Hyperacusis
Clinical Studies and
Effect of Cognitive Behaviour Therapy
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Hyperacusis is a type of decreased sound tolerance where the individual has decreased loudness discomfort levels (LDL), normal hearing thresholds and is sensitive to ordinary environmental sounds. Persons with hyperacusis frequently seek help at audiological departments as they are often affected by other audiological problems. Regrettably, there is neither a consensus-based diagnostic procedure nor an evidence-based treatment for hyperacusis.

The principal aim of this thesis was to gain knowledge about the clinical condition hyperacusis. The specific aim of Paper I was to compare hyperacusis measurement tools in order to determine the most valid measures for assessing hyperacusis. Items from a constructed clinical interview were compared with the LDL test, the Hyperacusis Questionnaire (HQ) and the Hospital Anxiety and Depression Scale (HADS). LDLs were significantly correlated with the anxiety subscale of the HADS. A third of the 62 investigated patients scored below the previously recommended cut-off for the HQ. The results suggest that HQ and HADS in combination with a clinical interview are useful as part of the assessment procedure in patients with hyperacusis.

The aim of Paper II was to further investigate the patient group with respect to individual characteristics, psychiatric morbidity and personality traits. It was shown that anxiety disorders and anxiety-related personality traits were over-represented, which suggests common or cooperating mechanisms. Avoidance behaviour proved to be very common in the patient group, as was being unable to work due to hyperacusis.

In Paper III it was investigated in a randomized controlled trial whether Cognitive Behaviour Therapy (CBT) could be helpful for patients with hyperacusis. The effect of CBT for hyperacusis was assessed with measures of LDLs, symptoms of hyperacusis and of anxiety and depression, fear of (re)injury due to exposure to sounds, and quality of life, compared to a waiting list control group. There were significant group effects for a majority of the measures with moderate and strong effect sizes within- and between groups. After assessment the waiting list group was also given CBT, and was then reassessed with similar effects. The results were maintained for 12 months, concluding CBT to be potentially helpful for these patients.
To my father Enn 1936-1989
List of Papers

This thesis is based on the following papers, which are referred to in the text by their Roman numerals.


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Introduction

Hyperacusis is when sensitivity to ordinary environmental sounds leads to restrictions in an individual's life. Many persons with hyperacusis seek help from audiology services as they are often affected by other audiological problems as well. Regrettably, there is neither a consensus-based diagnostic procedure nor an evidence-based treatment for hyperacusis.

Persons who experience hyperacusis differ widely with respect to symptom characteristics. This vignette of a fictitious person exemplifies the consequences of hyperacusis:

Elisabeth is a 42-year-old preschool teacher. Five years ago, after a very stressful few months at work, she noticed how she started to become more and more bothered by the sound level at her preschool. She experienced pain in her ears and started worrying about developing a hearing loss. At work, she tried to control the problem by asking the children to lower their voices, and guided them into playing in a way that loudness was experienced as softer to her. At home, she made an effort to keep her surroundings quiet by not listening to music, and she started using the text function on the TV so that she could turn off the sound completely. After a while she also started wearing earplugs; at first only at work but gradually more and more often, to the point where she always wore them. Unfortunately, wearing earplugs made it difficult to continue with some of her activities such as singing in the church choir and attending social gatherings like family activities. Elisabeth now stays at home a lot and often feels sad. She is not able to work full-time which makes her even more inactive, and this also affects her financial situation. Family members complain about her not taking part in their activities, such as going out to restaurants with them, which makes her even sadder. When seeking help from an audiology specialist, she describes her life as being “destroyed by sounds”.

Hyperacusis

Definition and clinical characteristics

Hyperacusis is defined as “unusual intolerance to ordinary environmental sounds” [132], and it characterises persons who are sensitive to everyday
sounds such as music, mechanical sounds and paper noises [4]. These individuals report fear of sounds, extensive use of ear protection devices and avoidance of environments perceived as loud, such as their workplaces and locations for social activities [90; 12]. When work is avoided, a likely consequence is decreased income, which may further affect many aspects of an individual's life. The use of earplugs is, not surprisingly, associated with communication difficulties [111]. Other likely consequences of avoiding sounds and environments that are considered loud are isolation, depressed mood and problems in maintaining relationships. Sounds and environments that are considered loud constitute aspects of life thought by many to be essential in maintaining a certain level of quality of life [36].

In audiological testing, a person with hyperacusis presents with reduced loudness discomfort levels (LDL), while hearing thresholds tend to be normal [2]. Loudness is the subjective perception of a sound [24] and changes with the properties of the sound such as frequency, or the presence of other sounds, but also with cognitive factors, and with the psychological and physical state of the listener [33].

The terminology used within this particular area of audiology is indistinct, with several existing definitions for describing reduced loudness tolerance. The specific term hyperacusis was first mentioned in the literature by Perlman, in 1938 [105], and other terms describing similar problems include "decreased sound tolerance" [62] and "hypersensitivity to sounds" [2]. Many different definitions also exist, such as "consistently exaggerated or inappropriate responses or complaints to sounds that are neither intrinsically threatening nor uncomfortably loud to a typical person" [73].

The term "phonophobia" is sometimes used, often to describe the sound sensitivity that accompanies migraine attacks [137]. However, the term "phobia" generally implies some kind of emotional reaction and it has been discussed as being an unfortunate term for this type of sound sensitivity [11]. Phonophobia may describe a person who suffers from a specific phobia for certain sounds, for instance thunder. To differentiate between hyperacusis and phonophobia, hyperacusis may be seen as an oversensitivity along the complete spectrum of frequencies, while phonophobia is an oversensitivity only to certain sounds, mostly independent of their intensity [112]. "Misophonia" is another type of sensitivity to sound that refers to a general dislike or an annoyance concerning a sound [63]. Another distinguishable type of sound sensitivity associated with cochlear hearing loss and outer hair cell dysfunction is "recruitment of loudness" [95]. Due to the present inner ear damage, recruitment of loudness is unlikely to be affected by sound avoidance or exposure.

Hyperacusis is furthermore not to be confused with noise annoyance and sensitivity, that constitute a specific research area, and have been studied from different perspectives, such as in connection with traffic noise [121; 79]. Most people are sensitive to noise [47].
This thesis focuses on individuals who suffer from unusual intolerance to ordinary environmental sounds, as defined by Vernon [131].

Diagnosis

There is no consensus-based standardized diagnostic procedure for hyperacusis [91]. An obstacle in diagnosing hyperacusis is distinguishing between different types of sensitivity to sounds, some of them described earlier. A probable reason for confusion is that sound sensitivity is treated by several disciplines, not just audiology but also neurology and psychiatry [11]. The term hyperacusis is in addition often used in audiological literature to describe many different types of sound sensitivity, making matters even more complicated, as not all persons who experience adverse effects of sound exposure actually suffer from hyperacusis.

When a person comes to an audiology department with complaints of hypersensitivity to sounds the person is normally examined at an outpatient clinic. Common procedure is to take a medical history, followed by audiological testing. Such testing takes place in soundproof test rooms, where pure tone audiometry is performed to detect hearing deficits. The loudness discomfort level (LDL) test is likely to be used for diagnosing hyperacusis, as it has been recommended both for diagnosis and as a treatment outcome measure [63; 99]. When taking the LDL test, the patient is presented with signals of various frequencies, first at a low volume, followed by gradually increasing volumes. The patient indicates when the volume becomes uncomfortable, either verbally or with some type of sign. An individual is suggested to meet test criteria for hyperacusis if LDLs are generally less than 90 dB in the measured ear [2]. The LDL test has been criticised for having problems with reliability, and also for being highly uncomfortable or even painful for persons suffering from hyperacusis [12]. Also worth considering is the fact that stimuli used in the test situation are restricted and easily controlled, and thereby different from those experienced in daily life [100].

A few questionnaires are available in clinical audiological practice. The Hyperacusis Questionnaire (HQ) has been used to measure hyperacusis severity in research [72] and has been translated into Swedish [17]. The Multiple-Activity Scale for Hyperacusis [27] and the Questionnaire on Hypersensitivity to Sound [101] are other questionnaires that are used internationally, but there are no Swedish versions of these instruments.

The above-mentioned audiometric measures have different characteristics. While the LDL test assesses the momentary discomfort of a defined tone of a certain volume, the questionnaires contain a wide variety of questions on the perception of various everyday sounds and the consequences with respect to social life. Expectedly, these measures correlate moderately or not at all [91], concluding an obvious need for improved diagnostics.
Pathophysiology

In the majority of patients with hyperacusis, no underlying medical cause can be found [10]. An attempt to categorize these patients divides possible causes of hyperacusis into four different groups: a) clinical conditions involving the peripheral auditory system; b) clinical conditions involving the nervous system; c) endocrine and infectious diseases, and d) unknown causes [66].

The clinical conditions involving the peripheral auditory system, e.g. Bell’s Palsy and Ramsay Hunt Syndrome, are rare (Baguley, p 16 in [99]). Conditions in this category have been questioned as direct causes of hyperacusis, as several include facial nerve dysfunction which innervates the stapedial reflex. Thereby, this type of sound sensitivity cannot be considered hyperacusis [10]. An audiological problem that results in sensitivity to sounds is the superior semi-circular canal dehiscence syndrome [93]. The syndrome is very rare and results in hearing loss, vertigo and so-called conductive hyperacusis. The syndrome is probably due to failure of bone development, resulting in a third window into the inner ear. This increases pressure between the normal windows, thereby causing sensitivity to sounds.

Further according to this division, clinical conditions involving the nervous system are assumed to include migraine and depression [66]. A proposed mechanism in hyperacusis is disturbed metabolism of 5-hydroxytryptamine (5-HT), also referred to as serotonin [88]. This is suggested because sound sensitivity is often present in conditions where this function is disturbed, e.g. migraine, depression and post-traumatic stress disorder [66].

Examples of endocrine and infectious diseases are Addison’s disease and Lyme disease [66]. The above are explanations for the onset of sound sensitivity, although whether hyperacusis is what is described is not definite.

Tinnitus is defined as a phantom auditory perception of sounds in the ear and/or head in the absence of an external source [3]. As tinnitus and hyperacusis are often assumed to be linked [12], an interesting theory, initially concerning the development of tinnitus, is the Jastreboff neurophysiological model (JNM) [63; 61]. Applied to hyperacusis, the so-called “central gain hypothesis” [61] views hyperacusis as a result of a problematic compensatory gain process in the auditory pathways. The increased gain or sensitivity in the central auditory system can be influenced by three factors: the amount of sound around the individual; the individual's mood; and how the person interprets incoming sounds, i.e. the meaning of the sounds to the individual. This activity is assumed to affect the limbic system and the autonomic nervous system for the particular sound. Hyperacusis has been linked to hyperactivity within the central auditory system [43] in a group of patients with a primary complaint of tinnitus. This was considered to be evidence of abnormal gain within the auditory pathway in this group of patients.
In some studies of animals, researchers argue that acoustic trauma affects the central gain by increasing it [102; 76]. This has been discussed as being contributory to hyperacusis, assuming that acoustic trauma is a possible start of hyperacusis symptoms [76]. However, these symptoms are associated with inner ear damage, which may point to another type of sound sensitivity than hyperacusis.

In one study, women with high levels of emotional exhaustion presented with lowered LDLs, compared to women with lower levels of emotional exhaustion, after performing a stressful task [50]. The possibility of stress as a factor in tinnitus has also been investigated [117]. Basic cortisol levels have been shown to be higher in tinnitus patients [52].

Prevalence

In one of the very few existing prevalence studies of hyperacusis, which was performed in the Swedish population, the point prevalence was 9 % for individuals responding via the internet and 8 % in a postal survey [4]. When excluding individuals with hearing impairment, which may imply suffering from loudness recruitment, the prevalence rates were 7.7 % ($n = 39$) and 5.9 % ($n = 28$), respectively. In this study, hyperacusis was elaborately defined with focus on finding proof that everyday sounds were perceived as annoying or unpleasant. Examples included for instance reactions to conversation and to paper noises. The researchers made clear that they were not asking about reactions to sounds most people perceive as loud, such as drilling machines, but to sounds that are experienced daily. Hence, this study focused on everyday sounds in accordance with the definition by Vernon [132].

Hyperacusis rates have been reported to be 15 % in the Polish population [120]. This was determined with a 13-item questionnaire mainly asking about tinnitus and tinnitus-related problems, but also about “presence or lack of hyperacusis”. It is unclear whether hyperacusis was further defined and also whether the 17 % of persons who reported suffering from hearing loss also reported hyperacusis, thereby resulting in the risk of including loudness recruitment and other types of sound sensitivity in the estimation.

In a Finnish study, the prevalence of self-reported hyperacusis was 17 % [48]. In this study, a person was considered to suffer from hyperacusis if answering “Yes” to the question “Are you particularly sensitive to loud sounds?” It is difficult to measure a problem such as hyperacusis reliably with only one question. There is a risk that this question measured sensitivity to sounds that probably actually were of a high volume, i.e. noise, thereby including individuals who did not meet the full criteria for hyperacusis.

The different prevalence figures reported seem to be related at least in part to differences in the definitions of hyperacusis that were applied. Regarding even the lowest prevalence rate mentioned above in the internet and postal study [4], there is reason to believe that some of the individuals who
were considered to suffer from hyperacusis actually did not do so. For instance, only 30% of them reported protecting themselves from everyday sounds, which seems unlikely based on clinical experience, as over-protection is very common.

Comorbidity

Somatic comorbidity

Hyperacusis has been reported to occur together with many different somatic problems and syndromes, including the diagnoses discussed in the pathophysiology section.

Another commonly mentioned example is autism. In one study, children and adolescents with autistic disorder presented with an 18% prevalence of hyperacusis, while in an age-matched group of non-autistic children there were no cases of hyperacusis [108].

Williams’ syndrome is a multisystem disorder characterized by dysmorphic facial features, mental retardation and a distinctive personality including elevated levels of anxiety [97]. Hyperacusis is commonly present in this patient group, with reported figures of up to 95% [73].

In migraine, the specific type of sound sensitivity called phonophobia is common [116]. There is evidence that persons with migraine are more sensitive to sounds both during and between attacks [135]. Sound sensitivity has also been reported together with traumatic brain injury [78].

Idiopathic sudden sensorineural hearing loss, or “sudden deafness”, is unexplained unilateral sensorineural hearing loss with onset over a period of less than 72 hours [107]. To these patients, sound may seem harsh and distorted, and they are generally recommended to wear earplugs. A clinical observation is that these patients are likely to present with hyperacusis.

The comorbidity between tinnitus and hyperacusis is large [10]. Between 40 and 60% of patients with tinnitus suffer from hyperacusis [64; 6], while up to 86% of patients with a primary complaint of hyperacusis also report tinnitus [2]. However, in the Swedish population-based prevalence study mentioned earlier, tinnitus was investigated both in the internet group and the postal group. In the internet group there was a reported prevalence of tinnitus of 21% among the persons who suffered from hyperacusis. This was significantly higher than in the persons who did not have hyperacusis. The postal group reported a 9% prevalence, the same as in the comparison group [4].

Comorbidity between different types of sound sensitivity such as hyperacusis and specific phobia for a certain sound, or hyperacusis and loudness recruitment, is theoretically possible. Further, a person suffering from a hearing loss could also suffer from hyperacusis, even if persons with hyperacusis are likely to have normal hearing.
Psychiatric comorbidity
In a study investigating psychiatric morbidity in patients with tinnitus and hyperacusis, 69 % of the patients had a present psychiatric diagnosis, with affective disorders diagnosed most frequently (57 %). The frequency of anxiety disorders was 43.5 % [37]. Hyperacusis has furthermore been reported in depressed patients [23].

In another study based on data from self-report questionnaires, it was concluded an association between hyperacusis and anxiety, as patients with both tinnitus and hyperacusis were significantly more likely to suffer from anxiety symptoms than patients with tinnitus alone [16].

In post-traumatic stress disorder, an exaggerated startle reaction to acoustic stimuli is common [22], and reducing this reaction with cognitive behaviour therapy (CBT) seems to be possible [42]. Also, there appears to be an abnormality in this patient group with respect to processing environmental auditory stimuli [89].

Furthermore, it has been reported that women with a high degree of sensitivity to noise exhibit more psychiatric symptoms when compared to women with lower degrees of sensitivity [122]. It can also be noted that psychiatric comorbidity has been investigated in patients with tinnitus, and depressive and anxiety disorders have been found to be common [140; 49]. Furthermore, there is a large co-occurrence between anxiety disorders and chronic pain [8].

Psychological aspects of hyperacusis

Personality
Studies of personality traits in patients with hyperacusis are sparse. In one study, patients with both tinnitus and hyperacusis had higher scores of trait anxiety, than patients with tinnitus alone [16].

Also, it has been reported that women who are highly sensitive to noise exhibit significantly more neurotic personality traits than women who are less sensitive [122]. Neuroticism refers to a tendency to respond with a negative emotional response to threat, frustration and loss [38]. Persons with higher levels of neuroticism are more vulnerable to many disorders such as depression and anxiety disorders [68]. The economic costs of neuroticism are immense, and it has been argued that neuroticism should be a primary target in psychological treatments [26]. A relation between noise sensitivity and traits of introversion has also been reported [130].

Avoidance behaviour
Some researchers have suggested that hyperacusis is maintained and exacerbated by avoidance of sounds [112; 132; 35]. This has actually been shown in an experimental study where persons with normal hearing, and without
hyperacusis, became more sensitive to sounds when they overprotected their ears [35], while exposure to low-level noise later desensitized these individuals.

In clinical practice with patients suffering from hyperacusis, the behavioural avoidance of sounds is likely to mainly consist of not spending time in environments perceived as loud. This avoidance behaviour may constitute a more or less profound change in the individual's life. Some persons who suffer from hyperacusis experience difficulties in their work environments and try to avoid them; sometimes their absence from work is even certified as being due to illness by their medical doctors. Others find contexts such as social gatherings to be problematic. Driving or riding in cars and taking the bus are often avoided by persons who are sensitive to sounds. The life of the sound-sensitive person sometimes becomes very inactive, as most activities in life include at least some degree of sound exposure [46; 2; 12]. In summary, it is easy to imagine the consequences of hyperacusis with respect to quality of life.

Other common behaviour among patients with hyperacusis that is aimed at avoiding loud sounds is the wearing of ear-protection devices in situations that are difficult to endure. This behaviour is also commonly observed in sound-sensitive persons in general [53]. By using ear-protection devices, unpleasant sounds are avoided and the fear of encountering possibly existing but unknown sounds is alleviated. Some individuals use earmuffs or cotton pads to shield themselves from sounds. As there is an enormous diversity of ear-protection devices on the market, it is possible to buy several kinds and brands with different degrees of protection. Many combine different ear-protection devices and some even develop their own, sometimes in peculiar shapes and forms, such as helmet-like devices, that may further complicate communication, social interaction and life in general.

The tendency to cope with something unpleasant by avoiding it is well described in different areas of psychiatry [110; 86]. It is also commonly found in the area of behavioural medicine, for example in those suffering from tinnitus [54; 16] and chronic pain [80; 18].

To understand how individuals with musculoskeletal pain develop chronic pain, the “fear-avoidance model” was developed [81]. In this model, fear of pain is central, and the behavioural response to pain is either to confront it or to avoid it. Confronting the pain leads to reduction of fear in the long run, while avoidance is hypothesized to lead to exacerbation of fear. There is evidence that pain-related fear and avoidance are important in the development of a chronic problem for a substantial number of patients with musculoskeletal pain [136]. This model could possibly be applied to hyperacusis, as fear of sounds, or fear of the anxiety and pain associated with sounds, seems to lead to avoidance behaviour, while confrontative behaviour might possibly reduce the fear.
Treatment

There are no established treatment guidelines for hyperacusis, or published randomized controlled trials (RCT) of any psychological treatment for hyperacusis. The care a patient is most likely to receive at an audiology department consists merely of an audiologic examination, including LDL measurement [12]. Some patients leave the clinic with prescribed, fitted earplugs with sound attenuation devices. Another common recommendation for these patients is to seek out support groups. Some form of professional counselling might be available at the audiology department or elsewhere, perhaps at a psychiatry department if depression or other psychiatric morbidity is present.

Tinnitus Retraining Therapy (TRT) [61] was developed to treat tinnitus, and has also been used to treat hyperacusis [34]. This method includes a medical evaluation, directive counselling, and sound therapy, where patients use a so-called masker, which is a device fitted into the ear that produces pink noise.

Other types of sound exposure treatments are being used in clinics. In one study patients with hyperacusis were exposed to pure tone stimuli for several hours a day for 15 weeks [103]. The patients had hearing loss and the sound stimulation was given on the frequency of the loss. Due to the existing hearing loss and other problems in this study, it is difficult to draw any conclusions, although significant effects on loudness scaling were obtained. Open-field sound exposure treatment has been shown to be possibly useful [85; 94].

Cognitive behaviour therapy (CBT)

According to the biopsychosocial model, disease should be considered biologically, psychologically and socially [32]. The biological part corresponds to the pathophysiology in the syndrome. In the psychological part, emotions and behaviours are accounted for. The social part refers to the social consequences of a problem. It has been argued that hyperacusis belongs within the framework of the biopsychosocial model [12].

In that context, CBT can be viewed as one of many existing psychological therapies. CBT has proven to be effective for a range of psychiatric disorders, and should be considered the psychosocial treatment of choice for many patients with mild to moderate psychiatric problems [125], including anxiety disorders [60]. CBT is also effective for a range of somatic problems such as chronic pain [96], tinnitus [55], and irritable bowel syndrome [84].

In CBT, there are two main orientations, based on two different theoretical frameworks. The first is the cognitive model, where the main target for analysis and treatment is the cognitive perception of events [14; 13]. Cogni-
Cognitive therapy emphasizes the importance of changing unrealistic thinking patterns. In cognitive therapy, an important term is “safety-seeking behaviours”, sometimes referred to simply as “safety behaviours” describing behaviours performed to avoid a perceived threat [110]. This type of avoidance behaviour is considered a crucial factor in maintenance of e.g. anxiety disorders, as disconfirmation of threat is unlikely when an individual has performed this type of behaviour.

The other theoretical framework in CBT, the behavioural, is based on the laws of learning. Respondent and operant conditioning comprise principles for learning and perpetuating behaviours [124]. Respondent conditioning refers to the establishment of a response by pairing a neutral stimulus with an unconditioned stimulus until the neutral stimulus elicits a conditioned response. Operant conditioning refers to the type of learning in which an individual's behaviour is modified by its consequences. The types of consequences are positive reinforcement and negative reinforcement, both of which increase the frequency of behaviour, and positive punishment and negative punishment, both of which decrease behaviour [124].

Mowrer’s two-factor theory of avoidance combines these two types of conditioning and makes it possible to understand the development of avoidance behaviours [98]. In the respondent conditioning part of the theory there is a pairing of a neutral stimulus with an aversive unconditioned stimulus. Due to the aversive nature of the unconditioned stimulus, the conditioned stimulus will elicit a conditioned emotional reaction from this point on, which serves to motivate operant responses. Responses that terminate the conditioned stimulus are reinforced by termination of the aversive affect (negative reinforcement). The individual escapes an aversive internal state that is caused by the conditioned stimulus [98].

In clinical practice, the focus of CBT is on resolving current problems by means of teaching the patient about development, maintenance and treatment of the present syndrome, about applying methods of self-analysis concerning cognitions and behaviours, and about trying different techniques for dealing with the problem [133]. Treatments are in general short-term, with goal-oriented, structured sessions and an active therapist. The patient and the therapist collaborate in working with different techniques to alter cognitions and behaviours. Common techniques include exposure techniques, behavioural experiments and behavioural activation. Homework is assigned between sessions to improve treatment results. Treatments end with a recapitulation and formulation of a relapse prevention plan.

CBT in audiology

With regard to research on CBT in audiology, most studies concern tinnitus, although occasional studies are found in other areas such as vestibular dysfunction/dizziness [31].
Tinnitus is generally seen as a chronic condition that is difficult to treat [29], and the main intention of CBT treatment is therefore to alleviate suffering from tinnitus. “The aim of psychological interventions is not to “cure” or to eliminate the inner noise but to reduce tinnitus-related distress and increase quality of life” [41]. For the psychological problems associated with tinnitus, CBT is an effective treatment and is the treatment of choice [5; 55]. There are several similarities and high comorbidity between tinnitus and hyperacusis. As mentioned earlier, it has been proposed that they have a common origin, and the Jastreboff neurophysiological model suggests that both result from a maladjusted central gain, abnormally increased activity within the auditory pathways [63]. Behavioural avoidance may be important in perpetuating both problems, as there is a relationship with both tinnitus distress [54] and hyperacusis [16].

In manuals for CBT treatment of tinnitus, hyperacusis is sometimes also addressed as part of the problem [65]. Interestingly, sound stimulation does not seem to have any additive effect on CBT for tinnitus [57].

CBT model for hyperacusis
Several scientists have suggested CBT, together with information counseling, relaxation therapy and sound therapy, as a reasonable strategy to counter the anxiety and stress associated with hyperacusis, although this has not been verified in research [10; 16]. Exposure to sounds by itself is also advocated in the literature in order to alleviate hyperacusis [43].

Behavioural avoidance is crucial for developing anxiety disorders, and is also a key for treating them. From a CBT perspective it is easy to see similarities in behavioural avoidance in hyperacusis, tinnitus and anxiety disorders. Many patients that develop hyperacusis start to avoid certain environments, and use earplugs extensively. These behaviours protect them from the instant unpleasantness of certain sounds. Unfortunately, overprotection of the ears exacerbates hyperacusis [35]. Due to this, and to other possible consequences of avoiding sound, i.e. isolation, depressed mood and problems in maintaining relationships, it would be sensible that the main goal of CBT for hyperacusis would be to target the present avoidance behaviour.

To illustrate how hyperacusis is maintained by avoidance according to the earlier mentioned Mowrer’s two-factor model, consider Elisabeth, described in the clinical vignette. Elisabeth experienced pain in her ears at work. Her experience led to a respondent conditioning, pairing sounds at her workplace with pain in her ears and fear of hearing loss. To reduce the pain and fear, she used earplugs and avoided many situations with a perceived risk of exposure to uncomfortable sounds, leading to negative reinforcement of her avoidance behaviour. The frequency of occurring avoidance behaviour therefore increased. Instantly, the pain or the risk of experiencing pain decreased, but in the long run Elisabeth will be more afraid of sounds and this will also
affect her auditory gain, and her auditory system will become more sensitive to sound.

In treatment, it is essential for the patient to understand these principles in order to be able to work together with the therapist to reverse the adverse effects of avoiding normally loud sounds. A tentative CBT treatment approach for Elisabeth would be to start with an introduction to the CBT model for hyperacusis. Together with her therapist, she would then set her treatment goals, and plan for exposure therapy by exploring and arranging situations she avoids, in order to prepare for controlled and stepwise exposure. There would also be a plan for sound enrichment. The exposure therapy would take place in the sessions but also in between sessions, both at her workplace and at home. In addition, Elisabeth would learn applied relaxation, and later to restart the activities she had quit due to hyperacusis. Finally, there would be a recapitulation, and a relapse prevention plan would be constructed.
The Department of Ear, Nose and Throat diseases at the Uppsala University Hospital has extensive experience of treating tinnitus with CBT. The treatment is given by licensed psychologists either individually, in a group format, or via the internet. Many of the patients who take part in tinnitus treatment complain about hyperacusis, and in several cases hyperacusis is perceived as more troublesome than tinnitus.

The overall aim of the present project was to gain knowledge about the clinical condition of hyperacusis, defined as unusual intolerance to ordinary environmental sounds [132].

The specific aim of Paper I was to compare the hyperacusis measurement tools often used in audiological practice in order to determine the most valid measures for assessing hyperacusis. A structured clinical interview for hyperacusis was also constructed and used to investigate the patient group.

A further aim of this project, which was addressed in Paper II, was to investigate the patient group further with respect to individual characteristics, and also to assess psychiatric morbidity and personality traits. It was hypothesized that individuals with certain psychiatric disorders, in particular those related to anxiety, would be over-represented among patients with hyperacusis. Psychiatric morbidity was assessed, based on the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV) [1] criteria in patients with hyperacusis. Furthermore, personality traits were assessed by means of the Swedish universities Scales of Personality (SSP).

In Paper III, the aim was to investigate whether CBT could be helpful for patients with hyperacusis. Due to the common comorbidity between tinnitus and hyperacusis, there is an intervention in the department’s ordinary tinnitus protocol that is aimed at treating co-existing hyperacusis. Up until now, this part of the tinnitus treatment has not been extensive, but since many patients refer to hyperacusis as their primary, and sometimes only audiological complaint, it was concluded that there was reason to extract, develop and evaluate the hyperacusis module. The effect of CBT for hyperacusis was assessed in a randomized controlled fashion, where the control group remained on the waiting list. The hypothesis was that patients with hyperacusis would benefit from CBT, as measured by loudness discomfort levels, hyperacusis symptoms, anxiety and depressive symptoms, fear of (re)injury due to exposure to sounds, and quality of life.
Methodology

Participants and procedures in Papers I, II and III

All studies were based on the same participants. The inclusion criteria were the same for all studies and were set according to the definition of hyperacusis by Vernon [132]: unusual intolerance to ordinary environmental sounds. To be included in this project, patients:

1. reported hyperacusis as their primary audiological problem
2. had average loudness discomfort levels less than 90 dB in at least one ear
3. presented with hearing thresholds of at least 40 dB in the best ear
4. were between 18 and 65 years of age
5. were able to understand and speak Swedish well
6. gave informed consent to participate

Patients were not included if they were suffering from psychotic disorder, severe depression, or answering affirmatively on questions about suicidality. The patient flow is presented in Figure 1.

In total, 81 individuals were screened for participation via telephone. Out of them, 73 were consecutive patients referred to the Department of Ear, Nose and Throat diseases at Uppsala University Hospital, and the remaining patients were self-referred to the same department as they had gained knowledge about the project by attending lectures given by members of the research team. All patients who were referred to a psychologist at the department with a primary complaint of hyperacusis during the timeframe of the trial, April 2005 to September 2009, were considered for participation.

Seven individuals in all were not included after the telephone screening. Three of them verbally reported a hearing impairment that was shown to be worse than 40 dB when consulting their medical journals. Two individuals were not included because tinnitus was their primary audiological problem, and two declined participation after receiving information about the study.

The remaining 74 patients were invited to undergo further evaluation at the department, and 62 of those were later included. The clinical evaluation included information about the project, a medical examination, audiological testing, a clinical interview and a structured psychiatric interview, the Mini-
International Neuropsychiatric Interview (MINI). The patients also completed several questionnaires. Out of the twelve patients not included, six had normal LDLs and two realised that tinnitus was their primary problem. One patient declined participation because he did not want to undergo LDL measurement. Three patients suffered from such severe psychiatric problems that it was concluded that they were likely to interfere with participation in the study. Two of these three patients answered the MINI questions about suicidality affirmatively, and the third patient had a psychotic disorder, and they were therefore immediately referred to a psychiatric clinic. The not included patients who were likely to benefit from ordinary CBT treatment at the present clinic were offered this.
Procedure in Paper III

After evaluating whether the patient was eligible for inclusion in the study, a carefully folded card, on which was written either "Treatment" or "Control", was randomly picked out of a box by the research leader. In this way patients were assigned to a treatment or control group.

The treatment group received CBT as soon as possible, in general starting within a week from randomization. The control group remained on the waiting list for six months, and were then evaluated again with audiological measures and with the questionnaires. Two patients did not return after the six months. The remaining 30 patients from the waiting list group were offered CBT treatment which they all accepted. After treatment, and again 12 months after completed treatment, all patients were evaluated by means of the audiological measures and the questionnaires.

In total, five patients dropped out of treatment at different stages, one after only one treatment session, two after two sessions, one after the fourth session, and one person did not show up for the 12-month evaluation. Three of these patients dropped out of the control group, and 2 dropped out of the treatment group. The reasons for drop-out were: personal crisis due to illness of a close relative in one case (drop-out after session 2), moving out of the country in one case (drop-out after session 4), and three were unknown.

Measures

Clinical characteristics

A clinical interview was constructed (See Paper I) that included questions about demography, onset and development of hyperacusis. Many of the questions aimed at investigating whether the comorbid conditions mentioned in the literature, including tinnitus, migraine and different sensitivities such as sensitivity to light, were present. There were also items investigating the patients’ behavioural avoidance of sounds, including whether they used ear protection and if so in which situations.

Audiological measures

Pure tone audiometry was performed using ascending technique [7] on an audiometer AC 40 from Interacoustics. Hearing thresholds were measured in dB (HL) at 125 to 8000 Hz. Pure tone averages (PTA) were defined as the average of the frequencies 500, 1000, 2000 and 3000 Hz for each ear. For loudness discomfort levels (LDL) for tones, the measured frequencies were 250, 500, 1000, 2000, 3000 and 4000 Hz. Patients were instructed to indicate
when the sound level became uncomfortable for them. If this occurred at an average level of 90 dB or less for the frequencies of 500, 1000 and 2000 Hz for at least one ear, the person was considered to meet test criteria for hyperacusis [2].

Psychiatric disorders

The Mini-International Neuropsychiatric Interview (MINI, Swedish version 5.0.0.) was administered to investigate psychiatric comorbidity. The MINI is a short, structured diagnostic interview with satisfactory validity and reliability when validated against more comprehensive systematic interviews [114; 123]. The MINI has been used in a wide range of both clinical and research applications including for tinnitus [87].

The MINI detects the most common DSM-IV and ICD-10 (International Statistical Classification of Diseases and Related Health Problems) psychiatric disorders: major depressive episode, dysthymia, manic episode, hypomanic episode, panic disorder, agoraphobia, social phobia, obsessive compulsive disorder, post-traumatic stress disorder, generalized anxiety disorder, psychotic disorder, alcohol abuse, alcohol dependence, drug abuse, drug dependence, anorexia nervosa and bulimia nervosa. It also assesses suicidality and anti-social personality disorder; the latter was not utilized in the present study.

Personality traits

The Swedish universities Scales of Personality (SSP) [45] was used to measure personality traits. The inventory consists of 13 scales with a total of 91 items, seven items for each scale. The scales are: 1) Somatic Trait Anxiety (STA), 2) Psychic Trait Anxiety (PsTA), 3) Stress Susceptibility (SS), 4) Lack of Assertiveness (LA), 5) Detachment (D), 6) Embitterment (E), 7) Trait Irritability (TI), 8) Mistrust (M), 9) Impulsiveness (I), 10) Adventure Seeking (AS), 11) Social Desirability (SD), 12) Verbal Trait Aggression (VTA), and 13) Physical Trait Aggression (PhTA).

The SSP has been standardized in a sex-stratified sample from the general Swedish population [45].

Hyperacusis severity

The Hyperacusis Questionnaire (HQ) was developed by Khalfa and colleagues for quantification and characterization of hyperacusis [72]. It consists of 14 items that can be divided into three subscales: attentional, social and emotional aspects.

The Swedish version by Blomberg and colleagues was used [17]. In that study, the translation was reviewed by two linguists and a psychologist who
was specialized in hearing disorders. No Swedish standardization exists for this questionnaire.

Symptoms of anxiety and depression
The Hospital Anxiety and Depression scale (HADS) [139] has been recommended for measuring anxiety and depression in patients with hyperacusis [12]. The HADS was developed for use with somatic patients and consists of 14 items that are divided into two scales with half of the items measuring anxiety and half measuring depression.

The test-retest reliability of the scale is $r = 0.80$. The internal consistency is $\alpha = 0.80 - 0.93$ for the anxiety scale and $\alpha = 0.81 - 0.90$ for the depression scale [82].

Quality of life
The Quality Of Life Inventory (QOLI) [36] has generally shown good psychometric properties in empirical studies, with test-retest reliability reportedly between $r = 0.80$ and 0.91, and internal consistency between $\alpha = 0.77$ and 0.89.

The QOLI consists of 17 items measuring satisfaction in particular areas of life considered important by the person in question. For instance, for questions about health, relations with relatives or economy, there are ratings of both importance and satisfaction. The scale has been used in studies of tinnitus [134].

Fear-avoidance
The Tampa Scale for Kinesiophobia (TSK) measures fear-avoidance beliefs and fear of (re)injury due to movement [92], and is a measure that has been used frequently in pain research. The TSK was adapted to hyperacusis, and is thereby aimed at measuring fear of (re)injury due to exposure to sounds.

The TSK for hyperacusis consists of 17 items. Each item is evaluated on a 4-point Likert scale with scoring alternatives ranging from “Strongly disagree” to “Strongly agree”. An example of an item is: “I’m afraid that I might injure myself if I expose myself to sounds”. The questionnaire has not been standardized in the present patient group.

Self-reported improvement
Drop-outs can be a major problem regarding being able to draw conclusions in RCTs. As a precaution, a self-report visual analogue scale was used to assess treatment response in the patients at every treatment session except
the first. This would make it possible to retrospectively analyse self-reported improvement in relation to number of treatment sessions completed.

Patients were instructed to make a single vertical mark on a horizontally oriented, 100-mm VAS labeled “not at all” at the far left and “a lot” at the far right, for the following three statements: 1) “How bothering is your sound sensitivity today?” 2) “How socially disabling is your sound sensitivity today?” 3) “How afraid/annoyed are you of/to sounds?”.

CBT treatment

Individual CBT treatment was given by a licensed psychologist with a minimum of two years basic training specifically in CBT. Four psychologists were engaged in this study and they all received supervision.

The treatment consisted of six sessions. The first session was 90 minutes long and the following five lasted for 45 minutes each. The first four sessions took place once a week for four weeks, while the time gap between the last sessions was increased to two weeks. This is common procedure in CBT, leaving the patient in charge of the treatment as more and more of the work is performed independently in the patient's natural setting.

General CBT principles were applied, such as Socratic questioning, goal-setting and homework. The session structure was similar in the different sessions, starting with a recap of the previous session, agenda setting, going through homework assignments and psychoeducation. After this, applied relaxation was practiced, followed by exposure to sounds, and in the final two sessions behavioural activation was also on the agenda. New homework was planned cooperatively. During the last treatment session, patients and their therapist summarized the therapy, evaluated the patients’ individual goals and constructed a relapse prevention plan.

The specific elements of the treatment were psychoeducation, sound exposure including sound enrichment, applied relaxation and behavioural activation. An overview of the treatment is presented in Table 1. To illustrate the different strategies, it will be referred to the treatment given to the fictitious patient Elisabeth.

Psychoeducation

The term psychoeducation refers to the education the patient is given in CBT, about the causes and effects of the patient's condition in general, and the structure and contents of the treatment [133].

In this study, psychoeducation consisted of education about the present CBT-model of the development of hyperacusis and about the treatment of hyperacusis according to this model. Patients also received information about sounds and sound levels and assessment of riskful sounds. The main
psychoeducative focus was on teaching the patient how avoidance of sound may affect fear/anxiety and the auditory system. Psychoeducative homework was given as written material, with essentially the same content that was presented by the therapist in the sessions.

Table 1. Overview of the CBT treatment.

<table>
<thead>
<tr>
<th>Session</th>
<th>Psychoeducation</th>
<th>Work in sessions</th>
<th>Homework assignments</th>
</tr>
</thead>
<tbody>
<tr>
<td>Session 1</td>
<td>CBT model for treatment Applied relaxation</td>
<td>Treatment planning Goal setting Applied relaxation 1</td>
<td>Psychoeducative material Applied relaxation</td>
</tr>
<tr>
<td>Session 2</td>
<td>CBT model for treatment Applied relaxation</td>
<td>Applied relaxation 2 Exposure therapy</td>
<td>Psychoeducative material Applied relaxation Exposure therapy Sound enrichment</td>
</tr>
<tr>
<td>Session 3</td>
<td>Risk assessment regarding sound Applied relaxation</td>
<td>Applied relaxation 3 Exposure therapy</td>
<td>Psychoeducative material Applied relaxation Exposure therapy Sound enrichment</td>
</tr>
<tr>
<td>Session 4</td>
<td>Applied relaxation</td>
<td>Applied relaxation 4 Exposure therapy</td>
<td>Psychoeducative material Applied relaxation Exposure therapy Sound enrichment</td>
</tr>
<tr>
<td>Session 5</td>
<td>Behavioural Activation Relapse prevention</td>
<td>Applied relaxation 5 Exposure therapy</td>
<td>Applied relaxation Exposure therapy Sound enrichment Behavioural Activation</td>
</tr>
<tr>
<td>Session 6</td>
<td>Behavioural Activation Relapse prevention</td>
<td>Exposure therapy Goal evaluation Relapse prevention</td>
<td>Applied relaxation Exposure therapy Sound enrichment Behavioural Activation</td>
</tr>
</tbody>
</table>

Sound exposure

Exposure to sound was executed in part as the traditional CBT-technique of graded exposure that is commonly used e.g. when treating specific phobias. Sound exposure also took place with a general purpose of environmental sound enrichment.
The first type of exposure targets the fear or anxiety reactions to sounds that many patients with hyperacusis experience. The patients first compiled situations including exposure to sounds hierarchically, assigning them numbers from 0 to 100 in difficulty to endure due to the loudness. Exposure then started around level 30 - 40 in the sessions, and patients continued with the same task at home, as homework. Homework was planned and registered, and the patient was instructed to perform it every other day. *Elisabeth* practiced dropping cutlery on plates. She started out in a session by dropping small teaspoons from a height of only a few centimetres, to gradually increasing the size and number of utensils and the height from which she dropped them.

The second type of exposure to sounds focused on increasing general sound levels at the patients’ homes, workplaces and other locations of importance to the individual patient. For *Elisabeth*, one part of this work was to lessen her use of ear protection devices in everyday situations, first by changing devices from her ordinary with a higher degree of protection, to a pair that protected her less, and later by making little cuts in her earplugs, thereby making protection less effective. Another part of this work was for her to listen to her favourite choir music at home, starting with a very low volume the first week but for a more extended period of time, around one hour per session. The volume was then gradually increased every week until *Elisabeth* reached a level that was considered normal by members of her family.

**Applied relaxation**

A shortened version of the technique of applied relaxation [141], as is used in the department’s ordinary treatment program in CBT for tinnitus [65], was practiced with the purpose of lowering the general level of stress, and making it possible to endure better in specific difficult situations. This technique starts with progressive relaxation, with the aim of teaching patients to be able to recognize when their muscles are tense. Further on in the manual, patients learn how to condition themselves to relax very quickly in specific situations. Patients learn to link relaxation to breathing, and also to a specific word chosen by the patient.

Homework consisted of practicing the different stages in applied relaxation at home or in other natural environments, several times a day. When *Elisabeth* returned to work, where stress reduction was much needed, she practiced applied relaxation several times a day, in combination with the task of overseeing the children when they were playing outdoors.
Behavioural activation

Also included in the treatment was a condensed version of the treatment manual for behavioural activation, that is normally used for treating depression [28]. As it is common to avoid different activities due to the sound environment, this part of the treatment aimed at gradually restarting activities patients had regretfully given up due to hyperacusis. This was supposed to result in a change in mood, but was also a way to expose the patients to different situations they avoided, thereby reducing fear of sound and restoring the auditory system.

Homework included restarting activities once perceived as positive in life; for instance, Elisabeth began going to restaurants again. She started by visiting small, quiet restaurants and continued with more difficult ones, until she was again able to visit a McDonald’s restaurant at lunchtime with her children.

Ethics

The International Clinical Trial registration number of this project is NCT01321814. The studies were approved by the Regional Ethical Review Board in Uppsala. All patients gave their written informed consent in accordance with recommendations from the Declaration of Helsinki [138].
Data analyses

Paper I
To investigate the degree of agreement between the LDL test, the HQ, the HADS and items in the clinical interview, Pearson correlation coefficients were calculated.

The t-test was used to compare mean differences between the right and left ears for the LDLs and hearing threshold values.

Paper II
The prevalences of psychiatric disorders, investigated with the MINI, were compared with data from the National Comorbidity Study [71] that had been adjusted to the male/female ratio present in the hyperacusis group of patients. Data were given as proportions with 95 % binominal confidence intervals.

The SSP data were adjusted for sex, and t-scores were calculated, with a mean of 50 and a standard deviation of 10 based on the Swedish normative sample [45]. Personality data were compared with t-tests.

Paper III
Demographic and baseline differences were tested with the t-test for continuous data and the chi-square test for categorical data.

The analysis of covariance (ANCOVA) was used to evaluate the treatment effect in the two groups. Post-treatment values for the treatment group were compared with pre-treatment values for the control group. The baseline measure was entered as a covariate to adjust for initial differences between groups. Twelve-month follow-up values were compared with post-treatment values to test if treatment effects were sustained.

Between- and within group effect sizes were measured with Cohen’s d, where a d of 0.2 was considered a small effect, 0.5 a medium effect and 0.8 and above a large effect [25].
Predictive Mean Matching was used to impute missing data, which was done for the primary outcome measure only, due to the low percentage of drop-outs.

P-values less than 0.05 were considered statistically significant for all analyses. Data analyses were performed using SPSS versions 18, 20 and 21 for Windows.
Results

Papers I & II

Demography

The majority of patients were female (76 %) and the average age was 40.2 years (SD = 12.2). Almost half of the patients (44 %) reported being on sick leave or retired due to hyperacusis. The patients were well educated; 41 (66 %) had a university education. Also, out of the 11 students, eight were university students. The most common profession, engaged in by 23 persons (37 %), was teaching.

The most interesting finding from the clinical interview was that a majority of the patients (89 %) had given up an activity due to developing hyperacusis. The most common activities reported were work-related, but recreational activities such as attending concerts, going to the cinema, and social activities such as visiting restaurants were also commonly avoided. Ear protection devices were used by 82 % of the patients in situations in which most people do not use such devices, such as when vacuuming or washing the dishes.

When exposed to sounds perceived as too loud, 53 % of the patients described a general sense of being uncomfortable, 47 % felt annoyed, 45 % felt pain and 32 % reported that they felt frightened. Twenty-nine per cent of the patients also described other reactions, the most common of which were stress and trouble concentrating. Demographic data are presented in Table 2.

Audiological results

Hearing thresholds were measured in dB (HL) at PTA at the frequencies 500, 1000, 2000 and 3000 Hz, for each ear. The patients had normal hearing levels with mean values of 10.6 dB (SD = 9.8) in the right ear and 12.3 dB (SD = 9.8) in the left ear. There was an expected significant difference in the hearing thresholds between right and left ears at the frequencies 125, 250, 500 Hz (\(p < 0.01\)) and 3000, 4000 and 6000 Hz (\(p < 0.05\)), with the right ear outperforming the left. Mean LDLs ranged from 69.3 dB (SD = 12.1) to 76.0 dB (SD = 11.7) at the frequencies 250, 500, 1000, 2000, 3000 and 4000 Hz for each ear.
Table 2. Results of the clinical interview, n = 62.

<table>
<thead>
<tr>
<th>Characteristics</th>
<th>N</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age in years, mean (SD)</td>
<td>40.2 (12.2)</td>
<td></td>
</tr>
<tr>
<td>Females / Males</td>
<td>47 / 15</td>
<td>76 / 24</td>
</tr>
<tr>
<td>MARITAL STATUS</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Married or Living with partner</td>
<td>33</td>
<td>53</td>
</tr>
<tr>
<td>Single</td>
<td>29</td>
<td>47</td>
</tr>
<tr>
<td>PROFESSIONAL STATUS</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Academic profession (e.g. teacher, lawyer)</td>
<td>41</td>
<td>66</td>
</tr>
<tr>
<td>Non-academic profession (e.g. factory worker)</td>
<td>10</td>
<td>16</td>
</tr>
<tr>
<td>Student</td>
<td>11</td>
<td>18</td>
</tr>
<tr>
<td>OCCUPATIONAL STATUS</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Working full-time</td>
<td>18</td>
<td>29</td>
</tr>
<tr>
<td>Student</td>
<td>10</td>
<td>16</td>
</tr>
<tr>
<td>Unemployed</td>
<td>3</td>
<td>5</td>
</tr>
<tr>
<td>Retired</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>Sick leave / disability pension, part- or fulltime due to hyperacusis</td>
<td>27</td>
<td>44</td>
</tr>
<tr>
<td>due to other problems</td>
<td>3</td>
<td>5</td>
</tr>
<tr>
<td>HYPERACUSIS</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Onset (sudden / gradual / don’t know)</td>
<td>21 / 37 / 4</td>
<td>34 / 60 / 7</td>
</tr>
<tr>
<td>Development over time (worse / the same / don’t know)</td>
<td>39 / 18 / 4 / 1</td>
<td>63 / 29 / 7 / 2</td>
</tr>
<tr>
<td>Laterality (right / left / both ears)</td>
<td>3 / 3 / 56</td>
<td>5 / 5 / 90</td>
</tr>
<tr>
<td>Type of sounds</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Crockery</td>
<td>57</td>
<td>92</td>
</tr>
<tr>
<td>Children crying</td>
<td>55</td>
<td>89</td>
</tr>
<tr>
<td>Cutlery</td>
<td>51</td>
<td>82</td>
</tr>
<tr>
<td>Electronic sounds</td>
<td>45</td>
<td>73</td>
</tr>
<tr>
<td>Signals</td>
<td>44</td>
<td>71</td>
</tr>
<tr>
<td>Applause</td>
<td>44</td>
<td>71</td>
</tr>
<tr>
<td>Hammer blow</td>
<td>40</td>
<td>65</td>
</tr>
<tr>
<td>Ventilation sounds</td>
<td>36</td>
<td>58</td>
</tr>
<tr>
<td>Vacuum cleaner</td>
<td>35</td>
<td>57</td>
</tr>
<tr>
<td>Dental drill</td>
<td>33</td>
<td>53</td>
</tr>
<tr>
<td>Paper rustling</td>
<td>25</td>
<td>40</td>
</tr>
<tr>
<td>Flushing water</td>
<td>24</td>
<td>39</td>
</tr>
<tr>
<td>Hair dryer</td>
<td>22</td>
<td>36</td>
</tr>
<tr>
<td>Birdsong</td>
<td>18</td>
<td>29</td>
</tr>
<tr>
<td>Other</td>
<td>54</td>
<td>87</td>
</tr>
<tr>
<td>Reaction to sounds</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Discomfort</td>
<td>33</td>
<td>53</td>
</tr>
<tr>
<td>Annoyance</td>
<td>29</td>
<td>47</td>
</tr>
<tr>
<td>Pain</td>
<td>28</td>
<td>45</td>
</tr>
<tr>
<td>Fear</td>
<td>20</td>
<td>32</td>
</tr>
<tr>
<td>Other (e.g. stress)</td>
<td>18</td>
<td>29</td>
</tr>
</tbody>
</table>

There were no significant differences between right and left ears for the LDL values.

The mean duration of hyperacusis was 11.9 years (SD = 15.0, range 1 to 60). Ten patients claimed they had always suffered from hyperacusis, and therefore the median value for hyperacusis duration was calculated to be 5.0 years, with the 25th percentile 2.4 years and the 75th percentile 14.3 years. The onset of hyperacusis had been sudden according to 59.7 % of the patients, while 33.9 % claimed that the onset was gradual and 6.5 % did not know.

Assessment of hyperacusis (Paper I)

Hyperacusis Questionnaire

The mean value for the HQ in the present group was 30.0 (SD = 5.9), with 21 (34 %) of the 62 patients scoring below 28, which has previously been suggested to identify patients with strong auditory hypersensitivity [72].

The Attentional and Social subscales correlated significantly with the total HQ ($r = 0.70, p < 0.001$ and $r = 0.81, p < 0.001$). There was also a significant correlation between the Attentional and Social subscales ($r = 0.40, p$
< 0.01). No significant correlations were found between the Emotional subscale and the total HQ, the Attentional subscale or the Social subscale.

The Hospital Anxiety and Depression scale
The mean score for the anxiety scale was 7.7 (SD = 4.2), and for the depression scale it was 6.3 (SD = 3.8). The cut-off for the scales is 8 [15]. Out of the 62 patients, 31 scored 8 or above on the anxiety scale, and on the depression scale 21 patients scored 8 or above.

Correlations between diagnostic tools
As reported in Paper I, the HQ correlated negatively with all LDL values except for 4000 Hz in the right ear, but not with any value in the left ear. There were significant negative correlations between LDLs and the HADS anxiety scale, but not with the HADS depression scale.

No significant correlations were found between the total score for the HQ and either of the HADS scales. The HQ emotional subscale correlated significantly with the HADS anxiety scale ($r = 0.30$, $p < 0.05$).

No clinically useful significant correlations were found between the HQ, or the subscales, and any item from the clinical interview, such as any of the reported reactions to sounds, sudden onset of hyperacusis or co-morbid tinnitus. Correlations were also explored between LDLs and items from the clinical interview, but no clinically useful significant correlations could be found.

Psychiatric comorbidity and personality traits (Paper II)

Psychiatric disorders
In all, 56 % of the patients suffered from a comorbid psychiatric disorder. Almost half of all the patients, 47 %, suffered from an anxiety disorder. The most common anxiety disorder in the group was social phobia, followed by agoraphobia. See Figure 2.

Eight patients (13 %) were taking antidepressant medication and four patients (6 %) were taking hypnotics. For complete results, see Paper II.
Personality traits

It was found in Paper II that the present patients reported significantly higher values than the normative sample on the scales Somatic Trait Anxiety ($t(60) = 5.61$, $p < 0.001$), Psychic Trait Anxiety ($t(60) = 2.70$, $p < 0.01$), Stress Susceptibility ($t(60) = 7.42$, $p < 0.001$), Embitterment ($t(60) = 4.28$, $p < 0.001$) and Trait Irritability ($t(60) = 2.79$, $p < 0.01$). They reported significantly lower values on the scales Impulsiveness ($t(60) = -2.28$, $p < 0.05$), Adventure Seeking ($t(60) = -2.66$, $p < 0.01$), Verbal Trait Aggression ($t(60) = -2.22$, $p < 0.05$) and Physical Trait Aggression ($t(60) = -3.19$, $p < 0.001$).

See Figure 3.
Effect of CBT (Paper III)

Demography

The ages for the 60 patients who participated in the treatment part of the project ranged from 18 to 61 years with a mean of 40.2 years (SD = 12.2). There were no statistically significant differences at baseline between the groups either for the variables of sex, age, tinnitus, or for any of the outcome measures. See Table 3.
Table 3. Means and standard deviations for baseline, pre-treatment (for the WL group only), post-treatment and follow-up measures.

<table>
<thead>
<tr>
<th></th>
<th>CBT</th>
<th></th>
<th>WL</th>
<th></th>
<th>Total</th>
<th></th>
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<td></td>
<td>M (SD)</td>
<td>N</td>
<td>M (SD)</td>
<td>N</td>
<td></td>
<td>N</td>
</tr>
<tr>
<td>Age</td>
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<td>30</td>
<td>42.1 (13.1)</td>
<td>30</td>
<td>60</td>
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<td>Married/single</td>
<td>14/16</td>
<td>30</td>
<td>18/12</td>
<td>30</td>
<td>60</td>
<td></td>
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<tr>
<td>Comorbid tinnitus</td>
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<td>30</td>
<td>26</td>
<td>30</td>
<td>60</td>
<td></td>
</tr>
<tr>
<td>Hearing Level Right ear</td>
<td>11.2 (10.8)</td>
<td>30</td>
<td>10.3 (9.0)</td>
<td>30</td>
<td>60</td>
<td></td>
</tr>
<tr>
<td>Hearing Level Left ear</td>
<td>13.3 (11.3)</td>
<td>30</td>
<td>12.0 (8.2)</td>
<td>30</td>
<td>60</td>
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<tr>
<td>LDL Right ear</td>
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<td></td>
<td></td>
<td></td>
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</tr>
<tr>
<td>Baseline</td>
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<td>72.22 (10.79)</td>
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<td>70.17 (12.19)</td>
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<tr>
<td>12-month follow-up</td>
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<td>60</td>
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<tr>
<td>LDL Left ear</td>
<td></td>
<td></td>
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<tr>
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<td>73.47 (10.73)</td>
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<td>72.94 (10.85)</td>
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</tr>
<tr>
<td>Baseline</td>
<td>29.77 (5.49)</td>
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<td>29.83 (6.33)</td>
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<td></td>
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<td>5.86 (3.53)</td>
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<td>6.07 (3.96)</td>
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<td></td>
</tr>
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<td>Baseline</td>
<td>6.63 (3.84)</td>
<td>30</td>
<td>5.80 (3.75)</td>
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<td>60</td>
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<td>Post-treatment</td>
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<td>4.68 (2.97)</td>
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<td>4.63 (3.83)</td>
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<td>54</td>
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<tr>
<td>Baseline</td>
<td>44.30 (8.29)</td>
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<td>42.87 (8.64)</td>
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<tr>
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</tr>
<tr>
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<td>32.89 (9.51)</td>
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<td>12-month follow-up</td>
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<td>31.67 (10.08)</td>
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<td>QOLI</td>
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<td></td>
<td></td>
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<td></td>
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<tr>
<td>Baseline</td>
<td>1.49 (1.60)</td>
<td>30</td>
<td>0.97 (2.04)</td>
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<td>60</td>
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<tr>
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<td>1.30 (2.00)</td>
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<td></td>
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<tr>
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<td>1.64 (1.95)</td>
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<td>56</td>
<td></td>
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<tr>
<td>12-month follow-up</td>
<td>2.36 (1.35)</td>
<td>28</td>
<td>1.61 (1.98)</td>
<td>27</td>
<td>55</td>
<td></td>
</tr>
</tbody>
</table>
Audiological results

The patients had mean hearing thresholds clearly within the range of normal hearing (PTA), of 10.8 dB (SD = 9.9) in the right ear and 12.6 dB (SD = 9.8). There was an expected significant difference in the average hearing thresholds between the right and left ears, with the right ear outperforming the left ($t = 2.76, p < 0.01$).

The mean LDLs ranged from 69.3 (SD = 12.1) to 76.0 (SD = 11.7) on average for the frequencies 250, 500, 1000, 2000, 3000 and 4000 Hz for each ear.

Effect of CBT

**Primary outcome measure**

* Loudness discomfort levels

The ANCOVAs showed a significant between-group effect on the LDL test for both ears (right ear $F(1, 57) = 14.2, p < 0.001$, left ear $F(1, 57) = 11.6, p < 0.001$). Without imputation, values were similar (right ear $F(1,55) = 12.5, p < 0.001$, left ear $F(1,55) = 10.1, p < 0.01$).

Between-group effect sizes (Cohen’s $d$) were moderate, $d = 0.67$ for the right ear and $d = 0.69$ for the left ear. The within-group effect sizes in the CBT group were moderate, $d = 0.56$ for the right ear and $d = 0.51$ for the left ear. See Table 4.

**Secondary outcome measures**

After controlling for differences in pre-test scores, there were significant group effects in favour of the treatment group on all secondary outcome measures except for the HADS anxiety scale. See Paper III for complete figures.

**Follow-up**

When the waiting-list group later received CBT, the treatment outcomes were similar. No significant differences were observed. Follow-up assessment was conducted 12 months after post-treatment measurement, and was completed by 55 patients. Improvements were maintained at the group level for all measures in the CBT group.

At follow-up the differences due to group on the HQ were significantly different in favour of the treatment group ($F (1, 52) = 5.3, p < 0.05$). On the HADS anxiety scale there was a significant improvement in the treatment group from baseline to follow-up, and also in the control group from pre-treatment to follow-up after they also had received CBT.

Furthermore, the degree of sick leave before treatment due to hyperacusis was 42%. At follow-up, 37% were still on sick leave due to hyperacusis.
Table 4. Effect sizes, Cohen’s d. WL-group received CBT after 6 mths on WL and was assessed thereafter.

<table>
<thead>
<tr>
<th></th>
<th>Within-group effect sizes</th>
<th>Between-group effect sizes</th>
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<tr>
<td></td>
<td>CBT</td>
<td>WL</td>
</tr>
<tr>
<td>LDL Right ear</td>
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<tr>
<td>Post-treatment</td>
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<td>12-month follow-up</td>
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<tr>
<td>LDL Left ear</td>
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<td>12-month follow-up</td>
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<td>Post-treatment</td>
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<td>0.01</td>
</tr>
<tr>
<td>12-month follow-up</td>
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<td>0.85</td>
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<td>0.15</td>
</tr>
<tr>
<td>12-month follow-up</td>
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<td>0.25</td>
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<td>HADS Depression</td>
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<tr>
<td>Post-treatment</td>
<td>0.46</td>
<td>0.16</td>
</tr>
<tr>
<td>12-month follow-up</td>
<td>0.59</td>
<td>0.16</td>
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</tbody>
</table>

Additional results

Visual Analogue Scale
The purpose of this measure was to make it possible, in the event of a large drop-out from treatment, to analyse whether the patients left treatment due to improvement or deterioration of their hyperacusis symptoms. There was a significant decrease in VAS ratings over time ($p < 0.05$ between session 1 and 2, and $p < 0.001$ between the subsequent sessions).
Differences between responders and non-responders

In an attempt to explore clinical significance of the LDL results, responders were considered to be individuals who had an LDL increase of at least 6 dB in one ear. Out of all the patients measured post-treatment (n = 56), including the patients who received CBT after the waiting list period, 32 individuals were found to have a positive change of at least 6 dB in one ear. At follow-up (n = 55) 35 patients were considered responders. No significant differences between responders and non-responders according to this estimation, could be found for data on age, sex, any personality trait as measured by the SSP, reaction to sound with fear, duration of hyperacusis, or any of the effect measures. However, there was a trend in the direction of responders also suffering from at least one anxiety disorder \( \chi^2 = 3.39 \) (\( p = 0.07 \)).
The overall aim of the present thesis was to obtain increased knowledge about the clinical condition hyperacusis, which was defined as an unusual intolerance to ordinary environmental sounds [132]. The more precise aims were to assess the effect of CBT as a remedy to decrease the symptoms of hyperacusis, to investigate clinical and psychiatric characteristics of the condition, and to compare tools for assessing hyperacusis. The investigations were performed in consecutive patients with a primary audiological complaint of hyperacusis, referred or self-referred, to the Department of Ear, Nose and Throat diseases at Uppsala University Hospital.

Effect of CBT for hyperacusis

Based on the randomized clinical trial presented in Paper III, it may be concluded that CBT can be helpful in treating patients with hyperacusis. Since there are presently no evidence-based psychological treatments for hyperacusis, this fact may instill hope in patients, their significant others, as well as in clinicians.

The results provide evidence for the hypothesis that sound-related avoidance behaviour in patients with hyperacusis maintains and exacerbates hyperacusis, and that CBT can alleviate suffering by focusing on decreasing this avoidance. The results were stable over 12 months, with effect sizes generally moderate immediately after treatment, and somewhat larger at follow-up, indicating that patients had continued applying the CBT principles after treatment ended.

According to behavioural theory, the mechanisms responsible for the development and maintenance of anxiety and avoidance in anxiety disorders are based on Mowrer’s two-factor theory [98]. This theory was also the base of the sound exposure therapy in CBT for hyperacusis, where avoidance of sounds would maintain the respondently conditioned response of fear or anxiety towards sounds. Although the patients’ compliance with the exposure therapy was not specifically measured, most of the sessions included exposure in vivo. The homework almost always included exposure therapy and sound enrichment, and the therapist and patient went through it carefully during the following session, ensuring that the patients actually did expose themselves to sounds.
Another likely mechanism for achieving change in CBT for hyperacusis is a change in cognitive appraisal of the consequences of exposure to sounds. The aim in the present study was to establish this by means of the psychoeducative elements and by the experience of exposure to sounds. Cognitive reappraisal has been shown to be an important factor in CBT for several anxiety disorders such as social anxiety disorder and panic disorder [39; 59].

In CBT for chronic pain, reductions in pain catastrophizing have been shown to be significantly related to improvements [21].

Sound exposure therapies are sometimes used in audiological practice, mainly to treat tinnitus, such as in tinnitus retraining therapy (TRT) [106; 58], and sometimes also to treat hyperacusis [34]. Possible advantages of CBT could be the focus on natural sounds in the exposure instead of sounds elicited by maskers, the commonly used devices that expose the patient to pink noise. Generalizability is likely to be better as the sounds are actually everyday sounds. Furthermore, in CBT there is no compliance problem with respect to wearing maskers for many hours each day. The chance of finding natural reinforcers is also greater if the patient conducts the exposure in a natural environment. The focus on returning to activities given up due to hyperacusis could also improve compliance in CBT, and of course also have the effect of mood elevation that was shown in this study.

Considering the numerous demanding tasks for the patients in both the evaluation and the different phases of CBT treatment, the drop-out rate in the present study was very low. Two individuals were lost before the treatment phase, and were therefore not included in the RCT analyses. Only four persons left the study prior to follow-up. One mechanism for the very low dropout rate may have been that a basic foundation in CBT is a distinct focus on therapist and patient cooperation with the aim of building an alliance and increasing motivation. This was operationalized in the present project by giving the patient a thorough rationale for the treatment, a reasonable explanation of the development of hyperacusis, and by cooperating in goal-setting and planning homework assignments. The exposure was also performed in a controlled and step-wise fashion, reducing the patients discomfort and increasing the likelihood of continuing treatment. Another tentative explanation might be that the patients’ motivation to adhere to treatment was high since they were aware of the scarcity of evidence-based treatments for their problems. Finally, the fact that treatment was performed in an audiology department with extensive experience of psychological treatment may have contributed to the high level of adherence.

Although there was a statistically significant difference on most measures before and after the treatment, patients were still sensitive to sounds after receiving CBT, and were probably not able to take part in all the activities they would like to. The degree of sick leave due to hyperacusis before treatment was 42 %. After treatment 37 % were still on sick leave due to hyperacusis. Although the aim in the present study was not to have the patients
return to work, it is worth considering that they attended only six sessions of
treatment over two months’ time. In many cases this limited amount of time
made a significant difference, but for some patients it is likely that six ses-
sions were too few. Extending the protocol over time would probably be
useful in some cases.

Clinical characteristics and behaviours in hyperacusis

As previously pointed out, from a CBT perspective the avoidance behaviour
in both hyperacusis and anxiety disorders is the key to maintenance of the
disorder and to treatment [98]. Behavioural avoidance has been investigated
before in hyperacusis [16], as well as in tinnitus [54]. The present patients
did present with high frequencies of behavioural avoidance, as measured by
their avoidance of different activities/environments (89 %) and over-
protecting their ears (82 %).

An important finding in the present investigation was the large number of
patients who were on sick leave from their workplaces due to hyperacusis.
This suggests that suffering from hyperacusis constitutes a problem which
has both economic and social consequences. Long-term sick leave has nega-
tive consequences for the individual in work situations, especially with re-
spect to salary and career advancement, and also in social situations [115; 20].

It was noticed that a large proportion of the patients were teachers, either
in schools or preschools. High noise levels are often seen as problems in
schools [9; 129]. In spite of this, the noise levels in schools are generally
within recommended levels [119]. Nevertheless, personnel in teaching envi-
nronments often consider the noise level a problem, which is likely to be due
to the sudden fluctuations in frequency and level of the noise [119]. Noise
exposure has also been found to result in stress in preschool employees
[118]. Around 30 % of the staff in preschools experience so-called burnout
syndrome, supported by measures of cortisol levels [118].

Several other relevant observations were made in the present sample
Firstly, almost half of the patients reported suffering from problems with
their jaws, possibly temporomandibular joint dysfunction. Such problems
have been shown to coexist with hyperacusis and also with tinnitus [56; 126]. Temporomandibular disorders have been treated with relaxation tech-
niques [104] and CBT [30; 83] with promising results. Secondly, half of the
present patients reported balance problems, which have also been reported in
individuals with tinnitus [127].
Psychiatric comorbidity and personality traits in hyperacusis

In agreement with the hypothesis, a high prevalence of anxiety disorders was found in the present patient group. In fact, almost every second patient could be diagnosed with an anxiety disorder, with social phobia, generalized anxiety disorder and agoraphobia being the most common. This figure is significantly higher than in extensive population-based studies [71; 70; 75; 74]. Furthermore, the MINI instrument which assesses on-going psychiatric morbidity, i.e. the point prevalence, was used in the present study, while the mentioned population-based studies have used instruments which assess symptoms during an extended time frame, commonly one year. It is therefore reasonable to believe that the difference between our patient group and the background population is even larger than these references suggest. The present results are in line with an earlier study of patients with hyperacusis [37], and also similar to a study performed in noise-sensitive women [121], and in accord with what has frequently been observed in clinical work.

Unfortunately, we have no information on the onset of anxiety disorders in relation to the first appearance of hyperacusis. Thus, we can only speculate as to whether the anxiety disorders had their onset before the hyperacusis, simultaneously, or afterwards. Nor do we have information concerning any possible changes in the presentation of anxiety disorders after the CBT. In an earlier study on psychiatric disorders in hyperacusis, it was argued that the existing psychiatric disorders present a risk factor for the development of hyperacusis [37]. Also, in a previous Swedish study on tinnitus, 45 % of the patients suffered from an anxiety disorder and it was concluded that most of them had this disorder before the onset of tinnitus [140].

Our patient cohort displayed higher levels of personality traits related to neuroticism as compared to a control group from the general population. This implies that patients with hyperacusis exhibit a greater than normal tendency to respond with negative emotional responses to threat, frustration and loss [38]. Persons with high levels of neuroticism are shown to be more vulnerable to both mental and physical disorders, e.g. depression, anxiety disorders and back pain [40; 26; 67]. This would reasonably also apply to hyperacusis.

High degrees of neuroticism also increase vulnerability to stress [44; 19; 113]. Stress has been suggested to be of importance in hyperacusis [109], and it has been shown that women with high levels of emotional exhaustion become more sensitive to sound after an acute stress task, than women with low levels [51]. In the present study, however, no standardized measure of stress was included. Nevertheless, perceived stress was reported as a reaction to sounds by a significant number of patients. In addition, the present patients presented with significantly higher scores on the SSP scale of stress susceptibility.
Assessment of hyperacusis

As previously stated, there is no consensus-based standardized diagnostic procedure for hyperacusis. This is quite natural as the most current definition of hyperacusis refers to when a patient reports discomfort for sounds that would be acceptable to most people with normal hearing [132]. This implies that the problems encountered by those affected are of a very subjective character. Individuals who agree with statements like: “Do you have a lower tolerance for noise than other people?” or “Do you consider yourself to be sensitive to everyday sounds?” experience suffering that is sufficient for a diagnosis of hyperacusis [4; 91]. Further assessments do not contribute to a firmer diagnosis, but are rather aimed at quantifying and characterizing the extent of the problems, above all at assessing clinical severity, and following the effect of treatment over time.

In Paper I, different measures were used to assess hyperacusis from this perspective, namely the LDL test, the HQ, and the clinical interview for hyperacusis. In addition, the HADS was used to assess anxiety and depressive symptoms. The results clearly confirmed findings from previous studies [91] suggesting that instruments tested in an attempt to pinpoint the core problem at best correlate moderately, and often not at all. When designing this study the LDL test was chosen as an independent outcome measure in the planned RCT, as it was possible to blind for the assessors. This assessment gives very momentary information on hearing discomfort with respect to standardized sound levels for each tested ear. The LDL test was correlated only moderately with the self assessment of HQ, and only for the right ear (Paper I).

Evaluation of hyperacusis using the HQ is worth a separate analysis. As part of the original construction of the instrument [72] 201 subjects were randomly selected from the general population and tested. The mean score for the self assessment instrument was 15 ± 6.7 out of a possible 42. The authors suggested that those individuals who scored significantly higher than the population mean, with a threshold value of 28.4, seemed to represent those with a strong auditory hypersensitivity. Therefore, a threshold level of 28 was suggested. A later study [91], however, showed that this value did not adequately discriminate those with clinical hyperacusis from those without, as about half of those with hyperacusis had scores below 28. This, together with our observation that about one third of our hyperacusis sample had scores below 28, suggests that the concept of a defined cut-off in HQ is inappropriate. Rather, HQ seems to be a relevant instrument as part of an integrated assessment in treatment programs or in clinical studies.
Ethical and methodological considerations

As there is no existing evidence-based treatment with established effect for hyperacusis, there is clearly a clinical need for attaining such a treatment. Doing so should be based on a solidly built knowledge-base where the effects of different treatment modalities are compared and studied in well-designed clinical trials. A randomized controlled trial (RCT) is the gold standard for such a trial. RCTs are used to test the efficacy and/or effectiveness of various types of interventions within a patient population. Randomization is needed to allow statistical hypothesis testing and to make sure unknown factors and factors that may have possible effect are evenly distributed between groups.

The randomization procedure can be performed in different ways. In the present RCT (Paper III), patients were randomly allocated to CBT or to a waiting list control group by the test leader picking a card labelled “Treatment” or “Control”. This method incurs a risk for allocation bias if the procedure of allocation is not adhered to properly [77]. In retrospect, central randomization with some kind of computerized alternative would have been preferable.

Another crucial point when performing an RCT is “blinding”, i.e. a procedure that prevents the study participant, or the outcome assessor, from knowing which intervention was received. However, blinding is sometimes inappropriate or impossible to perform. Such is the case if one of the randomized groups receives a treatment or other procedure where active participation of the patient is necessary. In the present study blinding of the intervention was not an issue since the patients were allocated to active treatment or a waiting list. However, with respect to outcome assessment we did choose a primary effect measure that allowed blinding, i.e. the LDL test. Thus, the audiologists who performed the LDL assessment were blinded both to group and phase of the study.

A core issue is that all study groups should be handled identically, except with respect to the core intervention under study. The choice of using a waiting list control group can be questioned from this perspective. First, it is possible that a nocebo-effect [69], which in this case could be related to negative assumptions or disappointments perceived by the patient who is in the waiting list group, may worsen his or her symptoms. However, there was no sign of deterioration in the effect measures in the waiting list group in this study. The waiting list design also involves an ethical component. All patients randomized to the control arm were, however, eventually offered CBT treatment, and the results from the CBT in this group were similar to those in the treatment arm once they received treatment. Second, there might have been differences in the extent of unspecific support given to patients who were allocated to active treatment compared to that given to those in the waiting list group.
An issue related to patient selection is that most patients included in the studies were referred to a highly specialized audiology department at a university hospital. This reasonably suggests that, on a group level, the study group experienced a higher degree of suffering than the average individuals with hyperacusis. Furthermore, the participants’ level of education was higher than expected from population data. The project took place in a city where there are two large universities, which may serve as an explanation. This is also in accordance with previous knowledge about highly educated individuals being more likely to seek healthcare [128]. Viewed together, these issues point to an external validity problem, as results obtained in a study generally apply only to persons with characteristics similar to those who have been studied. It must be emphasized, however, that most patients were recruited from a clinical setting, implying that individuals who were obviously suffering from their condition were included.

An ethical aspect is that in addition to the diagnostic procedures, parts of the CBT program contained elements that might have been difficult for the participants to endure, *i.e.* the element of exposure to sounds. In fact, patients with hyperacusis often find audiological testing to be highly uncomfortable, or even painful [12]. To minimize their discomfort in this project, the LDL testing started with low sound volumes and the entire procedure was carried out with caution so as not to exceed volumes that caused severe discomfort.

Clinical implications

In hyperacusis, hearing thresholds are normal, LDLs decreased, and the patient claims to be bothered by everyday sounds. When diagnosing hyperacusis, especially when the patient is referred for CBT treatment, it is advisable for clinicians to use the present clinical interview and the HQ. The HADS could be helpful for CBT treatment planning, but further investigation is needed. It is not common to investigate psychiatric problems in an audiological department, but the HADS could at least screen for anxiety and depressive symptoms. Audiological measuring, that is measuring hearing thresholds and LDL testing, may be necessary to exclude other problems and to evaluate treatment effects.

General advice for any audiology department would be to ensure that all staff members are educated about the adverse effects of over-protecting one’s ears. Medical doctors, nurses, audiologists and other staff members should stress the importance of patients exposing their auditory systems to normal sound levels. It is equally important to encourage patients to try not to alter their lives because of their condition, but to maintain their preferred habits as much as possible. This advice is in accordance with the CBT model.
for hyperacusis, and should be of at least some help and possibly prevent some cases of hyperacusis.

In those cases where an individual is restrained in life by sound intolerance, he or she may be helped by being referred to a psychologist with training in CBT. Where behavioural avoidance of sounds is present, CBT is likely to be of help. In patients who do not avoid sounds, some elements of CBT such as stress reduction and psychoeducative input can still be helpful in aiding the patient to keep away from falling into the trap of avoidance and thereby worsening the problem. A disadvantage of CBT, both in Sweden and internationally, is that not many professionals have adequate training in CBT. A way to overcome this could be to adapt the treatment protocol to be suitable for groups and for internet-administered treatment.

Future research

One single RCT is not enough to prove that CBT is a good treatment option for hyperacusis. First, replications are necessary, and particularly with longer follow-up periods including assessment of possible effects on sick leave and everyday life. Second, other cost-effective modes of delivering CBT are relevant topics. Here, group therapy and internet-based treatments are good options. Extending the protocol to include a focus on returning to work could also be a possibility for future research.

In the present study we did not include patients with significant hearing loss. With an aging population the number of such patients is expected to increase. Such individuals may suffer from both hearing loss and hyperacusis and sometimes also other types of sound sensitivity. In the clinical setting it can be observed that these individuals refrain from wearing their hearing aids due to the aversive experience of suddenly being able to perceive certain sounds. Not infrequently, patients also admit to feeling ashamed of their hearing loss and would rather not let other people know about it. This can be seen as an avoidance behaviour that exacerbates both hyperacusis and probably also some form of social anxiety. A sound hypothesis, and a possible aim for future research, is that this often over-looked group of patients would benefit from CBT.
Conclusions

Hyperacusis is an unexplored condition. The aim of this thesis was to contribute to the knowledge concerning assessment, comorbidity and treatment of hyperacusis. The main conclusions are:

- The Hyperacusis Questionnaire and the Hospital Anxiety and Depression Scale are useful tools in the diagnostic evaluation of hyperacusis, especially when considering patients for treatment with cognitive behavioural therapy.

- Anxiety disorders and personality traits related to neuroticism are over-represented in patients with hyperacusis.

- Cognitive behavioural therapy, with focus on exposure to sound, psychoeducation, applied relaxation and behavioural activation, is a promising treatment option for patients with hyperacusis. Treatment effects are maintained for at least twelve months.
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A doctoral dissertation from the Faculty of Medicine, Uppsala University, is usually a summary of a number of papers. A few copies of the complete dissertation are kept at major Swedish research libraries, while the summary alone is distributed internationally through the series Digital Comprehensive Summaries of Uppsala Dissertations from the Faculty of Medicine.