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Clinical Focus

New Hyperacusis Therapy Combines Psychoeducation, Sound Exposure, and Counseling

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ABSTRACT

Purpose: The aim of this study was to investigate the short- and long-term effects of a new cognitive sound exposure therapy (CSET) in patients with hyperacusis.
Method: A new therapy was developed to reduce hyperacusis using sound exposure combined with breathing and relaxation strategies from both acceptance and commitment therapy and cognitive behavioral therapy. Patients who were referred to the Speech and Hearing Centers located in Hengelo and Zwolle in the Netherlands and aged ≥ 18 years with hyperacusis as main complaint and no or mild hearing loss were included in this study. Patients were seen for CSET between June 2020 and August 2022. The sessions took place biweekly. Sessions ended when exposure reached a level with a maximum of 70–80 dB SPL. Short-term effects between the start and the end of therapy were based on tolerable level of sound exposure (dB SPL), subjective-level hinderance of hyperacusis, and sensitivity to sound using the Hyperacusis Questionnaire (HQ). The long-term effect was based on HQ 6 months after the end of therapy. Linear mixed-effects and regression models were applied to study outcomes over time.
Results: In total, 30 patients, 15 men and 15 women, aged between 24 and 76 years were included in this study. The mean number of sessions during therapy was 6 and ranged between 4 and 8. Results showed an increase of exposure level (mean change was +23.7 dB with an *SD* of 7.9, $p < .001$), a decrease in sensitivity to daily sounds (mean [*SD*] change was −1.6 [2.1], $p < .001$), and a decrease in HQ (mean [*SD*] change was −9.8 [4.9], $p < .001$), between the start and the end of therapy. There was no significant change in HQ after the end of therapy and 6 months later; mean (*SD*) change was 0.2 (4.3), $p = .81$.
Conclusions: The evaluation of CSET indicated a decrease in short- and long-term sensitivity to sound in patients with hyperacusis. Additionally, CSET has shown a positive impact, not only for the sounds used in the therapy sessions but also in transferring benefits to everyday sounds. The results of combining psychoeducation, sound exposure, and counseling are promising and warrant further evaluation.

Although sound is ubiquitous in our society, some people suffer from the constant presence of sounds. How people react differently to sound exposure and how they can adapt to noise have been studied for decades (Weinstein, 1978). However, if even everyday environmental sounds are perceived as being uncomfortably loud or intense, we speak of hyperacusis (Fackrell et al., 2017; Henry et al., 2022; Tyler et al., 2014).

Hyperacusis is common with an estimated prevalence around 9%, but the exact number differs depending on the definition used and varies between countries

Correspondence to Sandrien Thieren: CSET@pento.nl. ***Disclosure:*** *Analysis was performed by Paula van Dommelen from The Netherlands Organization for Applied Scientific Research (TNO). Research from TNO is conducted without any undue influence from commercial or other interests. Pento Speech and Hearing Centers provided support in the form of salaries for Paula van Dommelen but did not have any additional role in performing the analysis. Sandrien Thieren and Michel R. Benard are employed at Pento Speech and Hearing Centers, where they are involved in the treatment of individuals suffering from hyperacusis-related discomfort. The described treatment is administered at Pento Speech and Hearing Centers to those experiencing difficulties due to hyperacusis.*

(Andersson et al., 2002; Paulin et al., 2016). Hyperacusis is associated with high age, female sex, and high education (Paulin et al., 2016). Among other factors, hearing impairment and tinnitus were found comorbid with hyperacusis (Nelson & Chen, 2004). Khalfa et al. (2002) developed and normalized a Hyperacusis Questionnaire (HQ), which is frequently used in the Netherlands to diagnose hyperacusis.

The clinical challenge is that individuals with hyperacusis try to avoid sounds (Paulin et al., 2016). However, avoiding sounds to reduce hinderance of hyperacusis can have the opposite effect in specific circumstances. Typically, self-therapy of hyperacusis results in using hearing protection to limit exposure to environments with possible sound levels, which are considered too loud. The effect of noise reduction for extensive periods is twofold. It can possibly lead to neural attenuation for high-level sounds (Formby et al., 2003), and it can reduce low-level sounds, thus limiting healthy sound exposure. Hearing protection can potentially increase the hinderance of hyperacusis and, furthermore, it can induce temporary increased spontaneous neural activity (Schaette et al., 2012).

Since it is known that the perception of loudness can adapt, research focuses on finding the mechanisms involved in hyperacusis. Understanding these underlying mechanisms may lead to a therapy for hyperacusis (Auerbach et al., 2014). Although it has been shown that peripheral abnormalities can lead to the development of hyperacusis (Fournier et al., 2022), hyperacusis is considered as either increased sound-evoked neural activity (Wong et al., 2020) or increased central gain in the auditory pathway (Fackrell et al., 2017). Hyperacusis is associated with an increased sound-evoked activity in subcortical and cortical auditory regions. Central gain cannot explain hyperacusis in all cases (Assi et al., 2018). For example, Assi et al. (2018) investigated hyperacusis in athletes with sport-related concussions, suggesting that a type of hyperacusis, with normal peripheral hearing and absence of tinnitus, can be linked to concussion or mild traumatic brain injury. Hyperacusis can exert a significant influence on one's daily functioning. Some people grappling with hyperacusis regularly employ ear protection or opt out of social gatherings, potentially leading to social isolation.

A limitation of designing an effective therapy for hyperacusis is the finite knowledge of the underlying mechanism of hyperacusis (Wong et al., 2020). Although numerous studies were performed to find a therapy for hyperacusis (Attri & Nagarkar, 2010; Hawley et al., 2008; Jastreboff & Jastreboff, 2014; Jüris et al., 2014; Miani et al., 2001; Noreña & Chery-Croze, 2007; Silverstein et al., 2016; Valente et al., 2000), there is no universally accepted therapy (Assi et al., 2018; Fackrell et al., 2017; Jüris et al., 2014). Therapies to reduce hyperacusis may consist of cognitive behavioral therapy (CBT), tinnitus retraining therapy (TRT), counseling, hearing devices, pharmacological therapy, and surgery (Attri & Nagarkar, 2010; Dauman & Bouscau-Faure, 2005; Fackrell et al., 2019; Jüris et al., 2014; Miani et al., 2001; Noreña & Chery-Croze, 2007; Silverstein et al., 2016; Valente et al., 2000).

To date, most management strategies were assessed in patients who reported hyperacusis as either a secondary concern of as part of a set of symptoms. Research should prioritize assessing interventions in individuals for whom hyperacusis is the primary complaint (Fackrell et al., 2017). Finding a cure, however, is still eagerly anticipated in clinics (Fackrell et al., 2019). In the current study, we focused on reducing hyperacusis with a newly developed cognitive sound exposure therapy (CSET). The CSET is based on multiple separate therapies, to our best knowledge not applied as combined therapy before: exposure therapy (Jüris et al., 2014), psychoeducation on the auditory system and hyperacusis (Hawley et al., 2008; Jastreboff & Jastreboff, 2014), and clinical aspects from the CBT and acceptance and commitment therapy (ACT; Gloster et al., 2020; Jüris et al., 2014).

The aim of this study is to examine whether combining established therapies, namely, psychoeducation, sound exposure, and counseling, can be utilized as a treatment for people with hyperacusis, which would enable them to avoid silence and ear protection in situations when there is no risk of hearing damage and facilitate their participation in daily life. We studied the short- and long-term effects of CSET on levels of sound exposure, subjective-level hinderance of hyperacusis, and sensitivity to sound in patients with hyperacusis. Secondary outcomes were the association between sensitivity to sound and the duration of the complaints, the number of sessions, gender, and tinnitus.

Method

All patients of the CSET were initially referred to the Speech and Hearing Centers located in Hengelo and Zwolle in the Netherlands by general practitioners and ear, nose, and throat specialists for specialized audiological care. Patients were seen for CSET between June 2020 and August 2022.

Developing CSET

Before CSET was developed, patients with hypersensitivity to sounds received audiological examinations and, if necessary, received hearing rehabilitation. Subsequently, social work guidance was offered, providing tips and advice on coping with daily life challenges. For patients experiencing both hearing loss and hypersensitivity, the seamless use of hearing aids is often hindered by the amplification of bothersome sounds. If the hearing aids are not worn consistently, additional effort is required for effective communication, leading to reduced energy levels by the end of the day and exacerbating hypersensitivity concerns. The provided counseling to break this detrimental cycle has proven insufficient in some cases. Pento Speech and Hearing Centers has innovated a treatment specifically designed to alleviate hypersensitivity, for both patients with and without hearing loss. This article will specifically address patients with hypersensitivity and normal hearing. CSET consists of three main components, namely, psychoeducation during an information session, an intake by a clinical audiologist, and on average six therapy sessions facilitated by a social worker (SW).

Patients

Inclusion criteria of the patients were intolerance to sounds as the primary complaint and normal hearing thresholds or at most mild hearing loss in both ears (World Health Organization, 2021) defined as hearing threshold less than 35 dB HL averaged over 500, 1000, 2000, and 4000 Hz. Patients younger than 18 years of age or with tinnitus as the main complaint were excluded. Furthermore, patients with severe autism, untreated insomnia, (acquired) brain injury or damage, intellectual disabilities, or loss of memory and patients with ongoing psychological therapy were excluded. Figure 1 shows the patient journey from entry to 6 months after the end of therapy.

Entry

Clinical questionnaires were sent to the patient after receiving the referral to the Speech and Hearing Centers. The questionnaires included open and closed questions about their medical history, hearing situations, and hyperacusis complaints with the HQ and complementary open questions about auditory sensitivity. After completing the questionnaires, the patient had an appointment with a clinical audiologist. The HQ is a validated questionnaire to characterize sensitivity to sound (Khalfa et al., 2002). This questionnaire contains 14 questions and uses a 4-point rating scale, ranging from *no*, *yes a little*, and *yes quite a lot* to *yes a lot*. Patients filled out the same questionnaire at four different moments in time: at the entry, intake session (therapy start), after the last therapy session, and 6 months after the last therapy session; see Figure 1. An HQ score exceeding 28 points indicates hyperacusis. If the anamnesis reveals that the symptoms of hyperacusis were clearly present and the main complaint, despite an HQ score of less than 28, it was still assessed whether the patient is eligible for participation in the therapy. In case the patient had additional tinnitus complaints, the patient received the Tinnitus Functional Index (TFI) questionnaire as well (Meikle et al., 2012). Of the 30 patients, 14 patients had no tinnitus or tinnitus was not a prominent issue. The other 16 patients received the TFI. The mean score on the TFI is 51.20 (*SD* = 19.37), which is categorized as "moderate." The included patients who reported both hyperacusis and tinnitus identified hyperacusis as the primary concern. In patients showing signs of potential misophonia complaints, we utilized the Misophonia Screening List (van Loon et al., 2019). Additionally, we conducted a thorough anamnesis to pinpoint the specific sounds that the patient finds bothersome.

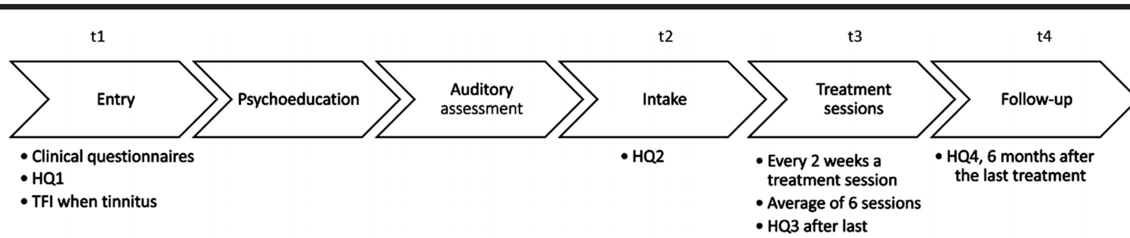
Psychoeducation

After entry, patients started with general psychoeducation about hyperacusis. Psychoeducation took place during a group session where they received information from the clinical audiologist and SW for 1.5 hr. If patients were not eligible for group therapy, for example, due to high sensitivity, they were given an individual appointment. The psychoeducation focused on the functioning of the hearing organ in general, the auditory pathway, and counseling and education on mechanisms involved in hyperacusis. The patients received advice on implementing sound enrichment, avoiding silence, and understanding the negative consequences of excessive use of hearing protection.

Auditory Assessment

After psychoeducation, hearing tests were performed. Each patient completed pure-tone audiometry

Figure 1. Infographic of the cognitive sound exposure therapy. HQ = Hyperacusis Questionnaire; TFI = Tinnitus Functional Index.



and speech audiometry testing, complemented with tympanometry testing if auditory assessment indicated a conductive component. Tests were performed according to current clinical standards (American Speech-Language-Hearing Association, 1988, 2005) and were taken in a soundproof booth designed for audiometric testing. The ear-specific pure-tone average (PTA) for each subject was calculated by averaging hearing thresholds at 500, 1000, 2000, and 4000 Hz obtained with pure-tone audiometry (World Health Organization, 2021). Speech stimuli were meaningful Dutch consonant–vowel–consonant words, to evaluate speech comprehension without background noise (Bosman & Smoorenburg, 1995). Patients with a hearing loss of PTA equal or above 35 dB HL in one or both ears were excluded from this study. After the auditory assessment, patients had an appointment with the clinical audiologist to determine the follow-up plan. Patients who met the criteria were invited into the study.

Intake

When the patient met the inclusion criteria to enroll CSET, the patient received an intake with a clinical audiologist. At this intake, the patient filled out the HQ2. The intake included psychoeducation and the selection of the five most disturbing sounds from the BBC Original Sound Effects Library (BBC, 1991), a database with daily sounds. In addition, the aspects from the psychoeducation were repeated and customized to the individual patient's needs.

To select the five sounds for the therapy from a database with 65 sounds, the patient was positioned 1 m from the loudspeaker (Yamaha MSP5). The sounds were played at a just-audible level using a clinical audiometer (Madsen Astera, Interacoustics AD528, or Kamplex Diagnostic Audiometer AD 27). The just-audible level was the sound level at which the patient could understand 50% of speech. The patients were observed closely, with special attention paid to bodily reactions such as a frown, a change in facial expression, a change in breathing, a cramped posture, or movement with hands or legs. The clinical audiologist increased the sound volume of the audiometer until just acceptable level for the patient. The audiologist instructed the patient to classify each presented sound as "no discomfort," "little discomfort," or "much discomfort." Subsequently, the audiologist documented both the presented level and the degree of discomfort for each sound, while any observable physical reactions were also noted. At the end of the intake, the patient selected the five most disturbing sounds. These five sounds were played each session of the exposure therapy CSET.

We mimicked the sounds that the patients found most disturbing as closely as possible from the database with daily sounds (BBC, 1991). Some examples of the used sounds were alarm, barking dog, crying baby, clattering of cutlery, running engine of a car, vacuum cleaner, extractor hood, bicycle bell, cymbal, and bass guitar. The duration of the each sample varied between 6 and 30 s and were played for the entire duration during the therapy sessions. If this database did not contain the most disturbing sounds, they were either recorded or edited at the clinic. Since the various sounds differ widely, from an impulsive sound like a firecracker bang to a slowly increasing in volume sound like a whistling tea kettle, we used a MATLAB script to adapt the stimuli such that the peak-to-peak levels of all sound samples were of equal intensity. We made the considered choice to set the normalization to peak-to-peak levels. By this method of sound normalization, we controlled for the maximum intensities to avoid the patients exposure to unexpectedly loud sounds.

Therapy Sessions

CSET was given by a clinical audiologist and an SW experienced in CBT, tinnitus, and hyperacusis. The therapy sessions comprised exposure to sound, incorporating clinical aspects from CBT and ACT, which encompassed emotion regulation, breathing, and relaxation strategies. During the initial therapy session with the SW, significant emphasis was placed on the process of getting acquainted, counseling, and the identification of potential other stressors that needed to be considered during the course of the therapy.

Subsequent therapy sessions with the SW took place every 2 weeks, with each session lasting approximately 1 hr. On average, six sessions were conducted, ranging from four to eight sessions. During each therapy session, the five selected sounds were played for the entire duration, with the sound duration varying between 6 and 30 s. In close collaboration with the patient, the SW manually adjusted the volume to attain a healthy maximum, attentively monitoring the patient's physical and emotional responses throughout the process. The sound level was gradually increased on the audiometer until a range of 70–80 dB SPL was achieved over the sessions. A safe listening level of 70–80 dB SPL was selected to ensure that there is no risk of causing harm to the hearing. An integral aspect of the therapy involved assigning homework to most patients, typically after the third therapy session. The homework entailed practicing with the sounds or engaging in activities at home that involve incorporating moments of recovery or relaxation. The assessment of subjective hinderance in patients was conducted during the sessions. Subjective hinderance encompasses the individual's personal perception and experience of discomfort, annoyance, or stress in response to sounds. This measurement involves evaluating the emotional and psychological impact that sounds may exert on an individual. Each following session started with an evaluation of the former session, including at least the experienced fatigue, changes in focus on sounds, and the recognition of the patient's reactions to sound. We used the self-rated number between 0 and 10 that the patient gave to their subjective hinderance.

At the last session, the SW played sounds from the database randomly at moderate sound level and repeated the five selected sounds multiple times. The SW played the five selected sounds also at the starting level and the final level to demonstrate the improvement resulting from the therapy. The patients filled out the HQ3 at the last therapy session to measure the effectiveness of the therapy.

Follow-Up

SW evaluated the patient's experiences of CSET by phone 4 months after the last session. The patient received the HQ4 to evaluate the long-term effects of CSET 6 months after the last therapy session.

Statistical Analysis

To investigate the association between exposure and subjective hinderance at the start, we applied linear regression analysis with subjective hinderance as dependent variable and start exposure as independent variable. To investigate the change in exposure or subjective hinderance between the start and the end of therapy, paired *t* tests were performed.

To investigate the change in HQ outcome between the time points (one model for each consecutive time point), we applied linear mixed-effects models with HQ as dependent variable and time as independent variable. To study if the change in HQ between intake and after the last therapy session depended on the duration of complaints (categorized into < 2 vs. ≥ 2 years), gender, or the number of sessions, we added the main effect of each of these factors and an interaction term of time by factor in the models (one factor per model). Multilevel models were used to encompass all available data over time. The analysis was performed in R Version 4.2.2. *p* values < .05 (two-sided) were considered statistically significant.

Ethics

This study was approved by the Medical Ethics Committee of the Isala Hospital, Zwolle, the Netherlands (Reference 200107). The therapy was explained to the patients. All patients included in this study had written informed consent.

Results

Of the 50 patients referred to the Speech and Hearing Centers for hyperacusis complaints, 11 patients did not meet the criteria. One patient was underage, two patients had acquired brain injury, and eight patients had hearing loss and/or were more affected by tinnitus than hyperacusis. Of the first 50 patients who participated in CSET, 30 patients met the inclusion criteria and agreed to participate in this study. Of these patients, 15 were women and 15 men, aged between 24 and 76 years old, and their native language was Dutch; see Table 1. Fourteen patients experienced hyperacusis, while 16 patients reported hyperacusis as their primary complaint alongside tinnitus. After the intake, there were no patients wearing hearing protection at times when there was no risk of hearing damage. The number of sessions varied from four to eight. The duration of the hyperacusis complaints at therapy start varied from 6 months to 20 years.

The Maximal Acceptable Sound Levels Before and After Therapy

Figure 2 shows the individual preferred maximum start sound level of the sounds presented to the patients before and after therapy. Each dot represents the average of the maximum sound level at the start (t2) and after (t3) therapy of the five sounds that were used in CSET for each patient. The black dots represent patients with

Table 1. Characteristics of the study population (*N* = 30).

Variable	*n* (%) or *M* (*SD*)
Gender	
Male	15 (50%)
Female	15 (50%)
Hyperacusis as the primary complaint	14 (46.7%)
Hyperacusis as the primary complaint with tinnitus	16 (53.3%)
Age (in years)	48.3 (14.5)
Duration of complaints (in years)	4.4 (4.5)
Number of sessions	6.0 (1.2)
Exposure (dB SPL) at the start of the therapy	53.6 (8.9)
Exposure (dB SPL) at the end of the therapy	77.4 (2.7)
Subjective hinderance at the start of the therapy	5.5 (1.5)
Subjective hinderance at the end of the therapy	3.9 (2.1)
HQ at entry (t1; *N* = 26)	27.3 (7.6)
HQ at the start of the therapy (t2; *N* = 30)	28.5 (6.5)
HQ at the end of the therapy (t3; *N* = 28)	18.9 (7.2)
HQ 6 months after the therapy (t4; *N* = 29)	18.8 (8.0)

Note. HQ = Hyperacusis Questionnaire.

Figure 2. The average of the maximal acceptable sound levels of the five selected sounds for each patient before (t2) and after therapy (t3). The black dots represent patients with hyperacusis, while the red dots denote patients with hyperacusis as the primary complaint along with tinnitus.

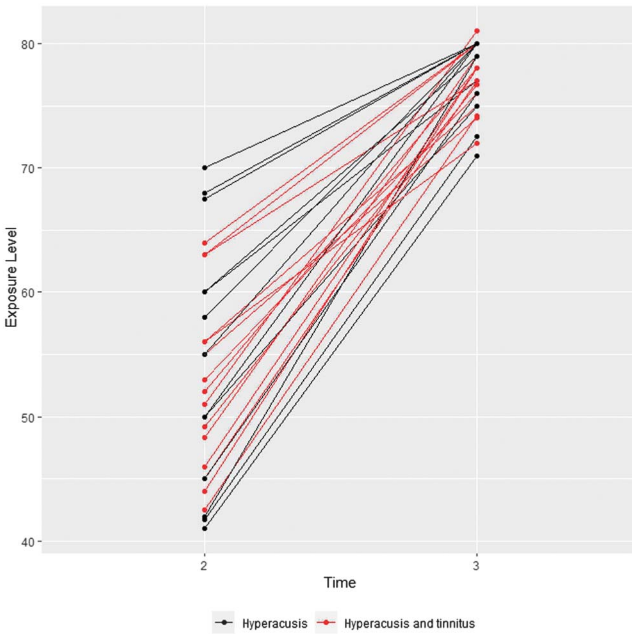
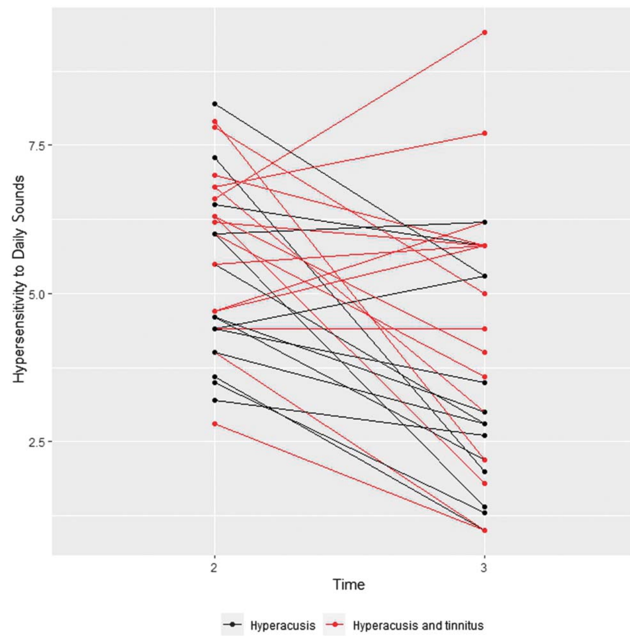


Figure 3. Self-rated number between 0 and 10 between the start (t2) and the end of therapy (t3). The black dots represent patients with hyperacusis, while the red dots denote patients with hyperacusis as the primary complaint along with tinnitus.



hyperacusis, while the red dots represent patients with hyperacusis as the primary complaint along with tinnitus. All 30 patients showed an increase in accepted sound after therapy. Before therapy, the mean start sound level was 54 dB SPL, while this was 77 dB SPL at the last session. The mean change was +23.7 (SD = 7.9, p < .001). Results showed an increase of accepted sound level such that the played sounds reached the maximum level of 71–81 dB SPL. When analyzing the group with hyperacusis (without tinnitus), the mean change was +22.4 (SD = 8.8, p < .001). For the group with hyperacusis and tinnitus, the mean change was +24.9 (SD = 7.1, p < .001). Inclusion of tinnitus as an interaction with the premeasurement in the multilevel model did not yield significance (p = .38).

Sensitivity to Daily Sounds

Figure 3 shows the individual self-rated number between 0 and 10 that patients gave to their subjective hinderance. The black dots indicate the patients with hyperacusis, while the red dots denote patients with hyperacusis as the primary complaint along with tinnitus. The mean decrease in sensitivity to daily sounds was −1.6 (SD = 2.1, p < .001) between the start and the end of therapy. In total, seven patients rated their hyperacusis higher at t3 than at t2. For the group with hyperacusis (without tinnitus), the mean change was −1.9 (SD = 1.7, p = .001). For the group with hyperacusis and tinnitus, the mean change was −1.3 (SD = 2.4, p = .04). The interaction involving tinnitus was not found to be significant (p = .46).

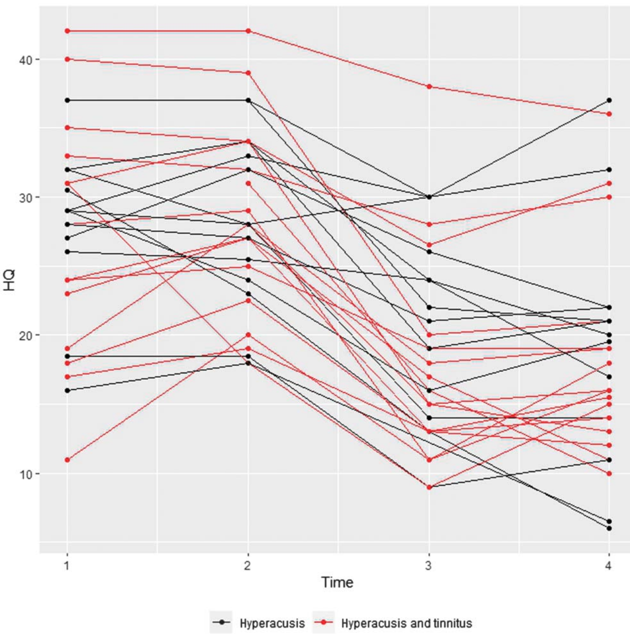
A significant negative association was found between exposure (dB) and subjective hinderance at the start, β (95% CI = −0.10 [−0.15, −0.05], p < .001). If the sound exposure at the start was 10 dB higher, subjective hinderance at the start was on average 1.0 point lower. A significant negative association between start exposure (dB) and HQ scores at therapy start, β (95% CI = −0.43 [−0.66, −0.20], p < .001), was found. If the exposure was 10 dB higher at the start, HQ at therapy start was on average 4.3 points lower.

HQ

Figure 4 shows the individual results of the HQ at entry (t1), intake (t2), after the therapy sessions (t3), and 6 months after t3 (t4). The black dots represent the patients with hyperacusis, and the red dots represent the patients with hyperacusis as the main complaint along with tinnitus. Twenty-six patients filled out the HQ at entry; 30 patients, at intake; 28 patients, after the therapy; and 29 patients, 6 months after the therapy.

There was no significant change in HQ between entry (t1) and intake (t2; β, 95% CI = 0.69 [−1.11, −2.49], p = .44). This indicates that, between entry and intake,

Figure 4. The results of the Hyperacusis Questionnaire at entry (t1), intake (t2), after the therapy sessions (t3), and 6 months after t3 (t4). The black dots represent patients with hyperacusis, while the red dots denote patients with hyperacusis as the primary complaint along with tinnitus. HQ = Hyperacusis Questionnaire.



there was no significant change in HQ in the absence of any therapy except psychoeducation. We observed a significant mean decrease in HQ between intake (t2) and after the therapy sessions (t3; β, 95% CI = −9.83 [−11.6, −8.02], $p < .001$), demonstrating that CSET reduced hyperacusis. The effect of CSET was stable between the last therapy session (t3) and 6 months later (t4), because we found no significant change in HQ (95% CI = 0.19 [−1.38, 1.75], $p = .81$).

For the group with hyperacusis (without tinnitus), we observed a significant mean decrease in HQ between intake (t2) and after the therapy sessions (t3; β, 95% CI = −8.5 [−11.2, −5.9], $p < .001$). For the group patients with hyperacusis as the main complaint and tinnitus, we observed a significant mean decrease in HQ between intake (t2) and after the therapy sessions (t3; β, 95% CI = −10.8 [−13.4, −8.2], $p < .001$). The interaction involving tinnitus was not found to be significant ($p = .21$).

The duration of complaints did not have a significant effect on the change in HQ between intake (t2) and after the therapy sessions (t3; β, 95% CI = −0.20 [−0.61, 0.22], $p = .35$). Furthermore, the analysis showed that the number of therapy sessions did not have a significant effect on the change in HQ between intake (t2) and after the therapy sessions (t3; β, 95% CI = 0.52 [−1.01, 2.05], $p = .50$). However, a higher number of sessions resulted in a lower decrease in HQ over time between intake (t2) and 6 months after the last therapy session (t4). Gender did not have a significant effect on the change in HQ between intake (t2) and after the therapy sessions (t3; β, 95% CI = 1.17 [−2.65, 4.99], $p = .54$).

Discussion

We introduced and evaluated a new therapy for hyperacusis, CSET. This exposure therapy was designed on three major pillars, namely, reducing hyperacusis by exposure therapy (Jüris et al., 2014), providing psychoeducation about the auditory system and hyperacusis (Hawley et al., 2008; Jastreboff & Jastreboff, 2014), and incorporating clinical aspects from CBT and ACT (Gloster et al., 2020; Jüris et al., 2014). In this study, 30 patients with hyperacusis were included with no or mild hearing loss in both ears. We observed short- and long-term effects on decrease of sensitivity to sound between the start and the end of therapy. Furthermore, long-term effects between the last therapy session and 6 months after the last therapy session were observed. CSET decreases sensitivity to sound in patients with hyperacusis and yields favorable effects on their daily existence.

Until now, multiple attempts have been made to implement therapies aimed at reducing the distress caused by hyperacusis, including CBT, TRT, counseling, use of hearing devices, pharmacological therapy, and surgery (Attri & Nagarkar, 2010; Dauman & Bouscau-Faure, 2005; Fackrell et al., 2019; Jüris et al., 2014; Miani et al., 2001; Noreña & Chery-Croze, 2007; Silverstein et al., 2016; Valente et al., 2000). Formby et al. (2015) expanded the auditory dynamic range for loudness for persons with sensorineural hearing loss. However, a universally accepted therapy specifically proven to be effective for normal hearing persons with only hyperacusis and without tinnitus has not yet been found. This emphasizes the critical necessity for a high-priority approach to develop a successful therapy for hyperacusis (Assi et al., 2018; Fackrell et al., 2017; Jüris et al., 2014).

Currently, CBT is a promising approach for treating hyperacusis, but this method is not suitable for all individuals experiencing the effects of hyperacusis (Jüris et al., 2014). Nonetheless, our findings indicate that psychoeducation alone is not enough to reduce the distress from hyperacusis. We observed that the HQ scores did not decrease between entry (t1) and intake (t2) after providing psychoeducation urging the need of a more sophisticated hyperacusis therapy.

Jastreboff and Jastreboff (2014) used TRT and sound therapy as therapy of hyperacusis. Counseling included their neurophysiological model containing sound enrichment applied by ear-level sound devices such as a sound generator or combination hearing aid with amplification and a sound generator. The desensitization approach showed an improvement in hyperacusis using TRT and sound therapy (Jastreboff & Jastreboff, 2014). Hawley et al. (2008) investigated noise generators with and without directive counseling. The results showed that the loudness-discomfort level increased by the therapy. The improvements of hyperacusis were greater in patients who used noise generators devices and directive counseling in comparison with patients who used only counseling or only sound generator or counseling with a placebo sound generator.

Besides CBT and TRT, other methods are described in literature to reduce the hyperacusis distress, such as acoustic training with sounds (Miani et al., 2001; Noreña & Chery-Croze, 2007), medication in a case of hyperacusis associated with depression (Attri & Nagarkar, 2010), or medication for unilateral hearing loss combined with hyperacusis (Valente et al., 2000). Silverstein et al. (2016) reported improved noise tolerance after surgery of the round and oval window in patients who suffered from severe hyperacusis and had not benefited from traditional therapy. Besides the various methods to find a cure, an effective therapy for hyperacusis was still eagerly anticipated in clinics (Fackrell et al., 2019).

Noreña and Chery-Croze (2007) showed that an enriched acoustic environment resulted in a decrease in auditory sensitivity. Their enriched acoustic environment represented a sequence of pure tones based on the cutoff frequency of the hearing loss and a weighting factor. They mentioned that several studies showed a reduction in auditory sensitivity by using chronic stimulation with broadband noise. However, stimulation with broadband noise had been proved to be slow and limited in effect (Dauman & Bouscau-Faure, 2005). In contrast to the mentioned studies, we chose not to use pure tones or noise. Instead, we used sounds that closely mimic daily sounds. Patients frequently report specific daily sounds to be unpleasant or annoying. CSET constitutes a synthesis of exposure therapy, psychoeducation, and components from CBT and ACT. Based on our current knowledge, these particular amalgamations had not been made prior to the development of CSET. Therefore, for CSET we utilized a wide range of different daily sounds to establish our database.

Our main finding is that CSET led to a significant increase in tolerated exposure level. Before therapy, the average start sound level was 54 dB SPL and, at the last session, the sound level had increased to an average of 77 dB SPL; see Figure 2. We observed an increase of accepted sound level such that the played sounds reached the healthy maximum level of 70–80 dB SPL in all patients. In the last session, the SW demonstrated the effect of CSET by playing the five selected sounds at the start sound level and the end sound level as empowerment for the patients. The therapy increased the exposure level to such an extent that patients are able to accept daily sounds other than those with which they have been trained. At the last therapy session, the SW also played random sounds from the database. The patient did not know which sound would be played and they could tolerate these randomly chosen daily sounds at a sound level of at least 65 dB. Thus, the therapy not only reduced auditory sensitivity to the selected sounds, but also to other sounds. These results were positive for patients suffering from hyperacusis.

The patients rated the presented sound from no hinderance to absolutely intolerable. We found a significant negative association between the sound exposure level and the subjective hinderance at the start of the therapy. The analyses showed that, if the subjective distress from hyperacusis was on average 1 point higher at the start, the exposure was 10 dB lower at the start. We have to interpret the results carefully because self-rating is subjective and can be influenced by issues in private or work situations and stress or well-being in general, which may have played a role. On the other hand, the self-rated number gives us information on how patients experience the presented sound. Toward the end of the therapy, the level of exposure has increased, and the self-rated score has decreased for the group results. However, some patients reported a higher subjective score even though they could all accept higher sound intensities; see Figure 3. Apparently, this subjective measure is for some patients influenced by other facets than hinderance from sound. The patients report to suffer less, even when the sounds are presented at a higher sound level, suggesting patients to be aware of the higher sound acceptance levels.

Furthermore, we found a significant negative association between the start sound level and the score on HQ at the intake. If the chosen sounds were required to be played at a lower initial volume, patients' HQ scores at intake were higher, indicating greater impact of hyperacusis. Patients with a low-mean HQ score at the intake could, on average, accept a louder start sound level. A lower score on the HQ was therefore an indication that the patient had less hyperacusis complaints and that sound exposure could have started at a higher sound level.

We used the HQ to investigate the sensitivity to sound at four different moments; see Figure 4. Patients filled out the HQ at entry, at the intake session, after the last therapy session, and 6 months after the last therapy session. The patients did only receive psychoeducation between entry and the intake sessions. On one hand, we anticipated that individuals with hyperacusis during the waiting period between therapies might experience less distress from their hyperacusis due to the reassurance of an upcoming therapy and their knowledge from the psychoeducation. On the other hand, since they did not receive any additional information or advice to cope with their hyperacusis, except psychoeducation, one would expect the level of distress from hyperacusis to remain constant since former research showed only providing information to be insufficient (Fackrell et al., 2017). As it turned out, the latter scenario was observed: no significant difference during the waiting period. The level of distress from hyperacusis remained stable between the entry and the intake without showing any significant changes.

The second main finding of this study is the significant decrease in HQ between intake (t2) and after the last therapy session (t3). This indicates that our therapy effectively reduces hyperacusis. See Figure 4; at group level, there was no significant change in the HQ scores between the last therapy session (t3) and after the follow-up 6 months later (t4). Therefore, we conclude that the effect of the therapy was persistent in the long term. However, a few individuals experienced an increase in the HQ after 6 months, suggesting a slight relapse, although not to the level observed prior to therapy. Furthermore, the duration of complaints (between 6 months and 20 years) did not have a significant effect on the change in score on the HQ between intake and at the last therapy session, demonstrating that CSET was effective regardless of the duration of hyperacusis complaints.

The number of sessions did not have a significant effect on the change in HQ scores between intake and at the last session of the therapy. However, results showed that a higher number of sessions resulted in a lower decrease in score on the HQ between the start and 6 months after the last therapy session. CSET has an average of six sessions, varying from four to eight sessions necessary to increase the accepted sounds to a level of 70–80 dB SPL. Patients that needed more sessions had more severe complaints (higher HQ scores) than patients that needed fewer sessions. This indicates that the CSET procedure correctly described more therapy sessions.

It has been reported that hyperacusis is more prevalent in females (Paulin et al., 2016), whereas other studies found no gender differences (Dauman & Bouscau-Faure, 2005). By chance, we had an equal distribution with 15 women and 15 men. Our analysis showed no significant effect of gender on the progress during the therapy. Despite the limited group size, we do not find any indication that gender plays a role in hyperacusis. Furthermore, all the patients in this study presented hyperacusis as their primary complaint. Of these 30 patients, 16 had tinnitus in addition to hyperacusis. The analysis showed that tinnitus played no role in the progress of the treatment.

There were several limitations in this study. First, the absence of a control group was due to the severity and duration of the complaints, as we made an ethical decision to refrain from including one. However, we compared our treatment with regular care, that is, psychoeducation. Between entry and intake, there was no significant change in HQ in the absence of any therapy except psychoeducation. This indicates that psychoeducation alone is insufficient to decrease the distress from hyperacusis. The duration of the complaints at therapy start varied from 6 months up to 20 years. Due to the extended duration of the complaints, we did not anticipate their sudden disappearance without therapeutic intervention.

Second, we did not randomize the patients across different SW, nor did we use a blind test. This is because the SW detects the emotions from the faces and bodies of the patients and provides relaxation tips, which makes blind testing impossible. Finally, we used only the HQ and not any other questionnaire, for example, the Hospital Anxiety and Depression Scale (Zigmond & Snaith, 1983), to evaluate the state of well-being before and after the therapy.

For future studies, it would be intriguing to investigate whether a cause can be identified for the increase in accepted sounds. There is a possibility that cognitive changes occur or that alterations take place in brain structures, leading to this phenomenon. Alternatively, parts of the therapy might result in the nonactivation of the limbic system by sounds that were previously bothersome. Understanding the underlying mechanisms responsible for the shift in sound acceptance could provide valuable insights into the therapy and management of hyperacusis. Studies such as voxel-based morphometry before and after therapy, high-speed videography of the face to detect sound-evoked pupil dilations, or differences in reaction time to sound before and after therapy might be useful (Makani et al., 2022). Moreover, it would be of interest to investigate if CSET can be implemented in hearing aids to facilitate a gradual habituation process to daily environmental sounds. Additionally, it would be valuable to explore the contribution of the three individual factors (psychoeducation, sound exposure, and CBT/ACT) to the therapy. Furthermore, it is recommended to clarify the influence of hearing loss on the therapy result.

Conclusions

CSET decreases short- and long-term sensitivity to sound in patients with hyperacusis. CSET has a positive impact by reducing auditory sensitivity, not only for the sounds used in the therapy sessions but also in transfer to daily sounds. This study indicates that combining psychoeducation, sound exposure, and counseling holds promise for patients with hyperacusis. Further evaluation of CSET is needed to gain more insight into the mechanism of hyperacusis and the contributions of psychoeducation, sound exposure, and counseling.

Data Availability Statement

The data that support the findings of this study are available from the corresponding author, S.T., upon reasonable request.

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