# HYPERACUSIS AND MISOPHONIA MEASURES: AN EXAMINATION OF THE PSYCHOMETRIC PROPERTIES AND THE IMPACT ON HEALTH

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# **DEDICATION**

This thesis dedicated to my daughter, Miray Ece, who reminds me daily of life's joys and beauty, and to my dear husband, Halil Ibrahim, whose unwavering love and support have been the foundation of my success. This accomplishment is as much yours as it is mine.

**DECLARATION OF ORIGINALITY** 

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#### **ABSTRACT**

# HYPERACUSIS AND MISOPHONIA MEASURES: AN EXAMINATION OF THE PSYCHOMETRIC PROPERTIES AND THE IMPACT ON HEALTH

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Hyperacusis is characterised by an increased sensitivity to everyday sounds, while misophonia involves strong emotional, behavioural and physical reactions to specific trigger sounds, such as chewing or tapping. Despite their significant impact, some questionnaires used to assess these conditions exhibit varying psychometric robustness, and the psychological links between these conditions and health outcomes—such as fatigue, stress, and sleep quality—remain incompletely understood.

This thesis addressed these gaps through a systematic review and three empirical studies. The systematic review assessed eight widely used questionnaires against COSMIN standards, identifying gaps in reliability and validity, which provided the basis for the subsequent empirical studies. The first study validated the English version of MisoQuest, a 14-item misophonia questionnaire, demonstrating adequate model fit in CFA but finding that a one-factor structure was not fully supported due to shared variance among some items. While it showed good convergent and discriminant validity, moderate correlations with fatigue indicated potential construct overlaps, requiring further investigation. The second study explored this relationship between misophonia and fatigue, identifying stress and sleep quality as partial mediators, highlighting the potential for targeted interventions. The final study validated the 4C Hyperacusis and Misophonia Management Questionnaires in clinical population, demonstrating a one-factor structure for the 4C Hyperacusis Management and two-

factor structure for the 4C Misophonia Management, both with acceptable model fit. The questionnaires also showed strong reliability, along with good convergent and discriminant validity. These tools assess patients' confidence in managing symptoms, particularly within the context of cognitive-behavioural therapy.

To summarise, the results of this thesis provide novel evidence of the assessment of hyperacusis and misophonia by validating questionnaires and examining the relationships between psychological factors and health outcomes.

#### **AUTHORSHIP DECLARATION**

This thesis is structured as a series of separate manuscripts, with Chapter two has already been published, while Chapter three and Chapter four have been submitted to journals and are currently under peer-review. Chapter five will be submitted to American Journal of Audiology.

# • Chapter Two:

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#### OTHER OUTPUTS DURING CANDIDATURE

# **Accepted Conference Presentations**

**Oral Presentation** "Mediation Analysis on the Correlation Between Misophonia and Fatigue, Sleep, Stress, Hyperacusis, and Mental Health: Preliminary Results." at the 36<sup>th</sup> World Congress of Audiology in Paris, 19<sup>th</sup> and 22<sup>nd</sup> of September 2024.

**Oral Presentation** "Validity and Reliability of Misophonia Impact Questionnaire in Misophonia Patients" at the 10<sup>th</sup> Misophonia Convention on November 2<sup>nd</sup> and 4<sup>th</sup> 2023, Albuquerque, USA.

**Oral Presentation** "Validity and Reliability of English Version of MisoQuest and Its Correlation with Misophonia Impact Questionnaire (MIQ), Hyperacusis Impact Questionnaire (HIQ) and Sound Sensitivity Symptoms Questionnaire (SSSQ)." at the 6<sup>th</sup> International Conference on Hyperacusis and Misophonia on July 1<sup>st</sup> and 2<sup>nd</sup>, 2022, London, UK.

**Poster Presentation** "Hyperacusis and Misophonia: A Systematic Review of Psychometric Measures" at the British Academy of Audiology Conference on November 18<sup>th</sup> and 19<sup>th</sup>, 2021, Manchester, UK.

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# **ABBREVIATIONS**

CFA	Confirmatory Factor Analysis
HIQ	Hyperacusis Impact Questionnaire
MIQ	Misophonia Impact Questionnaire
TIQ	Tinnitus Impact Questionnaire
SAD-T	Screening for Anxiety & Depression in
	Tinnitus and Sound intolerance
AMISOS-R	The Amsterdam Misophonia Scale- Revised
ННІ	Hearing Handicap Inventory
3D-WFI	Three-Dimensional Fatigue Inventory
SSSQ	Sound Sensitivity Symptoms Questionnaire
WLSMV	Weighted Least Squares Mean and Variance
RMSEA	Root Means Square of Approximation
SRMR	Standardized Root Mean Square Residual
TLI	Tucker Lewis Index
CFI	Comparative Fit Index
CMIN	Chi-square/ df
COSMIN	COnsensus-based Standards for the selection
	of health Measurement INstruments
ASD	Autism Spectrum Disorder
HQ	Hyperacusis Questionnaire

# **CHAPTER ONE: GENERAL INTRODUCTION**

#### Overview

In this introductory chapter, an overview is presented on the definitions, diagnostic challenges, prevalence, and underlying mechanisms of hyperacusis and misophonia. Hyperacusis is characterised by the perception of certain everyday sounds, such as household or environmental noises, as excessively loud or uncomfortable, leading to significant distress and disruption to daily activities, including social and occupational functioning. Different forms of hyperacusis have been identified: loudness hyperacusis, which involves perceiving specific sounds (e.g., voices, traffic, or household appliances) as intolerably loud, sometimes accompanied by physical symptoms such as ear fullness or spasms; pain hyperacusis (noxacusis), where certain sounds trigger sensations of pain (e.g., stabbing, throbbing, or burning), even at levels below those known to cause cochlear damage; and fear hyperacusis (phonophobia), which is dominated by a fear of sounds due to concerns about their potential impact, such as worsening tinnitus or hearing sensitivity.

Misophonia, in contrast, is defined by strong emotional, physiological, and behavioural reactions, such as anger, disgust, or anxiety, triggered by specific sounds. These are often sounds produced by humans (e.g., chewing, breathing) or objects (e.g., tapping, clock ticking), which are perceived as particularly aversive. Despite the overlap in some aspects, hyperacusis and misophonia have distinct triggers, responses, and underlying mechanisms, necessitating tailored approaches to their assessment.

A systematic review conducted as part of this thesis examined existing hyperacusis and misophonia questionnaires. This review later published (Kula et al., 2023) (see Chapter 2), evaluated the psychometric properties of widely used tools such as the Misophonia Questionnaire (MQ), MisoQuest, and Hyperacusis Questionnaire (HQ) against the COSMIN

standards. The findings revealed significant gaps in these measures, including inconsistencies in psychometric measurements and lack of cross-cultural validations. Building on this review, the thesis evaluates the English version of the MisoQuest, a 14-item questionnaire originally developed in Polish to measure misophonia. This study establishes the psychometric properties of the English MisoQuest, including its reliability, validity, and factor structure, while also exploring its relationships with audiological and psychological variables such as hyperacusis and fatigue. Additionally, the thesis explores the broader psychological and health impacts of these conditions, with a particular focus on the relationships between misophonia, fatigue, stress, and sleep quality. It further evaluates novel measurement tools aimed at assessing symptom management confidence, particularly within the context of Cognitive Behavioral Therapy (CBT), emphasizing their clinical applicability and relevance.

Towards the conclusion of this chapter, the scope of this thesis is outlined, including a systematic review and a series of empirical studies aimed at advancing the understanding and measurement of misophonia and hyperacusis. The subsequent chapters present a structured approach to addressing these gaps through systematic evaluations, psychometric validations, and new questionnaires validation. The rationale for each study is provided, alongside their specific aims, to illustrate how this body of work contributes to field of hyperacusis and misophonia and their relationship and assessment.

#### **Introduction to Misophonia**

# **Definitions and Key Characteristics**

Misophonia is a condition characterised by intense negative emotional and physiological responses to specific auditory and visual stimuli, often referred to as "triggers". As noted by Ferrer-Torres and Giménez-Llort (2022), the condition was initially identified in the late 1990s by audiologist Marsha Johson under the term "Sensitivity to Soft Sound" or "Selective Sound Sensitivity Syndrome". In 2002, Jastreboff and Jastreboff (2002) introduced the term

"misophonia" to describe negative reactions of limbic and autonomic systems triggered by the perception of specific sounds. Between June 2020 and January 2021, a multidisciplinary committee, including audiologists, psychologists, psychiatrists, and neuroscientists, conducted a systematic review of the existing literature using a modified Delphi method. Their aim was to establish a standardised definition of misophonia for the scientific community. The committee evaluated potential definitions based on clinical and scientific evidence, including only those statements that achieved at least 80% agreement among its members.

The final definition, published by Swedo et al. (2022), defines misophonia as follows:

Misophonia is a disorder characterised by a heightened sensitivity and reduced tolerance to specific sounds or related stimuli, known as "triggers". These triggers are experienced as unpleasant or distressing and elicit strong emotional, physiological, and behavioural responses that are atypical in most people. Misophonic reactions are not typically caused by the loudness of sounds but by their particular pattern or meaning to the individual. Triggers are often repetitive and are frequently, though not exclusively, associated with sounds produced by others, especially those made by the human body. Once a trigger is noticed, individuals with misophonia may find it difficult to distract from it, resulting in significant distress or impairment in areas such as social, occupational, or academic functioning. The severity of symptoms can range from mild to severe, and some individuals with misophonia are aware that their reactions are disproportionate to the situation. Symptoms commonly emerge in childhood or early adolescence.

Key characteristics of misophonia include a range of emotional, physiological, and behavioural responses to specific auditory (e.g., gum chewing, lip smacking or pen clicking) or visual cues (e.g., leg swinging) (Swedo et al., 2022). Emotional responses often involve intense anger, disgust, frustration, anxiety, or panic when exposed to these triggers (Holohan et al., 2023, Savard et al., 2022). Behavioural responses may include avoidance of triggering

situations, escape from the source of the sound, or even aggressive reactions, often driven by the intensity of the emotional distress (Lewin et al., 2024). Physiological symptoms associated with misophonia include muscle tension, increased body temperature, elevated blood pressure, and a heightened heart rate (Rouw and Erfanian, 2018).

These symptoms can significantly interfere with daily life, making it harder for individuals to maintain relationships, carry out work or studies, and take part in social activities. Over time, this can lead to withdrawal and social isolation (Hadjipavlou et al., 2008). This is reflected in a longitudinal study by Dibb and Golding (2022), which found that individuals with self-reported misophonia had significantly lower quality of life compared to the general population. By interfering with daily functioning and social interactions, misophonia creates challenges that extend beyond the distress caused by the triggers themselves. Recognising and understanding these complexities is crucial for improving support and outcomes for those affected.

#### Prevalence

The reported prevalence of misophonia varies considerably across populations, with studies reporting a wide range of affected individuals. For instance, a survey of 483 undergraduate students found that 22.8% frequently or consistently experienced irritation or sensitivity to specific sounds, such as eating, tapping, or nasal noises (Wu et al., 2014). Similarly, a study of 336 medical students in UK revealed that 49.1% exhibited clinically significant symptoms, with 37% experiencing mild symptoms, 12% moderate, and 0.3% severe levels of misophonia (Naylor et al., 2021a). In a large, nationally representative sample of the UK population, Vitoratou et al. (2023) estimated the prevalence of misophonia to be 18%.

Recent studies further illustrate the variability in prevalence across populations. Pfeiffer et al. (2024) found that 33.3% of German participants reported sensitivity to at least one specific misophonic sound. Within this sample, 21.3% experienced subthreshold symptoms, 9.9%

reported mild symptoms, 2.1% reported moderate to severe symptoms, and 0.1% reported severe to extreme symptoms. Dixon et al. (2024) reported that 78.5% of adults were sensitive to misophonic sounds, with 4.6% exhibiting clinical levels of misophonia.

Accurately determining the prevalence of misophonia is challenging due to the lack of standardised diagnostic criteria and variability in measurement tools, such as self-reports and structured interviews. Additionally, misophonia often co-occurs with other auditory and mental health disorders, leading to potential underdiagnosis or misclassification with other hearing-related conditions (Jastreboff and Jastreboff, 2015). These discrepancies highlight the need for robust, standardised methodologies to improve the understanding and quantification of this condition's prevalence. These issues, along with a detailed discussion of diagnostic criteria, will be address in the subsequent sections.

# **Underlying Mechanisms**

Initial theories about misophonia proposed hyperconnectivity between auditory and limbic systems, supported by evidence of structural differences in regions such as the left auditory cortex, bilateral hippocampus, and right anterior insula (Jastreboff and Jastreboff, 2002, Kliuchko et al., 2018). Early research suggested that emotional processing, rather than auditory sensitivity alone, play a critical role. For instance, San Giorgi (2015) observed heightened amygdala activity in response to misophonic triggers, indicating a stronger emotional response rather than over-responsivity to sound.

Functional neuroimaging studies have consistently linked the anterior insular cortex (AIC) and salience network in misophonia. Kumar et al. (2017) reported increased AIC activity in individuals with misophonia when exposed to trigger sounds, alongside heightened heart rate and galvanic skin response, indicating elevated emotional and physiological arousal. Schröder et al. (2019) similarly observed heightened activation in the right insula, anterior cingulate cortex (ACC), and right superior temporal cortex (STC) during audiovisual exposure to trigger

sounds. These findings suggest that trigger sounds engage both auditory processing regions and the salience network, contributing to physiological arousal and emotional reactivity.

Further research indicates that misophonic reactions are not solely mediated by an auditorylimbic response but involve higher-order mechanisms. For instance, Cerliani and Rouw (2020) identified the involvement of the orbitofrontal cortex (OFC) and its connections with the midcingulate cortex (MCC) and premotor areas, suggesting difficulties with emotional regulation. These findings provide an explanation for the compulsive nature of misophonic reactions and highlight the role of an indirect auditory-limbic pathway in interpreting the emotional significance of sounds rather than their physical characteristics. Eijsker et al. (2021) provided further evidence of structural abnormalities in misophonia, reporting increased white matter in the left inferior fronto-occipital fasciculus (IFOF), anterior thalamic radiation (ATR), and the corpus callosum. These findings suggest alterations in connectivity within networks associated with social-emotional processing, particularly the recognition of facial affect, and attention to affective information. Hansen et al. (2022) demonstrated the increased connectivity between auditory cortex and insula but with extending across multiple sensorimotor regions. They identified a potential neural basis for non-orofacial triggers in misophonia, showing significant connectivity differences between finger-related motor and somatosensory regions and the insula in individuals with higher misophonia scores. This challenges the idea that misophonia is limited to oral or nasal triggers. This might be important to understand the multitude presentation of misophonia. The aetiology of misophonia remains uncertain and complex. Understanding the relationship of sensory, emotional, and motor networks in misophonia could significantly enhance future assessment and diagnostic tools.

#### **Comorbid Conditions**

Misophonia frequently coexists with a variety of psychiatric, neurological, and auditory conditions, complicating its clinical presentation and management. Research indicates a strong

association between misophonia and psychiatric conditions such as obsessive-compulsive disorder (OCD), anxiety and depression as well as neurological disorders like Tourette's syndrome and auditory disorders (Ferrer-Torres and Giménez-Llort, 2022). Comorbidity rates appear vary depending on the population studied, i.e., whether participants are from the general population, psychiatric settings or a misophonia-focused sample. For instance, Cusack et al. (2018) found a strong association between misophonia and OCD, particularly with obsessive traits, in both community and student samples. Anxiety sensitivity was also shown to mediate the relationship between OCD and misophonia severity, supporting the role of shared cognitive mechanisms in these conditions.

In a misophonia-specific clinical population, Jager et al. (2020) reported that 26% of individuals exhibited traits of obsessive-compulsive personality disorder (OCD), while 10% were diagnosed with mood disorders, 5% with ADHD, and 3% with autism spectrum conditions. These findings emphasise the diverse range of comorbidities associated with misophonia. Furthermore, from a community sample of 207 individuals, Rosenthal et al. (2022) demonstrated significant correlations between misophonia severity and anxiety disorders, OCD, borderline personality disorder, and depression. Notably, these psychiatric conditions were found to be strong predictors of misophonia symptoms, even after controlling for demographic variables. Further comparisons with clinical groups emphasise the psychiatric, rather than audiologic basis of misophonia, for example, Siepsiak et al. (2022) showed that 71% of individuals with misophonia had at least one psychiatric diagnosis, most commonly major depressive disorder, panic disorder, and suicidality.

The overlap between misophonia and other auditory conditions such as hyperacusis and tinnitus further complicates its clinical presentation (Meltzer and Herzfeld, 2014). Hyperacusis involves discomfort or pain from everyday sounds, while tinnitus is the perception of sound without an external source (Cederroth et al., 2020). Studies suggest that individuals with

misophonia often experience co-occurring auditory symptoms. Jager et al. (2020) reported that 2% of individuals with misophonia also had tinnitus, and 1% had hyperacusis. Conversely, among patients seeking help for tinnitus and/or hyperacusis, 23% were classified as having misophonia symptoms (Aazh et al., 2022a). These findings suggest that misophonia is linked to both psychiatric and auditory aspects, highlighting the need for integrated clinical approaches for its diagnosis and treatment.

# Diagnosis and Measuring Approaches

Misophonia is a complex condition with unclear diagnostic boundaries and significant comorbidities with other disorders, as highlighted by Taylor (2017). Although Swedo et al. (2022) provided a consensus definition, debates surrounding its classification and overlap with other conditions persist. Some researchers propose that misophonia should be classified as a new mental health disorder with specific diagnostic criteria (Schröder et al., 2013), while others suggest that it may be a symptom of broader psychiatric conditions (Ferreira et al., 2013) or linked to certain personality traits or disorders (Cassiello-Robbins et al., 2021). Supporting this perspective, Rosenthal et al. (2022) reported a high prevalence of psychiatric comorbidities, particularly anxiety disorders, among individuals with misophonia. From an audiological perspective, however, the current definition by Swedo et al. (2022) does not clearly differentiate misophonia from related conditions such as hyperacusis or noise sensitivity, partly due to the limited understanding of its underlying mechanisms (Aazh, 2023). This ambiguity has led to ongoing debate about whether misophonia should be categorised primarily as an audiological, psychiatric, psychological, neurological, or a distinct condition (Danesh and Aazh, 2020).

Given the limited literature, and the absence of a formal diagnostic framework for misophonia, this has resulted in a lack of clarity and consensus regarding its assessment and typical clinical presentations. Despite this, several diagnostic approaches have been developed, with individual clinics or researcher often adopting their own diagnostic criteria. Examples include the Duke Misophonia Interview (Guetta et al., 2022), the Oxford King's Structured Clinical Interview for Misophonia (Vitoratou et al., 2023), the Amsterdam UMC Revised Diagnostic Criteria (Jager et al., 2020), and the diagnostic criteria proposed by Aazh et al. (2023).

In terms of assessment through questionnaires, several tools have been developed to assess misophonia, each capturing specific aspects of the condition. However, variations in the definition of misophonia have resulted in tools that do not measure the same constructs or dimensions (Swedo et al., 2022). Among these, The Amsterdam Misophonia Scale [A-MISO-S; Schröder et al. (2013)] and the Misophonia Questionnaire [MQ; Wu et al. (2014)] are the most commonly used tools for assessing misophonia. The A-MISO-S was developed based on proposed diagnostic criteria, but its psychometric properties have not been reported in either the original or revised versions [AMISOS-R; (Jager et al., 2020)]. However, a study by Naylor et al. (2021) in the UK, involving 336 undergraduate medical students at the University of Nottingham, reported a unifactorial structure and good internal consistency for the AMISO-S scale. The Misophonia Questionnaire is a self-report measure designed to assess reactions to sounds and sound sensitivity severity. However, it has been criticised for being too broad, limiting its specificity to misophonia. Additionally, validation studies in affected populations remain incomplete, raising concerns about its suitability for accurately measuring misophonia (Siepsiak et al., 2020a).

Several psychometrically validated recent misophonia questionnaires have been developed to provide additional understanding of the assessment of misophonia. Multidimensional questionnaires, such as the Misophonia Response Scale [MRS; Dibb et al. (2021)], the Duke Misophonia Questionnaire [DMQ; (Rosenthal et al., 2021a)], the Selective Sound Sensitivity Syndrome Scale [S-Five; Vitoratou et al. (2021)], the Duke-Vanderbilt Misophonia Screening

Questionnaire [DVMSQ; Williams et al. (2022)], the Berlin Misophonia Questionnaire Revised [BMQ; Remmert et al. (2022)], and the Sussex Misophonia Scale [SMS; (Rinaldi et al., 2022, Simner et al., 2024)], capture various emotional, cognitive, and behavioral aspects of misophonia, offering broader understanding of the condition. In contrast, unidimensional tools like the MisoQuest (Siepsiak et al., 2020a) and the Misophonia Impact Questionnaire [MIQ; (Aazh et al., 2023)] have unique focuses. MIQ focuses on the impact of misophonia on daily life, while the MisoQuest assesses symptom severity, with a particular focus on anger reactions, and is based on earlier diagnostic criteria by Schröder et al. (2013). Although the MisoQuest has limitations in capturing all misophonia symptoms, its concise format and comprehensive psychometric validation make it a valuable tool for screening purposes in both research and clinical settings.

While several diagnostic approaches and psychometrically validated questionnaires have been developed, they often focus on specific aspects of the condition due to complexity of this condition. This diversity in tools highlights the ongoing challenges in capturing the full complexity of misophonia and the need for standardised diagnostic criteria and comprehensive assessment methods.

# **Introduction to Hyperacusis**

# **Definitions and Key Characteristics**

Hyperacusis is characterised by an increased sensitivity to everyday sounds, such as household or environmental noises, perceived as excessively loud or painful, leading to significant distress and interference with daily life, including social, occupational, and recreational activities (Aazh et al., 2016, Aazh et al., 2024b). A recent definition of hyperacusis, based on a Delphi survey and clinical consensus, includes reduced tolerance for sounds that are generally perceived as normal by most people or were previously perceived as normal by the individual prior to the onset of hyperacusis (Adams et al., 2021).

There are four main subtypes of hyperacusis based on its features, as suggested by Tyler et al. (2014): loudness, annoyance, fear, and pain hyperacusis. Loudness hyperacusis occurs when ordinary sounds at moderate levels, such as a restaurant noise, dog barking, sirens are perceived as overly loud, causing discomfort in routine situations. Annoyance hyperacusis involves a strong negative emotional reaction to sounds, even if they are not perceived as loud, some argue that this subtype may overlap with or describe misophonia instead (Enzler et al., 2021). Fear hyperacusis is characterised by anxiety or fear in response to certain sounds, potentially overlapping with phonophobia, highlighting the need to distinguish hyperacusis from both misophonia and phonophobia by focusing on general discomfort versus reactions to specific sounds. Pain hyperacusis involves physical pain, such as stabbing sensations, in response to sounds below the typical pain threshold and may occur with sudden and loud noises or even loud speech. Although this classification may help in diagnosis and treatment, the subtypes are not mutually exclusive, and individuals may experience features of more than one category. The latest definition consensus on hyperacusis by Adams et al. (2021) highlighted that annoyance and fear were common features of the condition, but pain was reported less frequently.

Demographic studies have further showed the complicated nature of hyperacusis. Associations with higher age, female gender, and higher education levels with hyperacusis are reported within the general population (Paulin et al., 2016). For instance, Jacquemin et al. (2022) found that women are more likely to report hyperacusis, aligning with broader tendencies in sound intolerance prevalence among females. However, a systematic review by Musumano et al. (2023) found no statistically significant gender differences in hyperacusis severity or prevalence. Based on data from 604 patients, the review concluded that males and females exhibit similar levels of hyperacusis, despite some studies reporting slightly higher prevalence among men. These findings highlight the importance of considering biological,

psychological, and methodological factors to gain a clearer understanding of the perception, prevalence, and impact of hyperacusis.

# Aetiology and Prevalence

Hyperacusis is most commonly caused by exposure to high levels of sound pressure but can also result from a variety of other factors, with the underlying cause often remaining unknown. It may develop spontaneously or after exposure to a sudden loud noise, such as fireworks (Assis et al., 2024). According to Singh and Seidman (2019), hyperacusis can arise from infectious diseases (e.g., Lyme disease, neurosyphilis), vascular abnormalities (e.g., aneurysms, Chiari malformation), central nervous system disorders (e.g., PTSD, autism), and endocrine issues (e.g., Addison's disease, hyperthyroidism). Disruption of the stapedial reflex, often following traumatic brain injury (TBI), as well as medication withdrawal (e.g., benzodiazepines) or nutrient deficiencies (e.g., magnesium or pyridoxine), may also contribute. Additionally, hyperacusis is frequently associated with conditions such as tinnitus, head injury, Ménière's disease, Bell's palsy, migraines, Williams syndrome, and autism spectrum disorder (Aazh et al., 2018).

The prevalence of hyperacusis varies widely across different populations and studies, ranging from 0.2% to 17.2% in the general population and from 4.7% to 95% among individuals with conditions such as Williams Syndrome, tinnitus, and autism (Ren et al., 2021). In children and adolescents, Rosing et al. (2016) reported prevalence rates between 3.2% and 17.1%. This substantial variation in prevalence highlights the need for standardised diagnostic criteria and consistent assessment methods to ensure reliable estimates and comparisons across studies.

# **Underlying Mechanisms**

Hyperacusis arises from changes in the brain's auditory processing, resulting in an increased sensitivity to everyday sounds. One widely accepted explanation is the Central Gain Model,

which suggests that reduced auditory input—such as from hearing loss—causes the brain to amplify sound signals as a compensatory mechanism. However, this overcompensation can lead to normal sounds being perceived as uncomfortably loud (Auerbach et al., 2019).

Functional imaging studies support this theory, revealing heightened activity in the auditory cortex, thalamus, and inferior colliculus among individuals with hyperacusis. These findings point to hyperactivity within the central auditory system (Zeng, 2020). Koops and van Dijk (2021) further demonstrated that hyperacusis is associated with increased sound-evoked activity in both subcortical and cortical auditory regions. Interestingly, this heightened activity was not restricted to frequencies affected by hearing loss but extended to unaffected frequencies. On the subcortical level, increased activity was observed in the medial geniculate body (MGB), inferior colliculus (IC), and superior olivary complex (SOC), while cortical activation was heightened in the primary, secondary, and association auditory cortices. These findings emphasise that overactivity in the auditory pathway is a key feature of hyperacusis.

Hyperacusis may also involve non-auditory regions of the brain such as orbitofrontal cortex (OFC), anterior cingulate cortex (ACC), and supplementary motor area (SMA), which are associated with attention, emotional responses, and motor reactions to sound (Gu et al., 2010). Interestingly, hyperacusis shares similarities with neuropathic pain conditions, such as allodynia, where normal stimuli elicit exaggerated responses. This suggests that hyperacusis affects, not only auditory, but also broader sensory and emotional networks (de Klaver et al., 2007). Wertz et al. (2024) recently also showed hyperacusis is associated with distinct brain activity, including increased activation in the left temporoparietal and dorsolateral prefrontal cortices. This heightened activation may reflect increased attention to sound intensity across all frequencies, highlighting the unique neural mechanisms of hyperacusis and its distinction from tinnitus alone.

Furthermore, structural brain differences have been consistently associated with hyperacusis. For instance, gray matter alterations have been observed in individuals with hyperacusis, even when these changes are influenced by hearing loss (Makani et al., 2022). However, hyperacusis can also occur in individuals with clinically normal hearing thresholds. In such cases, hidden hearing loss—potentially arising from various underlying aetiology—is believed to play a contributory role (Zeng, 2020). Notably, a recent study by Makani et al. (2024) showed that hyperacusis is correlated with smaller Supplementary Motor Area (SMA) gray matter volumes, regardless of hearing loss. Therefore, hyperacusis appears to result from a combination of overactivity in the auditory pathways, structural changes in brain regions involved in motor and emotional responses, and heightened connectivity between sensory and non-sensory areas. These findings underline the complexity of hyperacusis and the importance of further research to improve its diagnosis and treatment.

#### **Comorbid Conditions**

Hyperacusis often coexists with various somatic conditions. For example, children with autism spectrum disorder (ASD) commonly exhibit a hyperacusis prevalence of 18%, whereas no cases were identified among age-matched non-autistic peers (Rosenhall et al., 1999). Similarly, a meta-analysis by Williams et al. (2021) found that the current prevalence of hyperacusis in individuals with ASD was 41.42% based on questionnaires and 27.30% using objective measures, with a lifetime prevalence of 60.58%. In Williams syndrome, a multisystem disorder characterised by mental retardation and heightened anxiety, hyperacusis is highly prevalent, with rates reaching up to 95% (Klein et al., 1990).

The co-occurrence of tinnitus and hyperacusis has been widely documented. Among individuals attending tinnitus clinics with tinnitus as their primary complaint, approximately 40% also experience hyperacusis (Jastreboff and Jastreboff, 2000). Conversely, in those presenting with hyperacusis as their primary complaint, the prevalence of tinnitus has been

reported to be as high as 86% (Anari et al., 1999). Additionally, a Swedish study by Cederroth et al. (2020) involving 3645 participants found that hyperacusis was significantly associated with tinnitus, with odds ratios increasing with tinnitus severity. The occurrence of hyperacusis in individuals with severe tinnitus was as high as 80%, underscoring the strong relationship between the two conditions. Additionally, hyperacusis can co-occur with other sound tolerance conditions such as phonophobia and noise sensitivity, which often share overlapping characteristics (Henry et al., 2022).

Hyperacusis is also strongly associated with some psychiatric conditions. Jüris et al. (2013) reported that 56% of individuals with hyperacusis had at least one psychiatric disorder, with anxiety disorders being particularly prevalent (47%). Additionally, personality traits associated with neuroticism were significantly over-represented among this population. Sacchetto et al. (2022) reported that individuals with hyperacusis exhibit significant psychological distress, characterised by elevated levels of anxiety, depression, and somatic attention, which were correlated with their hyperacusis severity.

# Diagnostic Criteria and Measuring Approaches

The diagnosis of hyperacusis does not rely on a single definitive test but rather involves a combination of self-report questionnaires and clinical interviews to collect detailed information about sound sensitivities, medical history, and other relevant factors. Although diagnosing hyperacusis can be challenging, it typically includes assessing Uncomfortable Loudness Levels (ULLs) through pure-tone audiometry and utilising questionnaires to evaluate the severity of the condition (Coey and De Jesus, 2024). To improve diagnostic accuracy, researchers have proposed specific criteria, such as cut-off values of ULLmin  $\leq$  77 dB HL and a Hyperacusis Questionnaire (HQ) score  $\geq$  22 (Aazh and Moore, 2017). Furthermore, a novel approach using psychoacoustic ratings of natural sounds has interestingly demonstrated comparable accuracy to existing diagnostic methods (Enzler et al., 2021). Despite these advancements, the

prevalence of hyperacusis diagnoses in clinical settings remains significantly lower than reported in the literature, likely due to the lack of standardised diagnostic protocols (Theodoroff et al., 2024).

There are several self-report hyperacusis questionnaires designed to assess hyperacusis for different purposes. Among these, the Hyperacusis Questionnaire (HQ), developed by Khalfa et al. (2002), is the most commonly used. However, an independent validation study by Fackrell et al. (2015) found that the HQ does not provide a valid overall measure of hypersensitivity, particularly when applied to individuals with tinnitus.

The Multiple-Activity Scale for Hyperacusis (MASH) evaluates discomfort across 14 activities or environments (e.g., concerts, shopping centres) (Dauman and Bouscau-Faure, 2005), however its psychometric measurements have not been fully established (Kula et al., 2023). Another instrument, the Hyperacusis Handicap Questionnaire (HHQ), was designed to assess hyperacusis in individuals in tinnitus. However, its development process and evidence for validity are limited, having been on a clinical sample of 77 patients (Prabhu and Nagaraj, 2020). Another hyperacusis questionnaire, the Inventory of Hyperacusis Symptoms (IHS), was developed by Greenberg and Carlos (2018) to measure hyperacusis for both research and clinical use. The initial validation was conducted using an online general population reporting sound sensitivity, rather than a clinical sample. Subsequent clinical validation by Aazh et al. (2021a) indicated that the IHS demonstrates good internal consistency and reasonably high convergent validity, as shown by correlations with HQ scores and ULLs (uncomfortable loudness levels). However, IHS scores may also reflect the influence of comorbid conditions such as tinnitus, anxiety, and depression, which could impact its validity.

Two recent questionnaires, the Hyperacusis Impact Questionnaire [HIQ; (Aazh et al., 2024a)] and the Sound Sensitivity Symptoms Questionnaire [SSSQ; (Aazh et al., 2024a)] were specifically designed for clinical use. The HIQ assesses the impact of hyperacusis on daily life,

while the SSSQ evaluates the type and severity of sound intolerance symptoms, aligning with Tyler's categories of hyperacusis, but both tools are designed to use in clinical and research context. The latest valid and reliable tool, Hyperacusis Assessment Questionnaire, was developed by Raj-Koziak et al. (2023) to measure the hyperacusis sensitivity regarding loudness, fear and pain. Together, these tools represent a range of approaches for measuring hyperacusis, with ongoing efforts to refine their diagnostic and psychometric robustness.

# **Interim Summary**

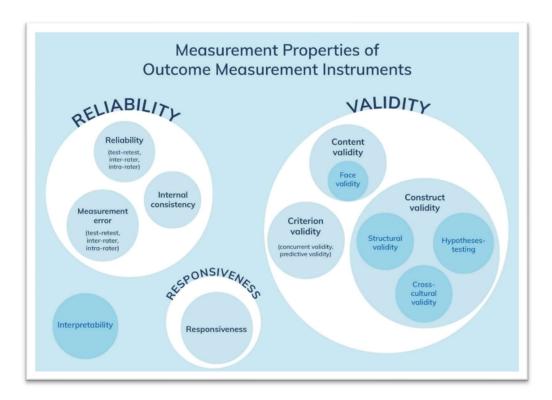
Misophonia and hyperacusis are complex and often overlapping conditions, characterised by heightened sensitivities to specific sounds, but they might differ in their triggers, responses, and underlying mechanisms (Henry et al., 2022). Misophonia is defined as a condition involving heightened emotional, physiological, and behavioural responses to specific auditory or visual triggers, often human-generated, such as chewing or pen-clicking (Swedo et al., 2022). These triggers cause significant distress and functional impairment, with symptoms typically beginning in childhood or adolescence (Ferrer-Torres and Giménez-Llort, 2022). The condition is associated with difficulties in emotional regulation and neural hyperactivity, particularly involving the anterior insular cortex (AIC) and salience networks (Kumar et al., 2021). Misophonia frequently coexists with psychiatric conditions, including obsessivecompulsive disorder (OCD), anxiety, and depression, and often overlaps with conditions such as hyperacusis (Jastreboff and Jastreboff, 2015). However, its diagnostic criteria remain argued due to uncertainty in its aetiology, and variations in measurement tools complicate accurate assessment. Recent psychometrically validated questionnaires, such as S-Five (Vitoratou et al., 2021), Duke Misophonia Questionnaire (Rosenthal et al., 2021), Berlin Misophonia Questionnaire (Remmert et al., 2022), which aim to capture the multidimensional nature of this condition and improve diagnostic reliability, but there is no gold standard measure due to its complex mechanisms.

Hyperacusis, by comparison, refers to an increased sensitivity to everyday sounds, such as household or environmental noises, perceived as excessively loud or painful, resulting in significant distress and disruption to social, occupational, and recreational activities (Aazh et al., 2016, Aazh et al., 2022b). Its varied presentations can be classified into subtypes, including loudness, annoyance, fear, and pain hyperacusis (Tyler et al., 2014). While the exact causes are not fully understood, hyperacusis is thought to involve overactivity in the central auditory system and structural brain changes in areas linked to emotional and motor responses (Zeng, 2020). Comorbidities, such as tinnitus and psychiatric conditions, are common, complicating its clinical presentation and management (Jastreboff and Jastreboff, 2015, Cederroth et al., 2020). Diagnostic criteria for hyperacusis are not well-established; however, measures such as the Uncomfortable Loudness Levels (ULLs) assessment tool, and tools like the Hyperacusis Questionnaire [HQ; (Khalfa et al., 2002)] are commonly used. Researchers have suggested specific criteria for identifying hyperacusis, including cut-off values of ULLmin ≤ 77 dB HL and a Hyperacusis Questionnaire (HQ) score  $\geq 22$  (Aazh and Moore, 2017). Additionally, newer psychometrically validated instruments, such as the Hyperacusis Impact Questionnaire [HIQ; (Aazh et al., 2024a)] and Sound Sensitivity Symptoms Questionnaire [SSSQ; (Aazh et al., 2024a)], [Hyperacusis Assessment Questionnaire; Raj-Koziak et al. (2023)] have been developed to assess various dimensions of hyperacusis severity and its impact.

Various questionnaires have been developed to assess hyperacusis and misophonia, aiming to capture their severity, impact, and associated symptoms. However, the quality and psychometric robustness of these tools vary, with some lacking rigorous validation. To ensure reliable and valid assessments, it is essential to evaluate these questionnaires using standardised criteria. Among the frameworks designed for this purpose, the COSMIN (Mokkink et al., 2018) is recognised as the gold standard for assessing the methodological quality of health measurement instruments.

# **COSMIN** and Psychometric Properties

The COSMIN —COnsensus-based Standards for the selection of health Measurement INstruments—was developed in the Netherlands in 2006 with the aim of setting standards for the methodological quality of studies evaluating the measurement properties of health status instruments. These standards have been continually updated and refined to reflect advancements in the field and to ensure their ongoing relevance to health measurement research. COSMIN identifies three key domains for evaluation: reliability, validity, and responsiveness (Mokkink et al., 2018). The validity domain comprises face and content validity, criterion validity (both concurrent and predictive), and construct validity. Construct validity includes studies employing classical hypothesis testing as well as those examining the dimensional structure of an instrument (also referred to as structural validity). Additionally, COSMIN highlights the importance of evaluating studies involving cross-cultural adaptation to different sociolinguistic contexts.



**Figure 1:** Measurement Properties of Outcome Measurement Instruments. Reprinted from the COSMIN website (https://www.cosmin.nl), © COSMIN, (Mokkink et al., 2010).

# **Overall Summary**

This chapter has provided an overview for understanding the conditions of misophonia and hyperacusis, emphasizing their distinct characteristics, diagnostic challenges, and the broader implications for assessment and management. Despite their overlapping symptoms, these conditions differ in their mechanisms, triggers, and impacts. This review highlighted the substantial gaps in the current understanding and measurement of these conditions. Existing assessment tools such as the Misophonia Questionnaire (MQ) and Hyperacusis Questionnaire (HQ), were evaluated for their psychometric properties, revealing inconsistencies in reliability and validity.

This chapter also explored the underlying mechanisms and comorbidities of misophonia and hyperacusis, linking them to psychological, neurological, and auditory processes. The prevalence and impact of these conditions are discussed, with a focus on their implications for daily functioning and quality of life. By setting the stage for the subsequent chapters, this chapter establishes the methodological context of this thesis, highlighting its aim to address critical gaps in the assessment and understanding of misophonia and hyperacusis.

# **Scope of Thesis**

The scope of this thesis is to address gaps in the understanding and assessment of misophonia and hyperacusis. By focusing on the validation of psychometrically robust tools and exploring the broader psychological and health dimensions of these conditions, this thesis aims to contribute to both research and clinical practice.

#### Aims of the Thesis

 To systematically review the psychometric properties of existing misophonia and hyperacusis questionnaires, identifying gaps in their methodological quality using COSMIN standards.

- To validate the English version of MisoQuest, establishing its reliability, validity, and structural properties, and exploring its relationships with related audiological and psychological factors.
- 3. To investigate the broader psychological and health impacts of misophonia, focusing on its associations with fatigue, stress, and sleep quality, and assessing the mediating roles of stress and sleep in these relationships.
- 4. To validate the 4C Misophonia and Hyperacusis Management Questionnaires, ensuring their clinical applicability and utility in evaluating patients' confidence in symptom management within CBT frameworks.

#### Structure of the Thesis

To address these aims, the thesis is organised as follows:

- Chapter 2 systematically reviews the psychometric properties of existing misophonia and hyperacusis questionnaires, including Hyperacusis Questionnaire (HQ), Inventory of Hyperacusis Symptoms (IHS), questionnaire on hypersensitivity to sound (GUF), Hyperacusis Handicap Questionnaire (HHQ), Short Hyperacusis Questionnaire, Amsterdam Misophonia Scale (A-MISO-S), MisoQuest, and Misophonia Questionnaire (MQ). The review evaluates these instruments against COSMIN standards, highlighting gaps in their psychometric properties.
- Chapter 3 focuses on the psychometric validation of the English version of MisoQuest, a 14-item tool initially validated in Polish. The chapter evaluates its reliability, validity, and factor structure, as well as its relationships with audiological (hyperacusis) and psychological (fatigue, stress and sleep) variables.
- Chapter 4 presents a cross-sectional study investigating the relationship between misophonia severity, fatigue, stress, and sleep quality. This chapter examines whether

stress and sleep mediate the relationship between misophonia and fatigue, highlighting the broader psychological and health impacts of misophonia.

• Chapter 5 reports on the validation of the 4C Misophonia and Hyperacusis Management Questionnaires. These tools assess patients' confidence in managing symptoms without reliance on avoidance behaviours, with their psychometric properties evaluated for reliability, structural validity, and clinical relevance, particularly within CBT frameworks.

Chapter 6, the concluding chapter, summarises the key findings of this thesis and provides a critical discussion of its methodological and clinical implications. These considerations are framed within the context of understanding misophonia and hyperacusis, addressing their measurement and management challenges. The chapter outlines potential directions for future research, focusing on psychometric tools and exploring the broader psychological and health impacts of these conditions. Collectively, this chapter aims to contribute to an understanding of misophonia and hyperacusis, advancing both research and clinical frameworks in the field

#### **CHAPTER TWO**

### Study 1. Hyperacusis and Misophonia: A Systematic Review of

#### **Psychometric Measures**

#### 2.1 Overview

This chapter systematically reviews the psychometric instruments used to measure hyperacusis and misophonia. While several instruments have been developed, their methodological quality has not been systematically evaluated in previous research. The primary aim of this chapter was to identify and critically assess studies examining the psychometric properties of these instruments, with particular reference to their adherence to the COSMIN standards.

A systematic search of five electronic databases identified 14 studies meeting the inclusion criteria, encompassing eight instruments—five for hyperacusis and three for misophonia. The methodological quality of the studies varied, ranging from very good to inadequate, and none fully met the COSMIN standards.

This chapter examines the findings in relation to their implications for research and clinical practice, highlighting significant gaps in the current literature. It emphasises the need for further research to enhance the psychometric rigour of these instruments and provides a foundation for the chapters that follow.

This systematic review has since been published in the Journal of the American Academy of Audiology (Kula et al., 2023).

#### 2.2 Research Paper

# Hyperacusis and Misophonia: A Systematic Review of Psychometric Measures

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Abstract

**Background:** Hyperacusis can be defined as an intolerance of certain everyday sounds, which

are perceived as too loud or uncomfortable and which cause significant distress and impairment

in the individual's day-to-day activities. Misophonia is defined as a high magnitude of

emotional and behavioural reaction to certain sounds produced by human beings, such as eating

sounds and breathing sounds. Several psychometric instruments have been developed to assess

symptoms and the impact of hyperacusis and misophonia; however, to the author's knowledge,

no study has evaluated and compared the methodological quality of the studies on psychometric

properties of the existing instruments.

**Purpose:** This article systematically reviews the research studies assessing the psychometric

properties of the instruments used for hyperacusis and misophonia and assess the quality and

appropriateness of the methodologies used.

Research Design: Systematic review.

Data Collection and Analysis: A systematic literature search was performed using five

electronic literature databases (PubMed, Scopus, PsycINFO, Google Scholar and Web of

Science). Studies were included if they were written in English and reported information about

the psychometric properties of instruments measuring hyperacusis or misophonia symptoms or

their impact. The quality of the studies and that of the psychometric instruments were evaluated

using the consensus-based standards for the selection of health measurement instruments

(COSMIN) tool.

**Results:** The title and abstracts of 916 articles were screened and 39 articles were selected for

full-text evaluation, with 14 articles meeting the inclusion criteria. From these 14 articles, eight

different instruments (5 for hyperacusis and 3 for misophonia) were identified and reviewed

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comprising: (1) Hyperacusis Questionnaire (HQ), (2) Inventory of Hyperacusis Symptoms

(IHS), (3) questionnaire on hypersensitivity to sound (GUF), (4) Hyperacusis Handicap

Questionnaire (HHQ), (5) Short Hyperacusis Questionnaire, (6) Amsterdam Misophonia Scale

(A-MISO-S), (7) MisoQuest, and (8) the Misophonia Questionnaire (MQ).

Conclusion: None of the papers reviewed reported all the information required to meet the

COSMIN standards. The studies' methodological quality varied between 'very good' and

'inadequate' depending on their grade on the COSMIN tool. There is a need for further research

on the psychometric properties of the instruments included in this review.

Key Words: Hyperacusis, misophonia, psychometric instruments and properties

Hyperacusis can be defined as an intolerance of certain everyday sounds, which are perceived as too loud or uncomfortable and which cause significant distress and impairment in the individual's day-to-day activities (Aazh et al., 2022a). Other definitions of hyperacusis are largely in agreement with this definition, with some differences in details (Adams et al., 2020, Aazh et al., 2016). Tyler et al. (2014) described four categories of hyperacusis comprising (1) loudness hyperacusis, (2) fear hyperacusis, (3) pain hyperacusis, and (4) annoyance hyperacusis. There is some overlap between annoyance hyperacusis and misophonia. A recent consensus study described that misophonia is characterized by the experience of unpleasant or distressing emotions when exposed to certain sounds generated by another individual, especially (but not exclusively) those produced by the human body (Swedo et al., 2022). In misophonia, the specific pattern or meaning of the sound to the individual as opposed to its loudness seem to be the key contributing factor to the individual's reaction. Individuals with misophonia often experience suffering, distress or cannot tolerate sounds associated with oral functions (e.g., chewing, eating), nasal sounds (e.g., breathing and sniffing), as well as non-oral/nasal sounds (e.g., pen clicking, keyboard typing, clock ticking) (Swedo et al., 2022).

Prevalence estimates range from 2% to 15.2% for hyperacusis (Andersson et al., 2002, Smit et al., 2021) and 6% to 49.1% for misophonia (Zhou et al., 2017, Naylor et al., 2021, Wu et al., 2014). It is likely that the discrepancy in prevalence reports is related the differences in study populations and the way that hyperacusis and misophonia were assessed and diagnosed.

Several psychometric instruments have been developed and applied in research and clinical practice to evaluate hyperacusis and/or misophonia. The methodologies used to design and evaluate the psychometric properties of these instruments (e.g., validity, reliability, sensitivity to change) are very diverse. The two important psychometric properties are reliability and validity which are essential for choosing suitable instruments for research or clinical purposes (Sürücü and Maslakci, 2020). Reliability comprises measures of internal consistency (degree

of interrelatedness among the items), test-retest reliability (consistency of scores obtained at different times), inter-rater reliability (consistency of scores obtained by different raters), and measurement error the systematic and random error of a patient's score that is not attributed to true changes in the construct to be measured (Gillespie, 2015). Validity is defined as the extent to which an instrument measures what it claims to measure (Altheide and Johnson, 1994) and comprises (1) content validity (the degree to which the questions on the instrument represent the construct that it seeks to measure (Creswell, 2002), (2) construct validity (the extent to which the instrument validly measures the construct it purports to measure), (3) structural validity (the degree to which the scores of the instrument is an adequate reflection of the dimensionality of the construct to be measured), (4) hypotheses testing (the degree to which scores on the instrument are consistent with hypothesized relationship with other instruments), (5) cross-cultural validity (the degree to which items on a translated or adapted measure correspond to the performance of the original items), and (6) criterion validity (the degree to which scores correspond with a gold standard measure).

Studies assessing the psychometric properties of hyperacusis and misophonia instruments have used inconsistent methods. For example, participants in some studies were recruited from hospital patient referrals (Aazh et al., 2021, Bläsing et al., 2010, Fioretti et al., 2015) while others from the general population or university students (Khalfa et al., 2002, Naylor et al., 2021). Some of these instruments are validated in languages other than English and the English versions although published, have not been validated (Siepsiak et al., 2020). In addition, among the published papers there are some discrepancies regarding reporting of the important psychometric properties of the instruments they evaluated or developed. As the result of these discrepancies, it may not be clear to many audiologists whether the psychometric properties of the existing hyperacusis and misophonia questionnaires meet the standards required for them to be used effectively in research and/or clinical practice.

To develop a greater understanding of the reliability and validity of the existing hyperacusis and misophonia instruments, a systematic review of the literature can be extremely informative. Systematic reviews provide a summary of the strengths and weaknesses of the existing questionnaires, appraise the methodological quality of published studies, and discuss the differences between them (Green, 2005); the results of which, can guide clinical practice and research.

Consensus-based standards for the selection of health measurement instruments (COSMIN) were developed to provide a comprehensive methodological tool for assessing the methodological quality of patient-reported outcome measures (Prinsen et al., 2018). COSMIN is an initiative of an international multidisciplinary team of researchers with a background in epidemiology, psychometric, medicine, qualitative research, and health care, who have expertise in the development and evolution of outcome measurement instruments. They developed the COSMIN risk of bias checklist that can be used in systematic reviews to assess the methodological quality of the studies included to the review (Mokkink et al., 2010, Terwee et al., 2012).

The present study aimed to systematically review the psychometric properties of the existing hyperacusis and misophonia questionnaires, summarise their strengths and weaknesses, and appraise the methodological quality of published studies against the criteria set by COSMIN tool (Prinsen et al., 2018, Terwee et al., 2012).

#### Methods

This systematic review was conducted in compliance with the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) guideline (Moher et al., 2010) and it was registered with the PROSPERO database (<a href="https://www.crd.york.ac.uk/prospero">https://www.crd.york.ac.uk/prospero</a>; registration number: CRD42021235539).

#### Inclusion and Exclusion Criteria

The following inclusion criteria for articles were applied: 1) published in English, 2) published in a peer-reviewed journal, 3) detailed the development or evaluated the measurement properties of instruments measuring hyperacusis or misophonia symptoms or their impact.

Articles were excluded if they: 1) were not indexed in a recognised database, 2) did not report at least one psychometric property as defined by the COSMIN checklist (information relating to the psychometric properties are presented in Appendix 1), iii) were a review, personal/expert opinions and manuals, guidelines, or reported animal studies and any unpublished and incomplete studies.

#### Search Strategy

An initial search was conducted on 29th January 2021. A systematic search was performed in the following electronic databases: PubMed, Scopus, Web of Science, PsycINFO and Google Scholar. We entered a specific search term strategy in each search engine (see Table 1), searching in articles topics, titles, abstracts, and keywords. The database search was conducted without setting any limits in terms of the publication date of the studies. If possible, filters were applied to find related articles in the English language only and with humans only. The reference lists of any relevant articles were checked throughout the process to ensure that any related studies were not missed. Searches were last updated on 29th April 2021. Prior to submitting the final revision of this article on 17th June 2022, a quick search was conducted to double check if any new studies have emerged with regard to the questionnaires reviewed in this article which did not show any new relevant studies.

**Table 1.** Search term strategies applied in databases

	Construct	Instrument	<b>Psychometric Properties</b>
Search	Hyperacus* OR Misophon*	Assess* OR	Psychometr* OR Valid*
Terms	OR "Reduc* sound	measur* OR	OR "Reliab* OR
	intolerance" OR "Noise	Questionnaire OR	Sensitiv* "internal
	Sensitivity" OR "Sound	instrument* OR	consistency" OR "Factor
	Intolerance" OR "Sensory	self-report OR	Analysis"
	intolerance" OR "Sound	inventory OR	_
	Sensitivity" OR "Selective	instrument OR	
	Sound Sensitivity Syndrome"	Checklist	
	OR "Soft Sound Sensitivity		
	Syndrome" OR "aversive		
	sounds" OR "trigger sounds"		
	OR "decreased sound		
	tolerance"		

#### Selection Criteria

After the removal of duplicates articles, one reviewer (FK) screened titles and abstracts to identify eligible articles. The reference list of any relevant articles was also reviewed by the first author. Then, two reviewers (FK and HA) screened the full text of the articles independently. The decision regarding the inclusion/exclusion of studies was made as a result of two reviewers' judgment about the selection of the articles and to verify inter-rater reliability of the full text screening, we calculated the Kohen's kappa value which was 0.65 indicating substantial agreement between the two reviewers (Viera and Garrett, 2005). Any disagreements were resolved by the third reviewer (MC).

#### Data Extraction

Psychometric properties including content validity, structural validity, internal consistency, reliability, hypothesis testing for construct validity, cross-cultural validity, measurement error, criterion validity and responsiveness were extracted from studies in line with recommendations specified in the COSMIN guidelines (Mokkink et al., 2010). Other extracted information was country of origin, number of samples, gender, study population, and instrument-related factors

such as construct measured, number of items, range of total score, and response options. All data were extracted by the first author in May 2021.

#### Evaluation of methodological quality

Two reviewers independently applied the COSMIN checklist for all included studies according to the recommended guidelines. Discrepancies of opinions were resolved by consensus between the two reviewers or, if the agreement was not achieved, disagreements were discussed and resolved through consultation with the third reviewer. No one graded any of their own papers.

The methodological quality of studies and their psychometric properties were assessed using the COSMIN checklist (Prinsen et al., 2018) as shown in Appendix 1. Based on this assessment we reported whether the above mentioned nine domains were assessed or adequately reported by various studies on psychometric properties of the hyperacusis and misophonia questionnaires.

#### Evaluation of Psychometric Properties of the Included Instruments

Each measurement property was rated by applying a four-point COSMIN risk of bias scale (4= 'very good', 3= 'adequate', 2= 'doubtful', 1= 'inadequate'). Consistent with COSMIN instructions, the overall quality rating for each measurement property was determined by taking the 'worst score counts' method (i.e., the lowest rating of any of the items in a given category) (Terwee et al., 2012). For the next step, the result of individual studies on measurement properties was also evaluated against COSMIN 2018 updated criteria for good measurement properties (Appendix 1). The assessment resulted in rating for each questionnaire: sufficient (+), insufficient (-), or indeterminate (?). We used this information to create a table that demonstrates whether the key nine psychometric properties were reported for each questionnaire and if they meet the COSMIN criteria.

Inter-rater between the two reviewers was 82.0% (Kappa: 0.73) for the risk of bias ratings, and 84.5% (Kappa= 0.82) for the measurement properties, indicating substantial agreement between the two reviewers (Viera and Garrett, 2005).

#### **Results**

#### Study Selection

A total of 1040 articles were identified through the initial search (Figure 1), and ten additional articles were identified through a review of citations. After removing duplicates, 916 articles were screened based on their title and abstract, and 39 articles were selected for full-text assessment. As a result of this full-text evaluation, 25 of the 39 articles were removed because they focused on the different constructs or did not report any psychometric property defined by the COSMIN checklist. In addition, one of the articles was not included this study because it was published in a predatory journal (Rice et al., 2021). Fourteen articles were included in this review, and from these 14 articles, eight different hyperacusis and misophonia instruments were evaluated (Aazh et al., 2021, Bläsing et al., 2010, Erinc and Derinsu, 2020, Fackrell et al., 2015, Fioretti et al., 2015, Greenberg and Carlos, 2018, Khalfa et al., 2002, Meeus et al., 2010, Naylor et al., 2021, Oishi et al., 2017, Prabhu and Nagaraj, 2020b, Siepsiak et al., 2020, Tortorella et al., 2017, Wu et al., 2014). See the PRISMA flow diagram in Figure 1.

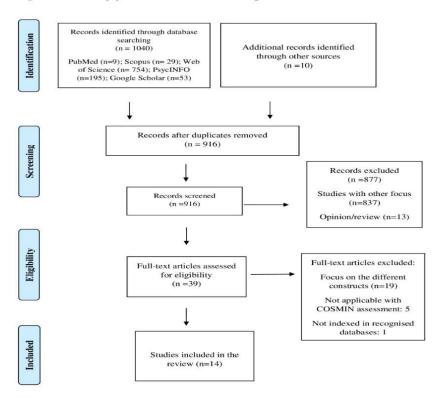


Fig. 1. Flowchart of paper selection based on PRISMA guidance.

#### Study and Participant's Characteristics

Table 2 summarizes the characteristics of the included studies. The eligible studies were published from 2002 to 2020. Approximately 20% of the studies were conducted in the UK (Aazh et al., 2021, Fackrell et al., 2015, Naylor et al., 2021), 13% in the USA (Greenberg and Carlos, 2018, Wu et al., 2014), and Italy (Fioretti et al., 2015, Tortorella et al., 2017). The rest of the studies were conducted in India (Prabhu and Nagaraj, 2020), Belgium (Meeus et al., 2010), Germany (Bläsing et al., 2010), Japan (Oishi et al., 2017), Turkey (Erinc and Derinsu, 2020), Poland (Siepsiak et al., 2020) and France (Khalfa et al., 2002). The most used questionnaire reported was the Hyperacusis Questionnaire developed by Khalfa in 2002

(Khalfa et al., 2002). All questionnaires were developed to assess or diagnose hyperacusis or misophonia.

Table 2 also shows the participants' characteristics of the studies included to this review. Sample sizes for these studies ranged between 46 and 705 individuals from the general population and/or clinical population. Most studies included clinical populations (n=9), and two studies reported student populations, with the remainder utilising individuals from the general population (n=3).

#### Psychometric Instruments for Hyperacusis and Misophonia

**Table 3** provides a summary of the description of the questionnaires including five hyperacusis and three misophonia instruments. All measures utilised the Likert type scales using 3 to 5 points scale. In addition, the structure of the included instruments varies greatly; five measures have three factor-structure (Bläsing et al., 2010, Khalfa et al., 2002, Prabhu and Nagaraj, 2020, Tortorella et al., 2017, Wu et al., 2014), two measures have one factor-structure (Naylor et al., 2021, Siepsiak et al., 2020), and the other measure has a five factor-structure (Greenberg and Carlos, 2018).

Table 2. Summary of study characteristics

#	Author and	Sample	Study Population	Sample Age	Gender	Country	The measure of Hyperacusis or
	Year	(N)		Range (years) or overall rates			Misophonia
1	Aazh et al, 2021	100	Clinical population- patients attended a tinnitus and hyperacusis clinic	21 to 81	48 Female 52 Male	UK	Inventory of Hyperacusis Symptoms
2	Blasing et al, 2010	91	Clinical population- patients suffered from tinnitus	15 to 76	36 Females 55 Males	Germany	GÜF: hypersensitivity to sound
3	Erinc and Derinsu, 2020	529	General population	18 to 73	320 Female 209 Male	Turkey	Hyperacusis Questionnaire
4	Fackrell et al, 2015	264	Clinical population- data collected from tinnitus studies	24 to 85	158 Male 106 Female	UK	Hyperacusis Questionnaire
5	Fioretti et al, 2015	117	Clinical Population- Patients with tinnitus complaints	14 to 88	53 Female 64 Male	Italy	Hyperacusis Questionnaire
6	Greenberg and Carlos, 2018	469	Patients attending an online support group or social media sites	34.8	40% Male 58% Female 2% not disclosed	USA	Inventory of Hyperacusis Symptoms
7	Khalfa et al, 2002	201	General population	17 to 72	132 Female 69 Male	France	Hyperacusis Questionnaire

8	Meeus et al, 2010	46	Clinical Population	21 to 81	14 Female 32 Male	Belgium	Hyperacusis Questionnaire
9	Oishi et al, 2017	215	Clinical population	Group A: 59.2	GroupA:52.7% Female	Japan	Hyperacusis Questionnaire
				Group B: 63.4	GroupB:46.6% Female		
10	Naylor et al, 2020	336	University medical students	18 to24	73%Female	UK	The Amsterdam Misophonia Scale
11	Prabhu and Nagaraj, 2020	77	Clinical Population (Participants with tinnitus complaints)	20 to55	36 Female 41 Male	India	Hyperacusis Handicap Questionnaire
12	Siepsiak et al, 2020	705	Clinical Population (Mixed group for misophonia and other health conditions patients)	18 to 68	86.2% and 80% Female for each phase	Poland	MisoQuest
13	Tortorella et al, 2017	117	Clinical Population (Participants with a primary complaint of tinnitus)	23 to 82	49 Female 68 Male	Italy	The Short Hyperacusis Questionnaire
14	Wu, 2014	483	Undergraduate university students	18 to 54	404 Female 79 Male	USA	Misophonia Questionnaire

Table 3. Description of questionnaires

#	Measure	Construct Measured	Structure	Number of items	Response Options	Total Range
1	Hyperacusis Questionnaire (HQ)	Hyperacusis	Three factors	14	4-Point Likert Scale (0= no, 3= yes, a lot)	0-42
2	Inventory of Hyperacusis Symptoms	Hyperacusis	Five-factor structures	25	4- Point Likert scale	25-100
3	GÜF: (questionnaire on hypersensitivity to sound)	Hyperacusis	Three factors	15	4- Point Likert Scale	0-45
4	Amsterdam Misophonia Scale	Misophonia	One Factor	6	5-Point Likert scale	0-24
5	Hyperacusis Handicap Questionnaire	Hyperacusis	Three factors	21	3-Point Likert Scale	0-84
6	MisoQuest	Misophonia	One Factor	14	5- Point Likert Scale	14-70
7	Misophonia Questionnaire	Misophonia	Three factors	17	4- Point Likert Scale	0-68(for the first two sections)
8	Short Hyperacusis Questionnaire	Hyperacusis	Three factors	6	4- Point Likert Scale	0-24

#### The Methodological Quality of the Included Studies

Table 4 summarises methodological quality ratings for the 14 studies included to the review. All the studies reported more than one psychometric property. In addition, all studies reported internal consistency. Most studies described hypotheses testing for construct validity (11/14) and structural validity (8/14). Only a small number of studies included psychometric data on cross-cultural validity (2 studies), reliability (1 study), and measurement error (1 study). No information was retrieved on responsiveness and criterion validity in any study.

#### Psychometric Properties of the Included Instruments

Table 5 presents ratings for each psychometric instrument. The psychometric properties extracted from the studies were evaluated against the criteria for good psychometric properties on the COSMIN. None of the instruments could be fully evaluated over all nine psychometric properties as the necessary data was not always reported.

Table 4. Methodological quality ratings of each study based on COSMIN.

		<u> </u>			d on COSMIN.	D 11 1 111:	3.5	G • .	TT (1	ъ .
#	Instrument	Study	Structural Validity	Internal consistency	Cross- cultural validity\ measurement invariance	Reliability	Measurement error	Criterion validity	Hypotheses testing for construct validity	Responsiveness
1	HQ	Khalfa et al., 2002	V	V	N	N	N	N	A	N
2	HQ	Erinc and Derinsu, 2020	V	V	D	N	N	N	A	N
3	HQ	Oishi et al., 2017	N	V	N	N	N	N	D	N
4	HQ	Fioretti et al., 2015	N	V	D	N	N	N	N	N
5	HQ	Meeus et al., 2010	I	V	N	N	N	N	D	N
6	HQ	Fackrell et al., 2015	V	V	N	N	N	N	V	N
7	IHS	Greenberg and Carlos, 2018	N	V	N	N	N	N	V	N
8	IHS	Aazh et al., 2021	N	V	N	N	N	N	A	N
9	ННQ	Prabhu and Nagaraj, 2020	N	V	N	N	N	N	D	N

10	SHQ	Tortorella et al., 2017	N	V	N	N	N	N	D	N
11	GUF	Blasing et al., 2010	D	V	N	N	N	N	A	N
12	MQ	Wu et al., 2014	D	V	N	N	N	N	N	N
13	MisoQuest	Siepsiak et al., 2020	A	V	N	D	D	N	A	N
14	A-MISO-S	Naylor et al., 2020	A	V	N	N	N	N	N	N

COSMIN rating: V: Very Good; A: Adequate; D: Doubtful; I: Inadequate; N: Not reported by the study authors

**Table 5.** Ratings for each psychometric property quality per instrument based on COSMIN.

	Structural validity	Internal consistency	Cross- cultural validity	Measurement invariance	Reliability	Measurement error	Criterion validity	Hypothesis testing for construct validity	Responsiveness
HQ									
Khalfa et al., 2002	-	-	NR	NR	?	NR	NR	NR	NR
Erinc and Derinsu, 2020	?	+	+	NR	NR	NR	NR	+	NR
Oishi et al., 2017	NR	+	?	NR	NR	NR	NR	NR	NR
Fioretti et al., 2015	NR	+	?	NR	NR	NR	NR	NR	NR
Meeus et al., 2010	-	?	?	NR	NR	NR	NR	+	NR
Fackrell et al., 2015	+	+	NR	NR	NR	NR	NR	?	NR
HIS									
Greenberg and Carlos, 2018	NR	+	NR	NR	NR	NR	NR	+	NR
Aazh et al., 2021	NR	+	NR	NR	NR	NR	NR	+	NR
MQ									

Wu et al., 2014	?	?	NR	NR	NR	NR	NR	?	NR
MisoQuest									
Siepsiak et al., 2020	+	+	NR	NR	+	?	NR	+	NR
нно									
Prabhu and Nagaraj, 2020	NR	+	NR						
A-MISO-S									
Naylor et al., 2020	-	+	NR						
GUF									
Blasing et al., 2010	?	+	NR	NR	NR	NR	NR	+	NR
Short-HQ									
Tortorella et al., 2017	NR	-	NR						

Note. COSMIN Rating: (+) 'sufficient', (-) 'insufficient', (?) 'indeterminate', NR= not reported by the study author

#### **Discussion**

The purpose of this systematic review was to evaluate the quality of psychometric properties of the current hyperacusis and misophonia instruments (until April 2021) using the COSMIN guidelines. The COSMIN checklist is a well-known tool and has been developed in conjunction with other existing guidelines for systematic reviews, such as the Cochrane Handbook for systematic reviews of intervention (Johnston et al., 2019), the PRISMA statement (Shamseer et al., 2015) and the Grading of Recommendations Assessment, Development and Emulation (GRADE) principles.

To our knowledge, this is the first systematic review to evaluate the measurement properties of instruments designed to measure hyperacusis or misophonia across a range of healthcare contexts and settings. This review identified eight measures (five for hyperacusis and three for misophonia) and 14 studies on the psychometric properties of these instruments. In general, the methodological quality of the included studies in this review varied between "very good" and "inadequate" across all psychometric properties based on the COSMIN tool. None of the identified instruments has reported all nine psychometric properties recommended by COSMIN.

## The Methodological Quality of the Included Studies and Psychometric Properties of the Instruments

According to the COSMIN guideline (2018), content validity is considered an important measurement property of an instrument. However, none of the included articles reported using adequate methods to assess content validity. One explanation is that the constructs of hyperacusis and misophonia are not fully understood. Therefore, it was not possible to rate this following the COSMIN recommendation; however, all the questions within the various

questionnaires appeared to have good content validity. Good content validity refers to base on our own assessment of the questionnaires, not on the COSMIN criteria. When reviewing the items in the questionnaires, appeared that they were all the items were measuring the construct they purport to and had good content validity from our perspective. In addition, the questionnaires have been designed by clinicians and/or researchers working with patients who experience hyperacusis and/or misophonia, so they were in a good position to create relevant items.

In terms of structural validity, six studies did not report any psychometric data. The rest of the studies methodological quality for structural validity varies between "very good" and "inadequate" according to COSMIN risk of bias checklist assessment. This mainly was due to studies only reporting exploratory factor analysis (EFA) without confirmative factor analysis (CFA). To test the factor structure, CFA or item response theory (IRT) analysis are preferred according to the COSMIN checklist (Mokkink et al., 2018).

None of the instruments reported on all three psychometric properties within the domain of reliability (reliability, internal consistency, and measurement error). Only one measurement instrument (MisoQuest) reported reliability with measuring interclass correlation coefficient (ICC), while all instruments reported internal consistency with receiving a very good score for study quality. Although measurement error is clinically important because as more error is introduced into the score, the lower reliability will be, only one article that tested MisoQuest reported it.

None of the studies reported information on criterion validity. As there is no universally accepted gold standard to measure hyperacusis and misophonia, this feature of criterion validity could not be reported in this review. In addition, cross-cultural validity was reported in two studies (Erinc and Derinsu, 2020, Fioretti et al., 2015) with doubtful ratings. However, five

studies (Khalfa et al., 2002, Bläsing et al., 2010, Greenberg and Carlos, 2018, Siepsiak et al., 2020, Wu et al., 2014) included in this review did not conduct cross-cultural validity because the measures were developed and validated in the original language.

Hypotheses testing for construct validity was reported in 11 studies (78.6%) with ratings of either very good, adequate, or doubtful. Only four studies (Aazh et al., 2021, Bläsing et al., 2010, Greenberg and Carlos, 2018, Meeus et al., 2010), reported both convergent and discriminant validity according to COSMIN risk of bias assessment. Except for these four studies, the remaining studies had limited evidence for construct validity.

**Table 5** gives information about the results of each study on the different measurement properties, and it was rated as sufficient (+), insufficient (-), or indeterminate (?) following COSMIN criteria for good measurement properties. There is insufficient evidence within the included papers to making a judgment on their overall quality. Therefore, we chose not to summarize the results and thus not to grade the total level of evidence per psychometric instruments.

There are some other hyperacusis questionnaires used in clinics and research, but these were not reviewed as their relevant publications did not provide the psychometric properties required by COSMIN. One questionnaire, for example, is the Multiple-Activity-Scale for Hyperacusis (MASH), by Dauman and Bouscau-Faure (2005). The development procedure and metrics were not reported in this paper, so it was not possible to review its psychometric properties.

Several newly developed hyperacusis and misophonia questionnaires were not included in this review as the results of their psychometric properties were not published in a peer-review journal at the time our literature search (Aazh et al., 2022, Rosenthal et al., 2021, Dibb et al., 2021). Therefore, it was not possible to evaluate them with the COSMIN checklist in this review. Future reviews should assess the questionnaires which were published from April 2021.

In this systematic review, the populations within included studies varied, with both clinical and non-clinical samples. Clinicians desiring to select measures for clinical use should consider how generalizable the results are to the intended population, taking into account the populations from which the data in these studies were generated. For example, IHS (Greenberg and Carlos, 2018) appears to be internally consistent in both clinical and non-clinical populations. The MisoQuest (Siepsiak et al., 2020) is internally consistent for the clinical population. Hyperacusis Questionnaire originally developed by Khalfa et al. (2002) was internally consistent for just general population and Fackrell et al. (2015) investigated the validity and reliability of the HQ in a population who had tinnitus. They found the HQ to have high internal consistency (Cronbach's alpha= 0.88) but confirmatory factor analysis revealed that the proposed three-factor, and an alternative one-factor structure were poor. Therefore, HQ does not seem to work well within a tinnitus population. Future studies should endeavour to use clinical population of patients with hyperacusis or misophonia when developing questionnaires.

#### Implications for Future Research

Given the recent measures being adapted for use in other countries and languages, we believe that there is a need for appropriate and more testing for cross-cultural validity. Studies with different cultural groups should perform factor analyses for multiple groups and complete measurement invariance or DIF (differential item functioning) to give information on whether the measures are equivalent when used in different cultures/languages. For example, MisoQuest was developed in Polish, and validation has only been performed in a Polish population. Therefore, for future directions, validation and cross-cultural evaluation of MisoQuest are needed for other countries and different languages.

Regarding structural validity, future studies should perform factor analyses using CFA (confirmatory factor analysis) or IRT (item response theory) for seven instruments (HQ, IHS,

Hyperacusis Handicap Questionnaire [HHQ], short HQ [SHQ], questionnaire on hypersensitivity to sound [GUF], Misophonia Questionnaire [MQ], Amsterdam Misophonia Scale [A-MISO-S]).

To gain a comprehensive picture of reliability, all elements of reliability should be assessed. Internal consistency has been assessed for all instruments, but future studies should assess test-retest, interrater, and intra-rater reliability for HQ, IHS, SHQ, GUF, MQ, MisoQuest, A-MISOS and HHQ. Measurement error also need to be assessed for all eight instruments.

We also believe that future studies measuring content validity should state more explicitly how they evaluated content validity and follow COSMIN criteria when developing and reporting a new measure. This may include exploring the relevance, comprehensiveness, and comprehensibility of the measure among a sufficient sample of participants and professionals, which could lead to more credible evidence of its content validity.

All the available questionnaires regarding hyperacusis and misophonia are designed for adults and therefore may not be appropriate for use in children and adolescents. Therefore, future studies are needed for the development of new questionnaires in these specific groups.

Responsiveness is defined as the ability of the psychometric instrument to detect change over time in the construct measured (Mokkink et al., 2018). This review showed that responsiveness to change has not formally been tested for hyperacusis and misophonia questionnaires. However, HQ and A-MISO-S have been used in several interventional studies and appear to be sensitive to change (Nolan et al., 2020, Aazh et al., 2020, Aazh et al., 2019, Aazh and Moore, 2018b, Aazh and Moore, 2018a, Schroder et al., 2017, Juris et al., 2014, Beukes et al., 2018) (scores have changed following treatment). This provides some evidence for responsiveness. More systematic studies are needed to further explore responsiveness to

change and the cut off for meaningful or clinically significant change in hyperacusis and misophonia questionnaires.

#### Conclusion

This study systematically reviewed publications that evaluated the psychometric properties of eight hyperacusis and misophonia instruments using COSMIN guidelines (i.e., HQ, IHS, HHQ, SHQ, GUF, MQ, A-MISO-S and MisoQuest). Evidence concerning psychometric properties was limited and no single measure of hyperacusis and/or misophonia was found to meet all nine methodological quality standards according to the COSMIN guideline. There is a need for further research on the psychometric properties of the instruments included in this review.

#### **Conflict of Interest**

No conflict of interest has been declared by the authors.

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## **CHAPTER THREE**

Study 2. Evaluation of the Psychometric Properties of the English

MisoQuest and Its Relationship with Audiological and Psychological

Factors

### 3.1 Overview

As outlined in the systematic review, existing psychometric instruments for hyperacusis and misophonia have been evaluated inconsistently, leaving gaps in their methodological quality. The review highlighted that no studies had specifically assessed the English version of MisoQuest, a 14-item questionnaire originally developed and validated in Polish. To address this gap, the aim of this chapter was to evaluate the validity and reliability of the English version of MisoQuest for use in English-speaking populations.

This chapter particularly focuses on examining the psychometric properties of the English MisoQuest, including its internal consistency, measurement error, and test-retest reliability. Confirmatory Factor Analysis (CFA) was performed to assess the factor structure, while convergent and discriminant validity were evaluated by correlating MisoQuest with related and unrelated constructs. A large sample of adult participants provided a robust dataset, and follow-up interviews were conducted to further support the findings.

The results demonstrated strong internal consistency and high test-retest reliability, supporting the reliability of the English MisoQuest. CFA confirmed the multidimensional structure, with an adequate model fit. Convergent validity was evidenced by moderate to strong correlations with other misophonia measures, while weak correlations with non-misophonia measures supported discriminant validity. However, moderate correlations with hyperacusis

and fatigue measures were unexpected and are discussed in relation to potential construct overlap.

This chapter discusses the implications of these findings, highlighting the strengths of the English MisoQuest while addressing its limitations. By establishing its psychometric properties, this chapter contributes to the overall aim of the thesis to improve the measurement and understanding of misophonia in English-speaking populations.

## 3.2 Research Paper

# **Evaluation of the Psychometric Properties of the English MisoQuest and Its Relationship with Audiological and Psychological Factors**

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#### **Abstract**

**Objective:** The primary aim was to establish the validity and reliability of the English version of MisoQuest, a 14-item misophonia questionnaire initially validated in the Polish language.

**Design:** Reliability was examined through internal consistency, measurement error, and test-retest reliability. Validity was assessed using confirmatory factor analysis (CFA), with convergent and discriminant validity evaluated by correlating MisoQuest with other relevant measures. Participants completed the MisoQuest at two-time points and participated in follow-up interviews.

**Study Sample:** A total of 451 adult participants, with an average age of 36.4 years (SD = 12.8), were included in the study.

Results: The internal consistency of the English MisoQuest was very good ( $\alpha$  and  $\omega$  = 0.93) and the test-retest reliability was high (ICC = 0.89). While CFA indicated an adequate model fit, it did not fully support a one-factor structure, as some items shared variance even when controlled for the latent variable. Convergent validity was evidenced by moderate to strong correlations with established misophonia questionnaires. Weak correlations with non-misophonia measures indicated discriminant validity. Unexpectedly, moderate correlations were found between MisoQuest and Hyperacusis Impact Questionnaire (r = 0.34; p < 0.01) and Three-dimensional Fatigue Inventory (r = 0.31; p < 0.01).

**Conclusion:** The study found that the English version of MisoQuest is a reliable and valid tool for measuring misophonia within the English-speaking adult population.

### 1. INTRODUCTION

Misophonia is characterised by profound emotional, physiological and behavioral reactions towards specific sounds or visual stimuli, as defined by Swedo et al. (2022). Common triggers for misophonia involve sounds produced by others, particularly those linked to oral functions (e.g., chewing, eating), nasal sounds (e.g., breathing and sniffing) and repetitive sounds such as tapping or clicking. When individuals with misophonia are exposed to the trigger sounds, they often experience emotional reactions (e.g., anger and irritation), bodily sensations (e.g., increased heart rate and tense muscles), and behavioral responses (e.g., avoidance and aggressive behaviors, asking people to stop making the noise) (Jager et al., 2020, Dibb et al., 2021, Edelstein et al., 2013). The prevalence of clinical misophonia (i.e., people for whom symptoms of misophonia cause a significant burden) has been reported in 18.4% of a representative sample of the UK adult population (Vitoratou et al., 2023). However, in the US, a recent study by Dixon et al. (2024) found that 4.6% of the adult population experience clinical levels of misophonia.

Research indicates that misophonia is frequently associated with conditions such as obsessive-compulsive disorder and trauma-related disorders (Wu et al., 2014, Schröder et al., 2013). Approximately 28% of individuals diagnosed with misophonia also exhibit one or more psychiatric disorders (Jager et al., 2020). Misophonia co-occurs with a diverse range of anxiety disorders, with nearly 57% of sampled participants meeting the criteria for at least one anxiety-related disorder (Rosenthal et al., 2022). It is estimated that 12% of misophonia patients also experience tinnitus, with 4% of them also presenting hyperacusis (Rouw and Erfanian, 2018). Some studies have suggested that individuals with misophonia also report hearing loss (Enzler et al., 2021b, Siepsiak et al., 2022, Aazh et al., 2022). Dibb and Golding (2022) reported that individuals with misophonia have difficulties in emotional well-being and social functioning.

They reported that compared to quality of life, individuals with misophonia showed increased levels of fatigue as measured by the Misophonia Response scale (Dibb and Golding, 2022).

A valid and reliable measurement tool is essential for accurately assessing misophonia severity and ensuring consistent results across studies, which enhances the credibility and comparability of research. The MisoQuest questionnaire, consisting of 14 items, was initially validated in a Polish sample (Siepsiak et al., 2022). However, a recent systematic review by Kula et al. (2023) emphasized the need for further evaluation of MisoQuest's psychometric properties, especially within an English-speaking population, using the COSMIN framework (Mokkink et al., 2018).

This study aims to validate the English version of the MisoQuest by evaluating its structural validity, reliability, measurement error, and convergent and discriminant validity according to COSMIN standards (Mokkink et al., 2018). Our data collection involved a three-stage approach. Initially, participants engaged in an online survey, completing the measurements at two separate points two weeks apart. A subsample of participants who completed both stages of the survey were invited to the third stage, which was an interview to complete the same survey, using identical measures, but this time administered by the researcher. More details are provided in the methods section.

Based on existing research, we hypothesised the following for the validation of the English MisoQuest. While Siepsiak et al. (2020) performed a confirmatory analysis using Graded Response Model to confirm the factor structure of the MisoQuest, they did not use Confirmatory Factor Analysis (CFA). Although measurement error was addressed in the original study, our study focuses on analysing the standard error of measurement. Furthermore, both convergent and discriminant validity are examined, aspects not fully explored in the initial validation. We expected the English version to demonstrate internal consistency (Cronbach's  $\alpha$  and McDonald's  $\omega > 0.70$ ), strong test-retest reliability similarly reliability levels as reported

in the original scale's development, and low measurement error. For convergent validity, we anticipated moderate to strong correlations between the English MisoQuest and other misophonia-related measures (e.g., AMISO-R, MIQ, SS4), and for discriminant validity, weak correlations with constructs unrelated to misophonia, such as hearing loss, hyperacusis, mental health, sleep and stress.

## 2. MATERIALS AND METHODS

## 2.1 Ethics approval and consent to participate.

Ethics approval for the study was obtained from the University of Surrey Research Integrity and Governance Office (Reference: FHMS-21 22 083 EGA). All participants provided online consent after being informed about the study's objectives, procedures, and their right to withdraw at any time.

## 2.2 Participants

The sample size was determined based on a 25:1 participant-to-item ratio for the MisoQuest questionnaire, resulting in a minimum target of 350 participants (Hair et al., 2019). Although some scholars recommend a minimum ratio of 5:1 (Comrey and Lee, 2013), a larger sample size can improve factor stability and result generalizability. For test-retest reliability, a minimum of 100 participants were required to minimize statistical error and improve reliability estimates (COSMIN guidelines; Mokkink et al. (2018).

The survey link was distributed via university email lists, social media (Instagram, X), and the Hashir International Specialist Clinics' online newsletter. Additionally, it was shared within Facebook support groups and through nonprofit organizations (e.g., SoQuiet). From 475 initial respondents, only those who passed an attention check and met standard response time criteria were included in the analysis, yielding a final sample of 451. This quality control process aligns with recommendations to exclude careless responses (Arias et al., 2020, Chandler et al., 2020), and we additionally exclude outliers with response times more than three standard deviations

from the mean (Leys et al., 2013). Initially, 475 individuals were recruited for the study. After applying the attention check and processing time criteria, a final sample of 451 respondents was included in the analysis. In the study's second phase for test-retest reliability, 154 participants took part.

The inclusion criteria for participants included being at least 18 years old and having a good understanding of English (self-reported). Exclusion criteria were if they had a significant cognitive impairment or severe visual impairment (this was communicated with them in the study invitation and information sheet).

## 2.3. The translation process

According to our agreement with the developer of MisoQuest, we used the English version of MisoQuest that was translated from the original Polish version and published as a supplement to their original paper (Siepsiak et al., 2022). For this translation, two Polish researchers translated the questionnaire from Polish to English. Following this, a third person, a native speaker familiar with misophonia, corrected the translation for linguistic accuracy. They also approved that there was a back-translation of MisoQuest during the process.

### 2.4 Procedures

The study, conducted between May and July 2023, invited participants to complete an online survey on sound sensitivity, hosted on Qualtrics (Provo, USA, <a href="https://www.qualtrics.com">https://www.qualtrics.com</a>). After consenting and receiving study information, participants provided demographic details (age, gender, education, ethnicity, religion, employment and completed questionnaires assessing misophonia, hyperacusis, tinnitus, mental health, hearing, stress, sleep, and fatigue (see Section 2.5 for details). Supplementary Figure 1 illustrates the stages of participant involvement. Although follow-up interviews were planned after the test-retest phase, only 14 participants agreed to participate.

## 2.4.1 Test-Retest Reliability

To assess test-retest reliability, participants completed the survey twice over a two-week interval, as recommended by Franzen et al. (1987). A total of 154 participants engaged in this phase, providing data on the stability of responses over time.

## 2.4.2 Assessing the effect of administration mode (self-administered vs interview-based)

Following the test-retest phase, participants were invited to an optional online interview to compare self-administered responses with those gathered in a researcher-administered setting. Fourteen participants took part in this third phase, which occurred immediately following the second phase. This allowed for a comparison between self-administration and researcher-administration of MisoQuest.

## 2.4.3 Screening Questions for tinnitus and hearing

Participants completed screening questions for tinnitus. Those with tinnitus were asked to complete the Tinnitus Impact Questionnaire [TIQ; Aazh et al. (2022a]. All participants answered a hearing status question, and those indicating hearing loss completed the Hearing Handicap Inventory [HHI; Cassarly et al. (2020)].

### 2.5 Measures

The primary measure in this study was MisoQuest. Additional measures were administered to evaluate MisoQuest's convergent and discriminant validity:

**MisoQuest** (Siepsiak et al., 2020a): This 14-item questionnaire uses a five-point Likert scale (1 = "strongly disagree" to 5 = "strongly agree") to evaluate sensitivity to bothersome sounds. Originally developed in Polish, it demonstrated a Cronbach's α of 0.96 and ICC of 0.84 in reliability tests. The authors of MisoQuest (Siepsiak et al., 2020a) acknowledged that they translated the questionnaire into English, and we employed this English version in our study. A cut-off value of 61 has been established to identify individuals who may be experiencing misophonia by Siepsiak et al. (2020b).

The Amsterdam Misophonia Scale- Revised (Jager et al., 2021): A 10-item scale with a 0-4 rating for misophonia severity, where scores of 0-10 indicate subclinical, 11-20 mild, 21-30 moderate, and 31-40 severe symptoms. The English version of AMISO-R demonstrated a Cronbach's  $\alpha$  of 0.81 (Naylor et al., 2021) and was included to assess the convergent validity of MisoQuest.

**Misophonia Impact Questionnaire** (Aazh et al., 2023): An 8-item scale measuring the impact of misophonia, with items 0-3 (0 = 0-1 day, 1 = 2-6 days, 2 = 7-10 days, 3 = 11-14 days), with a total score range of 0-24. Scores  $\geq$  12 indicate significant impact (Erfanian and Aazh, 2023). The MIQ demonstrated strong reliability, with a Cronbach's  $\alpha$  of 0.94 (Aazh et al., 2023). This measure was used to assess convergent validity.

Sound Sensitivity Symptoms Questionnaire (Aazh et al., 2024a): 5 items explore several sound sensitivity symptoms, including loudness hyperacusis, pain or discomfort hyperacusis, annoyance hyperacusis/misophonia, and fear hyperacusis, over two weeks. For each item, a score of 0, 1, 2, or 3 is assigned to the response categories of 0 to 1 day, 2 to 6 days, 7 to 10 days, and 11 to 14 days, respectively. A score of five or more indicates a clinically significant sound intolerance problem. In a clinical sample, the SSSQ demonstrated a reliability coefficient of Cronbach's  $\alpha = 0.87$  (Aazh et al., 2024a). SS4 (item 4 in SSSQ) specifically evaluates the severity of misophonia symptoms by assessing how often participants felt angry or anxious when exposed to specific sounds associated with eating noises, lip smacking, sniffling, breathing, clicking sounds, and tapping over the past two weeks. Consequently, we employed SS4 to measure convergent validity. The sum of items 1, 2, 3, and 5 quantifies hyperacusis symptoms, and this part was used to assess discriminant validity.

Hyperacusis Impact Questionnaire (Aazh et al., 2024a): An 8-item tool assessing the hyperacusis impact with items scored 0-3 based on frequency (0 = 0-1 day, 1 = 2-6 days, 2 = 7-10 days, 3 = 11-14 days), giving a total score range of 0-24. Scores of 12 or higher indicate

a clinically significant impact. The HIQ showed high reliability, with a Cronbach's  $\alpha$  of 0.93 (Aazh et al., 2024a). This instrument was applied to assess discriminant validity.

**Tinnitus Impact Questionnaire** (Aazh et al., 2022b): A 7-item measure assessing tinnitus impact daily activities, mood, and sleep. Each item is rated 0-3 to indicate frequency (0 = 0-1 day, 1 = 2-6 days, 2 = 7-10 days, 3 = 11-14 days), yielding a total score range of 0-21. Internal consistency is high, with Cronbach's  $\alpha$  = 0.89 (Aazh et al., 2022b). Scores below 5 indicate no impact, 5-6 mild impact, 7-8 moderate impact, and 9 or higher severe impact. TIQ was used to assess discriminant validity.

Screening for Anxiety & Depression in Tinnitus (Aazh et al., 2024a): The SAD-T contains four items that match those for the physical health questionnaire (PHQ-4; Kroenke et al., 2009). Two items relate to the experience of anxiety, and two relate to the experience of depression. Each item is scored 0-3, corresponding to 0-1 day, 2-6 days, 7-10 days, and 11-14 days. The total score ranges from 0 to 12, with scores of 4 or more indicating symptoms of anxiety and/or depression. The SAD-T has high reliability (Cronbach's  $\alpha = 0.91$ ; Aazh et al. (2023b) and was used to assess discriminant validity.

Hearing Handicap Inventory (Cassarly et al., 2020): A 10-item measure assessing the social and emotional impact of hearing loss. Responses are scored as "yes" (4), "sometimes" (2), or "no" (0), with total scores ranging from 0 to 40. Score ranges: 0-8 (no handicap), 10-24 (mild-moderate), 26-40 (severe). Higher scores indicate greater perceived handicap. The HHI shows high internal consistency (Cronbach's  $\alpha = 0.95$ ; Cassarly et al. (2020) and was used to assess discriminant validity.

A single-item measure of stress symptoms (Elo et al., 2003): A one-item questionnaire assesses the stress symptoms. The scores range from 1-5. Higher scores showed greater stress levels. The single-item measure has been found valid in terms of content and criterion validity

by Elo et al. (2003). We used this questionnaire to examine the discriminant validity because it assesses stress symptoms.

**Single Item Sleep Quality Scale** (Snyder et al., 2018): One item measure that evaluates sleep quality. The scores "0": Terrible, "1-3": Poor, "4-6": Fair, "7-9": Good, "10": Excellent. The test-retest reliability of this measure was found to be 0.74 in stable patients with depression and 0.62 in insomnia patients during a 4-week period, and it demonstrated good convergent and discriminant validity. We used this item to assess the discriminant validity due to its focus on measuring sleep quality.

Three-Dimensional Fatigue Inventory (Frone and Tidwell, 2015): The 3D-WFI is an 18item tool assessing fatigue across three dimensions: physical, mental, and emotional (six items
each). Responses are on a five-point Likert scale (1 = every day to 5 = never). Internal
consistency for each dimension is high, with reliability estimates exceeding 0.90 (Frone et al.,
2018). The 3D-WFI was used to assess the discriminant validity of the English version of
MisoQuest.

## 2.6 Data Analysis

Descriptive statistics and normality tests, including the Kolmogorov-Smirnov test, were calculated for all administered measures. Normality was assessed visually through histograms and quantitatively to ensure data met parametric assumptions. All analyses, except CFA, were conducted in IBM SPSS version 28.0. The structural validity of the English MisoQuest was assessed by CFA using IBM SPSS Amos version 28, following initial analysis in AMOS, a reassessment was conducted in R using "lavaan" package with the Weighted Least Squares Mean and Variance adjusted (WLSMV) estimator to account for the ordinal nature of the data. Model fit was evaluated using Root Means Square of Approximation (RMSEA), Standardized Root Mean Square Residual (SRMR), Tucker Lewis Index (TLI), Comparative Fit Index (CFI), Goodness of fit indexes (GFI), CMIN (Chi-square/ df). Following Schermelleh-Engel et al.

(2003), a good model fit was defined by SRMR  $\leq$  0.05; RMSEA  $\leq$  0.05, TLI  $\geq$  0.97, CFI  $\geq$  0.97, GFI  $\geq$  0.95 and  $\chi^2/df \leq$  2.

Reliability analyses included internal consistency (Cronbach's  $\alpha$  and McDonald's  $\omega$ ), testretest reliability, and measurement error. Internal consistency was considered acceptable with  $\alpha$  or  $\omega$  values > 0.7(Hayes and Coutts, 2020). Cronbach's  $\alpha$  and McDonald's  $\omega$  have a value between 0 and 1, where a value greater than 0.7 is considered acceptable (Raykov and Marcoulides, 2011). Confidence intervals (CIs) for Cronbach's  $\alpha$  were determined using biascorrected and accelerated (BCA) bootstrap with 1,000 bootstrap samples. To provide a comprehensive understanding of MisoQuest's reliability, we also reported model-based measurement error, such as item reliability within the measurement model, which offers additional information beyond what is provided by Cronbach's  $\alpha$  and McDonald's  $\omega$ .

Test-retest reliability over a two-week interval was assessed by intra-class correlation coefficient (ICC), using a two-way mixed model with absolute agreement, with ICC > 0.75 indicating good reliability (Fapta and Ms, 2015). Measurement error was further analysed using the standard error of measurement (SEM), the minimal detectable change (MDC95), and Bland-Altman analyses. The SEM assesses the variability within subjects when measuring them repeatedly as a group (Haley and Fragala-Pinkham, 2006). The MDC95 represents the smallest detectable change exceeding measurement error, indicating a significant change with 95% confidence (Schuck and Zwingmann, 2003). Bland-Altman analyses were used to identify systematic bias between repeated measurements. The Bland-Altman plot shows agreement between two test occasions and highlights potential outliers. If the 95% confidence interval (CI) of the mean difference includes zero, it suggests no significant systematic bias. The 95% limit of agreement (LOA) was used to examine natural variation, with narrower LOA indicating greater stability (Bland and Altman, 1986).

Convergent validity of the English MisoQuest was assessed by Pearson correlation coefficients between MisoQuest scores and other misophonia measures (AMISOS-R, MIQ, SS4). Discriminate validity was examined by correlating MisoQuest scores with measures unrelated to misophonia (HIQ, SSSQ excluding item 4, SAD-T, TIQ, sleep and stress measures, 3D-WFI, HHI). Correlation strength was categorised as strong (>0.5), moderate (0.30–0.49), or weak (0.10–0.29) (Cohen, 1992). Moderate to strong positive correlations supported convergent validity, while weak correlations supported discriminant validity.

Finally, a paired t-test was used to compare MisoQuest scores between self- and researcheradministered conditions in the third stage, assessing the effect of administration mode.

### 3. RESULTS

## 3.1. Participants' Characteristics and Descriptive Statistics

Demographic data of the 451 respondents who completed the survey are provided in the Supplementary Table 1. Most participants identified as female (N=272, 60.3%), white (N=346, 76.7), married (N=243, 53.9%), employed full-time (N=235, 52.1%), not having any religion (N=184, 40.8%), having a bachelor's degree (N=165, 36.6%). The average age of the sample was 36.46 years (SD = 12.81).

Participants were recruited from the general population, student groups, and support communities for hyperacusis, misophonia, and tinnitus. For detailed descriptive statistics and findings for each questionnaire and item in the English version of the MisoQuest, please refer to Tables 2 and 3.

Table 1: Descriptive Statistics and Reliability of Administered Questionnaires

Questionn	N	Mean	Min	Max	Skewness	Kurtosis	K-S	K-S	Reliability
aires		(SD)					Statistics	p-	(95% CI)
								value	
MisoQues	451	48.90	14	70	-0.40	-0.03	0.07	.08	0.93 (0.92-
t		(12.30)							0.94)
HIQ	451	9.60	0	24	0.37	0.11	0.09	.20	0.90 (0.89-
		(6.12)							0.91)
MIQ	451	9.30	0	24	0.39	-0.67	0.11	.05	0.93 (0.92-
		(6.82)							0.94)
TIQ	173	7.73	0	21	0.44	-0.79	0.12	< .05	0.92 (0.91-
		(6.03)							0.94)
SAD-T	451	4.85	0	12	0.48	-0.61	0.12	< .05	0.87 (0.85-
		(3.38)							0.89)
SSSQ	451	6.60	0	18	0.46	-0.31	0.14	0.06	0.80 (0.77-
		(4.26)							0.83)
AMISO-	451	18.81	0	40	-0.15	-0.52	0.14	< .05	0.92 (0.91-
R		(9.20)							0.93)
ННІ	87	21.17	0	40	-0.24	-0.39	0.09	0.20	0.84 (0.78-
		(9.40)							0.88)
Stress	451	3.60	1	5	-0.67	-0.48	0.15	< .01	Not
		(1.09)							applicable
Sleep	451	5.45	0	10	-0.47	-0.73	0.32	< .01	Not
		(2.10)							applicable
3D-WFI	451	66.25	18	90	-0.48	-0.13	0.09	.20	0.95 (0.95-
		(15.27)							0.96)

*Notes:* HIQ, Hyperacusis Impact Questionnaire; MIQ, Misophonia Impact Questionnaire; TIQ, *Note.* Tinnitus Impact Questionnaire; SAD-T, Screening for Anxiety and Depression in Tinnitus; SSSQ, Sound Sensitivity Symptoms Questionnaire; AMISO-R, The Amsterdam Misophonia Scale- Revised; HHI, Hearing Handicap Inventory; Stress: A single item measure of stress symptoms; Sleep, Single Item Sleep Quality Scale; 3D-WFI, Three- Dimensional Fatigue Inventory; N = Number of participants; K-S: Kolmogorov Smirnov.

Table 2: Descriptive Statistics, Factor Loadings and Item Reliability for MisoQuest items

Item Mean		Min	Max	Skewness	Kurtosis	Factor	R <sup>2</sup> (Item	
	(SD)					Loadings	Reliability)	
						(λ)		
MisoQuest	3.72	1	5	63	33	0.76	0.58	
Item 1	(1.10)							
MisoQuest	3.78	1	5	72	15	0.69	0.48	
Item 2	(1.08)							
MisoQuest	3.41	1	5	42	78	0.71	0.51	
Item 3	(1.17)							
MisoQuest	3.52	1	5	42	79	0.73	0.53	
Item 4	(1.19)							
MisoQuest	3.55	1	5	46	80	0.72	0.53	
Item 5	(1.19)							
MisoQuest	3.25	1	5	23	93	0.62	0.38	
Item 6	(1.21)							
MisoQuest	3.50	1	5	53	48	0.72	0.50	
Item 7	(1.22)							
MisoQuest	3.24	1	5	31	95	0.64	0.41	
Item 8	(1.22)							
MisoQuest	3.42	1	5	31	97	0.68	0.47	
Item 9	(1.23)							
MisoQuest	3.74	1	5	70	38	0.76	0.58	
Item 10	(1.16)							
MisoQuest	3.59	1	5	56	71	0.74	0.55	
Item 11	(1.22)							
MisoQuest	3.62	1	5	56	82	0.75	0.57	
Item 12	(1.26)							
MisoQuest	3.11	1	5	01	-1.15	0.66	0.47	
Item 13	(1.28)							
MisoQuest	3.44	1	5	47	75	0.72	0.53	
Item 14	(1.19)							

*Note.* N (number of participants) = 451

The study included 451 participants across various groups: 'General' (205), 'Student' (109), 'SoQuiet (misophonia charity)' (61), and 'Other Support Groups' (76, including social media, tinnitus patients, and a tinnitus charity).

3.2. Prevalence of Misophonia, Tinnitus, Hyperacusis, Hearing Loss, Sleep Disturbance, and Mental Health Symptoms among all respondents

Approximately 21.7% of respondents (98/451) were identified as having misophonia based on the MisoQuest, with a higher prevalence in females (60/98). A Chi-Square test showed a significant difference in misophonia prevalence between females and males ( $\chi^2$  (1) = 16.52, p < .001). Among those with misophonia, 32.1% reported tinnitus, and 16.7% reported hearing loss. Misophonia prevalence also varied by employment status: 10% among students, 18% among employed individuals, and 5% among retired individuals, with a significant association found ( $\chi^2$  (2) = 19.48, p < .005). Regarding tinnitus, 39% of participants reported experiencing it, with varying levels of impact (9.3% mild, 12.1% moderate, 42.2% severe). Hyperacusis had a clinically significant impact on 35.5% of participants, while 20% reported hearing loss, with 44% experiencing bilateral loss. Sleep disturbance varied, with 18% reporting poor sleep and 34.1% reporting good sleep quality. This sleep-specific questionnaire is separate from the tinnitus measure and assesses sleep quality in general, regardless of tinnitus. Additionally, 61% of participants showed possible symptoms of anxiety and/or depression based on SAD-T. Misophonia prevalence varied by the questionnaire used: 21.7% with MisoQuest, 36.6% with MIQ, and 80% with AMISOS-R, which also categorised symptoms as 34.1% mild, 35.5% moderate, and 10.2% severe to extreme.

## 3.3. Structural Validity

The initial CFA model, based on Siepsiak et al. (2020a), was tested on the present dataset to confirm the one-factor structure of 14 items. Although this structure was supported, model fit was poor:  $\chi^2 = 295.73$ ,  $\chi^2/df = 3.84$  (above the recommended threshold of 2), RMSEA =

0.07 (exceeding the 0.05 limit), CFI = 0.94, and TLI = 0.93 (both below the 0.97 cut-off), though SRMR was acceptable at 0.04. Modification indices were then used to address misspecifications, resulting in improved fit indices:  $\chi^2 = 180.53$ , df = 72,  $\chi^2$ /df = 2.50, RMSEA = 0.05, SRMR = 0.03, CFI = 0.97, and TLI = 0.96 (Please see Supplementary Figure 2). In a subsequent analysis, the model fit was reassessed using the WLSMV estimator to better align with the ordinal nature of the data, yielding slightly better fit indices compared to the modified model:  $\chi^2$ /df = 2.41, SRMR = 0.04, RMSEA = 0.05, CFI = 0.97, TLI = 0.97.

## 3.4. Reliability and Measurement Error

Cronbach's α and McDonald's ω for the English version of the MisoQuest were both robust, each with a value of 0.93 in this sample. Factor loadings for each item were analyzed to assess their relationship with the underlying latent construct. These loadings, along with the corresponding item reliability (R²) values, are presented in Table 5. The factor loadings, ranging from 0.62 to 0.76, are considered moderate to very good, indicating that the items effectively represent the intended construct. The R² values, which range from 0.38 to 0.58, suggest moderate to good reliability, confirming that a significant portion of the variance in each item is explained by the latent factor, thus supporting the overall reliability and validity of the MisoQuest scale.

The ICC analysis showed good test-retest reliability for the English MisoQuest (ICC= 0.89, 95% CI [0.85, 0.92]) using a two-way mixed effects model for absolute agreement. The SEM was 3.19, indicating the standard deviation of an observed score around the true score, and the MDC<sub>95</sub> was 8.81, reflecting the minimum detectable change. Bland-Altman analysis revealed no significant systematic bias, with the mean difference close to zero. Eight outliers fell outside the 95% limits of agreement (LOA), which were narrow, indicating minimal variability and high stability of the MisoQuest scores. This narrow range suggests small, consistent differences between repeated measures, supporting the instrument's reliability (**Please see Supplementary** 

**Figure 3**). The assumption of normality for the mean differences in the Bland-Altman analysis was also met, validating the results.

## 3.5. Convergent Validity

The convergent validity of the English version of MisoQuest was evaluated by calculating Pearson correlation coefficients between the AMISOS-R, MIQ and SS4. The convergent validity of the English MisoQuest was supported by strong positive correlations with the AMISOS-R (r = 0.65, t (449) = 18.11, p < 0.01), and moderate positive correlations with the MIQ (r = 0.66, t (449) = 11.29, p < 0.01) and SS4 (r = 0.659, t (449) = 10.84, p < 0.01).

## 3.6. Discriminant Validity

Table 3: Discriminant validity: correlations coefficients

	HIQ	SS1235	SAD-T	TIQ	Sleep Quality	Stress	ННІ	3D-WFI
MisoQuest	<i>p</i> < 0.01	r = 0.25 p < 0.01 N = 451	<i>p</i> < 0.01	p=0.931	p = 0.04	<i>p</i> < 0.01	p = 0.57	<i>p</i> < 0.01

*Note*. HIQ, Hyperacusis Impact Questionnaire; SS1235, items 1,2,3, and 5 on Sound Sensitivity Symptoms Questionnaire; SAD-T, stress and depression; TIQ, Tinnitus Impact Questionnaire; HHI, Hearing Handicap Inventory; HHI, Hearing Handicap Inventory. The number of participants is indicated by N.

**Table 3** reveals a weak positive correlation between the English version of MisoQuest and SS1235, SAD-T, stress and sleep quality measures and very weak, non-significant negative correlations with TIQ and HHI, supporting the discriminant validity of the English version of MisoQuest. Contrary to our expectations, there were positive moderate correlations between MisoQuest and HIQ and 3D-WFI.

### 3.7. Mode of administration of the MisoQuest Results

The participant-administered and researcher-administered MisoQuest were compared using a paired t-test. The mean score for participant-administered was 53.31 (SD = 11.16), and for

researcher-administrated was 53.88 (SD=13.0). The paired t-test analysis indicated no statistically significant difference between the two methods, as the mean difference was -2.43 (95% CI [-5.3, 0.49]), t (12) = -1.7, p=0.97. The effect size, Cohen's d, was estimated at -0.44, with a 95% confidence interval ranging from -0.95 to 0.08. In addition, a correlation analysis revealed a strong positive correlation between the scores of participant-administered and researcher-administered MisoQuest, with a correlation coefficient of 0.91 (p<0.01). This suggests a high degree of agreement between the two methods of delivery. These results indicate that when participants completed the questionnaire alone, it did not significantly impact the scores, and the two methods demonstrated a strong correlation.

### 4. DISCUSSION

The study aimed to assess the psychometric properties of the English MisoQuest following COSMIN guidelines (Mokkink et al., 2018) for translated questionnaire. CFA initially indicated a one-factor structure consistent with the model proposed by Siepsiak et al. (2020a). However, the initial model fit was poor, so modifications were made to improve model fit. These changes addressed shared variance among items with similar emotional and behavioural responses to trigger sounds, improved the overall model fit. Nonetheless, since these adjustments were data-driven rather than theory-based, they might suggest potential multidimensionality within the measure. Subsequent reassessment using the WLSMV estimator, which accounts for the ordinal nature of the data, yielded improved yet still adequate fit compared to the modified ML model, with persistent evidence of multidimensionality. These findings highlight that further adjustments may be needed in future studies to better align the model with theoretical support.

The internal consistency of the English version of MisoQuest with Cronbach's  $\alpha$  and McDonald's  $\omega$  of 0.93, closely aligns with the original scale developer's reported Cronbach's  $\alpha$  of 0.96 by Siepsiak et al. (2020a), highlighting the robustness of the questionnaire's reliability

across different sample characteristics. Our study found a test-retest reliability ICC value of 0.89 for the English version of MisoQuest, slightly higher than the original developer's reported value of 0.84, indicating strong consistency in responses over a two-week period.

While Siepsiak et al. (2020a) addressed measurement error in the original Polish version of MisoQuest, and Raymond and Butler (2024) have previously examined convergent and discriminant validity in the English version, our study provides a comprehensive psychometric analysis following COSMIN standards. This approach offers a robust evaluation of the English MisoQuest, supporting its suitability for clinical and research applications.

Our examination of convergent validity showed moderate to strong correlations between MisoQuest scores and those from AMISOS-R, MIQ, and SS4, indicating that these instruments assess related constructs. However, discrepancies were observed in the proportion of participants diagnosed with misophonia across these tools, which relates to diagnostic or differential validity rather than convergent validity. Specifically, AMISOS-R identified a higher prevalence than both MisoQuest and MIQ, likely due to differences in design and conceptual frameworks. Each tool approaches misophonia measurement uniquely: AMISOS-R focuses on experiences triggered in the past three days, MisoQuest has no specified timeframe, and MIQ assesses the impact of misophonia on patients' lives over a two-week period. Therefore, these differences reflect the diverse approaches used in each questionnaire. Future studies should explore whether a distinction between misophonia as a trait or a disorder can be made using MisoQuest and AMISOS-R and how these might be related to the impact of misophonia on the patient's life measured via MIQ.

The English version of MisoQuest demonstrates reasonably good discriminant validity, as evidenced by its weak correlations with tinnitus impact, hearing loss, sleep, stress, depression, and anxiety. However, there was a moderate correlation between MisoQuest and HIQ that assesses hyperacusis. This means that MisoQuest may be influenced by hyperacusis symptoms.

Hyperacusis is different to misophonia and is characterized by the perception of certain everyday sounds, such as domestic noise or noise in public places, as too loud or painful (Aazh et al., 2014). One explanation is that HIQ may be influenced by misophonia in addition to hyperacusis. However, this is unlikely as Aazh et al. (2024d) reported that the mean of HIQ score among patients with misophonia was 8.8 (SD=8.2) compared with 18.5 (SD=6.9) among patients with hyperacusis. This indicates that HIQ is reasonably specific in differentiating the impact of hyperacusis from misophonia. Another explanation for the moderate relationship observed between MisoQuest and HIQ is a possible overlap between misophonia and one form of hyperacusis (i.e., annoyance hyperacusis). According to Tyler et al. (2014) hyperacusis has four subtypes: loudness hyperacusis, pain hyperacusis, annoyance hyperacusis, and fear hyperacusis. Previous studies also suggest possible relationship or comorbidity between hyperacusis and misophonia (Aazh et al., 2022a, Aazh et al., 2023).

The English version of MisoQuest indicated a moderated correlation with fatigue measure (3D-WFI). This result is consistent with findings from Dibb and Golding (2022), who also reported a moderate correlation between misophonia and fatigue. There is a need for future studies to further explore the relationships between misophonia, hyperacusis and fatigue.

No significant correlation was found between hearing loss and misophonia, consistent with previous studies (Aazh et al., 2022a, Siepsiak et al., 2022). In terms of mental health, we observed a weak correlation between misophonia and anxiety-depression, differing from the moderate correlation reported by Wu et al. (2014), likely due to sample differences, as their study focused on students while ours involved a more diverse population. Similarly, we found a weak correlation between misophonia and stress, which contrasts with the moderate correlation reported by McKay et al. (2018). This discrepancy may be due to differences in participant demographics and the use of different questionnaires, as their study focused on U.S. participants and used the Misophonia Questionnaire (Wu et al., 2014) and Depression Anxiety

Stress Scale (Lovibond and Lovibond, 1995). These findings underscore the need for future studies to further investigate the relationship between misophonia and mental health and stress, particularly using validated tools such as MisoQuest.

Additionally, we found a weak but significant negative correlation between MisoQuest scores and sleep quality, indicating that higher misophonia symptoms are associated with poorer sleep. Although no prior studies have explored this relationship, research on hyperacusis has shown a connection between sound sensitivity and sleep problems, with 30% of participants reporting sleep issues (Fioretti et al., 2013). Further research should explore the impact of misophonia on sleep quality to expand our understanding of this relationship.

Some limitations should be considered when interpreting the outcomes of this study. First, female and male participant numbers in our study were unequal. This imbalance, however, is consistent findings from other online misophonia studies (Dibb et al., 2021), which also observed a higher proportion of female participants. In addition, Siepsiak et al. (2020a) who investigated the invariance based on gender, found that women demonstrated higher misophonia symptoms compared to men. However, it is important to note that in studies with representative samples of the population, there is no clear evidence that females score higher than males. Therefore, conclusions from convenience samples like ours should not be generalised to the whole population.

Single-item measures offer simplicity but also come with limitations. While test-retest reliability can be evaluated, internal consistency cannot be measured, as there is only one item. This limitation is especially important in cross-sectional studies like ours, where comprehensive reliability assessment is critical. For single-item measures, certain reliability aspects remain uncertain, so the findings should be interpreted with caution. Furthermore, single-item measures may be effective for straightforward constructs (e.g. sleep quality), but

as noted by Elo et al. (2003), may not fully capture more complex, multidimensional constructs

such as stress.

Lastly, we did not measure responsiveness to change for MisoQuest, which refers to ability

to detect changes over time in the construct (Mokkink et al., 2018). Therefore, further research

is needed to measure changes in MisoQuest scores before and after any intervention.

**Conclusions** 

The English MisoQuest is a reliable and valid tool for assessing misophonia in English-

speaking populations for both research and clinical use. High reliability was indicated by

Cronbach's α and McDonald's ω values of 0.93. CFA results showed a multidimensional factor

structure, and the model demonstrated adequate fit. Test-retest reliability was strong, with an

ICC of 0.89, factor loadings between 0.62 and 0.76, and item reliability (R<sup>2</sup>) values from 0.38

to 0.58. Convergent validity was shown through moderate to strong correlations with related

measures (AMISOS-R, MIQ), while discriminant validity was supported by weak correlations

with distinct constructs (TIQ, HHI, sleep, stress, and SAD-T). One limitation is the moderate

correlation with HIQ, a hyperacusis measure, suggesting a need for future studies to further

explore the relationship between misophonia and hyperacusis and to refine psychometric tools

for their differentiation.

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## **CHAPTER FOUR**

## Study 3. The Relationship Between Misophonia and Fatigue: The Role of Sleep Quality and Stress as Mediators

#### 4.1 Overview

As discussed in Chapter 1, the relationship between misophonia and fatigue has been examined in a study by Dibb and Golding (2022) found that individuals with misophonia reported lower energy and higher fatigue levels compared to those without misophonia. While this study highlighted an association between misophonia and fatigue, it did not account for potential mediating factors such as stress, sleep quality, hyperacusis, and anxiety/depression, which are known to influence fatigue. Building on this prior work and guided by clinical observations from my second supervisor, who noted frequent reports of fatigue among misophonia patients, this study sought to address these gaps.

This chapter uses the same sample as the previous chapter, where the fatigue measure was evaluated as part of the discriminant validity analysis of the MisoQuest. A positive moderate correlation between misophonia and fatigue was identified, which prompted further investigation. This chapter explores the relationship in depth, focusing on the mediating roles of stress and sleep quality. Separating the chapters allows for a clearer and more detailed examination of these mediating factors and their potential contributions to the experience of fatigue in individuals with misophonia.

## 4.2 Research Paper

# The Relationship Between Misophonia and Fatigue: The Role of Sleep Quality and Stress as Mediators

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## **Abstract**

**Objective**: This study aims to explore whether stress and sleep quality mediate the relationship between misophonia, characterised by intense emotional, cognitive and behavioural responses to specific sounds, and fatigue.

**Design:** A cross-sectional study using correlation, regression and mediation analyses was conducted to examine the mediators of fatigue in individuals with misophonia.

**Study Sample:** Data were collected from 451 participants (60.3% women, 37.5% men; M age = 34.61, SD = 11.61) via the Qualtrics platform. Participants completed assessments for misophonia (MisoQuest), fatigue (3D-WFI), hyperacusis (HIQ), sleep quality (SQS), stress, and anxiety/depression (SAD-T).

**Results:** Correlation analysis showed a positive relationship between misophonia severity and fatigue (r = 0.32, p < 0.01). Further, multiple regression analysis indicated that misophonia, sleep quality, and stress were significant predictors of fatigue, while hyperacusis, anxiety, and depression were not. Mediation analysis also revealed that stress and sleep quality partially mediated the relationship between misophonia and fatigue (p < 0.01 for both).

Conclusions: The severity of misophonia is correlated with fatigue, with stress and sleep quality identified as partial mediators. These findings suggest that addressing stress and improving sleep may reduce fatigue in those with misophonia. Future studies should explore interventions targeting these mediating factors.

### 1. Introduction

Fatigue is a complex and subjective sensation characterized by persistent feeling of exhaustion, lack of energy, or weakness that can occur during or after usual activities (Cornuz et al., 2006). Unlike tiredness, which is a common and short-lived experience that often disappears after rest, fatigue is not relieved by rest and can significantly impact physical, cognitive, and emotional functioning, making it difficult to perform daily tasks (Galland-Decker et al., 2019, Dittner et al., 2004, Ream and Richardson, 1996). Due to its chronic nature and significant impact on well-being, fatigue is an important concern in clinical practice and research.

Misophonia is a condition where individuals are abnormally sensitive to stimuli or specific types of sounds, known as triggers, such as chewing, breathing, or tapping (Swedo et al., 2022). These sounds can provoke intense emotional and behavioural reactions, including anger, anxiety and a strong desire to escape from these trigger sounds (Schröder et al., 2013). A longitudinal study by Dibb and Golding (2022) investigated the relationship between misophonia as measured using the Misophonia Response Scale (MRS) (Dibb et al., 2021) and fatigue as measured via the quality-of-life domain of the SF-36 (Framework, 1992). The study reported that individuals with self-reported misophonia (n=491) had significantly lower scores in the energy/fatigue domain of the SF-36 compared to those without misophonia (n=503), and this relationship remained consistent over time. However, their study did not account for possible confounding factors such as hyperacusis, stress, sleep, and anxiety/depression, which could have influenced the observed relationship between misophonia and fatigue.

Hyperacusis often overlaps with misophonia, complicating the experiences of those affected (Danesh and Aazh, 2020). Individuals with hyperacusis may find normal environmental sounds unbearably loud or even painful, significantly impacting their quality of life (Aazh et al., 2018). The relationship between misophonia and hyperacusis is complex and not fully understood,

though some research suggests a significant overlap (Jager et al., 2020). For instance, Aazh et al. (2022a) found that 23% of the 257 tinnitus and/ or hyperacusis patients reported misophonia symptoms. Furthermore, the likelihood of experiencing misophonia increased with the severity of hyperacusis. Brennan et al. (2024) revealed significant strong correlations between misophonia, as measured by the Misophonia Questionnaire (Wu et al., 2014), and hyperacusis, as measured by the Hyperacusis Questionnaire (Khalfa et al., 2002), among undergraduate and graduate students. Fatigue is a common symptom reported by patients with hyperacusis and items related to fatigue are included to most hyperacusis questionnaires (Greenberg and Carlos, 2018, Aazh et al., 2024a). Paulin et al. (2016) found that hyperacusis is comorbid with conditions such as exhaustion and chronic fatigue syndrome, suggesting that hyperacusis may contribute to both physical and mental exhaustion in those affected. Given the relationship between hyperacusis and misophonia, the connection between misophonia and fatigue reported by Dibb and Golding (2022) could have been influenced by hyperacusis. Since they did not assess hyperacusis, it remains unclear whether hyperacusis might have confounded their results.

Although studies assessing sleep disturbances among patients with misophonia are limited, poor sleep quality is a well-established contributor to fatigue (Lavidor et al., 2003). Some case studies suggest that people with misophonia often experience insomnia or poor sleep quality (Gowda and Avidan, 2023), but the specific relationship between sleep disturbances and misophonia, particularly whether these disturbances contribute to experience of fatigue in this population, has not been examined.

Stress can also play a key role in the relationship between misophonia and fatigue, as stress is a known contributor to both emotional distress and fatigue (Kop and Kupper, 2016). People with misophonia often experience high levels of stress when exposed to trigger sounds, which increases their emotional and physical reactions (Rouw and Erfanian, 2018). Recent research

shows that perceived stress is a strong predictor of misophonia severity, especially when associated with hyperarousal (Guetta et al., 2024). The stress and increased arousal may worsen the experience of fatigue (Kocalevent et al., 2011). In addition, stress can lead to the development or exacerbation of hyperacusis leading to further experience of exhaustion and fatigue (Hasson et al., 2013). Therefore, it is important to explore if stress impacts the relationship between misophonia and fatigue.

Research shows that anxiety and depression frequently co-occur with misophonia and are known to be associated with the experience of fatigue (Wu et al., 2014, Guetta et al., 2022, Roy-Byrne et al., 2002). However, whether anxiety and depression contribute to the observed relationship between misophonia, and fatigue reported by Dibb and Golding (2022) has not been explored.

To sum up, there is a need to explore whether stress, hyperacusis, sleep quality, anxiety and depression influence the relationship between misophonia and fatigue. The aims of this study were: (1) to examine the relationship between misophonia and fatigue; and (2) to assess whether this relationship remains significant when accounting for other factors, such as stress, sleep disturbances, hyperacusis, and anxiety depression.

## 2. Materials and Methods

## 2.1. Participants

A total of 451 participants were recruited through academic institutions, social media platforms (Instagram, LinkedIn and X), and subscribers of Hashir International Institute in the UK. Additionally, the study link was shared in Facebook Support Groups and via email lists of nonprofit misophonia and hyperacusis organizations. To verify the adequacy of the sample size, the G\*Power 3.1 software (G\*Power, Düsseldorf, Germany) was used to calculate the minimum required participants. The calculation, based on a significance level of 0.05, a

medium effect size of 0.15, and a statistical power of 0.95, indicated a need for at least 199 valid responses (Faul et al., 2007). Ultimately, 451 responses were included in the data analysis, confirming that the sample size is more than sufficient for the study.

Participants were eligible for inclusion in the study if they were at least 18 years old and self-reported having a good understanding of English. Exclusion criteria included significant cognitive impairments or severe visual impairments, as communicated in the study invitation and information sheet.

#### 2.2. Ethical Considerations

The study received approval from the University of Surrey Research Integrity and Governance Office and was conducted in accordance with the principles of the Declaration of Helsinki. All participants received detailed information about the study's purpose and procedures, and online informed consent was obtained before they completed the questionnaire. Only those who explicitly agreed to participate were given the questionnaire. Participation in the study was entirely voluntary, and the data collected were used solely for research purposes. Participants were free to withdraw from the survey at any point. Completing the questionnaire took approximately 20 minutes.

#### 2.3. Procedure

This present study was conducted using the Qualtrics platform (Qualtrics, Provo, USA, <a href="https://www.qualtrics.com">https://www.qualtrics.com</a>). Once consent was obtained, participants shared demographic information, such as age, gender, education level, ethnicity, religion, and employment status. Following this, they completed a set of assessments, including MisoQuest, a Fatigue Questionnaire (3D-WFI), a Hyperacusis Impact Questionnaire (HIQ), Mental Health (SAD-T), one item assessing sleep quality and a stress measure. For a comprehensive understanding of these measures, please refer to Section 2.4.

#### 2.4. Measurements

Misophonia was assessed through the 14-item MisoQuest questionnaire (Siepsiak et al., 2020a). Each item is rated on a scale from 1 ("strongly disagree") to 5 ("strongly agree") with the total score ranging from 0 to 70. The MisoQuest was developed and validated in Polish by Siepsiak et al. (2020a). It has been found to have good validity and reliability among English speakers by Kula et al. (2024).

Three-dimensional fatigue inventory [3D-WFI; Frone and Tidwell (2015)] was used to assess the participant's level of fatigue. The 3D-WFI consists of 3 dimensions with 18 items that assess people's responses to emotional, mental, and physical fatigue aspects of their daily living. A 5-point Likert scale is used to assess each of the items with options ranging from 1 (every day) to 5 (never). The possible total scores range from 18 to 90. The 3D-WFI has good internal consistency (Cronbach's alpha coefficient was 0.94 for physical fatigue, .95 for mental fatigue, and .96 for emotional fatigue (Frone and Tidwell, 2015)).

Hyperacusis was assessed through the Hyperacusis Impact Questionnaire [HIQ; (Aazh et al., 2024a)] which consists of eight 4-point self-reported items designed to assess the impact of hyperacusis on individuals. Each item involves assigning a score of 0, 1, 2, or 3 based on response categories indicating 0 to 1 day, 2 to 6 days, 7 to 10 days, and 11 to 14 days, respectively. The cumulative score ranges from 0 to 24, with scores of twelve or more indicating a clinically significant impact of hyperacusis. The validity and reliability of the HIQ have been demonstrated in preceding studies (Aazh et al., 2024a).

Sleep quality was assessed by the one-item Sleep Quality Scale (Snyder et al., 2018a) for sleep where 0 indicated terrible sleep and 10 indicated excellent sleep quality. In terms of stress, a single-item measure of stress symptoms (Elo et al., 2003) was used to measure stress experience. The scores range from 1 to 5, where higher scores connect with increased levels of stress. Single-item measures are often used in the research for their simplicity and validity (Allen et al., 2022).

The Screening for Anxiety & Depression in Tinnitus/ Sound Intolerance [SAD-T; (Aazh et al., 2023)] is used to assess an individual's mental health according to four items. Specifically, two questions focus on anxiety and the remaining two address experiences related to depression. Each item is evaluated on a 4-point scale ranging from 0 to 3. The number of item's responses is summed to indicate the severity of anxiety and depression with scores of 4 or more indicating symptoms associated with anxiety and/or depression.

## 2.5. Data Analysis

The data were anonymised before statistical analysis. All data analyses were conducted using IBM SPSS software, version 28.0. Descriptive statistics, means, SDs, 95% confidence analyses (CIs) and score range for all questionnaires were reported. For the statistical significance, the p-value was defined as p < 0.05.

Assumptions for Statistical Tests: Before conducting analyses, assumptions for normality, homoscedasticity, and multicollinearity were checked. Normality was assessed using the Shapiro-Wilk test, and multicollinearity was evaluated through variance inflation factors (VIF). No significant violations were found.

The following analyses were conducted to explore the link between misophonia and fatigue, hyperacusis, sleep, stress, and mental health.

1. **Pearson correlation analysis:** This was used to examine the bivariate correlations between misophonia symptoms and fatigue, hyperacusis, sleep, stress, mental health, age and gender. Following Cohen (1992a), effect sizes were interpreted as follows: values around 0.10 indicating a small effect, around 0.30 indicating a moderate effect, and around 0.50 indicating a strong effect. While correlations help identify the strength and direction of relationships between variables, they do not provide the predictive power of these relationships or account for potential confounding variables (Dowdy et al., 2011). To address these limitations and explore whether certain factors can predict

fatigue while controlling for other variables, a regression analysis was conducted as the next step.

- 2. Multiple Linear Regression: An enter method multiple-linear regression model (Tranmer and Elliot, 2008) was developed to predict fatigue with all variables, namely MisoQuest, HIQ, one-item sleep and stress measures and SAD-T. This model was adjusted for age and gender.
- 3. Mediation analysis: This analysis was performed by using PROCESS function V4.2 in SPSS V.28. The model 4 (model as a parameter in the PROCESS function) was used applying the 5000 bootstrapping method (Hayes, 2017). An effect was considered statistically significant if the bootstrap confidence interval did not include "0" (Preacher and Hayes, 2008). Fatigue total scores (3D-WFI) were entered as the dependent variable, while MisoQuest score was the independent variable. Stress and sleep total scores were included as the mediators with age and gender entered as covariates. Hyperacusis (HIQ) and anxiety/depression (SAD-T) were not included as mediators because the multiple linear regression analysis showed no significant relationship between these variables and fatigue. Additionally, stress and sleep were prioritized in the mediation model, as they are theoretically more relevant in explaining how misophonia relates to fatigue.

## 3. Results

## 3.1. General Characteristics of Subjects

The study sample primarily consisted of women (60.3%), followed by men (37.5%) and a small percentage identifying as non-binary (2.0%) or preferring not to disclose their gender (0.2%). Most participants identified as heterosexual (84.9%), with smaller groups identifying as bisexual (8.4%), homosexual (2.7%), or other sexual orientations (2.0%). In terms of education, the majority had at least a bachelor's degree (36.6%), with a significant portion

holding a master's degree (25.7%) or higher. Ethnically, the largest group was White British (48.8%), with notable representations from White Other (20.8%). Over half of the participants were married or living with a partner (53.9%), while 26.8% were single. The majority were employed full-time (52.1%), and the average age of the participants was 34.61 years (SD = 11.61).

## 3.2. Misophonia, Hyperacusis, Sleep Quality, Stress and Mental Health

**Table 1.** Means and Standard deviations of total scores on the Misophonia questionnaire (MisoQuest), Screening for anxiety and depression (SAD-T), Hyperacusis Impact Questionnaire (HIQ), Tinnitus Impact Questionnaire (TIQ), a single item sleep quality scale. A single-item measure of stress measure, Fatigue questionnaire (3D-WFI) among the participants

Questionnaire	N	M	SD	Possible Range
MisoQuest	451	48.90	12.30	0-70
SAD-T (Mental health)	451	4.85	3.38	0-12
HIQ (Hyperacusis)	451	9.60	6.12	0-24
Sleep quality scale	451	5.45	2.10	0-10
Measure of stress symptoms	451	3.60	1.09	1-5
3D-WFI (Fatigue)	451	66.26	15.27	18-90

*Note.* The number of patients was indicated by N.

**Table 1** shows the means and SDs of the total score of the self-report questionnaires completed by the study participants. Based on the score on MisoQuest, 21.7% of participants (98 out of 451) were classified as having misophonia. Based on the score on HIQ, 35.5% of participants (160 out of 451) were classified as being clinically impacted by hyperacusis. Based on the self-reported sleep quality, 0.4% of participants reported terrible sleep, 18% reported

poor sleep, 45.9% reported fair sleep, 34.1% reported good sleep, and 1.6% reported excellent sleep quality. Furthermore, based on the score on the SAD-T, 61% of participants (275 out of 451) exhibited possible symptoms of anxiety and/or depression.

## 3.3 Correlation Between Variables

**Table 2**. The correlation matrix presents the relationships between misophonia (as measured by the MisoQuest), hyperacusis (as measured by the Hyperacusis Impact Questionnaire, HIQ), anxiety/depression (as measured by the Screening for Anxiety and Depression in Tinnitus, SAD-T), fatigue (as measured by the 3D Fatigue Inventory, 3D-WFI), sleep quality (as measured by a one-item sleep measure), and stress (as measured by a one-item stress measure)

	Misop	Hypera	Anxiety-	Fatigue	Sleep	Stress	Age	Gender
	honia	cusis	depression		Quality			
Misophonia	1.00	.34**	.23**	.32**	14**	.25**	.03	08
Hyperacusis	.34**	1.00	.59**	.19**	13**	.25**	05	01
Anxiety-	.23**	.59**	1.00	.21**	20**	.37**	13**	.01
depression								
Fatigue	.32**	.19**	.21**	1.00	43**	.37**	01	08
Sleep	14**	13**	20**	43**	1.00	41**	08	.09
Quality								
Stress	.25**	.25**	.37**	.37**	41**	1.00	09*	08
Age	.03	-0.05	13**	01	08	09*	1.00	10*
Gender	08	01	.01	08	.09	08	10*	1.00

*Note.* p < .05, p < .01. Correlations are significant at the .01 level (2-tailed) and .05 level (2-tailed). N= 451

In **Table 2**, the correlation analysis revealed several key relationships. Fatigue showed a stronger positive association with misophonia compared to hyperacusis. On the other hand, hyperacusis demonstrated a stronger relationship with anxiety and depression than misophonia. Both misophonia and hyperacusis were negatively associated with sleep quality, indicating that more severe symptoms were linked to poorer sleep. Additionally, stress was positively associated with both misophonia and hyperacusis and showed a negative relationship with sleep quality. Fatigue was also negatively associated with sleep quality, indicating that higher levels of fatigue were linked to poorer sleep, and positively associated with stress. These findings show the relationship between misophonia severity and various factors such as fatigue, hyperacusis, mental health, sleep quality, and stress.

# 3.4. Regression Analysis

**Table 3:** The multiple regression results predicting fatigue, which was measured using the 3D Fatigue Inventory (3D-WFI). Misophonia was assessed via the MisoQuest, hyperacusis via the Hyperacusis Impact Questionnaire (HIQ), anxiety and depression via the Screening for Anxiety and Depression in Tinnitus (SAD-T), sleep quality via a one-item sleep measure, and stress via a one-item stress measure.

Predictor	В	SE (B)	β	t	p
Misophonia	0.273	0.054	.221	5.019	<.001
Hyperacusis	0.048	0.129	.019	0.374	.708
Anxiety-depression	0.090	0.237	.020	0.380	.704
Sleep Quality	-2.394	0.325	331	-7.357	<.001
Stress	2.258	0.669	.162	3.377	<.001
Age	0.042	0.049	.035	0.847	.398
Gender	-0.531	1.147	019	-0.463	.643

Note.  $R^2$ = .284, Adjusted  $R^2$ = .272, F (7,439) = 24.817, p < .001. (N=451)

The multiple regression analysis was conducted to examine the predictors of fatigue, with the results summarised in Table 3. The overall model was significant, F(7,439) = 24.817, p < .001, accounting for approximately 28.4% of the variance in the fatigue scores ( $R^2 = .284$ , Adjusted  $R^2 = .272$ ). Misophonia severity, sleep quality, and stress emerged as significant predictors of fatigue. Higher misophonia severity and stress levels were associated with increased fatigue, while better sleep quality predicted lower fatigue. In contrast, within the model, hyperacusis severity, anxiety/depression, age, and gender were not significant predictors, indicating these factors do not have a strong independent effect on fatigue.

# 3.5. Mediation Analysis

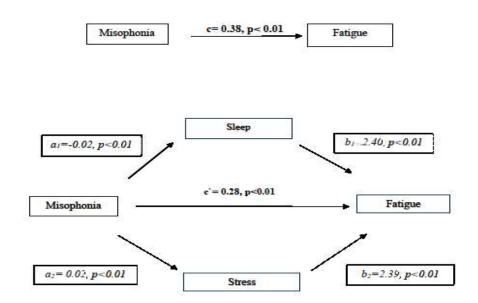
In this parallel mediation model (as shown in **Figure 1**):

- Path 1 (misophonia  $\rightarrow$  fatigue): The direct effect of misophonia on fatigue is represented by c' = 0.28, p < 0.01. This indicates that after accounting for the mediators (stress and sleep), misophonia still has a significant direct impact on fatigue.
- Path 2 (misophonia → stress → fatigue): The indirect effect through stress was statistically significant, with a path coefficient of a<sub>1</sub> = -0.02, p < 0.01 and b<sub>1</sub> = -2.40, p
   < 0.01. The indirect effect, calculated as a<sub>1</sub> x b<sub>1</sub> = 0.048, shows that stress negatively mediates the relationship between misophonia and fatigue.
- Path 3 (misophonia → sleep → fatigue): The indirect effect through sleep was also statistically significant, with a path coefficient of a<sub>2</sub> = 0.02, p < 0.01 and b<sub>2</sub> = 2.39, p
   < 0.01. The indirect effect through sleep is a<sub>2</sub> x b<sub>2</sub> = 0.0478, indicating that sleep quality positively mediates the relationship between misophonia and fatigue.

The **total effect** of misophonia on fatigue (before accounting for the mediators) was c = 0.38, p < 0.01. The fact that the direct effect remains significant even after including the mediators suggests that, while stress and sleep quality explain some of the variance, a substantial portion of the influence of misophonia on fatigue is direct and not accounted for by

these mediators. Moreover, it is important to note that while sleep and stress partially mediate the relationship between fatigue and misophonia, their indirect effects (0.048 and 0.0478, respectively) are relatively weak compared to the direct effect. Therefore, misophonia appears to have a more direct contribution to fatigue compared to the mediated influence of sleep and stress.

Figure 1: Parallel mediation analysis model for fatigue and misophonia by sleep and stress mediators (number of participants=451).



#### 4. Discussion

This study investigated the relationship between misophonia and fatigue, considering the mediating effects of stress and sleep quality. The results indicate that misophonia is a strongly associated with fatigue, even when the effects of hyperacusis, anxiety, depression, sleep quality, and stress are taken into consideration. Although sleep quality and stress were found to partially mediate this relationship, misophonia continued to exhibit a strong and significant direct effect on fatigue.

Dibb and Golding (2022) similarly found that people with misophonia experience higher levels of fatigue. Our study extends these findings by showing that the link between misophonia and fatigue remains strong even after accounting for other factors like stress, sleep problems, and mental health conditions such as anxiety and depression.

Our results showed a positive, moderate relationship between misophonia as measured by MisoQuest (Kula et al., 2024) and hyperacusis, as measured by HIQ (Aazh et al., 2024a). This finding is consistent with previous studies, such as Jager et al. (2020) and Brennan et al. (2024), which also reported an association between these two conditions.

We found a positive moderate correlation between anxiety and depression symptoms in individuals with misophonia, which aligns with previous research (Wu et al., 2014, Aazh et al., 2023, Siepsiak et al., 2020b). However, Quek et al. (2018) did not find a significant correlation between depression and misophonia in a sample of psychiatric patients, suggesting that the relationship may vary depending on the sample or methodology used. Interestingly, we observed a stronger correlation between hyperacusis (measured via the HIQ) and symptoms of anxiety/depression (as measured by SAD-T) compared to the correlation between misophonia (measured via MisoQuest) and these mental health symptoms. To our knowledge, no previous studies have directly compared the strength of these relationships. Additionally, a longitudinal study by Dibb and Golding (2022) found anger and disgust were the primary predictors of

misophonic responses, while anxiety and depression were not significantly associated with misophonia over time. This supports our observation that hyperacusis may provoke greater anxiety and depression, whereas misophonia may be more strongly linked with emotions such as anger and disgust. These findings point to the need for further research to understand the complex relationship between misophonia, hyperacusis and mental health conditions.

The findings of this study have significant implications for clinical practice, particularly for healthcare providers working with patients who have misophonia. Given the strong association between misophonia and fatigue, it is crucial that clinicians assess not only the symptoms of misophonia but also related factors such as fatigue, sleep quality, and stress levels. Interventions that address these issues could be particularly beneficial. Future research should explore whether improving stress and sleep quality can reduce the experience of fatigue and symptoms of misophonia.

Research has demonstrated that Cognitive-behavioural therapy (CBT) is effective in reducing the effect of misophonia symptoms (Jager et al., 2021, Gregory and Foster, 2023, Aazh et al., 2024c). Including strategies to address sleep disturbances and stress within CBT could further enhance its effectiveness in improving fatigue associated with misophonia. By integrating these evidence-based treatments, clinicians can more comprehensively address both the direct and indirect contributors to fatigue in individuals with misophonia and improve patients' overall quality of life.

In the regression analysis, which controls for other predictor variables within the model, age did not have an independent effect on fatigue. This contrasts with previous studies that generally suggest fatigue increases with age due to factors such as reduced physical function and chronic health issues. Research has shown that older adults often report higher fatigue levels related to mobility limitations and physical impairment (Matsumoto et al., 2024, Murphy and Niemiec, 2014). Other studies highlight that sleep disturbances, which become more

common with age, contribute to higher fatigue levels in older populations (Åkerstedt et al., 2018). The difference in our findings could be due to sample characteristics or indicate other lifestyle factors.

#### 4.2. Limitations and Future Directions

Although this study found some novel findings regarding the association between misophonia and fatigue, some caveats need to be considered. The cross-sectional design, which captures data at a single point in time, does not allow us to determine cause and effect. Future research should use longitudinal studies to better understand how misophonia, fatigue, sleep quality, and stress influence each other over time. Additionally, our sample was mostly female and from the UK, which may limit the generalizability of the findings to other populations. Future studies should include more diverse groups.

The generalizability of our findings may also be limited because the study did not focus on individuals with more severe misophonia symptoms or those with comorbid mental health conditions such as clinically diagnosed severe anxiety or depression. Future research should examine these subgroups to determine if the observed relationships remain consistent across different clinical populations.

## 4.5 Conclusion

This study demonstrated a relationship between misophonia and fatigue, with both factors influencing each other. The relationship was partially mediated by sleep quality and stress. While misophonia was moderately correlated with anxiety and depression, hyperacusis showed a strong correlation with these mental health factors, suggesting that hyperacusis may have a more direct link to anxiety and depression compared to misophonia. Despite demonstrating mediation, the direct link between misophonia and fatigue remained strong in both analyses.

Healthcare providers should consider these factors when supporting patients with misophonia, focusing on interventions that help manage not only the symptoms of misophonia

but also related issues such as poor sleep, high stress, hyperacusis and anxiety, and depression. Future research is needed to explore additional factors that may contribute to fatigue in

individuals with misophonia and to develop better targeted management strategies.

# **Data Availability Statement**

The datasets generated for this study are available on request to the corresponding author.

## **Conflict of Interest**

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

# **Author Contributions**

FBK: Conceptualization, Methodology, Data collection and analysis, Writing – original draft, Review & editing. MC and HA: Conceptualization, Supervision, Review & editing. All authors contributed significantly to the work and approved the final manuscript for publication.

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# **CHAPTER FIVE**

Study 4: Evaluating the 4C Misophonia and Hyperacusis Management

Questionnaires: Psychometric Validation in a Clinical Population

#### 5.1 Overview

This final empirical chapter builds on the systematic review in Chapter Two, which highlighted the need to validate psychometric tools in clinical populations for hyperacusis and misophonia. This chapter focuses on the evaluation of the 4C Hyperacusis and Misophonia Management Questionnaires, developed by my second supervisor and their team. These questionnaires were designed to assess patients' confidence in managing their symptoms without relying on avoidance behaviours, a crucial component of treatment planning, particularly in Cognitive Behavioral Therapy (CBT).

The 4C questionnaires were validated using data collected from patients clinically diagnosed with hyperacusis and misophonia at my second supervisor's clinic. This targeted validation ensures that the questionnaires are applicable to clinical practice and address a gap in existing instruments, which often examine symptom management strategies. By assessing patients' confidence in handling their symptoms, the 4C questionnaires provide clinicians with valuable tools for creating personalised and effective interventions.

# 5.2 Research Paper

# **Evaluating the 4C Misophonia and Hyperacusis Management**Questionnaires: Psychometric Validation in a Clinical Population

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#### **Abstract**

**Aim:** To evaluate the psychometric properties of the 4C Hyperacusis and Misophonia Management Questionnaires, designed to assess patients' confidence in managing symptoms of hyperacusis and misophonia, respectively without reliance on Safety Seeking Behaviours (SSBs).

Methods: Data were collected from a clinical sample of adult patients seeking help from a specialist audiology centre in the UK for distressing hyperacusis and/or misophonia, with 48 completing the 4C Hyperacusis Management Questionnaire and 40 completing the 4C Misophonia Management Questionnaire. Confirmatory factor analysis (CFA) was used to examine structural validity, while convergent and discriminant validity were assessed through correlations with these measures: Hyperacusis Impact Questionnaire (HIQ), Misophonia Impact Questionnaire (MIQ), Tinnitus Impact Questionnaire (TIQ), and the Screening for Anxiety and Depression in Tinnitus/Sound Intolerance (SADT). Reliability was evaluated using Cronbach's α and McDonald's ω.

**Results:** CFA indicated an acceptable model fit for the one-factor structure of the 4C Hyperacusis Management Questionnaire ( $\chi^2/df = 2.23$ , CFI = 0.97, TLI = 0.92, SRMR= 0.03, RMSEA = 0.16). For the 4C Misophonia Management Questionnaire, a two-factor structure provided a better acceptable model fit ( $\chi^2/df = 1.51$ , CFI = 0.99, TLI = 0.96, SRMR= 0.02, RMSEA = 0.11). Both questionnaires demonstrated high reliability, with Cronbach's  $\alpha$  values of 0.90 for the 4C Hyperacusis Questionnaire and 0.87 for the 4C Misophonia Questionnaire. Convergent validity was supported by significant negative correlations between the 4C Hyperacusis scores and HIQ ( $\rho$  = -0.41,  $\rho$  < .05) and between the 4C Misophonia scores and MIQ ( $\rho$  = -0.56,  $\rho$  < .05). Discriminant validity was indicated by low correlations with unrelated constructs, although the 4C Misophonia Questionnaire showed a higher correlation with

anxiety and depression ( $\rho$  = -0.46, p < .05) compared to hyperacusis scores. Age and gender were not significantly correlated with either questionnaire.

**Conclusion:** 4C Hyperacusis and Misophonia Management Questionnaires demonstrated good reliability and validity, supporting their use in assessing patient's confidence in symptom management, particularly within the context of Cognitive Behavioral Therapy (CBT).

#### 1. Introduction

Hyperacusis and misophonia are both classified as disorders of Decreased Sound Tolerance (DST) that negatively affects quality of life (Jastreboff and Jastreboff, 2015). Hyperacusis is characterised by an increased sensitivity to everyday sounds, such as household or environmental noises, perceived as excessively loud or painful, leading to significant distress and interference with daily life, including social, occupational, and recreational activities (Aazh et al., 2016, Aazh et al., 2024b). People with hyperacusis experience negative reactions to sounds based on their physical aspects, such as volume or frequency, often perceiving ordinary noises as intolerably loud (Salvi et al., 2022). Although there is no consensus on precise definitions, Tyler et al. (2014) proposed four types of hyperacusis are generally recognised in the literature: loudness hyperacusis, where every day sounds seem excessively loud; annoyance hyperacusis, which produces negative emotional reactions to sounds; fear hyperacusis, where sounds provoke avoidance behaviours due to fear; and pain hyperacusis, where sounds cause physical pain in the ears or surrounding areas like the face and neck. Prevalence estimates indicate that 15.2% of adults aged 45 to 70 experience symptoms of hyperacusis (Smit et al., 2021a).

Misophonia involves intense negative emotional, behavioural, and physical reactions to specific sounds (such as chewing or tapping) or stimuli, and is characterised by a reduced tolerance to these sounds, which may include oral, nasal, non-oral/ nasal sounds from people, as well as sounds produced by objects or animals (Swedo et al., 2022). The prevalence of individuals in the UK for whom misophonia symptoms cause a significant burden in their lives has been estimated at 18% (Vitoratou et al., 2023).

Existing hyperacusis and misophonia questionnaires, such as the Duke Misophonia Questionnaire (Rosenthal et al., 2021), MisoQuest (Siepsiak et al., 2020a), and Hyperacusis Impact Questionnaire (Aazh et al., 2024a) often include items with language reflecting

struggle, anger, and negativity. For example, the DMQ includes items such as "I hate being like this," and "I thought about physically hurting the person making the sound," illustrating both negativity and anger in response to trigger sounds. MisoQuest further emphasises anger and struggle with items like "I start feeling anger the moment I see a thing/animal/person that might make an unpleasant sound" and "I believe that my reactions to sounds are exaggerated, but I can't get rid of them". Similarly, the HIQ has items such as "Feeling anxious when hearing loud noises" and "difficulty in carrying out certain day-to-day activities/ tasks in noisy places", framing the condition as a source of continuous struggle. By contrast, the 4C Hyperacusis and Misophonia Management Questionnaires use language that promotes acceptance, ability, and confidence. Rather than focusing on challenges caused by symptoms, these questionnaires include items such as "How confident are you that you can enjoy your life fully despite your hyperacusis or misophonia?" and "How confident are you that you can do all of the above without using any avoidance behaviour?" This constructive approach encourages individuals to focus on their strengths and their ability to manage their condition.

Patients with hyperacusis and misophonia often do not prioritise therapies like cognitive behavioural therapy (CBT) as their first option for managing their condition, despite evidence supporting its effectiveness by Jager et al. (2021). Instead, many initially prefer medical solutions, hoping for a definitive cure. When no medical solution is available, they frequently rely on avoidance and ritualistic behaviours to cope. These behaviours might include overusing ear protection, avoiding social situations, leading clear of specific environments or individuals, or instructing others to reduce noise. While these strategies may offer temporary relief, they can limit long-term adaptation. Shifting from these avoidance behaviours to engaging in therapy often requires a significant adjustment. A key obstacle is patients' lack of confidence in their ability to replace these familiar coping mechanisms with new approaches. For instance, a person with misophonia may feel, "I cannot control my anger even with CBT; it is easier to

avoid shared mealtimes," while someone with hyperacusis may think, "Avoiding loud sounds helps me avoid pain, but I doubt CBT can provide the same relief." Addressing this confidence gap is essential to help patients engage in therapy (Aazh, 2024). Therefore, tools that foster confidence and motivation are essential for facilitating this transition. Building on similar challenges identified in tinnitus management, the 4C Tinnitus Management Questionnaire was developed by Aazh et al. (2024d) to measure patients' confidence in managing tinnitus within a CBT framework. Its psychometric evaluation demonstrated excellent reliability (Cronbach's alpha = 0.91) and validity, establishing it as a robust tool to assess confidence. Following this approach, the 4C Hyperacusis and Misophonia Management Questionnaires were developed at the Hashir Research Institute for Misophonia, Tinnitus and Hyperacusis in the UK. These questionnaires aim to assess patients' confidence in managing their symptoms and to support audiologists and clinicians in treatment planning, particularly in the context of CBT.

The 4C Hyperacusis and Misophonia Management Questionnaires were designed with two main goals. Firstly, it uses language that encourages patients to focus on their ability to manage the challenges of sound sensitivity. This approach is based on principles from motivational interviewing and self-determination theory, which suggest that fostering a sense of autonomy and capability can increase motivation and engagement in therapy (Markland et al., 2005). For example, the 4C Hyperacusis Management Questionnaire includes items such as, "How confident are you that you are able to carry out your day-to-day tasks despite your hyperacusis?", Similarly, the 4C Misophonia Management Questionnaires include items like "How confident are you that you are able to socialise and relax even when you hear the trigger noises?" By prompting patients to reflect on their confidence in coping with their symptoms, the 4C aims to strengthen their intrinsic motivation to participate in therapy and to explore ways to lead completing exists despite hyperacusis and misophonia. Secondly, the 4C differentiates between patients who are confident in managing their symptoms without relying

on avoidance behaviours and those who may use avoidance as a coping strategy. For example, the 4C Hyperacusis Management Questionnaire includes items such as, "How confident are you that you can do all of the above without using any avoidance behaviour (e.g., ear protection, avoiding certain situations)?" Similarly, the 4C Misophonia Management Questionnaire includes items like, "How confident are you that you can do all of the above without using any avoidance behaviour (e.g., use of background noise, ear protection, telling people to stop the noise, mimicry, or avoiding certain situations or individuals)?". This distinction is important for tailoring interventions that address specific coping methods and encourage more adaptive responses to sound triggers (Britton et al., 2011).

The aim of this study was to evaluate the psychometric properties of the 4C Hyperacusis and Misophonia Management Questionnaires by examining their construct validity, reliability, and structural validity, including both convergent and discriminant validity.

## 2. Methods

## 2.1. Ethical Approval

This study followed ethical guidelines to respect participant rights and maintain confidentiality. Ethical approval was granted by the University of Surrey Ethics Committee (Project ID: FHMS 21-22-147 EGA). The clinical data used in this research were collected from the Hashir International Specialist Clinics & Research Institute for Misophonia, Tinnitus and Hyperacusis.

## 2.2.Study Design and Questionnaires

## Study Design

This study examined clinical records from 285 patients who attended the Hashir International Institute for Misophonia, Tinnitus, and Hyperacusis between 2021 and 2024. The goal was to evaluate the reliability and validity of the 4C Misophonia and Hyperacusis Questionnaires for adults.

## 4C Hyperacusis and Misophonia Management Questionnaires

The 4C questionnaires measure how confident patients feel about managing their hyperacusis and misophonia symptoms. Each question in the 4C questionnaires is scored from 0 to 10, where 0 means "Not confident at all" and 10 means "Very confident." Question 4 specifically addresses patients' confidence in managing symptoms without using avoidance behaviours (such as avoiding certain situations or using background noise), which reflects a higher level of coping. As avoidance behaviours are often linked to less adaptive management strategies, the inclusion and weighting of Question 4 highlight its importance in evaluating patients' overall confidence.

To calculate the total score, the sum of Questions 1 to 3 is multiplied by the score for Question 4, and the result is divided by 3. This scoring method was designed to emphasize the value of managing symptoms without reliance on avoidance strategies, as higher scores on Question 4 indicate more adaptive and self-reliant coping. The total score ranges from 0 to 100, where 0 represents no confidence and 100 represents full confidence in managing symptoms actively. This approach aligns with the theoretical framework of self-determination theory, which suggests that autonomous and confident management is essential for long-term success (Deci and Ryan, 2013). Patients who rely heavily on avoidance strategies typically score lower, reflecting less confidence in managing symptoms effectively (See Tables 1 and 2 for detailed items of these questionnaires)

**Table 1: 4C Hyperacusis Management Questionnaire** 

Instructions: Please complete based on your confidence level. Circle the number that best represents the confidence level on a scale of 0–10, where 0 means "Not confident at all" and 10 means "Very confident."

Adult Version: Patient Self-Assessment	Rating Scale (0–10)
1. How confident are you that you are able to carry out your day-	0 1 2 3 4 5 6 7 8 9 10
to-day tasks despite your hyperacusis?	

2. How confident are you that you can socialise and relax despite	0 1 2 3 4 5 6 7 8 9 10
your hyperacusis?	
3. How confident are you that you can enjoy your life fully despite	012345678910
your hyperacusis?	
4. How confident are you that you can do all of the above without	012345678910
using any avoidance behaviour (e.g., ear protection, avoiding	
certain situations)?	

# **Table 2: 4C Misophonia Management Questionnaire**

Instructions: Please complete based on your confidence level. Circle the number that best represents the confidence level on a scale of 0–10, where 0 means "Not confident at all" and 10 means "Very confident."

Adult Version: Patient Self-Assessment	Rating Scale (0–10)
1. How confident are you that you are able to carry out your day-to-	0 1 2 3 4 5 6 7 8 9 10
day tasks even when you hear the trigger noises?	
2. How confident are you that you are able to socialise and relax	012345678910
even when you hear the trigger noises?	
3. How confident are you that you can enjoy your life fully despite	012345678910
your misophonia?	
4. How confident are you that you can do all of the above without	0 1 2 3 4 5 6 7 8 9 10
using any avoidance behaviour (e.g., use of background noise, ear	
protection, telling people to stop the noise, mimicry, or avoiding	
certain situations or individuals)?	

# **Convergent and Discriminant Validity:**

The following questionnaires, routinely administered during the initial assessment of patients at the Hashir International Institute, were used to validate the 4C questionnaires.

# 1. Misophonia Impact Questionnaire [MIQ: Aazh et al. (2023)]

This questionnaire was used to assess the impact of misophonia on the patients. MIQ has eight items, and each item is rated on a scale from 0 to 3, corresponding to response options

for frequency: 0 to 1 day, 2 to 6 days, 7 to 10 days, and 11 to 14 days, respectively. Total scores range from 0 to 24, with scores of 12 or higher indicating a clinically significant impact from misophonia (Erfanian and Aazh, 2023). The MIQ has demonstrated high reliability, with a Cronbach's alpha of 0.94 (Aazh et al., 2023).

# 2. Hyperacusis Impact Questionnaire [HIQ, (Aazh et al., 2024a)]

The HIQ consists of eight items that evaluate the impact of hyperacusis on patients. Responses for each item are scored as 0, 1, 2, or 3, corresponding to the frequency categories of 0 to 1 day, 2 to 6 days, 7 to 10 days, and 11 to 14 days, respectively. The total score can range from 0 to 24, with scores of 12 or higher indicating a clinically significant level of impact from hyperacusis and in a clinical sample, the HIQ demonstrated strong reliability, with a Cronbach's alpha of 0.93 (Aazh et al., 2024a).

# 3. Tinnitus Impact Questionnaire [TIQ, Aazh et al. (2022a)]

TIQ consists of seven items that designed to evaluate how tinnitus affects a patient's daily activities, mood, and sleep. Each item is rated on a scale from 0 to 3, representing the frequency of impact over a two-week period: 0–1 day, 2–6 days, 7–10 days, and 11–14 days, respectively. Total scores range from 0 to 21, with higher scores indicating greater impact. The TIQ has strong internal consistency, with a Cronbach's α of 0.89 (Aazh et al., 2022b). For interpretation, scores below 5 indicate no impact, scores of 5–6 reflect mild impact, 7–8 suggest moderate impact, and scores above 9 indicate severe tinnitus impact. This questionnaire employed to assess discriminant validity of 4C misophonia management and hyperacusis management Questionnaires.

4. The Screening for Anxiety and Depression in Tinnitus/ Sound Intolerance [SAD-T, (Aazh et al., 2024a)]

The SAD-T questionnaire includes four items assessing mental health: two items measure anxiety symptoms, and two address depression-related experiences. Each item is scored on a

4-point scale from 0 to 3, based on frequency (0 to 1 day, 2 to 6 days, 7 to 10 days, and 11 to 14 days). Scores are summed, with a total of 4 or more indicating symptoms of anxiety and/or depression (Aazh et al., 2024a).

# Diagnosis of Misophonia and Hyperacusis

Audiologists at the Hashir International Institute conducted clinical interviews to diagnose patients with misophonia and hyperacusis. For a misophonia diagnosis, patients had to show at least one of the first four characteristics listed in Table 3, along with all characteristics from 5 to 9. A hyperacusis diagnosis required at least one of the first four characteristics listed in Table 4, along with all characteristics from 5 to 9.

# Table 3: Misophonia Diagnostic Criteria

- 1. Sound intolerance adversely affects day-to-day activities and tasks.
- 2. Sound intolerance adversely affects mood.
- 3. Sound intolerance adversely affects resting or relaxing.
- 4. The person uses avoidance behaviours or rituals in order to cope with the sounds that bother them.
- 5. The person's main complaint is decreased tolerance to oral and nasal sounds (e.g., eating, chewing gum, lip smacking, mouth noises, breathing) or repetitive sounds, including repetitive tapping or rustling sounds made by humans or machines (e.g., finger tapping).
- 6. The person perceives specific sounds as disgusting, offensive, or annoying.
- 7. The person's primary reaction to sounds is not aural pain or physical discomfort in their ears due to excessive loudness of the sounds.
- 8. The person's sound intolerance is not better explained by their general attitude toward noise and environmental noise pollution (e.g., noise from neighbours, nearby airports, traffic, workshops, plumbing, or air conditioning).
- 9. The symptoms are not better explained by other underlying medical or psycho-social conditions.

*Note:* Misophonia was diagnosed if the person showed at least one of the first four characteristics listed above in addition to characteristics 5-9 (i.e., (item 1 or 2 or 3 or 4) + item 5 + item 6 + item 7 + item 8 + item 9)

# **Table 4: Hyperacusis Diagnostic Criteria**

- 1. Sound intolerance adversely affects day-to-day activities and tasks.
- 2. Sound intolerance adversely affects mood.
- 3. Sound intolerance adversely affects resting and relaxing.
- 4. The person uses avoidance behaviors or rituals in order to cope with the sounds that bother them.
- 5. The person perceives some sounds as too loud, uncomfortable, or painful.
- 6. The person's main complaint is not decreased tolerance to oral and nasal sounds (e.g., eating, chewing gum, lip smacking, mouth noises, breathing) or repetitive sounds, including repetitive tapping or rustling sounds made by humans or machines (e.g., finger tapping).
- 7. The person's sound intolerance is not better explained by their general attitude toward noise and environmental noise pollution (e.g., noise from neighbors, nearby airports, traffic, workshops, plumbing, or air conditioning).
- 8. If the person uses hearing aids, their sound intolerance is experienced even when the hearing aids are not being used.
- 9. The symptoms are not better explained by other underlying medical or psycho-social conditions.

*Note:* Hyperacusis was diagnosed if a person showed at least one of the first four characteristics listed above in addition to characteristics 5-9 (i.e., (item 1 or 2 or 3 or 4) + item 5 + item 6 + item 7 + item 8 + item 9).

#### 2.4. Data Analysis

Before analysing the data, all records were anonymised. IBM SPSS (version 28.0) was used for all analyses except for confirmatory factor analysis (CFA), which was performed in R studio (version 2024.09.0) using the Lavaan package with the WLMSV (the weighted least square mean and variance) estimator for this specially designed for ordinal data (Rosseel,

2012). Descriptive statistics were calculated for all questionnaires, including means, standard deviations, and ranges. The normality of each measure was assessed visually using histograms and with the Shapiro-Wilk test to confirm that the data met the necessary assumptions for parametric analyses. In all analyses, the significance threshold was set at  $\alpha = 0.05$  unless specified otherwise. Correlations were interpreted according to Cohen's (1988) guidelines: correlation values of (0.10), (0.30), and (0.50) were classified as weak, moderate, and strong, respectively (Cohen, 1992).

The non-parametric Mann-Whitney U test was conducted to compare total scores between male and female participants. This test was chosen due to the non-normal distribution of total scores of the included questionnaires. Additionally, the correlation between age and total 4C Hyperacusis and Misophonia scores was evaluated using Spearman's correlation coefficient (Spearman's rho),

# Confirmatory Factor Analyses

CFA was conducted using the WLSMV estimator to assess the structure of the English versions of the 4C Hyperacusis and Misophonia Management Questionnaires. A one-factor model fit was hypothesised for each questionnaire, consistent with the structure of the 4C Tinnitus Management Questionnaire, which previously demonstrated a one-factor structure (Aazh et al., 2024b). Fit indices were used to check how well the data fit the hypothesised model. The Root Mean Square Error of Approximation (RMSEA) was used, where values of 0.05 or less indicate a good fit and values between 0.05 and 0.08 are acceptable The Standardised Root Mean Square Residual (SRMR) was also used, with values of 0.05 or less indicating a good fit. Other indices included the Tucker Lewis Index (TLI) and Comparative Fit Index (CFI), both of which need to be 0.97 or higher to indicate good fit. The Goodness of Fit Index (GFI) was also included, with values of 0.95 or higher suggesting strong model fit. The Chi-square/df ratio (CMIN) was used as well, with a threshold of 2 or less indicating good

fit. These fit criteria follow recommendations by Schermelleh-Engel et al. (2003), who advise that SRMR  $\leq$  0.05, RMSEA  $\leq$  0.05, TLI  $\geq$  0.97, CFI  $\geq$  0.97, GFI  $\geq$  0.95, and  $\chi^2/df \leq$  2 indicate a well model fit.

#### Reliability

McDonald's  $\omega$  and Cronbach's  $\alpha$  were used to evaluate the overall reliability of the 4C Hyperacusis and Misophonia Management Questionnaires for the adult sample, with 95% confidence intervals calculated through bias-corrected and accelerated (BCA) bootstrapping based on 1,000 samples. Cronbach's  $\alpha$  greater than 0.7 are considered indicative of acceptable internal consistency (Taber, 2018).

# Convergent and Discriminant Validity

We assessed the convergent and discriminant validity of the 4C Hyperacusis and Misophonia Management Questionnaires by examining their associations with related and unrelated measures. For the 4C Hyperacusis Management Questionnaire, convergent validity was tested against the Hyperacusis Impact Questionnaire (HIQ), where moderate to high correlations were expected, indicating that both measures address hyperacusis-related aspects. Discriminant validity was assessed using the Screening for Anxiety and Depression in Tinnitus/Sound Sensitivity (SADT), the Tinnitus Impact Questionnaire (TIQ), and the Misophonia Impact Questionnaire (MIQ). Low correlations with these measures were expected, suggesting that the 4C Hyperacusis Management Questionnaire does not overlap with unrelated areas.

For the 4C Misophonia Management Questionnaire, convergent validity was tested using the Misophonia Impact Questionnaire (MIQ), expecting moderate to high correlations as both questionnaires focus on misophonia. Discriminant validity was assessed using SADT, TIQ, and HIQ, with low correlations anticipated, showing that the 4C Misophonia Management Questionnaire is specific to misophonia and does not overlap strongly with other areas.

Spearman's correlation coefficient (Spearman's rho) was used to evaluate the convergent and discriminant validity of both the 4C Hyperacusis and Misophonia questionnaires, using the total scores of each questionnaire to assess structural validity.

#### 3. Results

# 3.1.Sample Characteristics

A total of 285 patients' clinic records were included in the study; however, there were only 40 completed 4C Misophonia Management Questionnaires from adults. This reduction occurred because the 4C Misophonia Management Questionnaire was administered exclusively to patients who reported clinically significant misophonia symptoms during their assessment, limiting the eligible sample size. The average age of these 40 patients was 33.83 years (SD = 14.06), with an age range from 17 to 68 years. The sample was composed of 60% female (n = 24) and 40% male (n = 16) participants. According to diagnostic criteria described in the methods section and Tables 1-2, 33 of the 40 patients (82.5%) were diagnosed with misophonia. Among those diagnosed, 11 patients (27.5%) had both hyperacusis and misophonia, 1 patient (2.5%) had both misophonia and noise sensitivity, and 1 patient (2.5%) had a combination of tinnitus, hyperacusis, misophonia, and noise sensitivity.

Out of a total of 285 patients, 48 adults completed the 4C Hyperacusis Management Questionnaire (this drop due to the questionnaire being administrated only patients reported hyperacusis symptoms). The average age of these patients was 40.92 years (SD = 14.3), with 54.2% male (n = 26) and 45.8% female (n = 22). Among them, 6 patients (12.5%) were diagnosed with only hyperacusis, while 22 patients (45.8%) had both tinnitus and hyperacusis symptoms. Additionally, 8 patients (16.7%) had both hyperacusis and misophonia, 1 patient (2.1%) presented with hyperacusis, misophonia, and noise sensitivity, 3 patients (6.3%) had tinnitus, hyperacusis, and noise sensitivity, and 1 patient (2.1%) exhibited a combination of tinnitus, hyperacusis, misophonia, and noise sensitivity.

The Mann-Whitney U test indicated no statistically significant difference between male and female participants on either the 4C Hyperacusis Management Questionnaire or the 4C Misophonia Management Questionnaire.

Additionally, Spearman's correlation analysis found no significant relationship between age and total scores on either questionnaire. For the 4C Hyperacusis Management Questionnaire, age was not significantly correlated with total scores,  $\rho = .22$ , p = .127, and gender showed a weak, non-significant association with scores,  $\rho = -.27$ , p = .067. For the 4C Misophonia Questionnaire, there was no significant correlation between age and total scores,  $\rho = -.17$ , p = .285, or between gender and scores,  $\rho = -.12$ , p = .445. These findings suggest that age and gender do not significantly impact total scores for either the 4C Hyperacusis or the 4C Misophonia Questionnaire in this sample, although there is a small, non-significant pattern of higher scores in males.

# 3.2.Psychometric Analyses

Table 5 presents the descriptive statistics for the items in the 4C Hyperacusis and Misophonia Management Questionnaires. The total scores for both questionnaires exhibit non-normal distributions, confirmed through visual inspection (histograms and Q-Q plots) and statistical measures. Despite this, the factor loadings for individual items remain good. For the 4C Hyperacusis items, factor loadings range from 0.75 to 0.94, while the 4C Misophonia items range from 0.62 to 0.92.

**Table 5:** Descriptive Statistics and Factor Loadings of the 4C Hyperacusis and Misophonia Management Questionnaires.

Questionnaire	Item	Mean (SD)	Min	Max	Skewness	Kurtosis	Factor
							Loading
							$(\lambda_{\rm std})$ -
							One
							Factor
							Model

		T			1		1
4C	Item 1	5.00 (2.97)	0	10	19	87	0.75
Hyperacusis							
Management	Item 2	3.82 (2.86)	0	10	.56	51	0.84
Questionnaire							
	Item 3	2.94 (2.90)	0	10	.68	67	0.94
	Item 4	2.59 (2.71)	0	9	.88	22	0.85
	Total	7.93 (3.53)	0	81	1.65	1.90	
	(N=48)						
4C	Item 1	3.49 (2.71)	0	10	.76	.01	0.62
Misophonia							
Management	Item 2	2.90 (2.70)	0	10	.85	.04	0.92
Questionnaire							
	Item 3	3.53 (2.708)	0	10	.34	64	0.72
	Item 4	1.80 (2.05)	0	9	1.49	2.88	0.91
	Total	9.44 (15.71)					
	(N=40)						

*Note.* N = Number of patients

# 3.2.1. Confirmatory Factor Analysis

The results of the confirmatory factor analysis (CFA) for the **4C Hyperacusis Management Questionnaire**, show an acceptable model fit with one factor structure based on the criteria by Schermelleh-Engel et al. (2003). The relative chi-square ( $\chi^2/df = 2.23$ ) is slightly above the threshold of 2, indicating acceptable. The CFI (0.97) meets the recommended threshold, supporting an acceptable fit, while the TLI (0.92) falls below the ideal cut-off of 0.97. The SRMR value of 0.029 is well within the acceptable range ( $\leq 0.05$ ), suggesting minimal residual error. However, the RMSEA value of 0.16, with a 90% confidence interval of 0.00 to 0.34, exceeds the recommended threshold of 0.05.

Initially, a one-factor structure was tested for the **4C Misophonia Management Questionnaire.** While some fit indices indicated reasonable fit, others suggested potential misspecifications. The CFI (0.91) and TLI (0.72) were below the recommended thresholds,

and the relative chi-square ( $\chi^2/df = 4.90$ ) exceeded the acceptable cut-off of 2, indicating a poor fit. Additionally, the RMSEA value of 0.32, with a 90% confidence interval of 0.16 to 0.49, was significantly above the ideal threshold of  $\leq 0.05$ , suggesting misfit in the model. The SRMR (0.086) was also slightly above the acceptable cut-off of 0.08. To address these issues, a two-factor structure was tested, and this provided a much better fit to the data. The CFI (0.99) and TLI (0.96) exceeded the recommended thresholds, and the relative chi-square ( $\chi^2/df = 1.51$ ) was within an acceptable range. Although the RMSEA value (0.11) remained above the preferred threshold, the SRMR (0.018) indicated good fit.

# 3.2.2. Reliability

The reliability of these measures was found to be excellent, with Cronbach's Alpha and Omega values of 0.90 (95% CI [0.87–0.93]) for the 4C Hyperacusis 0.87 (95% CI [0.83–0.91]) for the 4C Misophonia Management, respectively.

### 3.2.3. Construct Validity

For the **4C Hyperacusis Management Questionnaire**, convergent validity was evaluated by examining its correlation with the Hyperacusis Impact Questionnaire (HIQ). As shown in **Table 6**, a significant negative moderate correlation with the HIQ ( $\rho$  = -0.41, p < .05) suggests that as hyperacusis management improves (higher scores on the 4C), the impact of hyperacusis (lower HIQ scores) decreases. This indicates that both measures capture similar aspects of hyperacusis, supporting convergent validity for the 4C Hyperacusis Management Questionnaire.

Discriminant validity was assessed through correlations with the Tinnitus Impact Questionnaire (TIQ), Misophonia Impact Questionnaire (MIQ), and Screening for Anxiety and Depression in Tinnitus (SAD-T). The low and non-significant correlations with these measures (e.g.,  $\rho = -0.08$  with TIQ,  $\rho = 0.15$  with MIQ, and  $\rho = -0.26$  with SAD-T) indicate that the 4C

Hyperacusis Questionnaire does not strongly relate to distinct constructs, such as tinnitus, misophonia, or anxiety and depression, supporting its discriminant validity.

**Table 6:** Correlations among items and total scores of 4C Hyperacusis Management Questionnaire, HIQ, TIQ, MIQ, and SADT.

	HIQ	TIQ	MIQ	SADT
4C Hyperacusis Management-Total	41*	08	.15	26
4C Hyperacusis Management -Q1	56*	17	.16	34*
4C Hyperacusis Management -Q2	52*	15	.04	40*
4C Hyperacusis Management -Q3	45*	09	.06	39*
4C Hyperacusis Management -Q4	33*	03	.20	23

*Note*. Number of patients = 48; (\*: statistically significant at p < .05). HIQ, Hyperacusis Impact Questionnaire; TIQ, Tinnitus Impact Questionnaire; MIQ, Misophonia Impact Questionnaire; SADT, the Screening for Anxiety and Depression in Tinnitus/Sound Sensitivity.

For the **4C Misophonia Management Questionnaire**, convergent validity was evaluated by its correlation with the Misophonia Impact Questionnaire (MIQ). As shown in Table 7, a significant negative high correlation with the MIQ ( $\rho = -0.56$ , p < .05) suggests that higher misophonia management scores are associated with lower misophonia impact, indicating that both questionnaires measure related aspects of misophonia, thus supporting convergent validity.

Discriminant validity was assessed using the Hyperacusis Impact Questionnaire (HIQ), Tinnitus Impact Questionnaire (TIQ), and the Screening for Anxiety and Depression in Tinnitus (SAD-T). The 4C Misophonia Questionnaire showed low or non-significant correlations with the HIQ ( $\rho$  = -0.16) and TIQ ( $\rho$  = 0.10), suggesting it does not strongly overlap with hyperacusis or tinnitus impact measures, which supports its discriminant validity regarding these conditions. However, the moderate significant correlation with SAD-T ( $\rho$  = -0.46, p < .05) indicates an association between misophonia management and anxiety and

depression symptoms. This finding suggests that the 4C Misophonia Questionnaire may share common elements with psychological constructs like anxiety and depression, which impacts its discriminant validity.

**Table 7:** Correlations among items and total scores of 4C Misophonia Management Questionnaire, HIQ, TIQ, MIQ, and SADT.

	HIQ	TIQ	MIQ	SADT
4C Misophonia Management-Total	16	.10	56*	46*
To Misophonia Management Total	.10	.10	.50	.10
4C Misophonia Management -Q1	13	.03	31	34*
4C Misophonia Management -Q2	04	.29	52*	43*
TO MISOPHONIA MANAGEMENT - Q2	.04	.27	.52	.43
4C Misophonia Management -Q3	34*	10	54*	67*
4C Misophonia Management -Q4	12	.24	54*	38*
4C Misophonia Management -Q4	12	1.24	54	36

*Note*. Number of patients = 40; (\*: statistically significant at p < .05). HIQ, Hyperacusis Impact Questionnaire; TIQ, Tinnitus Impact Questionnaire; MIQ, Misophonia Impact Questionnaire; SADT, the Screening for Anxiety and Depression in Tinnitus/Sound Sensitivity.

### 4. Discussion

This study evaluated the psychometric properties of the 4C Hyperacusis and Misophonia Management Questionnaires in patients with hyperacusis and misophonia, focusing on reliability, validity, and confirmatory factor structure analyses. Our results indicate strong reliability and convergent validity for both questionnaires, showing that they are useful for measuring patients' confidence in managing their symptoms.

# Confirmatory Factor Analysis

The confirmatory factor analysis (CFA) results for the 4C Hyperacusis and Misophonia Management Questionnaires provide evidence regarding their structural validity. The one-factor structure of the 4C Hyperacusis Management Questionnaire showed an acceptable model

fit overall, with indices such as the comparative fit index (CFI) and standardized root mean square residual (SRMR) meeting recommended thresholds, indicating the model effectively captures the covariance structure with minimal residual error. However, some fit indices, particularly RMSEA had a higher value than recommended threshold by Schermelleh-Engel et al. (2003).

In contrast, the one-factor model of the 4C Misophonia Management Questionnaire initially exhibited poor fit, with several indices falling below recommended thresholds. Testing a two-factor model for the 4C Misophonia Management Questionnaire resulted in a significant improvement in fit, with most indices exceeding recommended cut-offs. This improvement suggests that the questionnaire captures two distinct but related dimensions of confidence in managing misophonia symptoms. However, it is important to note that the two-factor model was derived through a data-driven approach rather than being guided by prior theoretical justification. While this approach enhanced the model fit, caution is required when interpreting the findings.

The poor RMSEA values observed for both questionnaires may partly reflect the small sample sizes (n = 48 for hyperacusis; n = 40 for misophonia). RMSEA is known to overestimate model misfit in smaller samples, making it less reliable in this context (Taasoobshirazi and Wang, 2016, Kenny et al., 2015). By contrast, CFI and SRMR, which are less sensitive to sample size (Shi et al., 2022), indicated acceptable fit for the 4C Hyperacusis Management Questionnaire and improved fit for the 4C Misophonia Management Questionnaire under the two-factor model.

Both questionnaires demonstrated high internal consistency, with Cronbach's  $\alpha$  and  $\omega$  values above 0.87, supporting their reliability. For convergent validity, the 4C Hyperacusis Management Questionnaire showed a moderate negative correlation with the Hyperacusis Impact Questionnaire (HIQ), and the 4C Misophonia Management Questionnaire had a strong

negative correlation with the Misophonia Impact Questionnaire (MIQ). These negative correlation results also indicate that greater confidence in symptom management is associated with a lower perceived impact, aligning with research demonstrating that self-management skills and Cognitive Behavioural Therapy can effectively reduce impact of misophonia and hyperacusis burden (Mattson et al., 2023, Aazh et al., 2024c).

# Discriminant Validity

The discriminant validity results revealed some unexpected findings. The 4C Hyperacusis Questionnaire showed low correlations with unrelated measures, suggesting that it specifically assesses hyperacusis management with minimal overlap with tinnitus and misophonia. Furthermore, our hypothesis anticipated a low correlation between hyperacusis and anxiety and depression; however, no significant correlation was identified in the analysis. This outcome supports the discriminant validity of the questionnaire, although it contrasts with previous studies that have reported associations between hyperacusis symptoms and anxiety and depression (Jüris et al., 2013, Paulin et al., 2016). Another reason for this could be that the SAD-T, used in this study for discriminant validity, is a screening tool designed to identify symptoms of anxiety and depression rather than providing a comprehensive assessment.

In contrast, 4C Misophonia Management Questionnaire demonstrated a different pattern in terms of discriminant validity, particularly in relation to anxiety and depression. The questionnaire showed low correlations with the HIQ and TIQ, supporting our hypothesis and indicating it effectively distinguishes misophonia from hyperacusis and tinnitus. However, a moderate to high correlation was observed between the 4C Misophonia Management Questionnaire and SAD-T, contradicting our expectation of a low correlation. This suggests that the questionnaire may not fully differentiate between misophonia-specific management and anxiety and depression. This outcome aligns with prior research an association between

misophonia and anxiety and depression (Rouw and Erfanian, 2018, Cassiello-Robbins et al., 2021).

# Demographic Findings

Our analysis of age and gender differences revealed no significant correlations with total scores on the 4C Hyperacusis Management Questionnaire. This result aligns with Musumano et al. (2023), in a systematic review study reported no significant gender differences in hyperacusis levels across studied populations. While some studies by Paulin et al. (2016), Ren et al. (2021) suggest that hyperacusis sensitivity may increase with age, this discrepancy could be due to differences in the focus of questionnaires. Many studies assess symptom severity, whereas the 4C Hyperacusis Management Questionnaire specifically measures coping confidence. This distinction may explain why demographic factors like age and gender did not significantly impact 4C scores, as confidence in managing symptoms may be less influenced by these factors than symptom sensitivity.

Similarly, for the 4C Misophonia Management Questionnaire, age and gender showed no significant correlations, aligning with findings from Brennan et al. (2024) found no relationship between age and misophonia. However, our results contrast with studies by (Erfanian et al., 2019, Stalias-Mantzikos et al., 2023), which reported higher misophonia severity scores among females. These differences may reflect variations in study design, sample characteristics, or the specific focus of each questionnaire. Further research with larger and more diverse samples is warranted to clarify the influence of demographic factors on both misophonia management and severity.

### Limitations and Future Directions

This study has some limitations. While the small sample size may limit the generalisability of the CFA results, and limitation prevented the use of latent variables to comprehensively evaluate convergent and discriminant validity. Latent variables provide a more robust approach

than direct measures, as they reduce measurement error, ensure factorial validity by requiring proper measurement models, and avoid untested assumptions characteristic in total score calculations (Cheung et al., 2024). However, a notable strength of this study is the use of data from a clinical population, which enhances the practical relevance and applicability of the 4C questionnaires for patients with hyperacusis and misophonia. Future research should address these limitations by employing larger sample sizes and evaluating the questionnaires using latent variable modelling to provide a more comprehensive assessment.

### 5. Conclusions

This study provides an evaluation of the psychometric properties of the 4C Hyperacusis and Misophonia Management Questionnaires, which can help assess patients' confidence in managing hyperacusis and misophonia symptoms. Both questionnaires demonstrated high reliability, with Cronbach's alpha and omega values exceeding 0.87, indicating consistent measurement.

Overall, these findings support the 4C Management Questionnaires as valuable tools for clinicians to assess management confidence. Future research with larger and more diverse samples will help confirm and refine these questionnaires, increasing their usefulness in providing tailored support for patients with hyperacusis and misophonia.

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# **CHAPTER SIX**

# **General Discussion and Conclusions**

#### 6.1 Overview

The final chapter of this thesis will summarise the findings of the previous chapters, including the systematic review and three empirical studies, and discuss their implications within the context of existing literature. The overarching aim of this thesis was to contribute to the understanding of hyperacusis and misophonia by addressing critical gaps in their assessment and broader psychological impacts. This chapter will highlight contributions of the research, address its strengths and limitations, and outline directions for future studies.

As outlined in Chapter One, hyperacusis and misophonia are complex conditions, with hyperacusis involving a heightening sensitivity to everyday sounds and misophonia characterised by strong emotional, behavioural, physical reactions to specific trigger sounds or stimuli, both significantly impairing daily functioning and quality of life (Swedo et al., 2022, Aazh et al., 2016). Despite increasing recognition of their impact, these conditions lack consistent diagnostic frameworks and a gold standard tool for clinical assessment and research (Ferrer-Torres and Giménez-Llort, 2022). The validation of some existing tools across different languages and cultural contexts is also insufficient, limiting their applicability in diverse populations. In addition, current instruments have generally focussed on symptom severity but fail to address key aspects such as symptom management and coping strategies, which are essential for effective treatment planning. The psychological impact of misophonia, particularly its relationship with fatigue, has received limited attention in existing literature. These gaps provided the rationale for this thesis and informed its objectives.

Chapter Two presented a systematic review of psychometric measures for hyperacusis and misophonia, evaluating eight widely used instruments, Hyperacusis Questionnaire, Inventory

of Hyperacusis Symptoms, questionnaire on hypersensitivity to sound, Hyperacusis Handicap Questionnaire, Short Hyperacusis Questionnaire, Amsterdam Misophonia Scale, MisoQuest, and the Misophonia Questionnaire, against COSMIN standards. The review included research papers published up to April 2021 and identified significant methodological gaps, including inconsistencies in reliability, validity, and cross-cultural applicability based on COSMIN. These findings emphasised the need for psychometrically robust instruments tailored for use in clinical populations, setting the stage for the subsequent empirical studies.

The overarching aims of the thesis were:

- 1. To systematically evaluate the psychometric properties of existing questionnaires for hyperacusis and misophonia, identifying gaps in their methodological quality.
- To validate the English version of MisoQuest, establishing its reliability, validity, and relationships with audiological and psychological variables, including hyperacusis and fatigue.
- To investigate the mediating roles of stress and sleep quality in the relationship between misophonia and fatigue, contributing to a broader understanding of the psychological impacts of misophonia
- 4. To validate the 4C Hyperacusis and Misophonia Management Questionnaires, designed to assess patients' confidence in managing their symptoms without reliance on avoidance behaviours, with a focus on clinical applications in Cognitive Behavioural Therapy (CBT).

This thesis contributes to advancing the understanding of hyperacusis and misophonia by providing validated tools, addressing broader psychological impacts, and highlighting clinical applications. A summary of the findings of each chapter will be discussed in the following section.

# **6.2 Summary of Findings**

The first study (Chapter 2) conducted a systematic review of the psychometric properties of existing hyperacusis and misophonia questionnaires, evaluating their adherence to the COSMIN standards. The review identified eight instruments, including the Hyperacusis Questionnaire (HQ), MisoQuest, and Amsterdam Misophonia Scale (A-MISO-S), among others. While some tools demonstrated good internal consistency in specific studies, significant gaps were found in the reporting of reliability, validity, and cross-cultural applicability. Importantly, no instrument fully met the COSMIN standards. These findings underscored the need for more robust and clinically relevant psychometric tools, providing a foundation for the subsequent empirical studies.

The second study (Chapter 3) validated the English version of MisoQuest, a 14-item misophonia questionnaire initially developed in Polish. The study involved 451 adult participants and demonstrated excellent internal consistency, high test-retest reliability and a confirmed multidimensional factor structure with adequate model fit through Confirmatory Factor Analysis (CFA). Strong convergent validity was established through moderate to high correlations with other misophonia measures, while weak correlations with non-misophonia constructs supported discriminant validity. However, moderate correlations with fatigue and hyperacusis raised questions about potential construct overlap, indicating the need for further exploration. These results validated the English MisoQuest as a reliable tool for assessing misophonia while highlighting the need for further research to better understand the correlation between misophonia and fatigue.

The third study (Chapter 4) investigated the relationship between misophonia and fatigue, with a particular focus on the mediating roles of stress and sleep quality. Using data from the same sample as the second study, correlation analysis revealed a positive moderate correlation between misophonia severity and fatigue, supporting findings from previous research (Dibb

and Golding, 2022). Mediation analysis further revealed that perceived stress and sleep quality partially mediated this relationship, indicating that these factors significantly contribute to the experience of fatigue in individuals with misophonia. However, the findings also suggested that the effect of misophonia on fatigue extends beyond the influence of stress and sleep quality. Interestingly, hyperacusis, anxiety, and depression did not emerge as significant predictors of fatigue in this context. These findings broadened the scope of the thesis by addressing the psychological and health impacts of misophonia, emphasising the importance of targeting stress and sleep quality in clinical interventions while acknowledging the need to explore other underlying mechanisms.

The final study (Chapter 5) focused on validating the 4C Hyperacusis and Misophonia Management Questionnaires, designed to assess patients' confidence in managing symptoms without reliance on avoidance behaviours. Data from 48 participants for the Hyperacusis Management Questionnaire and 40 participants for the 4C Misophonia Management Questionnaire demonstrated high reliability. CFA showed an acceptable model fit for the one structure 4C Hyperacusis, while the 4C Misophonia showed an acceptable better model fit in two factor structure. Even though the findings were based on a relatively small sample, both tools exhibited good convergent and discriminant validity, although the misophonia questionnaire showed a high correlation with anxiety and depression. These tools fill a critical gap in clinical assessments, providing a means to evaluate patients' confidence in managing their symptoms and supporting personalised treatment planning, particularly within CBT frameworks.

### 6.3 Research and Clinical Contributions

This thesis is primarily psychometric in nature, focusing on the validation and evaluation of measurement tools. Validity reflects how well an instrument measures what it is intended to measure, while reliability refers to the consistency of these measurements and their resistance

to measurement error (Mohajan, 2017). Without these essential properties, measurement instruments risk producing misleading conclusions, undermining the credibility and accuracy of research findings, and limiting their applicability in clinical and academic settings (Cohen et al., 2017). This issue is particularly critical in diagnosis of hyperacusis and misophonia, which relies generally on questionnaires (Jastreboff and Jastreboff, 2014). Ensuring that these tools are valid and reliable is essential for accurate assessment and meaningful clinical interventions. Therefore, this thesis makes a significant contribution by evaluating and highlighting the psychometric properties of these tools in line with COSMIN standards (Mokkink et al., 2018), providing a rigorous framework to ensure their methodological and clinical robustness.

This thesis makes contributions to both research and clinical practice by addressing key gaps in the assessment and psychological impacts of hyperacusis and misophonia. One of the contributions is the validation of the English version of MisoQuest, a widely used tool originally developed in Polish (Siepsiak et al., 2020), following COSMIN standards. This validation ensures that the questionnaire can reliably assess misophonia severity in English-speaking populations, enabling cross-cultural research and facilitating consistent comparisons across diverse contexts.

In addition to English MisoQuest, this thesis introduces the 4C Hyperacusis and Misophonia Management Questionnaires, specifically designed to measure patients' confidence in managing their symptoms without relying on avoidance behaviours. These questionnaires address a critical gap in existing tools, which generally focus on symptom severity. By validating these tools in clinical populations, this research provides clinicians and researchers with practical instruments that align with the principles of Cognitive Behavioural Therapy (CBT) and support the development of personalised treatment strategies (Aazh et al., 2016).

Beyond psychometric contributions, this thesis also explored the relationship between misophonia and fatigue, a topic that has received limited attention in the literature. The findings demonstrate that stress and sleep quality partially mediate this relationship, offering key findings the broader psychological and health impacts of misophonia. These results emphasise the importance of addressing stress and sleep disturbances as part of a comprehensive treatment approach. Although causality could not be demonstrated, the results of the study suggest that the impact of misophonia on fatigue may extend beyond these mediating factors, highlighting the need for future research to explore additional underlying mechanisms.

By combining rigorous psychometric evaluations with an exploration of the psychological impacts of misophonia and hyperacusis, this thesis advances the understanding of these conditions. The validated tools and findings on fatigue help improve how hyperacusis and misophonia are assessed and managed to support better clinical outcomes for individuals affected by these conditions.

### 6.4 Challenges and Limitations

While this thesis provides important insights, certain limitations must be acknowledged. The systematic review (published in 2023) included only psychometric measures published up to April 2021, excluding tools developed or validated after this date. As of December 2024, there have been an additional at least ten studies published assessing hyperacusis or misophonia.

In this thesis, I adhered to COSMIN standards for the systematic review and two psychometric empirical studies. However, I was unable to properly assess content validity based on these standards. Content validity refers to how well a measurement instrument represents the concept it is designed to measure and is crucial in developing new questionnaires, particularly in ensuring agreement among experts (Beckstead, 2009). Furthermore, as I did not develop a new questionnaire in my thesis, and the existing questionnaires often lacked detailed reports on content validity in the included articles, it was

challenging to evaluate their content validity comprehensively. This is something that I aim to pursue in the future.

Another psychometric property that I was not able to address in this thesis, including within the systematic review (Chapter 2), relates to the notion/construct of 'responsiveness'. Responsiveness refers to the ability of an instrument to detect meaningful changes over time in the construct being measured. In other words, it reflects how accurately a measure can capture changes in outcomes specific to the effects of a treatment (Mokkink et al., 2018). To our knowledge, none of the questionnaires included in the systematic review evaluated this psychometric property. Additionally, responsiveness could not be assessed in the empirical studies conducted for this thesis, as these studies did not involve an intervention-based design requiring pre- and post-treatment measurements.

Within this thesis, there are several methodological limitations that should be noted, with the specific limitations of each study discussed in their respective chapters. One important contextual factor is the timing of data collection for Studies 2 and 3, which occurred during the global COVID-19 pandemic. Recruitment was conducted online through charities and social media support groups for individuals with hyperacusis and misophonia. As a result, we relied solely on self-reports and were unable to independently diagnose participants with misophonia. Proper diagnosis would have ideally required structured clinical interviews alongside the questionnaires, which was not feasible under the circumstances. The absence of a clinically verified sample further limited our ability to evaluate the sensitivity and specificity of the measures. Additionally, in Study 3, comparisons between individuals with and without clinically confirmed misophonia could have provided more comprehensive insights into group differences, particularly in understanding the relationship between misophonia and fatigue, thereby enhancing the robustness of the study.

The sample size for the final empirical study in my thesis was limited due to time constraints within my PhD funding. As a result, the findings presented in this chapter are based on a relatively small sample. However, data collection is ongoing, and the work is planned to be submitted for publication at a later date.

Additionally, all studies included in this thesis a higher proportion of female participants. This is consistent with research indicating that females are more likely to report symptoms of misophonia, with studies suggesting that over 82% of individuals self-identifying with the condition are women (Claiborn et al., 2020). In contrast, findings regarding hyperacusis show no significant gender differences, with a relatively balanced distribution between males and females (Musumano et al., 2023). These patterns suggest that the gender distribution in this thesis may reflect the characteristics of the conditions studied rather than recruitment bias, although the potential impact on generalisability should still be acknowledged.

# 6.5 Directions for Future Research

While this thesis has contributed valuable insights and extended previous research in the field of hyperacusis and misophonia, it has also highlighted several implications for future research.

The findings from this thesis in the systematic review suggest that none of the hyperacusis and misophonia questionnaires research studies published up to 2021 adhered fully to COSMIN standards. Recent psychometric tools for misophonia, such as the Misophonia Response Scale (Dibb et al., 2021), the Duke Misophonia Questionnaire (Rosenthal et al., 2021), the Selective Sound Sensitivity Syndrome Scale (S-Five; Vitoratou et al., 2021), the Duke-Vanderbilt Misophonia Screening Questionnaire (DVMSQ; Williams et al., 2022), the Berlin Misophonia Questionnaire Revised (BMQ; Remmert et al., 2022), the Sussex Misophonia Scale (SMS; Rinaldi et al., 2022; Simner et al., 2024), and the Misophonia Impact Questionnaire (MIQ; Aazh et al., 2023), were not included in this review. Similarly, for

hyperacusis, tools like the Hyperacusis Impact Questionnaire [HIQ; (Aazh et al., 2024a)], the Sound Sensitivity Symptoms Questionnaire (SSSQ; Aazh et al., 2024a), and the Hyperacusis Assessment Questionnaire (Raj-Koziak et al., 2023) were also not assessed. Future research should evaluate whether these recently developed tools meet COSMIN standards and rigorously assess their validity, reliability, and responsiveness. Evaluating these tools will provide a clearer understanding of their psychometric properties and improve their applicability in both clinical and research contexts. Such efforts are essential for advancing the assessment and management of hyperacusis and misophonia.

Future research should also prioritise content validity, an essential psychometric property and complex steps in COSMIN (Mokkink et al., 2010), particularly through expert consensus, to ensure new questionnaires accurately represent the intended constructs. Intervention-based studies assessing responsiveness, including pre- and post-treatment measurements, are also necessary to evaluate the effectiveness of psychometric tools in detecting changes over time.

To address methodological limitations, future research should include diverse and representative samples with balanced gender participation to improve generalisability. Longitudinal studies are also essential for understanding the progression of hyperacusis and misophonia over time, identifying predictive factors, and establishing causal relationships. These studies could provide useful insights into the progression of these conditions and help design more effective interventions.

By addressing these areas, future research can build on the findings of this thesis to enhance the development, validation, and application of psychometric tools in the fields of hyperacusis and misophonia.

#### 6.6 Overall Conclusions

This thesis focuses on the psychometric assessment of hyperacusis and misophonia, critically evaluating existing questionnaires and validating tools in accordance with COSMIN standards. Through a systematic review and three empirical studies, it highlights the limitations of current instruments, validates tools, and explores understudied psychological aspects, particularly the relationship between misophonia and fatigue.

The systematic review identified significant gaps in the reliability, validity, and adherence to COSMIN standards among widely used questionnaires for hyperacusis and misophonia. Building on these findings, the English version of MisoQuest was validated, demonstrating its psychometric robustness and suitability for use in English-speaking populations.

In addition, this thesis introduced and validated the 4C Hyperacusis and Misophonia Management Questionnaires, which were developed to assess patients' confidence in managing symptoms without reliance on avoidance behaviours. In clinical populations support their potential application in treatment planning, especially within Cognitive Behavioural Therapy (CBT) frameworks.

The thesis also addresses the relationship between fatigue and misophonia, an area previously overlooked in the literature. By evaluating the partially mediating roles of stress and sleep quality, it contributes to the understanding the psychological impacts of misophonia. The findings reveal a strong and significant link between misophonia and fatigue, emphasizing the need for comprehensive treatment approaches that incorporate stress and sleep management to improve outcomes for individuals with misophonia.

By addressing the limitations of existing tools, validating measures, and examining the correlation between misophonia and fatigue, this thesis contributes significantly to the field. It provides a foundation for future research to enhance psychometric assessments and improve clinical care for individuals affected by hyperacusis and misophonia.

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#### **APPENDICES**

### Appendix A: University Ethics Committee Approval Letter (Study 1 and 2)

# RESEARCH & INNOVATION SERVICES SURREY.AC.UE/RIS



Research Integrity & Governance Office (RIGO) 4<sup>th</sup> floor Senate House Guilaford

Surrey GU2 7XH UK

T: +44 (0) 1483 68 9103/2051

ethics@surrey.ac.uk www.surrey.ac.uk

Fatma Betul Kula Psychology Faculty of Health and Medical Sciences

03 May 2023

Dear Fatma,

#### EGA ref: FHMS 21-22 083 EGA Amend 1

Project Title: Assessment of Reliability and Validity of the English version of MisoQuest using an online survey in the UK and the correlation between Misophonia, Hyperacusis, Tinnitus, Stress, Sleep and Fatigue.

On behalf of the Ethics Committee, I am pleased to inform you that your request for an Amendment to the above protocol has been considered and has received a favourable ethical opinion on the understanding that you continue comply with the relevant University policies, ethical and professional standards and any applicable regulatory requirements, and have completed all mandatory training provided by the University of Surrey. Please be advised that the Ethics Committee and/or RIGO can audit research projects to ensure that researchers are abiding by the University requirements and guidelines.

If the project includes distribution of a survey or questionnaire to members of the University community, researchers are asked to include a statement advising that the project has been reviewed by the University's Ethics Committee.

Please follow guidelines below and note that all research activity must comply with current University guidance regarding the Covid19 pandemic:

https://www.surrey.ac.uk/coronavirus/researchers/research-university-ethics-committee-approval

Date of confirmation of ethical opinion: 10th February 2022

Date of favourable ethical opinion of amendment to protocol: 3<sup>rd</sup> May 2023

The list of amended documents reviewed and approved by the Committee is as follows:

Document	Version	Date
notification-of-amendment-form_Fatma Kula_FHMS PSYCHOLOGY	Version 1	14/04/23
Ethics Governance Application (EGA) Form (Human Research)_ Fatma Kula FHMS PSYCHOLOGY_v3	Version 3.0	14/04/23



#### **Appendix B:** University Ethics Committee Approval Letter (Study 3)



RESEARCH & INNOVATION SERVICES
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Dr Hashir Aazh Psychology Faculty of Health and Medical Sciences Research Integrity & Governance Office (RIGO) 4th floor Senate House Guildford, Surrey GU2 7XH UK

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E; ethics@surrey.ac.uk W: www.surrey.ac.uk

22 February 2024

Dear Hashir,

#### EGA ref: FHMS 21-22 147 EGA Amend 1

Study Title: Evaluating the effectiveness of therapy for tinnitus, hyperacusis and misophonia conducted in face-to-face sessions as well as internet-based

On behalf of the Ethics Committee, I am pleased to inform you that your request for an Amendment to the above protocol has been considered and has received a favourable ethical opinion on the understanding that you continue comply with the relevant University policies, ethical and professional standards and any applicable regulatory requirements, and have completed all mandatory training provided by the University of Surrey. Please be advised that the Ethics Committee and/or RIGO and the HTA license Designated Individual (or those appointed by them) can audit research projects to ensure that researchers are abiding by the University requirements and guidelines.

If the project includes distribution of a survey or questionnaire to members of the University community, researchers are asked to include a statement advising that the project has been reviewed by the University's Ethics Committee.

Date of confirmation of ethical opinion: 27th May 2022

Date of favourable ethical opinion of amendment to protocol: 22<sup>nd</sup> February 2024

The list of amended documents reviewed and approved by the Committee is as follows:

Document	Version	Date
Notification of amendment form		
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**Appendix** C: Definitions and criteria for good measurement properties by COSMIN guidance. *Note*. The content presented in Appendix C of the thesis has been included as Appendix 1 in this thesis (chapter two) to comply with the journal's formatting guidelines. The table is reproduced from the COSMIN guidance, using their definitions and criteria.

Measurement Property	Definition	Rating	Criteria
Structural Validity	The degree to which the scores of a PROM are an adequate reflection of the dimensionality of the construct to be measured	+ + ?	CTT: CFA: CFI or TLI or comparable measure >0.95 OR RMSEA <0.06 OR SRMR <0.082 IRT/Rasch: No violation of unidimensionality3: CFI or TLI or comparable measure >0.95 OR RMSEA <0.06 OR SRMR <0.08 AND no violation of local independence: residual correlations among the items after controlling for the dominant factor < 0.20 OR Q3's < 0.37 AND no violation of monotonicity: adequate looking graphs OR item scalability >0.30 AND adequate model fit: IRT: $\chi$ 2 >0.01 Rasch: infit and outfit mean squares $\geq$ 0.5 and $\leq$ 1.5 OR standardized values > -2 and <2 CTT: Not all information for '+' reported IRT/Rasch: Model fit not reported
		-	Criteria for '+' not met
		+	At least low evidence for sufficient structural validity AND Cronbach's alpha(s) $\geq 0.70$ for each unidimensional scale or Subscale
Internal Consistency	The degree of the interrelatedness	?	Criteria for "At least low evidence4 for sufficient structural validity" not met
among the items	-	At least low evidence4 for sufficient structural validity AND Cronbach's alpha(s) < 0.70 for each unidimensional scale or subscale	
Reliability	The proportion of the total variance in the measurements which	+	ICC or weighted Kappa ≥ 0.70
	is due to 'true'†	?	ICC or weighted Kappa not reported

	differences between	-	ICC or weighted Kappa < 0.70
Measurement Error	patients The systematic and random error of a patient's score	+	SDC or LoA < MIC5
	that is not attributed to true changes in the	?	MIC not defined
construct to be measured		-	SDC or LoA > MIC5
Hypotheses testing for construct validity  The degree to which the scores of a PROM are consistent with hypotheses (for instance with regard to internal relationships, relationships to scores of other instruments, or differences between relevant groups) based on the assumption that the PROM validly measures the construct to be measured	the scores of a PROM are consistent with hypotheses (for	+	The result is in accordance with the hypothesis
	regard to internal relationships, relationships to scores of other	?	No hypothesis defined (by the review team)
	-	The result is not in accordance with the hypothesis	
Cross-cultural validity\measurement invariance	The degree to which the performance of the items on a translated or culturally adapted PROM are an adequate reflection	+	No important differences found between group factors (such as age, gender, language) in multiple group factor analysis OR no important DIF for group factors (McFadden's R2 < 0.02)
	of the performance of the items of the original version of the PROM	?	o multiple group factor analysis OR DIF analysis performed
		-	Important differences between group factors OR DIF was found
Criterion validity	The degree to which the scores of a PROM	+	Correlation with gold standard $\geq 0.70$ OR AUC $\geq 0.70$
are an adequate reflection of a 'gold standard'		?	Not all information for '+' reported
		-	Correlation with gold standard < 0.70 OR AUC < 0.70
Responsiveness	The ability of a PROM to detect	+	The result is in accordance with the hypothesis 7 OR AUC ≥ 0.70
	change over time in the construct to be measured	?	No hypothesis defined (by the review team)
	to oc incasured	-	The result is not in accordance with the hypothesis 7 OR AUC < 0.70

### **Appendix D:** English MisoQuest (Study 1 and 2)

MISOQUEST – A QUESTIONNAIRE FOR ASSESSING DECREASED SOUND TOLERANCE

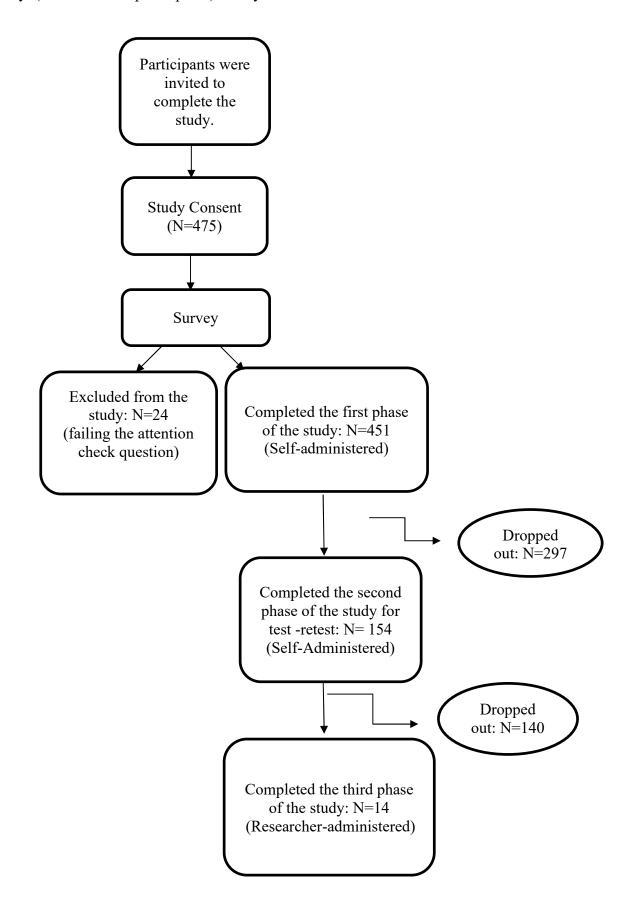
authors: Siepsiak, M., Śliwerski, A., Dragan, W. Ł

Some people are less sensitive to certain sounds, while other people are more sensitive to certain sounds. Are there any sounds which you find particularly burdensome? Please indicate how much you agree or disagree with the following statements using the following scale:

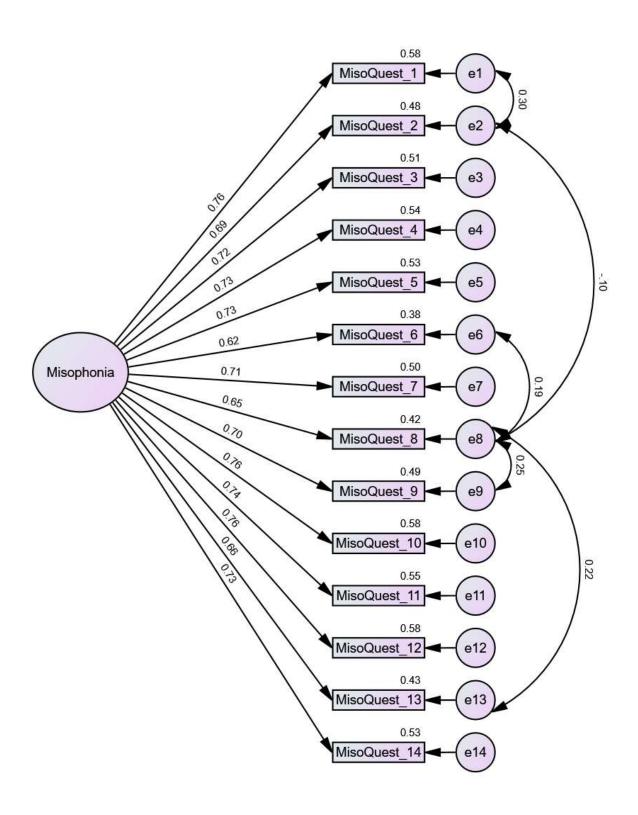
- 1 I definitely do not agree
- 2 I do not agree
- 3 Hard to say
- 4 I agree
- 5 I definitely agree

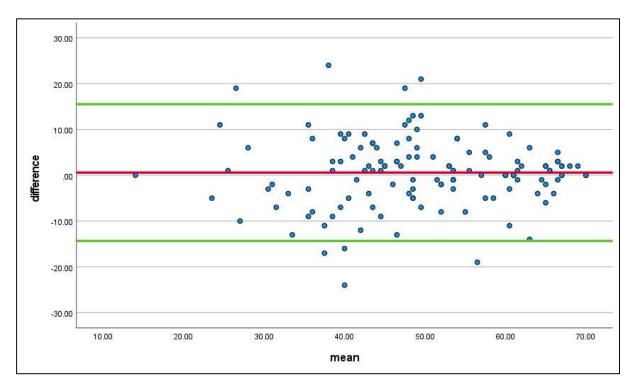
1	Some sounds bother me so much that I have difficulty controlling my emotions.	1	2	3	4	5
2	Unpleasant sounds make me feel overwhelmed.	1	2	3	4	5
3	I become anxious at the mere thought of an unpleasant sound.	1	2	3	4	5
4	I believe that my reactions to sounds are exaggerated, but I can't get rid of them.	1	2	3	4	5
5	When I hear unpleasant sounds, I start sensing emotions in my body (e.g. I sweat, feel pain, feel pressure, my muscles tens).	1	2	3	4	5
6	I start feeling anger the moment I see a thing/animal/person that might make an unpleasant sound at any time.	1	2	3	4	5
7	I put a lot of effort into controlling emotions when I hear an unpleasant sound.	1	2	3	4	5
8	If I can, I avoid meeting with certain people because of the sounds they make.	1	2	3	4	5
9	I find some sounds made by the human body unbearable.	1	2	3	4	5
10	I feel that my mental state worsens if I cannot leave a place where there's an unpleasant sound.	1	2	3	4	5
11	I often think about how to drown out unpleasant sounds.	1	2	3	4	5
12	Some unpleasant sounds make me instantly angry.	1	2	3	4	5
13	I am scared that unpleasant sounds may impact my future.	1	2	3	4	5
14	When meeting with other people, I am sometimes irritated because of unpleasant sounds that are present.	1	2	3	4	5

**Appendix E:** SF 1: Flowchart of the participant involvement process in the three phases of this study. (N= number of participants)- Study 1



Appendix F: SF 2: Factor structure of the English version of MisoQuest from CFA (Study 1)





**Appendix G:** SF 3: Chapter 3- Bland-Altman plot (Study 1)

Bland-Altman plot of the total misophonia score of the English version of MisoQuest. The plot demonstrates the agreement between two time points and highlights potential outliers. Each individual is depicted on the graph by showing the average value of their two assessments on the x-axis and the difference between those assessments on the y-axis. The mean difference represents the estimated bias, while the standard deviation (SD) of the differences indicates the variations around this mean (with outliers being values above 1.96 SD<sub>diff</sub>). The reference lines display the average difference between time one and time 2 (red-colored solid line) and the 95% limits of agreement for the mean difference (green-colored solid lines).

# **Appendix H:** Three-Dimensional Fatigue Inventory (Study 1 and 2)

**Physical Fatigue**: Involves extreme physical tiredness and an inability to engage in physical activity.

During the PAST 12 MONTHS, how often did you	Everyday	At least once a week	At least once a month	Less than once a month	Never
1. feel physically exhausted at the end of the day?					
2. have difficulty engaging in physical activity at the end of the day?					
3. feel physically worn out at the end of the day?					
4. want to physically shut down at the end of the day?					
5. feel physically drained at the end of the day?					
6. want to avoid anything that took too much physical energy at the end of the day?					

Mental Fatigue: Involves extreme mental tiredness and an inability to think or concentrate.

During the PAST 12 MONTHS, how often did you	Everyday	At least once a week	At least once a month	Less than once a month	Never
7. feel mentally exhausted at the end of the day?					
8. have difficulty thinking and concentrating at the end of the day?					
9. feel mentally worn out at the end of the day?					
10. want to mentally shut down at the end of the day?					
11. feel mentally drained at the end of the day?					
12. want to avoid anything that took too much mental energy at the end of the day?					

**Emotional Fatigue**: Involves extreme emotional tiredness and an inability to feel or show emotions.

During the PAST 12 MONTHS, how often did you	Everyday	At least once a week	At least once a month	Less than once a month	Never
13. feel emotionally exhausted at the end of the day?					
14. have difficulty showing and dealing with emotions at the end of the day?					
15. feel emotionally worn out at the end of the day?					
16. want to emotionally shut down at the end of the day?					
17. feel emotionally drained at the end of the day?					
18. want to avoid anything that took too much emotional energy at the end of the day?					

## **Appendix I:** Amsterdam Misophonia Scale – Revised (Study 1)

Review your experience from hearing your misophonia sounds *in the last 3 days*. Read instead of "sounds" your most disturbing misophonia sounds and instead of "emotion" your typical emotion. Choose the answer that is most applicable for you.

1.	How many times do you spend a day (thinking about) these sounds?	0 hour 0	< 1 hour 1	1-3 hour 2	3-8 hour 3	>8 hour
2.	To what extent do you focus on these sounds?	not 0	1	moderate 2	severe 3	extreme 4
3.	To what extent do you experience impairment due to these sounds?	not 0	mild 1	moderate 2	severe 3	extreme 4
4.	How intense is your feeling of irritability/anger when you hear these sounds?	not 0	mild 1	moderate 2	severe 3	extreme 4
5.	To what extent do you feel helpless against these sounds?	not 0	mild 1	moderate 2	severe 3	extreme 4
6.	To what extent are you suffering from these sounds?	not 0	mild 1	moderate 2	severe 3	extreme 4
7.	To what extent are you suffering from the avoidance of these sounds?	not 0	mild 1	moderate 2	severe 3	extreme 4
8.	To what extent are the sounds limiting your life (work, household etc.)?	not 0	mild 1	moderate 2	severe 3	extreme 4
9.	To what extent are you avoiding specific places or situations because of the sounds?	not 0	mild 1	moderate 2	severe 3	extreme 4
10	To what extent can you shift your attention when you are hearing these sounds?	always 0	usually (75%) 1	sometimes (50%)	seldom (25%) 3	never 4
To	otal score:				•	•

**Appendix J:** Misophonia Impact Questionnaire- Adult (Study 1 and 3)

Please answer each item to the best of your ability as close to your experience as possible.

Over the last 2 weeks, how often would you say the following has occurred because of your intolerance to certain sounds related to eating, chewing gum, lip smacking, mouth noises, sniffling, breathing, clicking, and tapping? 2-6 7-10 1. Feeling anxious 0 - 111-14 day days days days 2. Unable to distract yourself from certain 0-1 2-6 7-10 11-14 sounds day days days days 3. Experiencing difficulties in your relationships 0 - 12-6 7-10 11-14 with family members or friends days day days days 0-1 2-6 7-10 11-14 4. Feeling angry day days days days 5. Finding it difficult to be around certain 0 - 12-6 7-10 11-14 individuals because of the noises that they make day days days days 0 - 17-10 11-14 2-6 6. Feeling irritated day days days days 7. Avoiding certain situations because of the 0-1 2-6 7-10 11-14 noises that you have to put up with day days days days 8. Experiencing low mood because of your 0 - 12-6 7-10 11-14 intolerance to certain sounds day days days days

# Appendix K: Sound Sensitivity Symptoms Questionnaire (Study 1)

Please answer each item to the best of your ability as close to your experience as possible.

Over the last 2 weeks, how often have you been bothered by any of the following problems?					
1. Having a problem tolerating sounds because they often seem "too loud" to you?	0–1 days	2–6 days	7–10 days	11–14 days	
2. Pain in your ears when hearing certain loud sounds?	0–1 days	2–6 days	7–10 days	11–14 days	
Examples: loud music, sirens, motorcycles, building work, lawn mower, train stations.					
3. Discomfort (physical sensations other than ear pain) in your ears when hearing certain loud sounds?	0–1 days	2–6 days	7–10 days	11–14 days	
4. Feeling angry or anxious when hearing certain sounds related to eating noises, lip smacking, sniffling, breathing, clicking sounds, tapping?	0–1 days	2–6 days	7–10 days	11–14 days	
5. Fear that certain sounds may make your hearing and/or tinnitus worse?	0–1 days	2–6 days	7–10 days	11–14 days	

**Appendix L:** Hyperacusis Impact Questionnaire (Study 1, 2 and 3)

Please answer each item to the best of your ability as close to your experience as possible.						
Over the last 2 weeks, how often would you say each of the following has occurred because of certain environmental sounds that seemed too loud to you but that people around you could tolerate well?						
Feeling anxious when hearing loud noises	0-1 days	2-6 days	7-10 days	11-14 days		
Avoiding certain places because it is too noisy	0-1 days	2-6 days	7-10 days	11-14 days		
3. Lack of concentration in noisy places	0-1 days	2-6 days	7-10 days	11-14 days		
4. Unable to relax in noisy places	0-1 days	2-6 days	7-10 days	11-14 days		
5. Difficulty in carrying out certain day-to-day activities/tasks in noisy places	0-1 days	2-6 days	7-10 days	11-14 days		
6. Lack of enjoyment from leisure activities in noisy places	0-1 days	2-6 days	7-10 days	11-14 days		
7. Experiencing low mood because of your intolerance to sound	0-1 days	2-6 days	7-10 days	11-14 days		
8. Getting tired quickly in noisy places	0-1 days	2-6 days	7-10 days	11-14 days		

Appendix M: Tinnitus Impact Questionnaire (Study 1 and 3)

Please answer each item to the best of your ability as close to your experience as possible.					
Over the last 2 weeks, how often would you a sound in your ears or head with no extern hissing)?	•	_		_	
1. Lack of concentration	0-1 days	2-6 days	7-10 days	11-14 days	
2. Feeling anxious	0-1 days	2-6 days	7-10 days	11-14 days	
3. Sleep difficulties (delay in falling sleep and/or difficulty getting back to sleep if woken up during the night)	0-1 days	2-6 days	7-10 days	11-14days	
4. Lack of enjoyment from leisure activities	0-1 days	2-6 days	7-10 days	11-14 days	
5. Inability to perform certain day-to-day activities/tasks	0-1 days	2-6 days	7-10 days	11-14 days	
6. Feeling irritable	0-1 days	2-6 days	7-10 days	11-14 days	
7. Low mood	0-1 days	2-6 days	7-10 days	11-14 days	

**Appendix N:** The Screening for Anxiety and Depression in Tinnitus/Sound Intolerance (Study 1, 2 and 3)

Please answer each item to the best of your ability as close to your experience as possible.							
Over the last 2 weeks, how often have you been bothered by any of the following problems?							
1. Feeling nervous, anxious or on	0-1 days	2-6 days	7-10 days	11-14 days			
edge							
2. Not being able to stop or control	0-1 days	2-6 days	7-10 days	11-14 days			
worrying							
3. Little interest or pleasure in doing	0-1 days	2-6 days	7-10 days	11-15days			
things							
4. Feeling down, depressed or	0-1 days	2-6 days	7-10 days	11-14 days			
hopeless							

## **Appendix O:** Hearing Handicap Inventory (Study 1)

<u>Instructions:</u> Please check "yes," "no," or "sometimes" in response to each of the following items. Do not skip a question if you avoid a situation because of a hearing problem. If you use a hearing aid, please answer the way you hear without the aid.

Item		Yes	Sometimes	No
		(4 pts)	(2 pts)	(0 pts)
Е	Does a hearing			
	problem cause you			
	to feel			
	embarrassed when			
	meeting new			
	people?			
E	Does a hearing			
	problem cause you			
	to feel frustrated			
	when talking to			
	members of your			
	family?			
S	Do you have			
	difficulty hearing			
	when someone			
	speaks in a			
	whisper?			
E	Do you feel			
	handicapped by a			
	hearing problem?			
S	Does a hearing			
	problem cause you			
	difficulty when			
	visiting friends,			
	relatives, or			
	neighbours?			
S	Does a hearing			
	problem cause you			
	to attend religious			
	services less often			
	than you would			
T.	like?			
E	Does a hearing			
	problem cause you			
	to have arguments			
	with family			
G	members?			
S	Does a hearing			
	problem cause you			
	difficulty when			

	listening to TV or radio?				
Е	Do you feel that any difficulty with your hearing limits or hampers your personal or social life?				
S	Does a hearing problem cause you difficulty when in a restaurant with relatives or friends?				
<b>TOTAL SCORE</b> = (sum of the points assigned to each of the items)					

## **Appendix P:** A Single Item Measure of Stress Symptoms (study 1 and 2)

Stress means a situation in which a person feels tense, restless, nervous or anxious or is unable to sleep at night because his/her mind is troubled all the time.

Do you feel this kind of stress these days?

- o Not at all
- Not really
- o Undecided
- o Somewhat
- o Very Much

### **Appendix R**: Single Item Sleep Quality Scale (study 1 and 2)

#### **INSTRUCTIONS:**

- The following question refers to your overall sleep quality for the **majority** of nights in the **past 7 days ONLY**.
- Please think about the quality of your sleep **overall**, such as how many hours of sleep you got, how easily you fell asleep, how often you woke up during the night (except to go to the bathroom), how often you woke up earlier than you had to in the morning, and how refreshing your sleep was.
- 1. During the **past 7 days**, how would you rate your sleep quality overall? (*Please mark only 1 box*)

