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**Tinnitus in Patients with Sensorineural Hearing Loss –  
Management, Quality of Life and Treatment Strategies**

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To my angels; Ramesh, Alma & Arvid



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## ABSTRACT

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Approximately 15% of Swedish people experience tinnitus, but only 2.4% experience severe problems. Treatment modalities for tinnitus vary, but the most common treatment is counseling. The majority of patients with tinnitus report some degree of hearing loss, and hearing aids have been used for many years in patients who suffer from both tinnitus and hearing impairment.

The aim of the present thesis was to investigate disease management, determine quality of life and identify treatment strategies for patients with tinnitus and sensorineural hearing loss.

The first two studies described here are retrospective, descriptive studies of patients who sought care for tinnitus and hearing loss at two Ear-Nose-Throat (ENT) clinics in Östergötland County, Sweden, during the years 2004 - 2007. Study I showed that 70% of the cohort had tinnitus; however, many did not initially receive a diagnosis of tinnitus. Information about vertigo, heredity for hearing loss and tinnitus, diabetes history, cardiovascular disease history and other factors related to health was often missing from the patients' medical records. The results could show that the overall scores using the Tinnitus Handicap Inventory (THI) were higher in female patients than in male patients. Although it is likely that hearing aids would be beneficial for the majority of these patients, 314 (44%) of the 714 total patients had hearing aids. Furthermore, the outcomes from study II demonstrated that a majority of the patients (61%) who were dissatisfied with the care they had obtained had no hearing aids. This finding may indicate that the fitting of hearing aids is an important treatment for patients with both tinnitus and hearing loss.

Studies III and IV were prospective studies. Data collection was based on patients who sought care for tinnitus and/or hearing loss at the ENT clinic in Linköping during 2012-2013. In study III, 92 patients were divided into two groups: one group contained individuals with both tinnitus and hearing loss, and the other group contained patients with only hearing loss. The patients were assessed using the Reading Span test, the Hearing in Noise Test (HINT) and three questionnaires (the THI, the Hearing Handicap Inventory for Elderly and the Pittsburgh Sleep Quality Index) at baseline and follow-up. The results from the age-matched subgroups (n=30+30) generated from the full clinical groups (46+46) showed significantly improved Reading Span test performance and sleep quality in patients with both tinnitus and hearing loss. Similar results were observed in our full clinical population (n=46+46). However, the interpretation of this finding is difficult due to age differences between the groups. In conclusion, hearing aid fitting had a significantly positive impact on working memory capacity and sleep quality in patients with both tinnitus and hearing loss compared with patients with only hearing loss.

In study IV, a brief Motivational Interviewing (MI) guide was integrated into the hearing rehabilitation process for 23 patients with both tinnitus and hearing loss, and they were compared against a control group (n=23) of patients with both tinnitus and hearing loss who underwent traditional hearing rehabilitation. The results showed that the patients who received the brief MI guide required fewer visits to complete their hearing rehabilitation compared with the patients in the control group. In addition, there was a significant difference in THI scores between the groups, which indicated that the intervention reduced tinnitus annoyance more in the MI group. Furthermore, both groups showed higher scores at follow-up compared with baseline on the International Outcome Inventory for Hearing Aids (IOI-HA) scale, which indicated that both approaches showed a positive effect on hearing aid satisfaction.

Study V was a retrospective, descriptive study that focused on a part of a Stepped Care model and included patients who participated in half-day tinnitus information meetings from 2004 to 2011 in the audiology clinic at Linköping University Hospital. A total of 426 tinnitus patients with complete questionnaires (the THI and the Hospital Anxiety and Depression Scale, HADS) were included in the study. The results showed significant decreases in scores on the THI and the anxiety module of the HADS before and after the information session. However, there were no statistically significant changes in the depression module of the HADS. In conclusion, this thesis underscores the importance of hearing impairment, cognitive variables and motivational procedures in the management of tinnitus. Multidisciplinary group information needs to be further validated.

## LIST OF ORIGINAL PAPERS

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This thesis is based on the following papers, which will be referred to in the text by their roman numerals (I-V). All papers are reprinted with permission from the publishers:

I. Zarenoe R, Ledin T. (2013). A cohort study of patients with tinnitus and sensorineural hearing loss in a Swedish population. *Auris Nasus Larynx*, 40: 41-45

II. Zarenoe R, Ledin T. (2014). Quality of life in patients with tinnitus and sensorineural hearing loss. *B-ENT*, 10: 41-51.

III. Zarenoe R, Hällgren M, Andersson G, Ledin T. (In press). Working Memory, Sleep, and Hearing Problems in Patients with Tinnitus and Hearing Loss Fitted with Hearing Aids. *Journal of the American Academy of Audiology*.

IV. Zarenoe R, Lindhe Söderlund L, Andersson G, Ledin T. (2016). Motivational interviewing as an adjunct to hearing rehabilitation for patients with tinnitus: a randomized controlled pilot trial. *Journal of the American Academy of Audiology*, 27(8):669–676.

V. Zarenoe R, Bohn T, Dahl J, Ledin T, Andersson G. on behalf of the Östergötland tinnitus team. Multidisciplinary group information for patients with tinnitus: an open trial. (Manuscript).

## ABBREVIATIONS

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<b>ABR</b>	Auditory brainstem response
<b>ACT</b>	Acceptance and Commitment Therapy
<b>ANOVA</b>	Analysis of variance
<b>CANS</b>	The central auditory nervous system
<b>CI</b>	Cochlear implant
<b>dB</b>	deciBel
<b>ENT</b>	Ear, Nose and Throat
<b>HADS</b>	Hospital Anxiety and Depression Scale
<b>HHIE</b>	The Hearing Handicap Inventory for the Elderly
<b>HINT</b>	Hearing In Noise Test
<b>HL</b>	Hearing level
<b>IOI-HA</b>	The International Outcome Inventory for Hearing Aids
<b>ISI</b>	Insomnia Severity Index
<b>ISO</b>	International Organization for Standardization
<b>ISSNHL</b>	Idiopathic Sudden Sensorineural Hearing Loss
<b>kHz</b>	kiloHertz
<b>MD</b>	Menière's disease
<b>MI</b>	Motivational Interviewing
<b>MRI</b>	Magnetic resonance imaging
<b>NIHL</b>	Noise induced hearing loss
<b>PTA</b>	Pure tone average
<b>PSQI</b>	Pittsburgh Sleep Quality Index
<b>QOLI</b>	Quality of Life Inventory
<b>SD</b>	Standard deviation
<b>SNHL</b>	Sensorineural hearing loss
<b>SOAE</b>	Spontaneous Otoacoustic Emission
<b>THI</b>	Tinnitus Handicap Inventory
<b>VS</b>	vestibular schwannoma
<b>WHO</b>	World Health Organization
<b>WM</b>	Working Memory
<b>WMC</b>	Working Memory Capacity

## INTRODUCTION

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Tinnitus is a common condition in Western populations (Axelsson & Ringdahl, 1989; Rosenhall & Karlsson, 1991; Sindhusake et al., 2003). The verb *tinnire* is derived from Latin and means to buzz, hum, jingle, or ring. The World Health Organization (WHO) has warned that hearing-related diseases including tinnitus will be one of the ten most prevalent disease categories in the near future (WHO, 2004). Tinnitus is an auditory symptom and is often associated with hearing loss. It may be described in many different ways, such as the presence of constant or pulsating high or low frequency sounds or sometimes more complicated sounds. The sound level may vary from being barely noticeable to very disturbing, and this perception varies both between and within individuals over time. Tinnitus most commonly occurs bilaterally (Andersson et al., 2005). Sensorineural hearing loss (SNHL) refers to hearing loss that results from damage to the cochlea or the auditory nerve. SNHL is most commonly associated with normal aging, a reduction in cochlear hair cells or damage to the auditory nerve. Other than cochlear implantation (CI), surgical treatments are not possible for patients with SNHL; therefore, hearing aid(s) are often recommended for these patients.

Although hearing loss is not life threatening, the resulting loss in speech recognition may have a significant impact on patient quality of life (Dalton et al., 2003). The same argument can also be made for patients who suffer from tinnitus (Corcetti et al., 2009) because tinnitus often has a negative impact on the physical and emotional well-being of patients. Tinnitus may cause increased stress levels, problems concentrating, sleep disturbances and a perceived reduced ability to hear. These disturbances may have negative effects on the individual's social life, relationships and ability to work (Henry et al., 2005; Kochkin, & Tyler 2008).

Hearing aid fitting has been a useful treatment for patients suffering from both tinnitus and hearing impairment (Searchfield et al., 2010). Unfortunately, patients with hearing loss are unaware of the possibility of improving tinnitus symptoms through amplification (Kochkin, 2007). There are many explanations for this low uptake of hearing aids. However, regardless of the reasons, the low uptake rates in patients with tinnitus and hearing loss is worrisome because this adverse combination may diminish quality of life more than either condition alone.

Furthermore, to increase interest in hearing aid usage in patients with tinnitus and hearing loss, Motivational Interviewing, which is based on patient-centered care, could be implemented as a useful tool in the hearing rehabilitation process.

The studies in this thesis aim to improve the scientific knowledge concerning not only tinnitus and its effect on patients with SNHL but also the most common investigations and rehabilitation

methods that are used in patients with tinnitus, as well as how tinnitus can affect cognitive processing in patients and the possibility of using MI as a motivational procedure in the management of tinnitus.

## BACKGROUND

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### **Tinnitus**

#### *A historical description*

Tinnitus has likely troubled humanity for ages. The first written account of medical treatment for tinnitus came from the Egyptians and Mesopotamians (Stephens, 1984). In ancient Greece and Rome, poetry was written that described tinnitus as a symptom of passionate love, jealousy and telepathy (Stephens, 1984).

The psychological aspects of tinnitus and the various psychological cures including rest, spa treatments and other similar remedies were investigated early (Stephens, 1984). Furthermore, Stephens recognized the importance of early treatment of tinnitus and believed that a long-term psychological consequence of tinnitus could be a change of the sound into auditory hallucinations.

Several authors (e.g., Vernon, 1981; Hazell, 1979) have attributed the following statement on masking, which is the earliest known statement on this feature, to Hippocrates: “Why is it that buzzing in the ears ceases if one makes a sound? Is it because a greater sound drives out the less?” In fact, this statement could have been the foundation for one of the most well-known tinnitus treatment models, namely Tinnitus Retraining Therapy (TRT) (Jastreboff & Hazell, 2004).

#### *Epidemiology*

Tinnitus is a common complaint in the global population ( Baguley et al. 2013). According to different studies with different age groups, the approximate prevalence of tinnitus is between 10 and 15% (Axelsson & Ringdahl, 1989; Rosenhall & Karlsson, 1991), (Cooper Jr, 1994; Scott & Lindberg, 2000; Andersson et al., 2002; Shargorodsky et al., 2010). Another common observation is that the prevalence of tinnitus increases with age (Baguley et al. 2013). Gender differences regarding tinnitus have been observed in many studies (Dineen et al., 1997; Shargorodsky et al., 2010; Seydel et al, 2013). In some studies, the female patients reported a greater tinnitus annoyance than male patients and they perceived more stress than men did (Seydel et al, 2013). Accordingly, there are several studies on tinnitus that show a slightly higher prevalence in females (Leske, 1981; Coles, 1984; Nondahl et al., 2007). However, the prevalence was greater in females below the age of 40 years, while tinnitus was more common in males between 40 and 70 years of age in other studies (Axelsson, 1999). Men have traditionally had

higher exposure to loud noise in the form of firecrackers, firearms (military), and noisy work environments. However, women are more severely bothered by their tinnitus (Axelsson & Ringdahl, 1989; Stouffer & Tyler, 1990).

Tinnitus can be divided into two categories. Objective tinnitus can be recorded objectively by a microphone or can be heard by another listener (Lustige, 2010). The sound can come from, for example, the carotid artery, auditory tube or temporomandibular joint (Noell & Meyerhoff, 2003; Crummer & Hassan, 2004).

The perceived localization of a patient's tinnitus can potentially be of diagnostic significance, particularly because unilateral tinnitus may be a symptom of an underlying vestibular schwannoma. In some previous studies, tinnitus was found to affect the left ear more commonly than the right ear, particularly in male patients (Meikle et al., 1984; Erlandsson et al., 1992). A possible explanation for the higher incidence of left-sided tinnitus has yet to be proposed. Furthermore, there is no evidence that left-sided tinnitus is more annoying than right-sided tinnitus (Andersson et al., 2005).

Tinnitus is often accompanied by some degree of hearing loss (Irvine et al., 2001; Sindhusake et al., 2003). This loss is usually a sensorineural impairment, either cochlear or retrocochlear, and can be due to aging, noise exposure or ototoxic drugs (Chung et al., 1984; Coles, 1984; Ahmad & Seidman, 2004). However, tinnitus may also be present in individuals with normal hearing (Stouffer & Tyler, 1990; Schaette & McAlpine, 2011); however, the difference in the annoyance level between the groups is unclear. Symptoms may originate in several different places in the auditory system and may have various causes, such as conductive hearing loss (e.g., otosclerosis and infections in the middle ear) or problems in the cochlea (e.g., Menière's disease, sudden sensorineural hearing loss, and presbycusis) (Billue, 1998). Reports have suggested that patients with normal hearing, as assessed by various clinical tests, may have cochlear damage or hearing loss at frequencies above 8 kHz (Weisz et al., 2005). The primary lesion in most cases of hearing loss resides in the hair cells and/or spiral ganglion neurons. Studies that have investigated the effects of noise exposure or ototoxic drugs have shown that damage to the inner or outer cochlear hair cells increases the threshold of the auditory nerve fiber (Dallos & Harris, 1978; Schmiedt & Zwislocki, 1980; Liberman & Mulroy, 1984; Devarajan et al., 2012).

The loudness of tinnitus sounds fluctuates in the majority of individuals (Erlandsson et al., 1992; Devarajan et al., 2012). The volume can be altered by exposure to loud sounds, nerve tension, increased blood pressure, lack of energy and some chemical substances, such as drugs, alcohol, caffeine and tobacco.

The current models to investigate the origin of tinnitus in humans argue that damage to hair cells can encourage an imbalance in lateral inhibition on other neuronal levels and can cause central plasticity (Eggermont, 2003).

*Lateral inhibition* occurs when the activity of an excited neuron reduces the activity of nearby neurons in the same area. A reduction in the spontaneous activity of nerve fibers with different characteristic frequencies in the hearing loss range could result in a reduction of lateral inhibition at more central levels. This reduced lateral inhibition of neurons induces hypersensitivity and hyperactivity in these neurons (Eggermont, 2003), which are highly likely to be interpreted as a sound stimulation (Eggermont & Roberts, 2004).

## **Theories**

### *Non-cochlear models*

The neurophysiological theory of tinnitus was first presented by Jastreboff et al. (1996). This theory involves auditory perception, emotional and reactive systems and a combination of peripheral and central dysfunction (Attias et al., 2002). Jastreboff et al. (1996) suggested that sound interpretation in tinnitus involves the limbic system and the autonomous nervous system (Jastreboff et al., 1996). The tinnitus sound is interpreted in a negative way, which makes the individual aware of something abnormal and allows the sound to be perceived as a distressing symptom. Several researchers have described that the interpretation of the tinnitus sound could be associated with an adverse episode in the individual's life (Jastreboff et al., 1996; Jastreboff & Jastreboff, 2000; Henry & Wilson, 2001).

Another theory suggests that tinnitus occurs from the adoption of a temporal pattern in the activity of the auditory nerve (Eggermont & Roberts, 2004). Calcium is very important for the cochlea and its hair cells (Zenner & Ernst, 1993), and increases in the amount of calcium in the hair cells could lead to amplified signaling to the brain. If this signaling occurs in a dysfunctional cochlea, it may lead to increased neurotransmitter release from inner hair cells and increased activity in the auditory nerve fibers in the form of cascade signaling (burst firings). The synchronization of activity in the small nerve fibers could cause the perception of the tinnitus sound (Baguley, 2002).

### *Cochlear models*

Tinnitus in individuals with normal hearing is often associated with a varying degree of cochlear dysfunction (Jakes et al., 1986; Satar et al., 2003; Shim et al., 2009). Some researchers believe that tinnitus can be measured objectively by measuring spontaneous otoacoustic emissions

(SOAEs); however, studies have shown that 38-60% of individuals with normal hearing could also have these measurable emissions (Penner, 1990; Kim et al., 2010).

The tectorial membrane can be clamped toward the inner hair cell's cilium due to toxic drugs or loud noises. This change can result in a depolarization of the inner hair cells (Jastreboff et al., 1996; Baguley, 2002; Ricci, 2003). The frequency of the tinnitus sound in these individuals is often matched to the frequency of their hearing loss (Eggermont, 2003). Damaged outer hair cells on the basilar membrane may sometimes contribute to the onset of tinnitus (LePage, 1987). The normal function of the outer hair cells is to enhance the sound before it is received by the inner hair cells. The outer hair cells also check the sensitivity of the inner hair cells' operating level by assessing the difference between the sound transmitted and the sound that the brain normally interprets as no sound (Baguley, 2002; Ricci, 2003). When the outer hair cells lose their mobility, they also lose the ability to control the normal function of the inner hair cells (Ricci, 2003). This loss of function modifies normal input such that what is typically interpreted as a normal state is now perceived as tinnitus (Baguley, 2002).

In the auditory cortex, all frequencies are tonotopically mapped to show the coding of the different frequencies at the basilar membrane. The tonotopic mapping reorganizes after an injury (Eggermont & Roberts, 2004); the normal functions of the neurons in the cortex are modified, which implies that these neurons do not respond to their own frequencies or to the frequencies from the non-affected area (Eggermont & Roberts, 2004).

### *Phantom perception*

Tinnitus is not the only phantom perception in humans and is similar to phantom pain (Goodhill, 1950). Cortical reorganization, such as that which occurs in the case of phantom pain, occurs in the auditory cortex after peripheral changes (Baguley, 2002; Weisz et al., 2007). Damages to specific parts of the hair cells can lead to a reduction in activity in the cortical area at the corresponding frequencies (Baguley, 2002). One consequence of this reorganization is that a disproportionate number of neurons become sensitive to frequencies in the upper and lower limits of the hearing loss (Dietrich et al., 2001). Spontaneous activity in these areas can be perceived as the tinnitus sound (Baguley, 2002).

### **Measurement of tinnitus**

By developing and improving the procedures for testing hearing ability, it has been easier to more precisely determine hearing thresholds. There have been many attempts to measure the sound, loudness and pitch of tinnitus (Penner & Klafter, 1992; Mitchell et al., 1993). Because

tinnitus is subjectively perceived, a direct measurement of the degree of tinnitus severity is often obtained through self-report questionnaires. The THI is among the most validated and useful method to measure the impact of tinnitus on a patient's life (Newman et al., 1996).

Because tinnitus can have an impact on different aspects of a patient's life, secondary questionnaires that are related to the patient's sleep, anxiety, depression and other health problems are commonly administered. Among those, the HADS (Zigmond & Snaith, 1983) and the Insomnia Severity Index (ISI) (Bastien et al., 2001) are the most commonly used in tinnitus research. The HADS and the Quality of Life Inventory (QOLI) measure patients' psychological mental health and provide a profile of patients' life situations, whereas the ISI describes the patients' sleeping habits. These questionnaires enable a better understanding of the patients' life situations, which can significantly impact the rehabilitation process.

### **Treatment models**

Despite the existence of several treatment models, there is no permanent cure for tinnitus.

Improving the circulation in the cochlea that may have been altered following certain types of insults or trauma can help in the recovery process (Hultcrantz, 1988); however, the results from another study showed that vasodilators do not alter tinnitus (Hulshof & Vermeij, 1987).

Different treatment options have been used in the management of tinnitus, including surgical, drug and psychological treatments. Surgery has been performed on patients when tinnitus is secondary to an underlying condition, such as otosclerosis or vestibular schwannoma, VS, (Andersson et al., 2005). Because both depression and anxiety are frequently present in patients with tinnitus, psychoactive drugs may suppress the annoyance of tinnitus.

Masking is another treatment that covers up or masks the tinnitus sound by providing the patient with an external, manufactured sound (Jastreboff & Hazell, 2004). However, the generators that produce the sound can only offer a limited range of different sounds, which do not satisfy the majority of the patients.

The list of alternative therapies (e.g., acupuncture, music therapy, various herbal therapies, and relaxation) is long and demonstrates the strong need of patients who suffer from tinnitus to find relief. Jastreboff and Hazell (2004) emphasized the importance of counseling and its effect on tinnitus patients (Jastreboff & Hazell, 2004). Listening and confirming patients' complaints about their complex problem and providing adequate advice that could reduce the tinnitus annoyance constitute an appropriate method that can be used by clinicians to assist patients. However, it is generally difficult to differentiate the effects of alternative medicine approaches such as counseling from the effects of the actual treatment administered to the patients.

In most cases, an individualized treatment strategy can provide the best results (Jastreboff et al., 1996; Noell, 2003; Kaldo & Andersson, 2004; Westin et al., 2011). TRT and cognitive behavioral therapy (CBT) can be used as treatment modalities in patients with tinnitus. TRT consists of two parts, namely directive counseling and sound enrichment, the latter of which is accomplished by a white noise generator. The importance of the use of a sound generator in TRT is not clearly described. Therefore, stimulations using every-day sounds have also been recommended. In this case, if the patient has a hearing impairment, hearing aids combined with an integrated sound generator are recommended. CBT is characterized by a focus on how thoughts, behavior and reactions affect each other. A successful treatment should eliminate the disturbance caused by the tinnitus and help patients accept and deal with their tinnitus. The purpose of hearing aid fitting in patients with both tinnitus and hearing loss is to reinforce sounds that patients have difficulty hearing due to their hearing loss and to provide external auditory stimuli that can mask the tinnitus. The use of hearing aids has become widespread, and they are currently offered in many clinics worldwide. Furthermore, hearing aid use in patients with both tinnitus and hearing loss is recommended (Jastreboff & Hazell, 2004). Modern hearing aids with advanced programs can suppress the background environmental sounds that can enhance tinnitus.

As the number of treatment options for patients suffering from tinnitus has increased, caregivers have searched for additional rehabilitation alternatives that may have a greater impact on patients' sensitivity to their tinnitus. One of these options is acceptance and commitment (ACT) therapy. The goal of ACT is to increase the quality of life rather than to try and remove the annoyance or pain sensation.

### *Tinnitus Retraining Therapy (TRT)*

The hypothesis of TRT is that two different processes of non-habituation create the perception and annoyance associated with tinnitus. A combination of counseling and sound therapy is used in TRT. Sound therapy, which can be conducted with or without an instrument, provides sound at the pinnae using a device that generates white noise. The purpose of a noise generator is to provide a background sound. In TRT, hearing aids are also used for patients with hearing loss. Initial evidence suggests that TRT can be an effective treatment for patients suffering from tinnitus (Henry et al., 2008). To implement TRT in clinical practice, clinicians use a combination of sound therapy within a strict framework and educational counseling according to a detailed procedure (Jastreboff & Hazell, 2004). Many studies have discussed the use and evaluated the effectiveness of TRT. However, controlled trials with validated outcome measures are needed to

support the efficacy of TRT (Phillips & McFerran, 2010). Phillips and McFerran (2010) referenced only one study with 123 participants that was published in two separate journals (Henry et al., 2006a), (Henry et al., 2006b). The results of that study suggested a considerable benefit of TRT in the treatment of tinnitus. However, Phillips and McFerran (2010) questioned the quality of that single study and suggested that the evidence was not robust enough for firm conclusions to be drawn.

### *Cognitive Behavioral Therapy (CBT)*

CBT is a treatment approach used to identify and modify behavior, thoughts and cognitions that are disruptive for the individual (Balow, 2001). The CBT approach is based on a cognitive-behavioral model of tinnitus (Henry & Wilson, 2001; Andersson et al., 2002; Andersson et al., 2005). Patients with tinnitus have reported difficulties concentrating and have claimed that their tinnitus is distracting. Through CBT, clinicians can help patients accept their tinnitus and assist them in ignoring the sound (Kaldo & Andersson, 2004). CBT can be conducted in small groups or individually and is usually provided over six to ten sessions that occur on a weekly basis (Kröner-Herwig et al., 1995; Martinez-Devesa et al., 2010).

### *Stepped Care model*

To reduce the annoyance of tinnitus, all identifiable factors contributing to tinnitus should be addressed during treatment (Andersson et al, 2005). Most treatments have their focus either on reversing the maladaptive changes that may occur in the auditory processing centers of the brain or reducing the patient's emotional response to their tinnitus. Due to many different factors underlying tinnitus and its various comorbidities, effective treatment requires a multidimensional approach (Daugherty & Wazen, 2010, Langguth et al, 2013). Patient health literacy has been the focus of several studies. Ferguson (2013) discussed the role of health literacy in the care of the patient and defined it as a level of intelligence and communication skills that a patient must have to make informed decisions regarding what is best for them (Ferguson 2013). Many researchers have been concerned about the lack of appreciation for patient health literacy. Therefore, an educational session that provides information about tinnitus and is offered by a multiprofessional team could promote acceptance of the condition by providing the patient with adequate knowledge and skills. A multidisciplinary management approach is often necessary and helpful for patients with complex symptomatology (Andersson et al, 2005). The use of basic techniques such as education, counseling and empathetic support along with pharmacological or other treatment protocols could lead to the optimal outcome.

A stepwise multidisciplinary treatment program consisting of counseling, cognitive behavioral therapy, and auditory stimulation was recently evaluated in a large randomized controlled trial (Cima et al., 2012). The specialized treatment program started with audiological measurements/treatment and counseling, followed by optional multidisciplinary group or individual treatment sessions for a period of 12 weeks. These sessions involved clinical psychologists, exercise therapists, physiotherapists, audiologists, social workers, and speech therapists. The results of this study underline the importance of an interdisciplinary, stepwise approach to the treatment of tinnitus. Compared with the standard treatment over an observation period of 12 months, the stepwise multidisciplinary treatment program showed significant improvements in the quality of life, the severity of tinnitus and the degree of disability caused by tinnitus (Cima et al., 2012).

### *Hearing Aids Fitting*

For many years, hearing aid fitting has been a useful treatment for patients suffering from both tinnitus and hearing impairment (Searchfield et al., 2010). The amplification of sound by hearing aids can increase the level of neural activity, which can reduce the gap between the tinnitus stimuli and the background neural activity (Parra & Pearlmutter, 2007; Searchfield, 2008). The use of hearing aids can also indirectly help patients with both tinnitus and hearing impairment by reducing the negative effects of tinnitus annoyance, regardless of the severity of the hearing loss (Surr et al., 1985; Carmen & Uram, 2002).

Recent hearing aid studies have verified the effects of the currently available technology and compared more sophisticated hearing aids with less sophisticated hearing aids used in the management of tinnitus (Trotter & Donaldson, 2008; Searchfield et al., 2010). Patients who used hearing aids combined with counseling obtained approximately twice the reduction in their tinnitus handicap than those who preferred only counseling (Aazh et al., 2009).

Despite the obvious benefits of using hearing aids, many patients with hearing loss do not consider hearing aids as a treatment option (Aazh et al., 2009).

## Hearing impairment

Tinnitus patients often report hearing loss. This loss is usually a sensorineural impairment (cochlear or retrocochlear) encompassing the entire spectrum of ear diseases, such as exposure to noise, the use of ototoxic drugs and the slow process of hearing impairment in presbycusis (Sindhusake et al., 2003; Sindhusake et al., 2004). The pathophysiological basis of the tinnitus that often coexists with SNHL will be discussed here. Inner ear diseases can lead to hearing loss and may also result in tinnitus (Hoffman & Reed, 2004). However, not everyone suffering from hearing loss will develop tinnitus, and not everyone who suffers from tinnitus has a hearing impairment (Kim et al., 2010). The findings from a recent study showed that 7.4 to 20% of tinnitus patients did not exhibit a hearing loss at any frequency of conventional pure tone audiometry (Shim et al., 2009). A recent study examined the neuroanatomical alterations associated with hearing loss and tinnitus in three patient groups: those with both hearing loss and tinnitus, those with hearing loss without tinnitus and normal-hearing controls without tinnitus (Husain et al., 2011). The findings showed that the individuals with only hearing loss had significantly less gray matter in the anterior cingulate, superior gyri and medial frontal gyri compared with those with both hearing loss and tinnitus. In addition, the authors found a further reduction in the superior temporal gyrus in the hearing loss group compared with the tinnitus group. The results of an investigation of the effects of hearing loss alone showed that the gray matter loss in the superior and medial frontal gyri in patients with hearing loss was similar to the normal-hearing controls. A loss in the fractional anisotropy values in the right superior and inferior longitudinal fasciculi, corticospinal tract, inferior fronto-occipital tract, superior occipital fasciculus, and anterior thalamic radiation in the hearing loss group compared with the normal-hearing patients was also shown in this study (Husain et al., 2011). Future research could explain why different tinnitus treatments are beneficial for some patients but have no effect on others. A relationship between the development of a tinnitus perception and the neural plasticity of the central auditory system (including the auditory cortex) often exists (Bauer et al., 2008; Engineer et al., 2011). According to Jastreboff and Hazell (2004), this imbalance of neural activity that can cause tinnitus-related changes affects type I and type II fibers of the auditory nerve (Jastreboff & Hazell, 2004). The result can be a bursting activity at the dorsal cochlear nuclei level in the brainstem that could lead to a disturbance of the afferent inputs to the cochlear pathways. After further amplification within the auditory pathways, this process may be perceived as tinnitus. However, not every individual subjected to the same process subsequently suffers from tinnitus. Generally, the answer to this phenomenon is not found in the psychophysical parameters of tinnitus (Baguley et al., 2013). Loud tinnitus or tinnitus sounds at a certain frequency could lead

to increased distress, and patients who experience more complex sounds tend to report greater problems (Dineen et al., 1997). Another possible explanation for this result could be that pre-existing psychological characteristics affect the way in which a patient reacts to tinnitus (Andersson et al., 2005).

For a general understanding of the association between tinnitus and hearing loss, it is important to discuss a number of otological pathologies to understand the various associated symptoms.

The main subtypes of lesions associated with hearing impairment in humans, based on the location of the lesion, are central and peripheral lesions, the latter of which are divided into sensorineural and conductive lesions.

Conductive hearing loss is caused by a disease or damage to the eardrum or middle ear and usually results in reduced sensitivity over the entire frequency range. The signal transmission from the middle ear to the inner ear decreases independently of the sound pressure level of the stimulus. Conductive hearing loss can be detected by audiometry, where an air-bone gap above 10 dB can indicate suboptimal transmission of sound between the middle ear and inner ear.

Diseases or damage to hair cells cause a reduction in sensory function. SNHL can also be detected by an audiometry reading that indicates no gap between the air and bone thresholds, i.e., the air-bone conduction is equal to the bone conduction. This result suggests that signal transmission from the middle ear to the inner ear functions well, but some other obstacle prevents the sound from being perceived by the brain. SNHL is the most common type of hearing impairment.

Hearing impairment can sometimes be due to a combination of conductive hearing loss and SNHL, termed *mixed hearing loss*, and can sometimes be due to damage to the central pathways, termed *central hearing loss*.

### **Working memory**

The concept of working memory (WM) was first introduced in the 1960s and 1970s (Baddeley & Hitch 1974). Until that point, the ability to briefly store information in memory was referred to as short-term memory and was described as passive. This definition has now changed and WM refers to our ability to store, process and use information in the moment (Baddeley & Hirsh, 2010).

WM is the system that manages the temporary storage and processing of information necessary for thought processes and language capabilities (Baddeley 2012). Complex cognitive tasks such as speech comprehension are performed while task-relevant information is maintained. WM enables more than one idea to be processed at a time and is crucial for the ability to solve more complex cognitive tasks. WM is active when people are focused on a task where they have to do

more than one thing at a time. WM is linked to our perception and is also dependent on a good memory where essential information can be recalled and used quickly.

### *Component model*

A theoretical WM model is the “component model” (Baddeley & Hitch 1974, Baddeley 1984). According to this model, there are four main components: a modality-free central executive component that resembles attention, a phonological loop that contains information in a speech-based (phonological) form, a visuo-spatial sketchpad that is specific for spatial and visual coding and an episodic buffer that is a temporary storage system, in which information from the phonological loop, the visuo-spatial sketchpad and long-term memory are maintained and integrated (Baddeley 2001, Repovs & Baddeley 2006).

The central executive component is involved in demanding cognitive tasks and when shifting attention. It enables the focus and division of attention (Repovs & Baddeley 2006). It is the most important and versatile component of the WM system; however, its role is not yet fully understood (Eysenck & Keane 2005). The phonological loop is the component that specializes in holding verbal information in the WM (Repovs & Baddeley 2006) and comprises two components: a phonological short-term storage and a sub-vocal repetition system. In the phonological short-term storage, preserved memories leave traces for a few seconds before fading, unless the memory is refreshed by processes in the subvocal repetition (Baddeley 2003). The most reliable method of measuring the storage capacity of the phonological loop is to repeat verbal stimuli in the correct order (Gathercole et al., 2004), e.g., using digit span (Geers 2003). The visuo-spatial sketchpad is a temporary sub-system in the WM where spatial and/or visual information may be stored and processed (Baddeley, 2003, Eysenck & Keane, 2005). The episodic buffer is a temporary system where information from different modalities is integrated and stored in a continuous representation (Baddeley 2003). These modalities may be from other components in the WM or may be information retrieved from long-term memory (Repovs & Baddeley 2006).

Research has shown that cognitive function, and therefore WM, declines with increasing age (Park 1999), and phonological ability declines when auditory stimulation decreases over time, i.e., due to an age-related loss in hearing (Andersson & Lyxell 1998, Andersson 2001, Hällgren et al., 2001). In hearing-impaired patients, the auditory portion of the sensory-perceptual system may be adversely affected, which leads to difficulties in analyzing and retrieving information from acoustic signals and results in an incomplete auditory sensory signal in the form of a false

signal to the WM. This false signal may lead to impaired phonological representations in the long-term memory of patients with hearing loss (Andersson 2001).

#### *Working memory and tinnitus*

Tinnitus can potentially affect cognitive processing and thereby WMC (Ricketts, 2005). The mechanisms of tinnitus and patients' reactions to tinnitus were studied by Tyler et al.(1992). Furthermore, Andersson and McKenna (2006) discussed the role of cognition in the experience of tinnitus and argued for a model based on the cognitive influences on tinnitus. This model proposed that when tinnitus becomes a significant problem for the patient, it interferes with the patient's cognitive processing on the following three levels: cognitive performance, emotional processing, and conscious appraisal (Andersson and McKenna, 2006). Cognitive factors, such as selective attention, autobiographical memory specificity, impaired performance on the color Stroop test, and decreased working memory capacity (WMC), have been studied in tinnitus (Andersson et al, 2000; Hallam et al, 2004; Stevens et al, 2007). The links between tinnitus and cognition are not clearly outlined, and there is no established theory that identifies the cognitive function that is most likely associated with tinnitus interference (Mohamad et al, 2015). Still, tasks that demand WMC are likely to be involved for patients suffering from tinnitus because external "irrelevant" sounds (i.e., ambient sounds) influence working memory task performance (Jones & Macken, 1993).

#### **Audiological rehabilitation**

The aim of audiological rehabilitation is to enable the patient to participate in daily activities despite their disability, and it is an intervention where instruction, counseling, hearing aid fitting and communication training are used to reduce the impact of hearing loss on the individual's life (Boothroyd 2007, Hull 2001). Audiological rehabilitation may be performed both individually and in groups. Some aspects, such as information, communication training and counseling, may be applied using a group design (Alpiner & McCarthy 2000). Moreover, Alpiner & McCarthy (2000) argued that audiological rehabilitation in groups could enhance the individual's perception of their hearing loss and their perception of the advantages of communication strategies.

Counseling is the central part of audiological rehabilitation and is divided into content counseling and personal-adjustment counseling (Clark & English, 2004). Content counseling aims to help the patient address her or his issues and concerns and overcome them during the rehabilitation. This process may include training or education prior to hearing aid fitting and is therefore, a significant aspect of the audiologist's profession. In contrast, personal-adjustment

counseling provides support and helps the patient manage the emotional impact of the information provided in the informational counseling (Clark & English 2004).

To ensure that an individual with hearing loss perceives a conversation satisfactorily, some elements, in addition to hearing aids, must be mastered by the patient to fill the gaps caused by the hearing loss (Alpiner & McCarty 2000). One of these elements is communication strategies (Danermark 2005). These strategies are important for the patient because a lack of communication skills could have negative consequences in the life of that individual.

#### *Conventional audiological rehabilitation*

The hearing clinic in Linköping, Sweden, is an example of a conventional audiological rehabilitation program and can be described as a series of visits where 1) during the first visit, the patient undergoes pure-tone audiometry and 2) subsequently, the patient receives information on the outcome while the audiologist gathers the medical case history. In the case of asymmetric hearing loss or vertigo, the patient is referred to an ENT clinician. Otherwise, if hearing loss is noted, the patient is offered hearing aid fitting, where at the first visit, the audiologist provides informational counseling on the hearing loss and hearing aids. In cooperation with the patient, the audiologist frames some key goals and forms a plan to achieve them. Based on the patient's hearing loss, the ability to handle the hearing aid and the preferences of the patient, the audiologist proposes hearing aids that may fit, according to Swedish guidelines (Arlinger et al., 1994, Smeds & Leijon 2000). If the patient approves of the selection, the audiologist adjusts the amplification. The follow-up visits comprise informational counseling, personal-adjustment counseling, functional evaluation and fine-tuning of the amplification.

However, typical hearing aid rehabilitation in Sweden may vary due to regional, economic and political guidelines, and the individual audiologist's strategies, and generally includes between three and five visits. In some cases, the number of visits and the time spent on counseling may vary from this norm.

#### *Patient-centered rehabilitation*

Educational programs for hearing rehabilitation purposes have been offered to patients with hearing loss for some time. A number of studies have investigated the effects of individual pre-fitting counseling/educational interventions in hearing aid users (Gusseklou et al., 2003; Kramer et al., 2005), whereas others have examined the effects of individual post-fitting counseling/education interventions (Taylor & Jurma 1999; Sweetow & Sabes 2006).

The model of patient care called *patient-centered rehabilitation* was presented in 1964 by Balint. He indicated that two perspectives were involved in rehabilitation: the clinician's interpretation of the health problem in terms of the indications and symptoms and the patient's perception of the rehabilitation in terms of the experience (Balint 1964). The collaboration between the clinician and the patient should result in the development of a common perception of the patient's needs.

The core component of the patient-centered care approach is communication between the health-care provider and the patient (Dwamena et al., 2012). In practice, a patient-centered model indicates a partnership between the clinician and patient that uses shared decision making and support/coping strategies (de Silva, 2014). One method that fits this theory is Motivational Interviewing (MI), which is a counseling method. Over the last three decades, MI has expanded to a wide range of fields, such as the abuse of alcohol, tobacco, or drugs, problems with diet, physical activity and diabetes, mental health and somatic medicine (Miller & Rose 2009). In the patient-centered model, clinicians do not see themselves as uncensored parts and neutral dispensers of therapy. They must build a meaningful relationship by attending carefully to their patient's problems in their daily life and guiding them to empowerment (Bechtel & Ness 2010).

### **Motivational Interviewing (MI)**

MI was developed in the 1980s and early 1990s by psychologists William R. Miller and Stephen Rollnick (Wagner & Connors 2010). MI was originally developed for patients in drug treatment; however, over the last three decades, MI has been used in many other fields. The process of developing MI may have been like the model itself, i.e., a gradual process of listening and reflecting to determine understanding and clarification. This approach has been supported by various theoretical models of human processes and behavioral change (Miller & Rollnick 1991). There are three definitions of MI, the first of which was generated by Miller in 1983. Miller described MI as "a way of talking with people to evoke and strengthen their personal motivation for change" (Miller 1983). Rollnick, William and Miller presented the second definition of MI in the 1990s as follows: "motivational interviewing is a directive, client-centered counseling style for eliciting behavior change by helping clients to explore and resolve ambivalence". The most recent definition of MI is "a collaborative, person-centered form of guiding to elicit and strengthen motivation for change" (Miller & Miller 2009, page 130). An additional definition of MI is a "collaborative, goal-oriented style of communication with particular attention to the language of change" (Miller and Rollnick, 2012, page 29). However, the most-recent definition of MI is a "MI is a person-centered counseling style for addressing the common problem of

ambivalence about change” (Miller & Rollnick 2013, page 21). This thesis is based on the definitions from 2012 and 2013.

MI has been observed to be effective for various forms of behavior change; therefore, it has expanded to a wide range of fields, such as the abuse of alcohol or problems with physical activity, cardiovascular disease and mental health (Bertholet et al., 2005, Schoener et al., 2006, Hardcastle et al., 2012). MI has become an increasingly common practice in health-care. MI applies person-centered skills within a flexible structure to help patient progress and individually motivated requests for change by exploring the positive and negative sides of change (Miller & Rollnick 1991).

### *Theories*

The Stages of Change model was developed separately and independently from MI in the 1980s (Prochaska & DiClemente 1983). The Self-determination Theory, which is based on four sub-theories (the cognitive evaluation theory, the organismic integration theory, the causality orientations theory and the basic needs theory) (Hagger & Chatzisarantis 2007), may offer a theoretical framework to understand how changes occur in MI (Ginsberg et al., 2002). The Self-determination Theory, which is focused on the belief that humans show persistent positive features and that they frequently show determination, action and commitment in their lives, may address personality development and self-motivated behavior (Deci & Ryan 1985). More specifically, MI may generate self-motivated behavioral changes by endorsing the internalization and combination of a new behavior that is more aligned with the individual’s broader values, goals and sense of self (Markland et al., 2005).

### *The spirit of MI*

Three key elements of MI are defined and include *the overall spirit of MI*; *collaboration* between the therapist and the patient/client, *evoking or drawing out* the patient’s thoughts, ideas on change and highlighting the *autonomy* of the patient (Rollnick et al., 2008, Miller & Rollnick 2013). Miller and Rollnick (2013) noted that any MI intervention should contain these elements. Using the term *collaboration*, Rollnick, Miller and Butler (2008) referred to the relationship between the therapist and the patient, which should be grounded in the point of view and the experiences of the patient. Thus, the risk of a more hierarchical relationship between the health-care professional and the patient is eliminated. However, collaboration does not mean that the health-care professionals must automatically agree with the patient on the nature of the problems or on changes that may be most appropriate to achieve optimal outcomes.

During MI, it is important that the MI practitioners activate the patient's own motivation via *evocations* of their own thoughts and ideas, rather than imposing the health-care professional's opinions to motivate and induce commitment to change (Rollnick et al., 2008). This process requires an understanding of the patient's perspective. The therapist should avoid convincing the patient of the need to implement changes in their lives. Again, his or her purpose is to "draw out" the patient's skills to implement the change thereby increasing the patient's own motivation. MI recognizes the power to induce change in the patient and empowers the patient's *autonomy* (Rollnick et al., 2008). All decisions on changes must be made by the patient. Thus, the patient is given responsibility for their decisions and actions. The therapist supports the patient by confirming that there are many *right ways* to change and by acknowledging that the change may occur in multiple ways.

### *The principles of MI*

MI involves four distinct principles that guide and strengthen the process: the expression of empathy, support of the patient's self-efficacy, rolling with resistance and developing discrepancies (Miller & Rollnick 2002).

*The expression of empathy* involves seeing the world through the patient's eyes and thinking or feeling about the world as the patient does. It is a central part and a defining feature of MI (Miller & Rollnick 1991). This approach provides the foundation for the patient to be heard and understood because the patient can then share their experiences honestly and in depth. The success of this process relies on the patient experiencing that the health-care professional is capable of seeing or feeling the world as they see or feel it.

Health-care professionals who practice MI believe that the patient is capable of accomplishing changes in their lives. However, the patient requires *support for their self-efficacy* to instill the hope that they can make these challenging changes (Miller & Rollnick 2002). The therapist should support the patient and help them believe in themselves by focusing on previous successes, which thereby encourages the skills and strengths that the patient already has.

Another critical part of MI is *rolling with resistance*. Resistance during the treatment process occurs when the patient experiences a possible conflict with the health-care professional in the sense that patient's view of a problem or solution does not match with the health-care professional's view. These experiences are based on the patient's ambivalence towards the change (Miller & Rollnick 2002). The health-care professional should avoid eliciting resistance by not confronting the patient. However, when resistance occurs, the health-care professional must reduce it, avoid a negative interaction and instead *roll with the resistance*. Every sign

(actions or statements) that demonstrates a resistance early in the treatment process should remain unchallenged.

To practice MI, the therapist uses specific techniques to bring the “MI spirit” to life, which demonstrate the four principles of MI and guide and provoke the patient to begin the process for change. Miller & Rollnick (2002) described these techniques as open-ended questions, reflective listening, affirmations, summarizing and eliciting change talk.

Using *open-ended questions*, the therapist allows the patient to do most of the talking, which expands the amount of information and invites elaboration on more deep thoughts on the change (Arkowitz & Miller 2008). Open-ended questions create forward energy that may help the patient explore the reasons for the change. *Reflective listening* helps the patient express their opinions more openly.

Acceptance involves affirmation that seeks and recognizes the patient’s strengths and efforts (Miller & Rollnick 2013). The therapist should affirm the patient often using statements that recognize the patient’s strengths. These statements encourage the patient to feel that the change is possible even when previous efforts failed. *Affirmations* involve reframing the behavior as evidence of positive patient qualities.

*Summarizing* the patient’s statements is a form of reflection where the therapist reviews what has occurred in all the counseling sessions. These statements may be used to shift attention from one direction to another or to prepare the patient to move on to the next subject (Miller & Rollnick 2002).

*Eliciting change talk* provides the patient with an exit from their ambivalence (Miller & Rollnick 2002) and involves statements that reflect desire, perceived ability, need, readiness, reasons and commitment to change (Arkowitz & Miller 2008). Research has shown a correlation between change talk and improved patient outcomes (Baer et al., 2008, Gaume et al., 2008).



## AIMS

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### *Study I*

To describe a large cohort of patients with tinnitus and SNHL in the Östergötland area.

To analyze the possible differences in examination methods and treatment models in different subgroups.

### *Study II*

To evaluate the quality of life in patients with tinnitus and SNHL, to investigate the patients' mental and physical health and to measure the level of satisfaction with the given care as perceived by the patients.

### *Study III*

To compare a group of patients with SNHL and tinnitus with a control group who only had SNHL (no tinnitus) regarding WM and hearing problems before and after hearing rehabilitation. The second aim was to investigate whether sleep problems have any effects on WM.

### *Study IV*

To test the effects of brief Motivational Interviewing (MI) as an adjunct to hearing aid rehabilitation for patients with tinnitus and SNHL.

### *Study V*

To evaluate the effects of multidisciplinary group information as part of a Stepped Care model using the Tinnitus Handicap Inventory (THI) and Hospital Anxiety and Depression Scale (HADS) before and after group informational counseling.



## MATERIALS AND METHODS

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Studies I and II were retrospective, descriptive studies based on data from patients who sought care for tinnitus and hearing loss at two ENT clinics in Östergötland County, Sweden, from 2004 to 2007 and who received a diagnosis.

Patients between 20-80 years of age with tinnitus and a pure tone average (PTA) lower than 70 dB HL were included in the study. Patients were excluded from the analyses if they had a CI, middle ear disorder, or hearing loss since birth or childhood. Multi-handicapped patients and those who did not speak fluent Swedish and required an interpreter at the ENT visit were also excluded.

Studies III & IV were prospective studies of patients who sought care for tinnitus and/or hearing loss at the ENT clinic in Linköping, Sweden, from September 2012 to March 2013. Patients who were between 40 and 82 years of age with SNHL and who had a pure tone average <70 dB HL in both ears were recruited for these studies.

In studies III & IV, all patients were first time hearing aid users. In study III, we started with a clinical sample of patients who fulfilled our study criteria and were willing to participate in the study based on the current waiting list of the clinic during the study period. However, it was apparent that the clinical cohort of patients with tinnitus and hearing loss were on average younger than the patients with only hearing loss. To age-match the two groups, repetitive elimination of the youngest remaining subject in the group that contained patients with both tinnitus and hearing loss and the oldest subject from the group with patients with only hearing loss was undertaken until the groups no longer differed significantly regarding age or PTA. This criterion was fulfilled at n=30 in each group. These groups were denoted as *Hearing Loss and Tinnitus* age-matched group (patients with both tinnitus and hearing loss) and *Hearing Loss* group (patients with only hearing loss) in the analysis. The clinical material (n=92) with completed follow-up was also collected for a third group of patients (denoted as *Hearing Loss and Tinnitus* non age-matched group, n=46), whose subjects had both tinnitus and hearing loss, and *Hearing Loss* group (n=46), which included subjects with only hearing loss.

In study IV, the patients who sought care for their tinnitus and were on the waiting list were randomly assigned to two groups: the intervention group and control group. A brief Motivational Interviewing (MI) program was used during the hearing aid fitting in 25 patients, whereas the remainder received the standard practice (SP), with conventional hearing rehabilitation during the hearing rehabilitation. This randomization was performed blindly by including every other patient from the waiting list to the MI group, which was conducted by our secretary at the

hearing clinic in Linköping. To improve the patients' hearing aid usage, MI techniques (Rollnick et al., 1999) were adopted, including open questions, reflective listening, summaries and affirmations (de Silva 2014). A specific manual based on Rollnick et al. (1999) and Miller & Rollnick (2012) was constructed for the MI (See table 1, study IV). The manual was used and the sessions for half of the group were recorded. These interviews were transcribed and then later analyzed together with a MI trainer. All patients were asked to answer four questions at the end of each visit (see table 2, study IV). Furthermore, four overlapping processes were assumed to work together to guide the patient to use the hearing aid. The first process was “Engaging”, which aims to develop a working alliance between the audiologist and the patient and leads to the second process. The second process was “Focusing” on one single behavior, namely using the hearing aids (table 1, study IV). The third process “Evoking” dealt with the patient's own motivation to use the hearing aid. The last process, “Planning”, indicates that the audiologist moved the discussion from the importance of daily use of the hearing aid to developing a plan to accomplish it (table 1, study IV).

Study V was a retrospective, descriptive study based on the data from patients who sought care for tinnitus at two hearing clinics in Östergötland County, Sweden, during 2004–2011 and were diagnosed with subjective tinnitus. In total, there were 426 adult patients included in the analysis of this study. All participants were recruited from hearing clinics in Östergötland and were registered as regular patients within the public health care system, which provided the diagnostic assessments and treatments. Patients were excluded from the analyses if they had a CI or a hearing loss since birth/childhood. Patients with a PTA less than 70 dB HL were eligible for the study. The exclusion criteria were the same as was described above for studies I & II.

## **Measures**

### *Audiometry*

Pure-tone audiometry using an audiometer is a standard clinical method to measure a patient's auditory sensitivity and can detect hearing thresholds over a range of frequencies (usually ranging from 125 Hz to 8 kHz). The procedure involves active participation from the patient. The equipment requirements for pure-tone audiometry are specified in IEC 60645-1 (IEC, 2001).

A patient's hearing threshold, as measured by an audiometer, is quantified in dB hearing levels (HL), which are defined from a standardized average hearing threshold for otologically normal subjects between 18 and 30 years old (ISO, 2004).

Hearing threshold measurements are performed by presenting an audible stimulus to the patient using earphones or a bone vibrator, which is standardized according to ISO- 8253-1 (ISO, 1989).

When a response to the stimuli is received, the stimuli level decreases by 20 dB until the patient does not respond. Then, the level is increased from an inaudible level in 5-dB steps. The threshold is determined when three out of a maximum of five levels are detected. The frequencies range from 125 Hz to 8 kHz. The result of the hearing threshold level test is described in terms of the dB HL (ISO, 2004). In the ENT clinics in Östergötland, the 4-frequency average (Pure-Tone Average, PTA) formula is used (i.e., the average of 0.5, 1, 2 and 4 kHz) to detect patients who have normal hearing thresholds at lower frequencies, but greater hearing loss in the high frequency range (Aniansson 1974, Smoorenburg 1992).

### *Self-assessment Instruments*

Self-assessment instruments, such as the EuroQoL 5D (EQ-5D) and the THI, were the basis of study II and III. In study IV and V, self-assessment instruments were used at the baseline and follow-up. To evaluate patients perceived tinnitus annoyance, sleep quality, hearing problems and hearing aid satisfaction before and after the hearing aid fitting, the THI, tinnitus background questions, the HADS, the Pittsburgh Sleep Quality Index (PSQI), the Hearing Handicap Inventory for the Elderly (HHIE) and the IOI-HA were used. In study III, the Reading Span Test and the HINT were used to evaluate patients' speech recognition and working memory capacity at the baseline and follow-up.

### *EuroQoL (EQ-5D)*

Data on quality of life were collected using the EQ-5D, which is a standard instrument to measure health outcomes that provides a simple descriptive profile and a single index value for health status.

The EQ-5D contains five questions about mobility, self-care, usual activities, pain and depression on a three-degree scale in addition to a VAS scale that assesses current health status. The EQ-5D is used for a wide range of health conditions and treatments, such as population health surveys and the clinical and economic evaluation of health care. The EQ-5D has been used in Swedish studies on audiological rehabilitation (Persson et al., 2008).

### *Tinnitus Handicap Inventory (THI)*

The THI is a tinnitus-specific, widespread, and validated questionnaire for quantifying the severity of tinnitus in patients' daily lives (Newman et al., 1998). Because of its wide use, the THI was recommended in a consensus document to be used as an outcome measurement in clinical trials to allow comparability across studies (Langguth et al., 2007). This questionnaire

has good psychometric characteristics (Newman et al., 1998) and is designed to evaluate the behavioral and treatment outcomes based on the emotional and physical aspects of the patients' health and lifestyles. The THI is self-administered, and includes 25-items that are scored on a 3-point scale (No = 0, Sometimes = 2 and Yes = 4). The total THI score is the sum of the scores for the following three subscales: functional, emotional, and catastrophic. Based on the total THI score, tinnitus sufferers can be classified into four categories that denote handicap severity: no handicap (0-16), mild handicap (18-36), moderate handicap (38-56) or severe handicap (58-100).

#### *Hospital Anxiety and Depression Scale (HADS)*

The HADS is a self-assessment scale developed by Zigmond and Snaith (1983) to be a compact and easily administered measure of depression and anxiety levels in patients seeking help for somatic illnesses. This instrument is useful for assessing the absence or presence of symptoms of anxiety and depression in patients. To differentiate the psychological or 'mood' state from physical conditions the test omits conceptually interlaced states, for example dizziness and headaches. This prevents mixing the psychological effects of tinnitus with the physiological effects. The HADS includes 14 items, where each of the questions has four response choices (0-3). There are two subscales, one for anxiety (HADS A) and one for depression (HADS D), which consist of seven items each. Higher scores indicate more symptoms. The results can be interpreted as follows: 0-7, no risk for anxiety/ depression (low); 8-10, anxiety and depression is possibly present; and  $\geq 11$ , anxiety/ depression exist (high).

#### *Questionnaire about life quality*

To assess the level at which the patients rank their own general health, another questionnaire was included that covered satisfaction with some aspects of their lives, such as their current physical health, physical activity, lifestyle, work and family situation, social cohesion, friendships, sleep, stress, personal development, interest and alcohol and tobacco consumption. This questionnaire is similar to the QOLI. In this study, the QOLI was not used to avoid having duplicate questions and to make it simple for the patients to respond by shortening the response time. The answers ranged from 1 (representing a bad condition) to 10 (representing a very good condition). Three open questions were designed to determine whether the patients also sought care outside the ENT clinic, if the patients were willing to change their health situation, and, in that case, how they were willing to change. The participants were also asked to rate the care they received at the ENT clinic.

### *Pittsburgh Sleep Quality Index (PSQI)*

Because sleep deprivation may affect cognitive abilities including concentration, it was important to measure the patients' perception of sleep. Therefore, the PSQI was used in this study. The PSQI is a self-assessed questionnaire that measures sleep quality and sleep disturbances over the previous month (Buysse et al., 1989). A total of 19 different items assess seven areas: subjective sleep quality, sleep latency, sleep duration, habitual sleep efficiency, sleep disturbance, the use of sleep medication and daytime dysfunction. Each question is answered 4-point scale (0-3), and the total score ranges from 0 to 21. The original authors of this questionnaire identified that a cut-off global score of less than 5 represents good sleep quality, whereas a total score higher than 5 denotes poor sleep quality (Buysse et al., 1989). The results may be provided both as a total score and as the individual scores for each of the seven different areas.

### *Hearing Handicap Inventory for the Elderly (HHIE)*

The HHIE is a questionnaire that measures the experience of hearing loss in the elderly by focusing on the psychosocial and emotional effects of hearing loss (Ventry & Weinstein 1982). The HHIE has shown good reliability (Ventry & Weinstein 1982, Weinstein et al., 1986), and several studies have confirmed that the HHIE has high reliability over repeated measurements (Öberg et al., 2007). The HHIE is divided into two subscales: social (comprised of 12 questions on whether the hearing loss has any impact on the social life of the patient) and emotional (comprised of 13 questions on the attitude of the patient towards his or her own hearing loss and how the patient perceives others dealing with their hearing loss; Ventry & Weinstein 1982). The results are scored on a 3-point scale (No=0, Sometimes=2 and Yes=4) where the minimum number of points is 0 and the maximum number of points 100. The HHIE may be administered either "face to face", where the audiologist asks the questions and fills in the answers, or as a "paper and pencil" test, where the patient fills the questionnaire in by him- or herself (Wienstein et al., 1986). Studies have suggested that the HHIE may be used as a measurement tool to evaluate the benefit from hearing aids after 3 weeks, 2 months, 3 months, 6 months and one year (Newman & Weinstein 1988, Malinof & Weinstein 1989, Abrams et al., 1992, Taylor 1993, Newman et al., 1993).

### *The International Outcome Inventory for Hearing Aids (IOI-HA)*

The IOI-HA is comprised of seven questions and evaluates the effectiveness and different aspects of patient satisfaction with the hearing aid. Each question has five response alternatives

and the score ranges from 1-5 where a higher global score represents more satisfaction (Cox et al., 2000). Each question covers different areas (daily use, benefit, residual disability, satisfaction, residual participation restrictions, impact on others and quality of life) to offer the most robust assessment possible. The results from a Swedish study showed that the translated version of the IOI-HA was as reliable as the original (Öberg et al., 2007).

#### *Reading span*

The Reading Span test is a measure of working-memory capacity (Daneman & Carpenter, 1980; Rönnerberg et al., 1989). Three-word sentences are presented as text on a computer screen word-by-word. The Reading Span test is performed in a quiet environment. Half of the sentences are meaningful and the others are absurd. Following each sentence, the subject's task is to respond "yes" (for a normal sentence) or "no" (for an absurd sentence). After a sequence of two to five sentences, the test leader indicates that the subject should begin to recall either the first or the final word for each sentence presented. The number of correctly reported words is used as the outcome measure and an estimate of working-memory capacity (max score = 28).

#### *Hearing in Noise Test (HINT)*

Speech recognition in noise was measured using the Swedish HINT sentences (Hällgren et al., 2006). For the HINT test, short everyday sentences are presented in noise, and the subject's task is to respond orally by repeating the sentence. The speech was presented at a fixed level of 65 dB SPL, and the noise level varied in 2-dB steps using an adaptive method to reach 50% correctly repeated sentences. The noise is stationary and spectrally shaped based on the long-term average spectrum of the speech material (Hällgren et al., 2006). Ten sentences were used for practice and twenty were used for the final test. The average signal-to-noise ratio was calculated and used as the outcome measure.

The HINT was performed as follows: the subjects were seated in a sound-attenuated room. The auditory stimuli of the HINT test were presented over a loudspeaker at a distance of one meter in front of the subject. Background noise was a permanent feature of this task.

## STATISTICAL METHODS

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Statistical analyses were performed using Microsoft Office Excel, Windows 2003 and Statistica 10 (Statsoft, Tulsa, OK, USA). The continuous data are shown as the mean  $\pm$  standard deviation (SD), and the categorical data are expressed as numbers with percentages. The distribution of the patients between the groups with two or more possible states was evaluated using the Chi-squared test, and the Yates correction was used in cases of 2x2 data. Between-group comparisons of the measurement variables with normal distributions were generated using Student's t-test, and comparisons of three or more groups in study II were assessed using one-way ANOVAs. In cases of paired measurements with binomial variables, we used the McNemar's test.

Because the data in study III and IV were not normally distributed (Shapiro-Wilks test), the data were analyzed using non-parametric methods. For the analyses, pairwise differences (e.g., before and after the hearing rehabilitation) were evaluated using the Wilcoxon paired rank sum test, and the differences between the groups were evaluated using the Mann-Whitney's U test. Moreover, in study V, we used a median value in one variable (for tinnitus duration) with a skewed data distribution.

The level of significance for all five studies was set to  $p < 0.05$ .

## ETHICAL CONSIDERATIONS

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The Eastern Regional Medical Research Ethic Committee located in Linköping, Sweden, approved studies I-IV, and the registration numbers are Dnr. M214-07 (study I and II) and Dnr. 2012/143-31 (study III and IV). The participants in studies II, III and IV were provided with written information on the study by mail. All data were handled confidentially and all analyses were conducted at the group level.

No ethical permission was sought for study V. This was due to the rules for monitoring existing treatment models within the public health care system and no additional procedures apart from regular clinical practice were evaluated (e.g., no randomization). The Declaration of Helsinki (WMA 2013) was followed.



## RESULTS

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### Study I

#### *Examinations*

Retrocochlear examinations were conducted in 372 patients, and MRI was the most common examination (table 3, study I). There was a significant difference between the patients with unilateral tinnitus and bilateral tinnitus regarding the use of the MRI examination ( $p=0.001$ ). Patients with asymmetric hearing loss (61%) underwent more retrocochlear examinations than patients with symmetric hearing loss (50%,  $p=0.014$ ).

#### *Treatments*

Of the 400 patients without hearing aids, 220 had unilateral tinnitus and 180 had bilateral tinnitus (table 4, study I). There were significantly more patients with bilateral tinnitus (49%) who had hearing aids compared with patients with unilateral tinnitus (39%,  $p=0.02$ ). A total of 219 patients had a PTA  $>20$  dB and did not have a hearing aid. All patients were examined by an ENT doctor, and 135 (20%) of the patients met with an audiologist with tinnitus training for further treatment (step 1). In total, 105 patients participated in the multidisciplinary group information on tinnitus, which is the step 2 in the Stepped Care model. The findings showed that after the step 2 there were 75 patients (71% of the participants in step 2) who needed additional treatment in the third step. There were 55 patients who visited a registered physical therapist (RPT), 35 a counselor and 31 a psychologist. Some of the patients were in need of several visits to various resources.

### Study II

#### *THI*

The female patients had a significantly higher total THI score than the male patients ( $p<0.05$ ) (Table 3, study II). For patients aged 20-40 years, the THI scores indicated a trend towards higher higher levels of self-reported tinnitus disability (THI). A similar result was found in patients with bilateral tinnitus and patients with bilateral hearing aids. However, non of these differences were statistically significant.

#### *EQ-5D*

Within each age group, a significant difference was seen when comparing the scores for “pain” and “anxiety/depression” with the scores of the other questions ( $p<0.05$ ) (table 4, study II). The

number of patients with self-care problems was low in all groups. There was no significant difference regarding the scores for the EQ-5D between the patients with and without hearing aids. Regarding the patients with the various diagnoses, there were no significant differences among the groups.

#### *Questions about patients' general health*

The youngest group (20-40 years old) had the lowest scores for the lifestyle question ( $p=0.015$ ) (table 5, study II). The results for the sleep question showed that the patients in the middle-aged group had the lowest scores ( $p<0.05$ ). A post-hoc analysis showed that the middle-aged group had lower scores than the oldest age group (Tukey's test,  $p=0.03$ ). The patient's age was correlated with their stress levels (ANOVA,  $p<0.05$ ), with the youngest age group having the greatest levels of stress. However, none of the pairwise comparisons were significant.

#### *Open questions*

The results from the three open questions showed that 101 (24%) of the patients did not perceive any receipt of treatment during their visits to our ENT clinics. Of the participants ( $n = 426$ ), 39 (9%) tried treatment options other than those offered by our ENT clinics. These alternatives included psychotherapy, chiropractic treatment, acupuncture, massage, and occlusal splints. The absence of hearing aids was more common in the group of patients who stated that they had not obtained any treatment at our ENT clinics ( $n = 107$ , 61%) than in the patients who thought the care that they had received was good/very good ( $n = 63$ , 42%;  $p = 0.001$ ). The patients who thought that the care they had received was good/very good showed a mean THI score of  $24.1 \pm 15.5$ , whereas the patients who stated that they had not obtained any treatment at our ENT clinics had a score of  $33.0 \pm 16.3$  (see table 6, study II), which was significantly higher than the former ( $p<0.05$ ). When comparing the male and female patients with respect to their satisfaction and THI scores, there was a significant difference within each treatment group.

Of the 426 respondents, a total of 159 responded to the question: "What do you think of the care you obtained at our ENT clinics?" (see table 7, study II). Many answers were related to hearing aids ( $n = 64$ ), with the majority of patients ( $n = 26$ , 40%) reporting that they did not receive any treatment, only hearing aids. Several patients stated: "I did not receive any treatment. The only treatment I received was a hearing aid.". Whereas, other patients ( $n = 23$ , 36%) perceived that their hearing aids improved their hearing (see table 7).

### Study III

#### *Analysis of the clinical material (n=92)*

The results showed that there were no differences between the groups at baseline, with the exception of the PSQI ( $p < 0.002$ ; see table 1 study III). The patients in the *Hearing Loss and Tinnitus* group had a higher PSQI score. Pre/post changes in the HHIE and HINT did not differ between the two groups ( $p < 0.61$  and  $p < 0.87$ , respectively), and both groups showed significant improvement at follow-up ( $p < 0.001$ ) compared to baseline. The PSQI score improved significantly in the hearing loss and tinnitus group ( $p < 0.001$ ), but not in the hearing loss group ( $p < 0.58$ ). The Reading Span was significantly improved in the *Hearing Loss and Tinnitus* group ( $p < 0.001$ ) at follow-up, but it was not improved in the *Hearing Loss* group ( $p < 0.44$ ). There was a significant difference between the two groups in the degree of improvement at follow-up ( $p < 0.001$ ). The patients in the *Hearing Loss and Tinnitus* group exhibited significantly improved THI scores at follow-up compared to baseline ( $p < 0.001$ ). In summary, the analyses of the non-age-matched groups demonstrated that, during the study period, the *Hearing Loss and Tinnitus* group underwent significantly larger changes in the Reading Span and PSQI than the *Hearing Loss* group. However, this interpretation is complicated due to the significant age differences between the groups.

#### *Analysis of the age-matched subgroups (n=60)*

After the groups were age-matched, the findings showed that there were no differences at baseline, with the exception of the PSQI (see table 2, study III). The patients in the *Hearing Loss and Tinnitus* group had significantly higher PSQI scores ( $p < 0.015$ ) compared to the patients in the *Hearing Loss* group. The pre/post changes were significant for both groups on the Reading Span and HHIE. However, the changes in the Reading Span were larger in the patients in the *Hearing Loss and Tinnitus* group ( $p < 0.001$ ; see figure 2, study III). Additionally, the results showed a significant decrease in the PSQI score in the patients in the *Tinnitus and Hearing Loss* group compared to the patients in the *Hearing Loss* group ( $p < 0.015$ ). The Reading Span scores at follow-up were higher in the tinnitus group (see table 2, study III). However, the between group effect size was moderate for the Reading Span ( $d = 0.52$ ), and the effect size for the PSQI ( $d = 0.07$ ) was very small. Therefore, for the pre/post improvement, similar differences were found in these ( $n = 30 + 30$ ) subgroups as in the full sample. The results of the THI revealed a significant improvement ( $p < 0.001$ ) at follow-up for the patients in the hearing loss and tinnitus age-matched group.

### Study IV

#### *Differences between the MI and SP groups at baseline and follow-up*

Furthermore, the findings showed that the number of visits during the rehabilitation process was significantly lower ( $p < 0.022$ ) in the MI group (average = 3.3) than in the SP group (average = 4.1). The level of self-reported tinnitus annoyance (THI) was significantly decreased in both the MI group and the SP group ( $p < 0.001$  and  $p < 0.006$ , respectively), and the between-group difference in the change in scores was statistically significant ( $p < 0.013$ ; Figure 2, study IV). The level of patient satisfaction concerning the hearing aids (IOI-HA) did not differ between the groups ( $p < 0.99$ ); however, each group showed significant improvements relative to the baseline at follow-up ( $p < 0.038$  and  $p < 0.026$ , respectively). The level of self-reported tinnitus annoyance (THI) decreased significantly (MI group,  $p < 0.001$ ; SP group,  $p < 0.006$ ) within each group between the baseline and follow-up. The analysis of the differences between the patients with bilateral and unilateral hearing aids in the two groups separately and in the full group did not show any differences concerning the number of visits in each group. The results obtained for the level of patient satisfaction concerning the hearing aids (IOI-HA) did not differ between the baseline and follow-up.

Moreover, to investigate any possible difference between the counseling models, the patients' perception of the interviewing models was studied using four questions at the end of each visit. The analysis of these data did not show any significant differences.

### Study V

Concerning the tinnitus duration in this study, the median value for the 373 patients was 5 years, whereas 33 patients described the duration of their tinnitus as "for a long time" and 20 did not answer the question. Our assumption is that the median value including the 33 patients who stated "for a long time" will remain at 5 years due to the observed data distribution.

The results for the self-reported sensitivity to sound showed that high sensitivity to sound was common in our study population, with 115 (27.0%) of the patients reporting being quite sensitive and 124 (29.1%) reporting being very sensitive.

The results showed significant changes in the THI scores ( $p < 0.001$ ) and for the subscale A in the HADS questionnaire ( $p < 0.014$ ) at the one-month follow-up. However, no significant changes were found for the subscale D in the HADS questionnaire ( $p < 0.18$ ; see table 2, study V). The THI scores were divided into four handicap categories according to the quartile ranges defined by Newman et al. (1998). There were significant improvements in the pre- to post changes for the different handicap categories at the one-month follow-up ( $p < 0.001$ ).

## DISCUSSION

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The main purpose of the present thesis was to investigate the disease management and identify the quality of life and treatment strategies in patients with tinnitus and sensorineural hearing loss. Tinnitus is common in patients with SNHL. It affects approximately 10-15% of the population and is a common condition among older adults (Sindhusake et al., 2003; Sindhusake et al., 2004). Hoffman and Reed compared the prevalence of self-reported tinnitus from several large epidemiological studies (Hoffman & Reed, 2004) and found that for patients aged 50 years and older, the estimated prevalence of tinnitus ranged from 7.6 to 20.1%. Our data from study I showed that the majority of patients (79%) were 50 years and older. The results showed that 1175 (70%) of the patients also had tinnitus (study I), which is consistent with previous studies that have reported similar frequencies (Axelsson & Ringdahl, 1989; Rosenhall & Karlsson, 1991; Cooper Jr., 1994; Scott & Lindberg, 2000; Andersson et al., 2002; Shargorodsky et al., 2010). The outcomes of this investigation show that the majority of patients who were dissatisfied with the care they obtained did not have hearing aids. The Stepped Care model could be used to lead patients to more individually adapted treatments and more accurate alternatives in an ENT clinic (study I). The findings also showed a significant difference within each age group between the scores for “pain” and “depression” and those for the other questions in the EQ-5D (study II). We observed that an absence of hearing aids was more common in the patients who stated that they had not obtained any treatment at our ENT clinic compared with the patients who thought that the care they received was good/very good. Furthermore, one of the primary outcomes (as a result from hearing rehabilitation) from this thesis is the significantly positive impact observed on speech recognition in a noisy environment (i.e., the HINT test) and on working-memory capacity (i.e., the Reading Span) in the patients with both tinnitus and hearing loss compared with the patients with only hearing loss (study III). One interesting outcome from study IV was the decreased THI scores in the MI group. An unexpected finding is that the use of a brief MI guide during the hearing rehabilitation process may shorten the number of visits with no loss in patient satisfaction (study IV). However, the aim of study IV was not to test this finding. Moreover, disease management is a central topic in this thesis. Therefore, we focused on the information meeting as a part of the multidisciplinary Stepped Care model (study V). Our results show a significant decrease in the THI scores at the one-month follow-up, which confirmed the effectiveness of this approach to reduce tinnitus annoyance.

**Methods discussion**

The results in this thesis showed that there was often a lack of information in the patients' medical records concerning vertigo, tinnitus, heredity for hearing loss and or tinnitus, and the patients' general health. Most of the information physicians obtain during a patient's visit remains in the physician's head in "a constantly expanding and reinterpreted database" (Tanenbaum, 1994). Therefore, it would be desirable to include an interview in which the physicians could clarify the reasons that they do not transfer specific information from a patient's visit to their medical record, which can remain in their head after the visits. This practice could obviously interfere with the development of an optimal care plan for the patients. It is not clear whether all of the patients received an adequate diagnosis because of the small size of some of the subgroups (study I). For example, it is possible that patients who received a diagnosis of bilateral SNHL should have been classified as having presbycusis or noise-induced hearing loss. This classification could affect the outcome of rehabilitation. Specifically, the patients who may need further treatments, therapy or analysis may not be able to obtain these services due to incorrect classifications. It is possible that the fact that some patients did not receive an adequate diagnosis further affected the outcome of our study. If all patients had received an accurate diagnosis, some of the groups, such as the presbycusis or ISSNHL groups, may have been larger, which could have enabled us to better analyze some of our findings. One of the limitations of retrospective studies (study I and II) is that the patient may not recall his/her condition correctly, i.e., the patients' experiences of tinnitus are measured long after they received their diagnosis, which may explain why the total THI scores indicated a mild degree of tinnitus annoyance in our cohort. Nevertheless, this finding is consistent with other studies (e.g., Gopinath et al., 2010), which suggests that the subjective distress associated with tinnitus may decrease over time and implies that some patients may habituate to tinnitus annoyance over time. Study III was prospective and the subjects were recruited from the waiting lists at our ENT clinic. Due to limited number of patients with obvious tinnitus annoyance during the study period, it was not possible to match the groups for age or PTA. This issue occurred despite the fact that a secretary at our clinic randomized the groups. Studies have reported an association between the performance on cognitive tests and factors such as age, gender and years of education (MacPherson et al., 2014). Therefore, the desired groups in future studies should be matched not only for the patient's PTA, age and gender but also for their education level. One limitation in study IV was the differences between the two groups regarding the THI scores and age. Although the patients were recruited randomly and the randomization process was preformed blindly by a secretary at our clinic, the patients in the MI group had lower PTA and

THI scores. Another limitation in study IV was the fact that the first author was the only MI-trained audiologist that could apply the treatment model to the hearing rehabilitation process. It would be desirable to involve several MI-trained audiologists in a future study to avoid possible biases in the rehabilitation process, while investigating the value of brief MI interventions as an adjunct to hearing rehabilitation.

In study V, the patients evaluated the effect of the group information meeting only a short time after the meeting (four weeks). However, it would be desirable to have a longitudinal follow-up at additional time points such as 6 or 12 months (Andersson et al. 2001) after the group information meeting to evaluate the long-term effects. Longitudinal studies could also lead to a better understanding of how tinnitus affects patients with impaired hearing, which could elucidate possible differences compared to patients with tinnitus and normal hearing. An additional limitation in study V was the fact that there was no control group who received usual care methods. An evaluation between the groups could reveal the possible benefits of the Stepped Care model.

## **Results discussion**

### *Examinations at the ENT clinics*

Unilateral SNHL associated with tinnitus and/or vertigo can be a sign of a retrocochlear lesion; therefore, these patients should be examined using retrocochlear examinations (Turner et al., 1984). Of the 159 patients with asymmetric SNHL and tinnitus, 61% were investigated using retrocochlear examinations. Of the 357 patients with unilateral tinnitus, 64% underwent retrocochlear examinations. Ideally, the percentage of patients receiving these examinations should be close to 100% because this exam is highly recommended to identify patients with a vestibular schwannoma (VS). The global incidence of VS is approximately 7.8 to 9.5 patients per million per year (Tos et al., 1992; Tos et al., 1998). Earlier retrospective studies have raised a concern about the validity of ABR as a screening tool for asymmetric SNHL (Wilson et al., 1992). In several recent reports, researchers have more clearly stated their reluctance to use ABR and have called for an end to ABR testing (Cueva, 2004; Fortnum et al., 2009; SBU, 2010).

### *The offered treatments*

The Stepped Care model is thought to be an effective treatment model that provides better access to optimal tinnitus-focused treatment and can make more resources available to patients with severe problems. In study I, the results could indicate that only a small group of patients who went to a half-day information meeting held by a multiprofessional team continued to more

resource-consuming treatments. However, with this method, the patients reached their optimal level of care with the appropriate caregiver more quickly. To meet with the wrong caregiver could waste the patient's time and could create a resistance in the patient to seek further treatment. The findings from a randomized controlled trial showed that a multidisciplinary approach could be an effective treatment method for patients with tinnitus, irrespective of the initial tinnitus severity. In addition, there were no adverse events in that trial (Cima et al., 2012). In this study, the number of patients who were included in the Stepped Care model was low. The fact that some ENT physicians lacked knowledge about the Stepped Care model could have affected the number of patients included in this treatment model. The Stepped Care model applied in the ENT clinics in Östergötland should probably have been introduced in a better way. A more desirable model is that after an initial contact with an ENT physician, the patient is referred to an audiologist who has tinnitus training and is a member of a multidisciplinary team to be the main caregiver for patients with both tinnitus and hearing loss.

The majority of the patients (56%) did not have hearing aids, even though it is likely that almost every patient with hearing loss was offered a hearing aid, which is the normal practice in Östergötland. Similar findings have been reported in other studies (Aazh et al., 2009). There are several possible explanations for why the number of patients with hearing aids was so low, such as the stigma associated with hearing aid use, the limitations of amplification to remedy the fundamental difficulty of understanding speech in the presence of background noise, and economic issues (Kochkin, 2007). Furthermore, most of the patients had high-frequency hearing loss and could, in quiet environments, handle a conversation despite their hearing impairment. Studies that have investigated gender effects have mostly found none (Jerram & Purdy, 2001). However, the 1998 study by Brooks and Hallam found that female participants were slightly more satisfied than male participants. In study IV, the number of male patients was higher than the number of female patients. Jerram and Purdy (2001) reported a greater use of hearing aids in patients with impaired hearing who accepted their hearing loss than in patients who denied their condition. This interesting fact suggests the need to study patient motivation before, during and after a hearing aid fitting, which is the subject of study IV.

The results show that 47% of the patients in study II were dissatisfied with the treatment they obtained at our ENT clinics. These patients had lower PTA and higher THI scores than those who were satisfied, and 61% of them had no hearing aids. This finding could indicate a need for individually focused treatment, therapy, and analyses in these patients. Hearing aids improve not only communication but also the tinnitus annoyance (Searchfield et al., 2010).

### *Measurement outcomes*

The results from study II showed that the female patients had a higher mean THI score compared with the males. However, it is not clear whether the greater tinnitus annoyance was due to the extent of hearing impairment or the resulting reduction in sensory input. This finding contradicts the findings by Gopinath et al. (2010).

The results from study IV, where all the subjects underwent hearing aid fitting, revealed that both groups showed significantly decreased levels of tinnitus annoyance, which is consistent with other studies (Hoare et al., 2012; Shepperd et al., 2009) that noted that counseling may provide a good explanation to the patient of the relationship between tinnitus and hearing loss. A correlation between the severity of tinnitus and psychological and general health factors has been previously reported (Crocetti et al., 2009). However, the results of the EQ-5D questionnaire in study II showed that the number of patients who perceived limitations or problems in their daily lives was low. These results are similar to the results from a previous report on patients with hearing impairment from the Östergötland, Jönköping, and Kalmar counties in Sweden (Persson et al., 2008).

Hallam (1996) reported a higher level of tinnitus annoyance in patients with severe sleep disturbances. In the cohort of patients in study II, the middle-aged group showed worse scores for sleep, whereas the other groups showed relatively better scores. However, the findings showed that the patients estimated their current general health to be quite good. Studies have investigated poor sleep quality and other types of sleep complaints, such as difficulties initiating sleep, difficulties maintaining sleep, and early morning awakening (Dluqaj et al., 2014). The authors reported that poor sleep quality rather than sleep-disordered breathing was associated with mild cognitive impairment. Hence, the PSQI questionnaire was used in study III with the intention of excluding patients who suffered from sleepiness that could affect their WM. The results revealed that patients with only hearing loss and no tinnitus also had difficulties with sleep, which is consistent with another study (Asplund 2003) that showed a greater extent of sleep problems in older patients. Nonetheless, sleep quality increased in both groups at follow-up following the completion of the hearing aid fitting. This benefit may be due to the amplification of sound that could facilitate listening during the day, which could be an effective method of reducing the gap between the tinnitus stimuli and the background neural activity in patients with both tinnitus and hearing loss (Searchfield 2008). Another outcome that confirms the impact of hearing aids in patients with hearing loss was the HHIE results that were also consistent with another study (Öberg et al., 2008). The fact that patients with hearing loss experienced a significant improvement is not an unusual finding. However, the interesting finding is that the

patients with both tinnitus and hearing loss showed improved PTA but similar hearing difficulties to the patients with only hearing loss and no tinnitus. The improvement in hearing problems was comparable between groups, which indicated that tinnitus might interfere with speech recognition and cause hearing difficulties (Møller 1997, Andersson et al., 2009).

The results of bilateral or unilateral amplification in patients with both tinnitus and hearing loss have varied (Del Bo & Ambrosetti 2007, Totter & Donaldsson 2008, Parazzini et al., 2011). Therefore, it is difficult to confirm the most appropriate treatment for these patients. However, in study III, no scientifically controlled analysis of the benefit of bilateral vs. unilateral hearing rehabilitation was possible because the patients were fitted with hearing aids based on clinical recommendations.

It is recognized that tinnitus may affect cognitive processing (Andersson & Mckenna 2006). By focusing on tinnitus, the patients pay less attention to other daily sounds, such as traffic, conversations and music, among others, implying that speech processing, which places a considerable burden on WM processes, may be even more difficult for an individual suffering from tinnitus. In study III, there was a statistically significant improvement ( $p < 0.001$ ) in the HINT scores in patients in both the aged-matched groups ( $n=60$ ) and our clinical material ( $n=92$ ) at follow-up when compared with the baseline. These improvements in the HINT scores were consistent with other studies (Klemp & Dhar 2008) and show the benefit of using hearing aids in a noisy environment. Therefore, hearing aids may facilitate speech recognition and relieve tinnitus annoyance.

To investigate whether working-memory capacity may be affected by tinnitus, the Reading span test was used in study III. The results showed higher scores on the Reading Span test at follow-up compared with the baseline in both aged-matched groups. Nevertheless, the patients with tinnitus showed a significantly larger improvement on this memory task (i.e., Reading Span test) from baseline to follow-up than the patients with only hearing loss. This finding is consistent with another study (Rossiter et al., 2006) that showed that working-memory capacity might be affected by tinnitus. These data may indicate that the hearing aid users with both tinnitus and hearing loss may benefit more from processing in noisy environments because tinnitus annoyance reduces signal processing (Ricketts 2005, Rudner et al., 2011). Hence, in patients with both tinnitus and hearing loss, more cognitive processing resources become available to overcome the extra processing of the artificial or distorted signals common in noisy environments, and they may receive and process more details in speech signals. Furthermore, the patient's individual cognitive abilities should be taken into account because they are critical in

providing signal processing, which is itself modified based on their hearing loss (Ng et al., 2013).

A brief MI guide was used in study IV to test whether it could be applied as a counseling model to the field of hearing aid rehabilitation. An unexpected finding in study IV showed that the number of visits was reduced with no loss in patient satisfaction during hearing rehabilitation. However, the aim of study IV was not to test this finding. Research has shown that successful counseling may reduce the perceived tinnitus handicap (Henry et al. 2006; Aazh et al., 2009; Westin et al., 2011). However, they have also emphasized that the patient's motivation and willingness to undertake the rehabilitation is essential for a successful outcome. If the patient is not sufficiently committed and motivated to undergo a long-term rehabilitation, the counseling sessions often become frustrating for both the patient and the audiologist. Person-centered care has been observed to be an important aspect to support the users of the health-care system (de Silva 2014), where an actual partnership is the core component and is preferred for decision making and supporting coping strategies. This model may strengthen personal motivation and commitment during the rehabilitation process. The outcome of the brief MI guide used in study IV indicated that it was possible to encourage the process of hearing aid fitting in patients with both tinnitus and hearing loss compared with conventional hearing aid rehabilitation.

Furthermore, this model may result in a faster rehabilitation process for the MI-guided patients and an economic gain for the hearing clinics by saving scarce resources. These promising results should be confirmed in further studies with larger groups and more MI-trained audiologists (Bean et al., 2012). However, it is important to clarify that the implementation of fewer sessions was not an outcome but rather a decision made by the clinician and the patient. Therefore, strong conclusions should not be drawn in this regard. It would have been better to have an equal number of sessions between the two groups even if we had found an advantage of MI on THI performance. Future studies should control for this factor.

Despite the fact that study IV was a pilot trial, the results could strengthen the need of more empirical evidence regarding patient-centered care and the probable application of MI in the hearing rehabilitation research field. However, the findings are preliminary and should be replicated in larger samples. Moreover, this model includes a more effective rehabilitation process where the patient plays an equal part and may be more committed to the program. The brief MI guide that was evaluated in this thesis may serve as a model for the patient.

To investigate the effects of the multidisciplinary group information session as a part of a Stepped Care model, two questionnaires (THI and HADS) were used in study V. The results showed a significant decrease in the THI scores in the patients one month after attending the

multidisciplinary group. Additionally, a significant, but small, change in the anxiety module of the HADS was found, in which the score was decreased from a mean of 6.8 at the baseline to 6.4 at the one-month follow-up. The findings from study V are consistent with previous studies that recommend a multiprofessional approach for tinnitus treatment to reduce tinnitus annoyance (Stephens et al., 1986; Coles & Hallam 1987; Baguley et al., 2013). However, there was no significant improvement in the depression module of the HADS. This could be because depression is often a significant co-morbidity in the subgroup of 'severe' tinnitus sufferers; however, these patients are only a small proportion of tinnitus sufferers. This could explain the failure of trials to show a significant effect in tinnitus samples (Martinez-Devesa et al., 2010). Furthermore, the current treatment strategies are diverse and often costly (Cima et al., 2009). By handling the information session in a larger group instead of seeing all patients individually, the resources become more available for the patients in need of more intensive therapy.

### **Future directions**

Hearing problems affect many individuals worldwide in everyday situations. The issues encountered raise the question of whether people with hearing impairment require hearing aids to maintain their quality of life or whether they can live without them. The next generation of hearing aid users is expected to place a higher demand on health care and require more equal partnership with their health-care professionals. Therefore, a more patient-centered approach that could strengthen the position of the patient in health care is highly desirable. Future patients will be more interested in the "software" of the rehabilitation process. Hence, further research focusing on the development of patient-centered hearing rehabilitation is as important as high-tech hearing aids and could be essential for the successful management of tinnitus in addition to hearing rehabilitation.

To investigate the processes of change and the genuine role of MI in hearing rehabilitation, more findings in larger trials are required. These trials are necessary to generate more knowledge on implementing patient-centered models, such as MI, to the field of hearing rehabilitation. By measuring the patient's expectations, motivations and willingness prior to undertaking rehabilitation, it may be easier to build a solid foundation for a successful outcome. The hearing rehabilitation process may need to be delayed until the patient is ready to continue with the hearing aid fitting. Audiologists should learn how to inspire the patient to a greater extent, remove their resistance and reduce the patient's ambivalence. These may be the biggest challenges for audiologists who are facing new patients with higher expectations. Therefore, there is a need for additional research on MI and how it can be applied as a counseling model in

hearing rehabilitation. Future work should focus on developing MI models for the field of hearing rehabilitation and to investigate the processes of change in hearing rehabilitation.

## CONCLUSIONS

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The conclusions of this thesis can be summarized as follows:

The effect of hearing aids on WM was investigated using the Reading Span test. The results showed that patients with tinnitus and hearing loss exhibit significantly improved concentration after hearing aid fittings which is a valuable finding and may indicate that more cognitive processing resources become available to manage speech signals.

Despite the obvious benefits of using hearing aids together with counseling, there were many patients (56%) who did not have a hearing aid. This finding may be due to patients' belief that nothing could be done about their situation. It is possible that all patients with tinnitus and hearing loss should consider being fitted for hearing aids when showing clinically significant hearing loss.

An unexpected finding showed that brief MI guiding may shorten the number of visits during hearing aid rehabilitation demonstrates the efficacy of MI as a communication technique. Nevertheless, the aim of study IV was not to test this finding. Yet, this result could be interpreted as a technique to improve the rehabilitation process with a greater focus on patient-centered care which eventually could generate an economic gain for hearing clinics by saving scarce resources.

The findings showed a need of improvement of information flow regarding patients' general health which were missing in their medical records. This could be a necessity that would prevent important information get lost and creates better opportunity to make an optimal care plan for the patients.

The overall improvement in the scores for the THI and the anxiety module of the HADS showed that a half-day information meeting held by a multiprofessional team could have a positive impact on the perceived tinnitus annoyance. Moreover, information sessions held in a larger group could apparently make more resources available for the patients in need of more intensive therapy (e.g., CBT).

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---

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## SVENSK SAMMANFATTNING

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”Patienter med tinnitus och hörselnedsättning; Hantering, livskvalité och behandlingsmodeller”

Upplevelsen av att höra ljud utan någon yttre källa är ett vanligt fenomen och kallas för tinnitus. Ungefär 15% av svenska befolkningen upplever tinnitus men bara 2.4% upplever allvarliga problem. Det kan även leda till sömnstörningar, psykisk stress, depression m.m. Tinnitus kan ibland jämföras med så kallade fantomsmärton. De flesta människor har någon gång upplevt tinnitus i öronen t.ex. efter ett konsertbesök, vid feber eller vid lockkänsla. Det kan finnas skadade nervstrukturer kvar, efter t.ex. en operation i hörselorganet som ”lurar” hjärnan att tro att det finns ett verkligt ljud. Hos de flesta patienter med tinnitus finns det en bakomliggande hörselnedsättning. Det kan vara antingen ett ledningshinder eller en sensorineural hörselnedsättning, bl.a. bullerskada eller Menière´s sjukdom. Tinnitus är sällan ett tecken på något farligt. Det är inte ett sjukdomstecken i sig, utan är oftast ett tecken på att något är fel i hörselsystemet.

Det finns olika behandlingsmetoder för patienter med tinnitusbesvär beroende på orsaken och patientens behov, bl.a. hörapparatutprovning, sjukgymnastik, kognitiv beteendeterapi och informationskurser. Hörselrehabilitering i form av hörapparat Anpassning har länge använts hos patienter med både tinnitus och nedsatt hörsel. Dilemmat med hörapparatutprovning är dock ofta att många patienter tackar nej till denna eller avbryter rehabiliteringen efter några få besök.

De två första studierna (I & II) som beskrivs i denna avhandling är retrospektiva, deskriptiva studier av patienter som sökt vård för tinnitus och hörselnedsättning på Öron-Näsa-Hals klinikerna i Östergötlands landsting under 2004-2007. Vi fann att det var en minoritet av patienterna som kommit i kontakt med Stepped Care-modellen som startades 2004 för att erbjuda effektivare process för patienter med tinnitus. Det visade sig att kvinnorna i studien i högre grad upplevde tinnitusbesvär, jämfört med männen. De flesta av deltagarna hade ingen hörapparat även om de kunde ha haft nytta av en sådan. Vidare visade resultatet att majoriteten av patienter som var missnöjda med den vård de fått på våra kliniker inte hade någon hörapparat.

Studierna III & IV var prospektiva och innehöll patienter som sökt vård för tinnitus och eller hörselnedsättning vid Öron-Näs-Hals kliniken i Linköping under 2012-2013. I studie III var det till slut totalt 92 patienter som undersöktes vid dels baseline och dels uppföljning (ca 3 månader efter avslutad hörselrehabilitering). Det visade sig att patienterna inte kunde matchas under rekryteringsfasen. Detta berodde på att vi valde kliniska patienter som stod på hörselvårdens olika väntelistor. För att få jämförbarhet mellan grupperna i den statistiska analysen gjordes

därför en successiv elimination av de mest extrema patienterna avseende ålder i respektive grupp. Till slut fanns det därmed två grupper kvar ( $n=30+30$ ); grupp 1 innehöll patienter med både tinnitus och nedsatt hörsel och grupp 2 patienter med endast nedsatt hörsel. Resultatet för de åldersmatchade grupperna visade en positiv effekt av hörapparater gällande arbetsminneskapacitet och taluppfattning i bullrig miljö, särskilt hos patienter med både tinnitus och nedsatt hörsel. Detta skulle kunna antyda att mer resurser frisläpps för kognitiva processer för att ta emot och bearbeta talsignaler hos patienter med tinnitus och hörselnedsättning. Dessa fynd kunde också erhållas när vi analyserade det totala materialet ( $n=92$ ), som vi hade innan åldersmatchningen gjordes.

I studie IV ingick till slut 46 patienter ( $n=23+23$ ) med både tinnitus och nedsatt hörsel. De delades slumpmässigt i två lika stora grupper; i grupp 1 användes tekniken MI som en del av hörselrehabiliteringen i syfte att öka acceptansgraden för att använda hörapparater medan i grupp 2 användes traditionellt samtal. Resultatet visade att MI kunde ha en positiv effekt på upplevelsen av tinnitus samt skulle kunna förkorta rehabiliteringsprocessen utan att ha någon som helst negativ effekt på patienternas nöjdhet. Detta skulle kunna innebära ekonomiska vinster för kliniker då resurserna kan användas effektivare.

Den sista studien (V) som beskrivs i denna avhandling är en retrospektiv, deskriptiv studie av patienter som sökt vård för tinnitus och hörselnedsättning på Öron-Näsa-Hals klinikerna i Östergötlands landsting under 2004-2011 och genomgick en halvdags informations tillfälle som en del av Stepped Care modellen vi använder i Östergötland. Till slut fanns det 426 vuxna patienter som fick ingå i analysen av denna studie. Den totala förbättringen av tinnitusbesvär, mätt i poäng för frågeformuläret THI och ångest modulen i frågeformuläret HADS visade att detta steg av vår behandlings modell (Stepped Care) var effektivt. Informationsmöte i en större grupp istället för att se alla patienter individuellt skulle förmodligen kunna frigöra mer resurser till patienter i behov av mer intensiv terapi.

Resultaten från denna avhandling talar för att hanteringen av patienter som lider av tinnitus och hörselnedsättning skulle kunna effektiviseras ännu mer, med fokus på individuella behov som måste uppmärksammas. Avhandlingen kan även rapportera resultat som visar att bland de som kände sig missnöjda med den vård de fått på öronkliniken i Linköping hade majoriteten ingen hörapparat. Fynden i studie III visar att hörapparater kan frisläppa resurser i hjärnan hos patienter som har både tinnitus och nedsatt hörsel som kan hjälpa till med taluppfattningen. Resultatet från

de två sista studierna visar att den traditionella samtalsmetodiken skulle kunna ändras till en samtalsmetodik med inslag av Motiverande Samtal och att Stepped Care modellen som är väl forankrad i tinnitusverksamheten i Östergötland kan förse patienterna med rätt vårdgivare på rätt vårdnivå.

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