Minimally Invasive Surgery for the Treatment of Hyperacusis

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**Objective:** To evaluate the efficacy of a minimally invasive surgical procedure in patients with severe hyperacusis.

**Study Design:** Prospective, longitudinal design.

**Setting:** Tertiary referral center.

**Patients:** Adult patients with history of severe hyperacusis.

**Intervention:** Using a transcanal approach, the round and oval window was reinforced with temporalis fascia or tragal perichondrium in six subjects (nine ears) and was subdivided into two groups (unilateral or bilateral reinforcement procedure).

**Main Outcome Measures:** Pre- and postoperative noise tolerance was measured using uncomfortable loudness level (ULL) test scores. In addition, a self-report hyperacusis questionnaire (HQ) was used to assess hypersensitivity to sound before and after the intervention.

**Results:** Analysis of the data reveals improved postoperative mean ULL test scores of 14 dB (confidence interval [CI], 70–98 dB) in the unilateral group. For the bilateral group, improved mean scores were 13 dB (CI, 63–88 dB) in the first ear and 8 dB (CI, 71–86 dB) for the second ear. Further, a negative linear trend was observed in the mean subjective scores for the HQ when both groups measures were analyzed together decreasing from a mean score of 32.0 (standard deviation [SD] = 3.32) preoperative to a mean score of 11.5 (SD = 7.42) after surgery. Postoperatively, the patients reported no change in hearing and improved quality of life after the procedure.

**Conclusion:** The results suggest that reinforcement of the round and oval window with temporalis fascia or tragal perichondrium may offer significant benefit for individuals with severe hyperacusis that has not responded to traditional therapy. ULL scores and self-report measures postoperatively demonstrate improved noise tolerance, high patient satisfaction, and enhanced quality of life.

**Key Words:** Hyperacusis—Loudness discomfort level—Oval window reinforcement—Round window reinforcement.

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Hyperacusis Questionnaire

The HQ used in this study is a non-validated English language adaptation of the German self-report measure for hypersensitivity to sound (14). It is composed of 15-items, evaluating three dimensions or subscales: cognitive behavior in relation to hyperacusis; somatic behavior or reaction linked to specific situation; and finally emotional reactions (21). Using a four-point Likert-type scale, respondents answer “no” (0 points), “yes, a little” (1 point), “yes, a lot” (2 points), and “yes, a lot” (3 points) for each item. The total possible score range is 0 to 45, with higher scores indicating higher sound hypersensitivity. The complete questionnaire is displayed in Figure 1.

Hospital Anxiety and Depression Scale

The HADS was used to screen for excessive depression and anxiety in participants. It is composed of a 14-item questionnaire and evidence of validation has been demonstrated for use in the outpatient setting (22). Seven questions specifically assess the severity of both anxiety and depression. The possible score range is from 0 to 21 for each subscale, with a score of 11 or higher for the subscale suggesting presence of the mood disorder (23).

Uncomfortable Loudness Level

The protocol for ULL testing in this study is described as follows: the participant sat in a sound booth facing the tester. First, pure tone thresholds at 250, 500, 1000, 2000, 3000, 4000, and 8000 Hz were obtained. For each tested frequency, a 1 second steady pure tone was presented via insert or headphone in an ascending order starting at 60 decibel (dB) hearing level (HL). The patient reported if the sound was either “ok” or “uncomfortably loud.” When the tone reached the uncomfortably loud level, the step size was decreased to determine the ULL within a 1 dB resolution. To ensure reliability, the ULL was measured twice at each frequency, and an average of the two presentations was reported. The normal reference level for the ULL is traditionally accepted at 100 dB HL, although normal hearing individuals have been found to have ULLs between 86 and 98 dB HL (24,25).

Surgical Technique

A trans-canal round window and oval window reinforcement under general anesthesia was performed similar to an approach for middle ear procedures. For hemostasis, a four-quadrant...
injection in the ear canal and tragus was made with 1% Lidocaine with 1:100,000 Epinephrine. Vertical canal incisions were made at 6 o’clock and 12 o’clock position and a standard tympanomeatal flap was elevated. The middle ear was entered and chorda tympani was preserved in all cases. If additional exposure was needed, the bony posterior external canal was drilled down with a 1.5 mm diamond burr or curetted to allow adequate visualization of the ossicular chain, round window niche, chorda tympani, facial nerve, and hypotympanum (Fig. 2A). When necessary, additional drilling of the bony round window niche was performed with a 1.0 mm diamond burr for exposure of the round window membrane. In the first two cases in this series, temporalis fascia graft was used. However, because of incision discomfort, tragal perichondrium graft was used in remaining cases. Through a separate 1 cm incision, a small piece of the perichondrium was taken leaving the tragal cartilage intact. The graft was flattened with the fascia press; 4 and 2 mm round grafts were obtained with a biopsy punch. The mucosa of the round window niche and the stapes footplate was scraped with a micro pick to facilitate tissue

FIG. 1. Hyperacusis questionnaire.
welding. The 4 mm graft was used for the round window and the 2 mm graft was placed between the stapes crura on the stapes footplate (Fig. 2B). A small piece of Gelfoam was placed over the round window graft to hold it in place against the round window. The tympanomeatal flap was re-approximated along the posterior canal wall and the canal was filled with mupirocin ointment. Fast absorbing suture was used to close tragal incision.

Statistical Analyses
For analyses, the demographic variables were expressed as means, standard deviation (SD), and the range. Because of the small sample size, parametric analyses were not used. Mean scores with 95% confidence intervals (CI) were calculated for the outcome variables at each interval visit and plotted to examine the trends of scores over time. Microsoft Excel (Microsoft Corporation, Redmond WA, USA), (2010) was used for all statistical analyses.

RESULTS
The sample was formed of six subjects: four women and two men whose mean age was 67 (SD = 9) years, ranging from 55 to 77 years. The sample was further subdivided into two groups depending on whether they underwent unilateral or bilateral reinforcement procedures. The audiometric measures for all participants are displayed in Table 1.

| Subject | Pre-op PTA (dB) | 1 Month Post-op PTA (dB) | 6 Months Post-op PTA (dB) |
|---------|----------------|--------------------------|--------------------------|
| 1       | 5              | 9                        | 10                       |
| 2       | 86             | 86                       | 86                       |
| 3       | 50             | 60                       | 55                       |
| 4A      | 26             | 29                       | 29                       |
| 4B      | 33             | 38                       | 38                       |
| 5A      | 5              | 8                        | 8                        |
| 5B      | 6              | 13                       | 13                       |
| 6A      | 23             | 33                       | 30                       |
| 6B      | 30             | 33                       | 36                       |

PTA indicates pure tone average; Post-op, postoperative; Pre-op, preoperative.

Unilateral Group
This group included three participants: one woman and two men. The mean duration of their hyperacusis was 30 (SD = 17) months, with a range between 24 and 36 months. Two had unknown causes for their hyperacusis and the third developed hyperacusis radiation for an acoustic neuroma radiation. Temporalis fascia was used for the round window reinforcement in two participants, while tragal perichondrium was used for one participant. After the treatment, all the participants reported subjective improvement as evidenced by an improved mean score of 28 (SD = 14) on the HQ (Fig. 3A). The mean ULL value at baseline was 77 dB (CI, ±12 dB) and at 1 month and 6 months postoperative was 94 dB (CI, ±10 dB) and 91 dB (CI, ±18 dB), respectively (Fig. 3B). These results indicate an improved postoperative mean ULL score of 14 dB in this group. The patients reported no change in hearing and improved quality of life after the procedure.

Bilateral Group
In this group, there were three women subjects, totaling six operated ears. In this sample, the mean hyperacusis duration ranged between 18 and 61 months, with a mean of 38 (SD = 17) months. All had unknown causes for their hyperacusis. Four operated ears had temporalis fascia for the round window reinforcement while two ears were reinforced with tragal perichondrium. Analysis of the ULL measures revealed a mean ULL of 69 dB (CI, ±5 dB) at baseline for the first ear. After 1 month, the mean ULL values were 81 dB (CI, ±9 dB) and 82 dB (CI, ±15 dB) at 6 months postoperatively. Mean ULL values were 75 dB (CI, ±2 dB) pre-op for the second ear and at 1 and 6 months after the procedure were 85 dB (CI, ±5 dB) and 83 dB (CI, ±11 dB) respectively (Fig. 4, A and B). After the treatment, two of the three participants reported subjective improvement in their sound hypersensitivity. The mean HQ score was 29.6 (SD = 6) preoperatively and 16.8 6 months after surgery (Fig. 5). One patient underwent revision surgery on the first operated ear to place more fascia because of her lack of improvement on ULL testing. There was minimal improvement noted in
both the ULL testing and HQ score after the revision for this participant. However, the participant stated their noise tolerance improved in social situations. Similar to the subjects in the unilateral group, none had subjective change in their hearing as a result of the procedure and there were no surgical complications. Furthermore, the participants stated their overall quality of life was improved after their procedure(s).

**DISCUSSION**

In 2014, Silverstein et al. (20) first reported marked improvement in sound intolerance in two patients after round and oval window reinforcement with temporalis fascia. The present results of this exploratory study suggest that round and oval window reinforcement may offer relief for individuals afflicted with hyperacusis. There was improvement in the postoperative ULL’s in both the unilateral and bilateral groups, suggesting improved sound tolerance. Most patients noted their greatest improvement after the first ear surgery. Further, subjective improvement was observed after the second ear surgery despite minimal improvement in psychoacoustic measures. While the second ear surgery does not show a significant increase in ULL, the participants’ subjective statements substantiate the benefit of second ear surgery. Similar phenomena where greatest benefit occurs after the first ear surgery has been observed in subjects who underwent bilateral stapedotomies (26). While we were not able to perform statistical testing because of our small sample size, a positive trend was noted with improvement of the ULLs when analyzing first ear and second ear surgeries.

Hyperacusis has been generally accepted to be related to an increase in auditory responsiveness arising from auditory neuronal degeneration. Previously published trials have used a variety of measures and protocols for the assessment of hyperacusis with variable results (16,27). Moreover, there is significant variability in the interpretation of the test data recommended for the
This study has several limitations. Because of the small sample size of our cohort, sample variability was increased and unfortunately we lacked statistical power. The use of a non-validated self-report questionnaire limits interpretation of the participants’ HQ scores in regards to sound hypersensitivity. In future studies, we may consider the Multiple-Activity Scale for Hyperacusis (MASH) as a tool to measure an individual’s level of annoyance in relation to hypersensitivity to sound (33) as well as the tinnitus handicap inventory to assess tinnitus improvement over time after the procedure. Performing the second ear surgery at 1 month made the ULL data from the un-operated ear more difficult to interpret and we were unable to control this variable when collecting data for the initial ear that was treated. In future studies, we plan to wait a full 6 months before performing the procedure on the second ear to allow for full data collection on each ear, thus enhancing the understanding of the potential benefit of the unilateral versus bilateral procedure. Another potential limitation is the possibility of a placebo effect in the treated subjects. When present, a placebo effect threatens a study’s internal validity. In the present study, the outcome measures used to assess the efficacy of the round and oval window reinforcement procedure were subjective. Therefore, response bias, the notion that a participant offered positive responses to the questionnaires just because they received an intervention or because they wanted to please the researcher must be considered. The use of a control group with no intervention, and the addition of objective measures in future studies may be helpful to determine the true effect of this novel minimally invasive procedure.

Our results offer supportive evidence that when the ULL is below 90 dB, minimally invasive surgery using reinforcement of the round and oval window with either temporalis fascia or tragal perichondrium may reduce noise sensitivity in individuals with intractable hyperacusis. The participants of this exploratory study reported diminished sound intolerance and improved quality of life. In patients with bilateral hyperacusis, there appears to be a greater improvement with noise tolerance after the first surgery. The second ear surgery added minimal benefit as evidenced by the ULL results. However, the participants stated they experienced decreased sound sensitivity after the second procedure. Further investigation of this innovative technique is needed.

**REFERENCES**

1. Baguley D, Anderson G. Hyperacusis: Mechanism, diagnosis, and therapies. Plural; 2007.
2. Blasing L, Kroener-Herwig B. Self-reported and behavioral sound avoidance in tinnitus and hyperacusis subjects, and association with anxiety ratings. *Int J Audiol* 2012;51:511–7.
3. Fackrell K, Fearnley C, Hoare DJ, Sereda M. Hyperacusis questionnaire as a tool for measuring hypersensitivity to sound in a tinnitus research population. *Biomed Res Int* 2015;2015:1–12.
4. Hannula S, Bloigu R, Majamaa K, Sorri M, Mäki-Torkko E. Self-reported hearing problems among older adults: prevalence and comparison to measured hearing impairment. *J Am Acad Audiol* 2011;22:550–9.

Otolaryngology, Vol. 37, No. 10, 2016
5. Andersson G, Lindvall N, Hursti T, Carlbring P. Hypersensitivity to sound (hyperacusis): a prevalence study conducted via the internet and post. Int J Audiol 2002;41:545–54.
6. Jastreboff MM, Jastreboff PJ. Decreased sound tolerance and tinnitus retraining therapy (TRT). Aust N Z J Audiol 2002; 24:74–81.
7. Sheldrake J, Diehl P, Schaette R. Audometric characteristics of hyperacusis patients. Front Neurol 2015;6:105.
8. Jastreboff PJ, Jastreboff MM. Tinnitus retraining therapy for patients with tinnitus and decreased sound tolerance. Otolaryngol Clin North Am 2003;36:321–36.
9. Henry JA, Jastreboff MM, Jastreboff PJ, et al. Assessment of patients for treatment with tinnitus retraining therapy. J Am Acad Audiol 2002;13:523–44.
10. Herbert S, Fournier P, Norena A. The auditory sensitivity is increased in tinnitus ears. J Neurosci 2013;33:2356–64.
11. Vernon J. Recruitment. When noise spells pain. The Voice 1991;12:37.
12. Knipper M, Van Dijk P, Nunes I, Rüttiger L, Zimmermann U. Advances in the neurobiology of hearing disorders: recent developments regarding the basis of tinnitus and hyperacusis. Prog Neurobiol 2013;111:17–33.
13. Fukaya T, Nomura Y. Audiological aspects of idiopathic perilymphatic fistula. Acta Otolaryngol Suppl 1988;456:68–73.
14. Nelting M, Rienhoff NK, Hesse G, Lamparter U. The assessment of subjective distress related to hyperacusis with a self-rating questionnaire on hypersensitivity to sound. [In German]. Laryngorhinootologie 2002;81:327–34.
15. Aazh H, McFerran D, Salvi R, Prasher D, Jastreboff M, Jastreboff P. Insights from the first international conference on hyperacusis: causes, evaluation, diagnosis and treatment. Noise Health 2014;16:123–6.
16. Juris L, Andersson G, Larsen HC, Ekselus L. Cognitive behaviour therapy for hyperacusis: a randomized controlled trial. Behav Res Ther 2014;54:30–7.
17. Silverstein H, Kartush JM, Barnes LS, et al. Round window reinforcement for superior semicircular canal dehiscence: a retrospective multi-center case series. Am J Otolaryngol 2014;3:286–93.
18. Nikkar-Esfahani A, Whelan D, Banerjee A. A occlusion of the round window: a novel way to treat hyperacusis symptoms in superior semicircular canal dehiscence syndrome. J Laryngol Otol 2013;127:705–7.
19. Dang PT, Kennedy TA, Gubbels SP. Simultaneous, unilateral plugging of superior and posterior semicircular canal dehiscences to treat debilitating hyperacusis. J Laryngol Otol 2014;128:174–8.
20. Silverstein H, Wu YH, Hagan S. Round and oval window reinforcement for the treatment of hyperacusis. Am J Otolaryngol 2014;26:158–62.
21. Khalfa S, Dubal S, Veuillette E, Perez-Diaz F, Jouvent R, Collet L. Psychometric normalization of a hyperacusis questionnaire. ORL J Otorhinolaryngol Relat Spec 2002;64:436–42.
22. Bjelland L, Dahl AA, Haug TT, Neckelmann D. The validity of the hospital anxiety and depression scale. An updated literature review. J Psychosom Res 2002;52:69–77.
23. Zigmund AS, Snarth R. The hospital anxiety and depression scale. Acta Psychiatr Scand 1983;67:361–70.
24. Wallen MB, Hasson D, Theorell T, Canlon B. The correlation between the hyperacusis questionnaire and uncomfortable loudness levels is dependent on emotional exhaustion. Int J Audiol 2012;51:722–9.
25. Goldstein B, Shulman A. Tinnitus-hyperacusis and the loudness discomfort level test—a preliminary report. Int Tinnitus J 1996;2:83–9.
26. Kazilevsky VE, Bailie NA, Dutt SN, Halik JJ. Functional results of 394 bilateral stapedotomies evaluated with the Glasgow benefit plot. Eur Arch Otorhinolaryngol 2010;267:1027–34.
27. Pienkowski M, Tyler RS, Roncancio ER, et al. A review of hyperacusis and future directions: part II. Measurement, mechanisms, and treatment. Am J Audiol 2014;23:420–36.
28. Hazell JW, Sheldrake JB. Hyperacusis and tinnitus. In: Aran J-M, Dauman R, editors. Proceedings IV International Tinnitus Seminar, Bordeaux 1991. Amsterdam: Kugler, Gbedini Publications; 1992, pp. 245–8.
29. Bentler RA, Cooley LJ. An examination of several characteristics that affect the prediction of OSPL90 in hearing aids. Ear Hear 2001;1:58–64.
30. Jastreboff PJ, Jastreboff MM. Decreased sound tolerance. In: Snow Jr J, editor. Tinnitus: Theory and Management. London, Hamilton: BC Decker; 2004. pp. 8–15. Ch. 2.
31. Katzenell U, Segal S. Hyperacusis: review and clinical guidelines. Oto Neurotol 2001;22:321–6.
32. Dawson J. A comparison of physical measurements of pure-tones, third-octave bands of noise and third-octave bands of speech to subjective judgments of audibility threshold, MCL and UCL for three normally hearing listeners (Independent Studies and Capstones, Paper No. 306). St. Louis, MO: Washington University School of Medicine, Program in Audiology and Communication Sciences.
33. Dauman R, Bouscau-Faure F. Assessment and amelioration of hyperacusis in tinnitus patients. Acta Otolaryngologica 2005;125:503–9.