	Scores of Uncomfortable loudness level and hyperacusis questionnaire	TRT benefitted a fair number of patients. Broadband noise generators and extended therapy are encouraging. Medically compromised people may not show improvement of hyperacusis
Thong et al (2011)	Prospective Cohort study	What is the efficacy of Tinnitus Retraining Therapy (TRT) in patients with hyperacusis?	197 patients	Loudness discomfort level (LDL) measurements, Structured interview with aid of questionnaires and visual analogue scores.	Improvement with Vienna tone was significantly higher than with environmental sounds alone (49% vs 35%, $P < .05$), especially when used in presence of hearing loss (61%)

Aazh et al(2016)	Cohort Study with Retrospective service evaluation survey	Is there significant difference between the effectiveness of TRT relative to a simplified version of TRT (sTRT)	Two groups of 12 consecutive patients who received TRT and sTRT each	Tinnitus Handicap Inventory and the Visual Analogue Scale	TRT and a simplified version of TRT (sTRT) show no difference
Lee et al (2019)	Ambispective Cohort study	Is the difference between pre- and post-TRT resting-state quantitative electroencephalography findings seen to show improvements in Tinnitus Handicap Inventory (THI) scores and numeric rating scale (NRS) scores?	The rs-qEEG data records of tinnitus patients	Audiometric tests and a pre-TRT rs-qEEG were done. Tinnitus Handicap Inventory (THI) was used	The brain activity of tinnitus patients varies with the degree of hearing loss or combined hyperacusis

Abouzari et.al (2020)	Prospective Cohort (Observational Longitudinal Analytic) study	What is the efficacy of a multi-modal migraine prophylaxis therapy for patients with hyperacusis?	Hyperacusis patients	Multi-modal step-wise migraine prophylactic regimen (nortriptyline, verapamil, topiramate, or a combination thereof) as well as lifestyle and dietary modifications	Hyperacusis patients show improvement with the views of various modes of migraine prophylactic therapy just as other known treatment option for hyperacusis like TRT.
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3.2 Quality Assessment

The Critical Appraisals Skills Programme for randomized controlled trials (CASP) (Carneiro et al., 2020) was used to assess the quality of the studies. It is a generic tool for appraising the strengths and limitations of any qualitative research methodology. It consists of 11 questions to assess the article in depth across each section to reduce bias. The questions in the tool are marked as "Yes", "No" or "Can't tell," depending on the question's requirement. The results of the quality assessment for all of the selected studies are provided in Tables 3.2, 3.3, 3.4 and 3.5.

Thong et al (2007)												
Silverstein et.al (2016)												
Bartnik et.al (2011)												
Hazell et al (2008)												
Purhal et.al (2020)												
Alam et.al (2017)												
Total % of Yes	100	100	100	100	77.77	44.44	77.77	100%	100	88.88	66.66	77.77

Table 3.4. Results of the quality assessment for all of the selected studies. Systematic Review checklist

	Can the study be relied upon?					Will the results help locally?		
	1. Did the review address a clearly focused question?	2. Did the authors look for the right type of papers?	3. Do you think all the important, relevant studies were included?	4. Did the review's authors do enough to assess quality of the included studies?	5. If the results of the review have been combined, was it reasonable to do so?	6. Can the results be applied to the local population?	7. Were all important outcomes considered?	8. Are the benefits worth the harms and costs?
Potgieter et.al (2020)	Green	Green	Green	Green	Yellow	Yellow	Green	Green
Spielmann et al (2014)	Green	Green	Green	Green	Green	Green	Green	Green
Total % of yes	100	100	100	100	50	50	100	100

Table 3.5. Results of the quality assessment for all of the selected studies. Diagnostic study Checklist:

	Are the results of the study valid?						Will the results help locally?		
	1. Was there a clear question for the study to address?	2. Was there a comparison with an appropriate reference standard?	3. Did all patients get the diagnostic test and reference standard?	4. Could the results of the test have been influenced by the results of the reference standard?	5. Is the disease status of the tested population clearly described?	6. Were the methods for performing the test described in sufficient detail?	7. Can the results be applied to your patients/the population of interest?	8. Can the test be applied to your patient or population of interest?	9. Were all outcomes important to the individual or population considered ?
Aazh et al(2016)									
Smit et.al (2022)									
Zhang et.al (2011)									
Total % of yes	100	100	100	0	100	100	100	100	



Yes



Can't tell



No

On analysis, as depicted in Tables 3.2,3.3,3.4 and 3.5 it was found that all the studies were of good quality, which includes 1 randomized control trial, 9 cohort studies, 2 literature reviews and 3 diagnostic studies. Seven out of 11 questions (question numbers 1, 2, 3, 5, 6, 7, 9) were answered as "Yes," for randomized control trial study (table 3.2), Six out of ten questions (question numbers 1, 2, 3, 4, 8, 9) were answered as "Yes," for all Cohort studies (table 3.3) six out of eight questions (question numbers 1, 2, 3, 4, 7,8) were answered as "Yes," for all Review articles (table 3.4), eight out of nine questions (question numbers,1,2,3,5,6,7,8,9) were answered as "Yes", for all diagnostic studies (table 3.5), indicating good quality appraisal across the literature selected. In randomized control trial studies, the research questions were addressed, all the participants included in the intervention group were treated equally, and the treatment effects were reported comprehensively. No participant was excluded at the conclusion. Whether results are applicable to the residents in the vicinity was not clearly stated. If the experimental intervention is giving better results than any of the presently used interventions was not clearly stated. All pertinent confounding factors were identified in 7/9(77.77%) of Cohort studies. Have they considered the confounding factors in the design and/or analysis. In comparison, 2/9(22.22%) studies have not clearly identified all important confounding factors and have not efficiently taken account of the confounding factors in the design and/or analysis. 7/9(77.77%) studies showed a complete follow-up of subjects. In comparison 2/9(22.22%) studies did not clearly state about complete follow up of subjects. Results of 8/9(88.88%) studies can be applied to the residents in the surrounding area. In comparison 1/9(1.11%) studies did not show a clear application of results in a local population. The results of 6/9 studies fit with other available evidences. In comparison, the results of 3/9 studies cannot be correctly fit with other available evidence. 7/9(77.77%) studies

show implications of their study for practice. In comparison 2/9(22.22%) studies does not clearly state the implications of its study for practice. In literature reviews 1/2 (50%) results of the review have been combined, and it was reasonable to do so, and the results can be applied to the population in the locality. In diagnostic studies, all the studies show the results of the test have not been clearly influenced by the results of the reference standard.

CHAPTER IV

DISCUSSION

This systematic review is to assess the efficacy of Tinnitus Retraining Therapy (TRT) in the treatment of hyperacusis. Hyperacusis and tinnitus have a common etiology as the same neurophysiologic mechanisms play a role in both. It is observed that tinnitus is often accompanied by hyperacusis. Also, the presence of hyperacusis has been used as a criterion for the categorization of patients for TRT. Many studies have been done wherein this highly effective and popular treatment modality has been utilized to address hyperacusis. Most authors have reported very encouraging results in treating hyperacusis using TRT and recommended the application of TRT for hyperacusis with or without tinnitus.

In this systematic review, fifteen articles were enrolled in the study after going through the specified methodology in article selection. Of these six were prospective cohort studies, three were retrospective cohorts, one was an ambiceptive cohort, two were diagnostic cross sectional analytical studies, one was a randomized control trials and two were systematic reviews. This study has provided data regarding the various results obtained by TRT when used in the treatment of hyperacusis.

4.1 Cohort studies

4.1.1 Prospective cohort studies

In his prospective longitudinal cohort study using minimal invasive surgery for the treatment of hyperacusis, Herbert Silverstein et al have agreed with Aazh et al that TRT is currently one of the treatment options for noise intolerance (Silverstein et al., 2016). The rate of efficacy of treatment results, however, is noted to vary with the

different treatment modalities like avoidance of provocative stimuli, cognitive behavioural therapy, TRT and hearing amplification (Silverstein et al., 2016).

Thong et al., in a study with a prospective cohort design of hyperacusis patients, investigated the efficacy of TRT in these patients (Thong et al., 2011). The authors have observed that hyperacusis is frequently found in patients who complain of tinnitus. Loudness discomfort level (LDL) measurements were used to confirm hyperacusis. All patients were subjected to audiometry. Questionnaire-supported interview, which is structured and visual analogue scores, were used to assess the improvement of hyperacusis following TRT. Of a total of 197 patients. Only a few had hearing aids (H.A.) fitted, even though a majority had hearing loss. It was found that these patients complained of worse effects of hyperacusis on life (Thong et al., 2011). Improvement was not related to age, gender, and the presence of hearing loss. This research reveals that only a relatively small percentage of patients with hyperacusis improved with TRT. Better results were obtained by using broadband noise generators and a longer duration of therapy. The study found that medically compromised hyperacusis patients show poor success using TRT (Thong et al., 2011).

In a prospective cohort study, Abouzari et al evaluated the multi-modal migraine prophylaxis therapy for patients with hyperacusis. They have opined, quoting several authors, that treatment options for hyperacusis include tinnitus retraining therapy as also avoidance of various treatment modalities (Abouzari et al., 2020).

Hazell et al. conducted a prospective cohort study among patients at the Tinnitus and Hyperacusis Centre, London (Hazell et al., 2002). This analysis of 187 cases of decreased sound tolerance (DST) revealed a variety of predisposing (trigger) factors. Treatment was done in a tinnitus retraining therapy programme, and the results were

encouraging. By the 6 month visit, loudness discomfort levels reached normal levels at 44.8%. It increased to 51.4% by the 15-month visit and 60.4% by the 25th-month visit. There was a fall in the mean number of life factors/activities affected by hyperacusis from 3.5 at the 1st visit to 1.1 at the 15 month visit. The study clearly supports the use of TRT in the treatment of hyperacusis. Further, the study also provides the valuable finding that the trigger factors do not affect the outcome. Jastreboff categorization was done for all patients. Jastreboff category 4 was assigned when the symptoms were enhanced by exposure to sound for an extended period in excess of 24 hours, and Jastreboff category 3 was when they were not. It was noted that the primary problem was hyperacusis. Except in 3.6% of patients, tinnitus was also present along with DST, though not as the dominant symptom. A total number of two to seven visits was required per patient. This was decided according to need and according to the TRT protocol. TRT protocol was strictly adhered to in the study. When it was required to explain and make the patient understand what he or she was experiencing, the Jastreboff model was presented. Patients belonging to Category 4 were treated in a similar manner, but the volume of the sound generator was increased very gradually. Hyperacusis was assessed by the number of life factors that were affected or interfered with. The results of this study confirm the previous findings of the same authors and also many others. This was proved by three factors: improvement in life factors previously affected by DST, reduction in LDLs and reduction in the distress caused by tinnitus (Hazell et al., 2002).

A prospective cohort by Turhan et al has evaluated the outcomes of masking, counselling, and a new sound therapy protocol in subjective tinnitus patients (Turhan et al., 2020). The rate of hyperacusis was 58.3%, 61.5%, 46.2%, 66.7%, and 60% in sixty chronic subjective tinnitus patients who were randomly assigned to five groups. Hyperacusis was present in 50% in group A(masker given), 62.5% in group B (masker

given with habituation) and 60% in group C (only habituation). It is seen that THQ1-2 and THQ1-3 score differences were significantly prominent in hyperacusis negative patients. The authors have stated that habituation to tinnitus was developed after the neurophysiologic model was proposed in the 1970s. The neurophysiologic model is clinically implemented as TRT. This is based on the habituation of the brain and body reactions to tinnitus.

Jastreboff has categorized tinnitus patients into five categories. It has been well documented that patients with hyperacusis (categories 3 and 4 of Jastreboff) show less response to treatment, whatever their level of tinnitus. This has been validated in this study. Hyperacusis was incident in 58.3% of this study; however, hyperacusis was not a criterion in patient selection in this study. Hence all groups showed a similar presence of hyperacusis. Both masking and habituation patients with hyperacusis also showed less progress, as reported by other authors. Hazell JWP et al have endorsed the recommendation of Jastreboff that in the presence of hyperacusis, a slow desensitization protocol and treatment of hyperacusis prior to the treatment of tinnitus is needed. They have also advocated the use of LDLs to monitor hyperacusis (Turhan et al., 2020).

Alam et al have used Jastreboff's categorization in their prospective cohort study. Of the five categories, category 3 included persons with Hyperacusis with or without tinnitus, hearing loss irrelevant, and having no kindling effect as in category 4. The treatment provided was counselling and desensitization using noise stimuli (sound treatment). Category 4 included persons with hyperacusis (intolerance towards loud sounds) with or without tinnitus and with a prolonged effect on symptoms after noise exposure. The treatment provided was intensive counselling and sound treatment. The efficacy of TRT using noise presentation from mobile phones was studied. It was found

that only 50% of patients showed improvement of tinnitus in category 3 and only 33% improved in category 4. This proves the effect of the presence of hyperacusis on tinnitus therapy using TRT and the need to address hyperacusis before tinnitus in patients where both are present (Alam et al., 2017).

4.1.2 Retrospective cohort studies

In a retrospective cohort study, Aazh et al. have compared TRT and a simplified version of TRT in the treatment of tinnitus in adults (Aazh et al., 2008). When used in the context of TRT, DST is used as a blanket term to include Hyperacusis and other forms of sound intolerance. It is to be noted that the therapy first focused on the DST, and the tinnitus was addressed more directly afterwards. In Simplified TRT WSGs were fitted using open molds, and sound enrichment using sound generators was advised.

The results showed there is no significant difference between the improvement in THI scores for patients with and without DST between TRT and a simplified version of TRT (sTRT) (Aazh et al., 2008).

Bartnik et al have studied epidemiology, audiological profile, and preliminary results of treatment in troublesome tinnitus in children in their retrospective cohort study (Bartnik et al., 2012). Here the authors have used the impact of tinnitus on the patient's life, presence or absence of hyperacusis, subjective hearing loss, the presence of prolonged worsening of tinnitus, and/or hyperacusis after exposure to loud sound as the factors by which patients are allotted to the different categories. In the categories of TRT mixing point-level close to the place where the sound from the instrument blends with the tinnitus, Hyperacusis was found to be present in categories 3 and may or may not be present in category 4. The main problem was hyperacusis in category 3 and was either tinnitus or hyperacusis in category 4. Children whose only problem was

hyperacusis and who showed abnormality in the tympanometry test were excluded from this study (Bartnik et al., 2012).

Zhang et al, in their retrospective study, have stated, referencing other authors, that hyperacusis frequently accompanies tinnitus (Zhang et al., 2011). In this study, they used the interview method for data collection. This method also helps determine the role of hyperacusis and misophonia in TRT. The process of TRT also includes sound therapy besides surveys for the purpose of symptom evaluation. According to the principle of TRT, the differences in the stimuli from the background are worked upon. This principle is based on the theory that the stimulus is not associated with the perceived signal (Zhang et al., 2011).

4.1.3 Ambiceptive cohort

In an acceptive retrospective cohort study by Lee et al, it was found that tinnitus patients show different brain activity patterns according to the degree of hearing loss or hyperacusis. Therefore, they have advised evaluating the part played by cortical activity (Lee et al., 2019).

4.2 Diagnostic cross sectional analytical studies

Smit et al, have reported two diagnostic cross-sectional analytic surveys which were done to analyze the tinnitus and the results and the factors that influence tinnitus (Smit et al., 2022). Hyperacusis was one of the comorbid conditions, which is described as "a sensitivity to sounds, that is the sounds are irritating and painfully loud when others hear them as normal". 58.3% of patients experienced hyperacusis in this study (Smit et al., 2022).

A diagnostic cross sectional analytic survey was done by Aazh et al to assess patients' judgements of the effectiveness of the tinnitus and hyperacusis therapies

offered in a specialist U.K. National Health Service audiology department (Aazh et al., 2016). TRT has an educational component which is used as the basis for developing the presentation. However, there is poor evidence to state that education alone is sufficient to manage tinnitus or Hyperacusis. An article which was selected in the review by Aazh et al. gives credence to the role of TRT in the effective management of hyperacusis.

The authors have mentioned that several authors such as Pienkowski et al in 2014 and Tyler et al in 2001 have recommended the use of the same in the management of tinnitus and hyperacusis. The hyperacusis questionnaire by Khalifa et al was used in this study (Aazh et al., 2016).

The authors warn that the outcomes of this study may not be representative of the whole sample of tinnitus and hyperacusis patients, and therefore findings need to be interpreted with some caution. The study provides the insight that counselling practice and training are required for audiologists (Aazh et al., 2016).

4.3 Randomized controlled trial

Scherer et al, in a randomized control trial, has compared TRT and standard of care in the treatment of tinnitus (Scherer & Formby, 2019). Before randomization, the participants were assigned to five categories. Of these, the IIIrd category indicated hyperacusis, no prolonged effect of noise exposure, and subjective hearing loss irrelevant; and the IVth indicated hyperacusis, the prolonged effect of noise exposure, and subjective hearing loss irrelevant. They found few differences among the standard of care, TRT, or partial TRT in the treatment of tinnitus. Each of these treatments for debilitating tinnitus resulted in clinically significant improvement in most treated individuals. However, the authors have admitted to several limitations in their study.

Improvement in hyperacusis has not been specifically mentioned (Scherer & Formby, 2019).

4.4 Systematic reviews

Potgieter et al. have done a scoping review on hyperacusis in children (Potgieter et al., 2020). Hyperacusis and TRT are two conditions which are often co-occurring. Hence, though TRT was originally designed for tinnitus, it can be suited to treat hyperacusis with good results. Tinnitus was found to be just as common in children with hyperacusis, followed by William's Syndrome and Attention Deficit Hyperactivity Disorder. Hence the question has been put forth by the authors for future research on whether hyperacusis is a symptom or a disease (Potgieter et al., 2020).

Grewal et al. have done a systematic review to compare the outcomes of tinnitus retraining therapy and cognitive behavioural therapy (Grewal et al., 2014). The severity of hyperacusis also increases from no significant hyperacusis from Grade 0-2, significant hyperacusis with no prolonged worsening after sound exposure in Grade 3 to hyperacusis manifesting as the dominant complaint with worsening on exposure to sound (Grewal et al., 2014).

4.5 Studies with primary complaint of hyperacusis

4.5.1 Treated with TRT

Thong et al. studied patients with hyperacusis and demonstrated improvement with TRT (Thong et al., 2011).

Patients with primary complaints of decreased sound tolerance were taken up for the study, and required data was collected. Though the majority of patients exhibited

concomitant tinnitus, this was never the dominant symptom. Improvement was noted with TRT. Predisposing (triggering factors) were also identified (Hazell et al., 2002).

A scoping review of hyperacusis in children was carried out. TRT was one of the methods of management. Significant benefits were noted, with 75% of children showing improvement in one study (Potgieter et al., 2020).

4.5.2 Treated with other modalities, but TRT also advocated

Surgery is advocated for treating hyperacusis in patients with superior semicircular canal dehiscence. The other modalities, including TRT, are recommended for treatment (Silverstein et al., 2016).

Patients with hyperacusis were treated with a multi-modal staged regimen for migraine prophylaxis. Drugs and other modifications in living patterns were advised (Abouzari et al., 2020).

4.6 STUDIES WITH PRIMARY COMPLAINT OF TINNITUS

4.6.1 Treated with TRT but improvement seen in hyperacusis

Sixty subjective tinnitus patients were randomly assigned to five groups. Total masking was implemented in four groups (groups 1, 2, 3, and 4), while combined habituation therapy and sound therapy were implemented in one group (group 5). Patients were evaluated with the tinnitus handicap questionnaire (THQ) at the beginning of the study (THQ1) and in the second (THQ2) and fourth months (THQ3) (Turhan et al., 2020).

A total of 30 male adult patients with tinnitus were enrolled for TRT. TRT is comprised of two activities, i.e. directive counselling and sound treatment. Tinnitus

treatment may be provided with the help of mobile phones, which are a cheaper substitute for costly noise maskers (Alam et al., 2017).

TRT and TRT in a simplified form were compared in tinnitus patients (Aazh et al., 2008).

A study considered 143 children with tinnitus aged under eighteen. All the children with troublesome tinnitus took part in the TRT. There were no other preselection criteria except the requirements for children to be treated for at least 6 months, and each of them had to be actively present at all follow-up visits. 3% had hyperacusis. TRT resulted in improvement (Bartnik et al., 2012).

Utilizing the patient records as well as the total number of rules can be used to improve treatment effects (Zhang et al., 2011).

In a study, comparison of pre-and post-TRT resting-state quantitative electroencephalography (rsqEEG) findings, rs-qEEG data recorded before and after TRT were compared. A remarkable improvement was noted in the Numeric Rating Scale and perception (Lee et al., 2019).

Two surveys were done to find the relation between tinnitus impact and tinnitus outcomes (Smit et al., 2022).

In a randomized clinical trial of 151 participants with 18 months of follow-up, average tinnitus distress decreased in all 3 groups (receiving TRT, partial TRT and Standard of care) (Scherer & Formby, 2019).

Three steps are involved in a study comparing TRT and Cognitive Behavioural Therapy (CBT). TRT is recommended as a long term therapy (Grewal et al., 2014).

4.7 Studies with primary complaint of tinnitus and hyperacusis

The authors of a diagnostic study state that research in favour of sound therapy for tinnitus or hyperacusis is scanty. Educational sessions are used in concomitance in most studies (Aazh et al., 2016).

This systematic review reveals that TRT can be managed efficiently with hyperacusis. Relief from tinnitus is achievable only after successful treatment of the associated hyperacusis, as evinced by the selected studies.

CHAPTER V

SUMMARY AND CONCLUSION

The efficacy of treatment of hyperacusis using tinnitus retraining therapy is yet to reach a consensus among audiologists. It is well established that hyperacusis may co-exists with tinnitus in a fairly good proportion of cases. The TRT protocol uses Jastreboff's categorization where hyperacusis is present in Categories 3 and 4. It is also reported by many authors that hyperacusis has to be addressed first in cases where it is present along with tinnitus. The therapy for tinnitus is to be initiated after control of hyperacusis is achieved. Several studies have noted the common etiology of these two annoying and, many a time, distressing disorders. This has led audiologists to attempt treatment of hyperacusis with TRT, which is based on the neurophysiologic model of Jastreboff and can be called the clinical implementation of the latter.

A randomized controlled study on the efficacy of TRT on hyperacusis has not been conducted so far. This systematic review may help audiologists to use TRT in alleviating hyperacusis, either occurring alone as a primary complaint or in association with tinnitus.

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