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Stapes hypermobility as a possible cause of hyperacusis

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ABSTRACT

Objective: Hyperacusis is a reduction of normal tolerances for everyday sounds. Although several publications have been produced demonstrating that minimally invasive surgical procedures may improve patient symptoms, the precise etiology of hyperacusis often remains elusive. This study describes 21 patients, 7 of whom stapes hypermobility is believed to be a mechanical genesis of their hyperacusis symptoms.
Study design: A prospective, repeated-measure single-arm design was used for this study.
Setting: All patients were evaluated and treated at a tertiary level otologic referral center.
Subjects and methods: 21 patients (Cohort A) with severe hyperacusis underwent oval and round window reinforcement. Seven patients (Cohort B) intraoperatively appeared to have subjective hypermobility of the stapes. Additional reinforcement of the stapes superstructure was performed in these patients.
Results: In Cohort A, loudness discomfort level (LDL) values improved on average from 72.7 dB to 81.9 dB. Hyperacusis questionnaire (HQ) scores improved from 30.1 to 14.7. Numeric Rating Scale scores (0 − 10) decreased from 8.5 to 4.0. In Cohort B, values similarly improved from an average of 72.4 dB to 88.2 dB. HQ scores improved from 35.8 to 18.9. Numeric Rating Scale scores fell from 10.0 to 3.7. Postoperatively there were no complaints of hearing loss. Sixteen out of 21(76%) reported improved quality of life and diminished symptoms of hyperacusis.
Conclusion: It is possible that patients suffering from hyperacusis may have a mechanical cause for their symptoms. Further research is necessary to clarify stapes mobility in patients with these symptoms. Excess temporalis tissue reinforcement of the stapes along with round window reinforcement shows promise as a minimally invasive surgical option for patients suffering from hyperacusis.

1. Introduction

Hyperacusis is an auditory phenomenon described as a hypersensitivity to everyday sounds. Individuals with this condition describe an unusual sensitivity to ordinary environmental noises such as music, the rustling of paper or plastic and clanking dishes [1]. The prevalence of hyperacusis reported in previously published literature is inconsistent, ranging between 5.9% and 17.2% of the population [2,3]. The mechanism of hyperacusis is not well understood but has been related to increased central auditory pathway gain resulting from acoustic overexposure [4]. While the etiology is often unknown, the most common known causes of hyperacusis reported in the literature include cochlear trauma, head injury, adverse medication reactions, hearing loss, aging, surgery, chronic ear infections, and autoimmune disorders [5]. Perilymphatic fistula (PLF), superior semicircular canal dehiscence and other lesions with third window physiology may also cause hyperacusis [6,7].

Currently, the range of treatment options for sound hypersensitivity includes avoidance of provocative stimuli, cognitive behavioral therapy (CBT), tinnitus retraining therapy, and hearing amplification with varied rates of efficacy [8]. CBT has been shown to be effective when exposure to sounds in a controlled and step-wise fashion, thereby reducing avoidance as well as audiological sensitivity [9]. Unfortunately, there is limited published data on the benefit of CBT in patients with hyperacusis and further investigation is warranted [4].

In our institute, we have investigated and published a series of patients with clinically diagnosed hyperacusis treated with a minimally invasive surgical procedure [10,11]. The procedure involves reinforcement of the round and oval window and patients showed improvement of objective measures of hyperacusis including loudness discomfort level (LDL) and a non-validated hyperacusis questionnaire. In our previous studies, the mobility of the stapes was not previously investigated before or during the operative treatment.

We present a series of 21 patients with severe hyperacusis who underwent round and oval window reinforcement. In a subset of 7 patients with hyperacusis, the stapes was perceived to be hypermobile

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and was treated with more extensive oval window reinforcement. The purpose of this study was to evaluate the effects of additional stapes reinforcement of a perceived hypermobile stapes in patients undergoing round and oval window reinforcement for hyperacusis as compared to minimal reinforcement of the oval window and reinforcement of the round window.

2. Materials and methods

Twenty-one patients with a history of severe hyperacusis underwent round and oval window reinforcement for the treatment of hyperacusis between January 1, 2015 and December 31, 2017. Institutional Review Board (IRB) approval had previously been obtained prior to initiating study-related activities. All patients were enrolled in an ongoing clinical trial to evaluate the effectiveness of round and oval window reinforcement for treatment of hyperacusis. Inclusion criteria have been previously described [10].

Prior to enrollment, subjects underwent a complete history and physical examination, high resolution CT scan of the temporal bones and a thorough audiometric workup. This included an audiogram with pure tone air and bone conduction thresholds, speech discrimination, tympanometry and Loudness discomfort level (LDL) testing. Patients were asked to self-report the severity of their hyperacusis on a 0–10 numeric rating scale. A self-report measure (HQ) was used to further assess the patient's hypersensitivity to sound. Patients were questioned regarding the presence of tinnitus before and after intervention. A self-report inventory Hospital Anxiety and Depression scale (HAD) was used to screen for mood disorders. Self-report and audiometric measures were repeated at 1 month following surgery.

2.1. Cohorts

From January 2016 through January 2017, accessing mobility of the stapes during the surgery was avoided to minimize chance of inner ear trauma. After January 2017, the mobility of the stapes was assessed after we found an obvious hypermobile stapes during a surgery. Excess reinforcement of the oval window and stapes superstructure was used in 7 patients (Cohort B) when the stapes was found to be hypermobile. Cohort A included 14 patients who were treated without investigating stapes mobility using minimal oval window reinforcement with one 2 mm piece of temporalis fascia. The round window niche was reinforced in both groups using three pieces of temporalis fascia shown in Fig. 1.

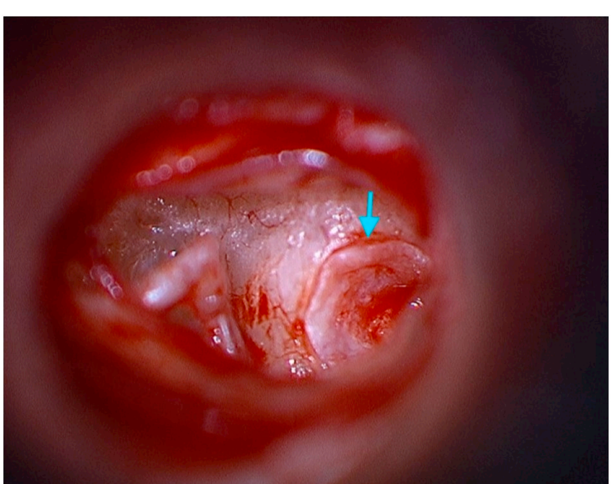

Fig. 1. Right ear with blue arrow showing temporalis fascia reinforcement of the round window. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

2.2. Hyperacusis questionnaire

This study uses a hyperacusis questionnaire that is a non-validated English adaptation of the German Gerauschuberempfindlichkeit (GUF), a German self-report measure for hypersensitivity to sound. 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. The total possible score range is 0 to 45, with higher scores indicating higher sound hypersensitivity.

2.3. Hospital Anxiety and Depression scale (HAD)

This scale was used to screen for excessive anxiety or depression in study participants. This 14-item questionnaire is composed of two subsets of questions ("a" and "d") designed to score the severity of both anxiety and depression. The possible score range is from 0 to 21 for each subscale. A score of 11 or higher in either subscale is suggestive of the presence of a mood disorder.

2.4. Hyperacusis Numeric Rating Scale

An 11-point numeric rating scale was used to allow patients to score the severity of their hyperacusis from 0 to 10.

2.5. Loudness discomfort level (LDL)

Pure tone thresholds at 250, 500, 1000, 2000, 3000, 4000, and 8000 Hz were first obtained. 3-tone pure tone averages (0.5 kHz, 1 kHz and 2 kHz) were then calculated. Loudness discomfort level testing proceeded starting at 60 dB HL and increased in increments of 5 dB HL. As the tone approached the uncomfortable loud level, the step size was decreased in order to determine the LDL with a 1 dB resolution. This process was performed twice at each frequency, and the average of the two LDLs is recorded for each ear separately.

2.6. Tinnitus

Subjects were questioned regarding a history of tinnitus and their answers were recorded in a binary fashion (tinnitus present: yes or no).

2.7. Surgical technique

All procedures were performed by the senior author (HS). A standard trans-canal approach under general anesthesia was performed similar to an approach for middle ear procedures. This has been previously described [10]. Both temporalis fascia and perichondria grafts have been used to reinforce the oval and round windows. In the first 6 cases (Cohort A) perichondrium and temporalis fascia was used while in the remaining cases temporalis fascia was found to be easier to manipulate than perichondrium. When the stapes was assessed for mobility, the ossicles were palpated using a 1.0 mm straight pick. The stapes was deemed hypermobile if during palpation the capitulum excursion was greater than half the width of the instrument tip. If the stapes was hypermobile then the stapes superstructure and oval window were reinforced with additional 6 to 8, 2 mm round temporalis grafts.

2.8. Statistical analysis

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. Microsoft Excel (Microsoft Corporation, Redmond WA, USA), (2010) was used for data analysis.

Table 1
Demographics of study participants.

Demographics	
Cohort 1	Cohort 2
Round and oval window reinforcement	Hypermobile stapes round and oval window reinforcement
Materials: temporalis fascia or tragal perichondrium	Materials: temporalis fascia
N = 16 subjects/ears	N = 7 subjects/ears
M = 6, F = 10	M = 4, F = 3
Average age: 60 (SD 13)	Average age: 65 (SD 5)

Table 2
Results. LDL = loudness discomfort level, HQ = hyperacusis questionnaire, PTA = pure tone average, HNS = Hyperacusis Numeric Rating Scale, HADS = Hospital Anxiety Depression Scale (A = anxiety score, D = depression score).

Results				
		Cohort A	Cohort B	All
	Patients (#)	14	7	21
LDL (dB)	Pre-op	72.7	72.4	72.6
	Post-op	81.9	88.2	83.8
HQ (0–45)	Pre-op	30.1	35.8	31.8
	Post-op	14.7	18.9	14.7
PTA (dB)	Pre-op	18.4	43.1	24.8
	Post-op	20.6	46.2	27.4
HNS (0–10)	Pre-op	8.5	10.0	9.1
	Post-op	4.0	3.7	4.0
HADS A (0–21)	Pre-op	7.3	9.0	7.7
	Post-op	5.5	7.3	6.2
HADS D (0–21)	Pre-op	4.9	9.5	5.3
	Post-op	4.7	4.9	4.8

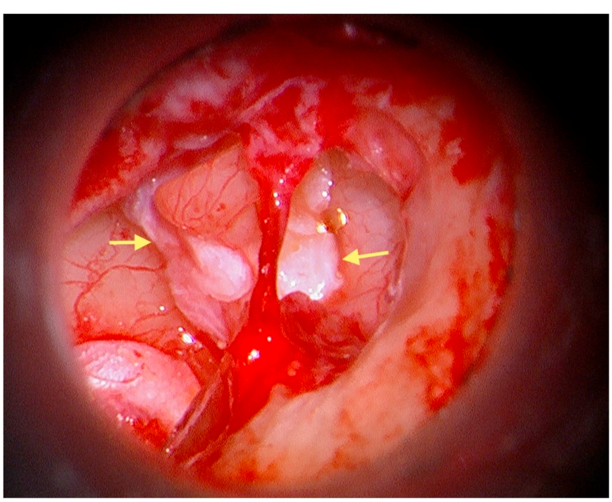

Fig. 2. Left ear with yellow arrows showing temporalis fascia additional reinforcement of the oval window and stapes superstructure as performed in the hypermobile stapes Cohort B. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

3. Results

The demographic information for these Cohorts is displayed in Table 1. Sixteen out of 21 (76%) patients had subjective improvement in their symptoms after surgery. The remaining 5 patients noted their symptoms were the same after the procedure. There was overall improvement in patients' symptoms including increased LDL, decreased HQ, and decreased HADS scores after surgical treatment and are listed in Table 2. The LDL increased from 72.6 dB to 83.8 dB post-operatively, hyperacusis questionnaire scores reduced from 31.8 pre-operatively to 14.7 post-operatively, and the HADS scores also showed improvement post-operatively in all but three patients because of incomplete data collection. Overall hearing outcomes showed a slight worsening in PTA with 24.8 dB before surgery and 27.4 dB after surgery, see Table 2 for detailed overall outcomes and by Cohort A and B.

3.1. Cohort A: round and oval window reinforcement

The Cohort A consisted of 14 patients (16 ears) whose stapes was not assessed in the operating room but did have oval and round window reinforcement as described and pictured in Fig. 1. Ten out of fourteen (71%) of patients noted subjective improvement of their symptoms. Seven patients endorsed a history of tinnitus preoperatively and one additional patient developed tinnitus postoperatively. These subjects did show improvement in both self-reported hyperacusis severity and HQ scores. HADS scores showed improvement in each subscale inventory. Average PTA was similar preoperatively 18.4 and postoperatively 20.6 and detailed audiometric results are displayed in Fig. 3. Examination of the HQ data shows that scores improved postoperatively in 12/14 subjects.

3.2. Cohort B: hypermobile stapes round and oval window reinforcement

Cohort B consisted of seven patients (seven ears) whose stapes were felt to be hypermobile and was reinforced with additional tissue as pictured in Fig. 2. Six of seven (85.7%) of patients noted subjective improvement of their symptoms. Three patients endorsed a history of tinnitus preoperatively and this was unchanged postoperatively. The post-operative HADS scores could not be calculated in three subjects due to incomplete data collection. In the remaining four subjects, preoperative and postoperative HADS scores decreased in category A and D as seen in Table 2. HQ scores improved in all seven subjects. No subjects developed tinnitus after surgery. One subject reported a hyperacusis severity of 10/10 both before and after surgery. This same subject also had an unexpected auditory outcome after surgery and had a large PTA shift (preop PTA 33, postop PTA 63) and PTA results were excluded from the Table 2 calculations. Postoperative auditory results for one of the patients in this cohort were not obtained due to lack of follow up but did note subjective improvement in symptom of hyperacusis after surgery, see Fig. 4 for auditory results.

It is our impression that whether there is a hypermobile stapes or not, the reduction in hyperacusis is better when extra tissue is placed around the stapes.

4. Discussion

In 2015 [10] and 2016 [11], Silverstein et al. reported patients with hyperacusis who experienced improvement in sound tolerance following round and oval window reinforcement with temporalis fascia. We postulate that acoustic or head trauma might injure the annular ligament of the oval window, leading to stapes hypermobility and hyperacusis. It is also possible that there is weakening of the annular stapes ligament secondary to aging or unknown factors. These patients were then treated with more extensive reinforcement of the oval window and stapes superstructure with temporalis fascia to help reduce the mobility of the stapes and therefore improve symptoms of sound intolerance. Despite the additional reinforcement, symptom and audiologic outcomes were found to be similar to those treated in Cohort A with the exception of one patient who had an unexpected rise in PTA after surgery.

4.1. Stapes mobility

Ossicular mobility has historically been a subject of interest among

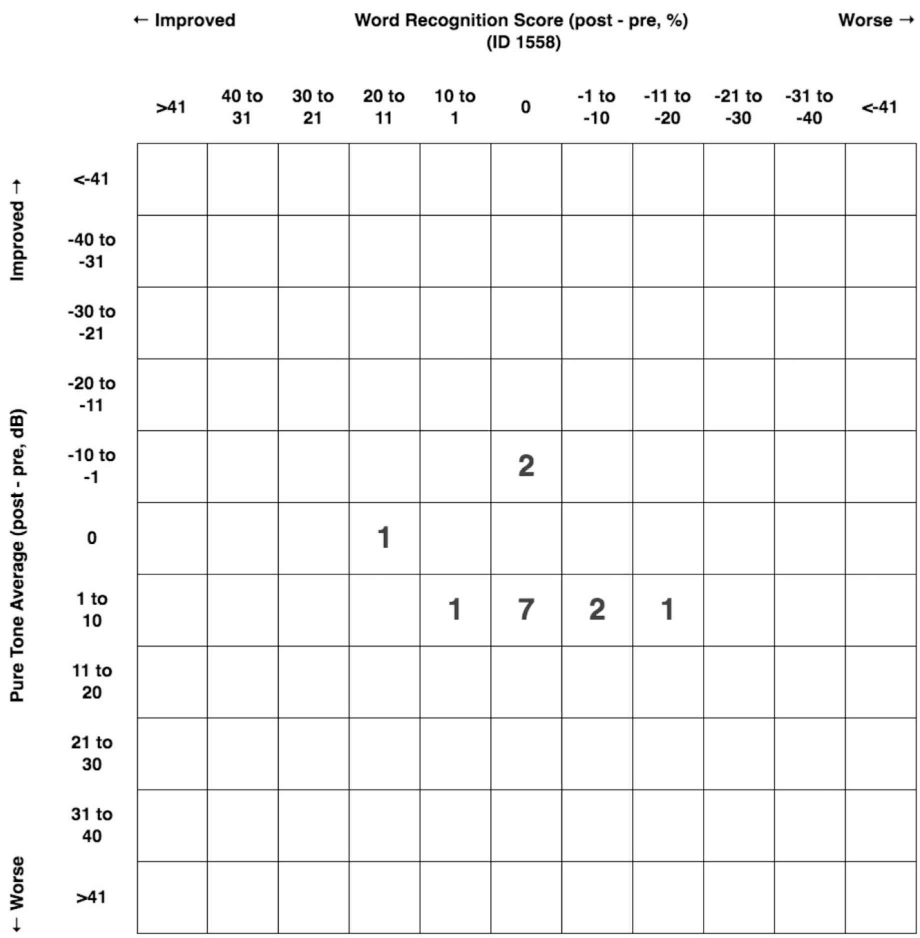

Fig. 3. Postoperative hearing outcomes in Cohort A.

otologists starting in the early 20th century with studies investigating ossicular mobility that were limited by the available technology of the day. The more recent development of sensitive equipment such as laser Doppler vibrometry (LDV) has permitted accurate measurements to be obtained both in vivo and in vitro. This led to a renewed interest in the pathophysiology of middle ear micromechanics. Greene et al. recently described the effects of high level, low frequency sounds on displacement of the stapes. The volume and frequency were designed to mimic percussive waves that occur in acoustic or blast trauma. The effects on the annular ligament were quantified using LDV [12].

Obtaining accurate measurements of stapes mobility has proven challenging. Huttenbrink recorded measurements on fresh postmortem ears by palpating the head of the stapes with a needle connected to a micrometer screw via a foil strain gauge. He found that complete annular ligament rupture occurred with 30–40 g of pressure and 0.5 mm of AP displacement of the stapedial head. Superior-Inferiorly directed forces induced annular ligament rupture with significantly less force but greater displacements [13]. Recently, laser doppler vibrometry (LDV) has been used to obtain accurate measurements of ossicular mobility in live human ear [14–17]. Greene et al. utilized LDV in embalmed cadaveric specimens to measure stapes displacement in response to high volume, low frequency sounds to model acoustic trauma from an explosion. They demonstrated that stapes displacement increases linearly with sound levels up to 150 dB SPL and that the sound-induced interpeak mobility of the capitulum is significantly larger than previously demonstrated in animal models. Displacement of the annular ligament approached saturation at 150 μm (0.15 mm) of displacement after which ligamentous injury was assumed to occur [12]. A relationship between hyperacusis and PLF has been previously established in the literature [18,19].

There are relatively few publications specifically evaluating stapes mobility. In addition, many published studies have performed measurements on human cadaveric specimens, and significant mechanical differences in sound transmission have been described between live and cadaveric human ears [20]. Despite these limitations, it seems that either mechanical or acoustic trauma may lead to annular ligament injury and potential PLF formation. A significant weakness of the current study is the lack of objective measurement of stapes mobility. In this study, a 1 mm straight pick was used for stapes palpation in each case. Those stapes felt to be hypermobile had a capitulum excursion greater than half the width of the instrument tip, which approximates the most conservative published stapes displacement necessary for annular ligament rupture [12,13].

4.2. Hyperacusis

Demographic reports of hyperacusis reveal that this abnormal sound sensitivity most commonly occurs bilaterally [21]. The majority of subjects in our series reported bilateral hyperacusis, although some identified a “worse” side. Surgical intervention was directed to the subject-identified “worst” side. Interestingly, improvement was reported in both ears after unilateral surgery and this was reflected in self-report measures postoperatively. Subjects also reported resuming social activities postoperatively which had previously been avoided. These included dining out, social interaction and no longer avoiding concerts or other public venues with loud sounds.

4.3. Hearing outcomes

Hearing outcomes were monitored and are reported in Figs. 3 and 4.

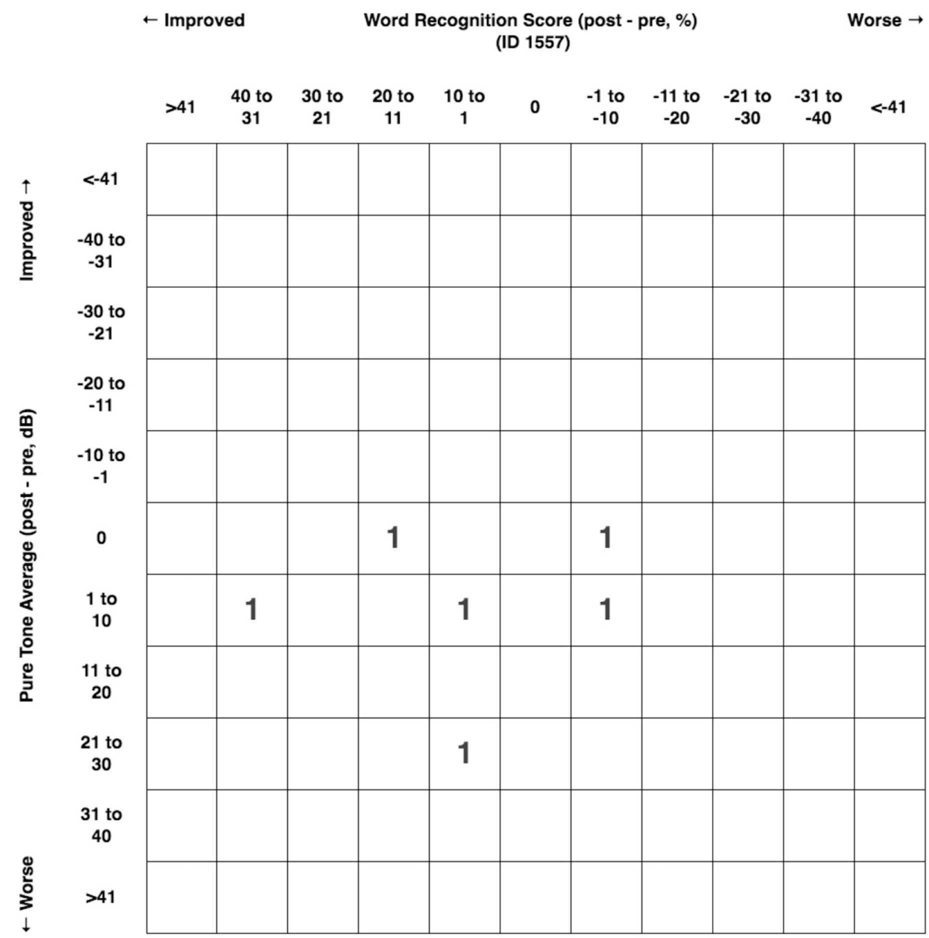

Fig. 4. Postoperative hearing outcomes in Cohort B.

Fig. 3 shows the post-operative changes in word recognition score (WRS) and pure tone average (PTA) for Cohort A. Fig. 4 identifies the number of participates in Cohort A who had changes in either PTA, WRS or both postoperatively. These are largely unremarkable but demonstrate that most subjects had a small change in PTA. The majority of these changes involved a diminished PTA but with less than a 10-dB change. Most of these were within test-retest parameters. Discrimination improved in two patients and three experienced worsening on follow up testing. Clinically, these patients did not notice a change in the clarity of their hearing or complain of hearing loss.

Fig. 4 shows the post-operative changes in word recognition score (WRS) and pure tone average (PTA) for Cohort B which shows a trend toward improvement in WRS (four participants). One subject experienced a dramatic improvement in WRS postoperatively. In this case it was felt that the subject's hyperacusis symptoms were so bothersome as to interfere with the ability to tolerate the 50-word CNC list used to calculate discrimination. We postulate that postoperative improvement in hyperacusis symptoms permitted a more accurate assessment of discrimination.

One subject in Cohort B experienced a shift in PTA from 33 to 63 postoperatively. There were no complaints of a change in hearing and the loss was identified on postoperative testing 1 month after surgery. There were no intraoperative complications noted during these procedures and the cause of the loss was not identified. It is possible that an undetected perilymph fistula could have been caused by injury to the annular ligament during palpation of the hypermobile stapes. Forces required to disrupt the oval window annular ligament have been previously quantified [13]. In each instance, annular ligament disruption was accompanied by stapedial tendon rupture. Neither perilymph leakage nor tendon disruption was observed during any of the procedures in our series.

4.4. LDLs

On average, there was improvement in the LDLs in both cohorts, suggesting an improvement in tolerance to sound. The average improvement following surgery was larger in the hypermobile stapes Cohort B, although these data are underpowered to show a statistically significant difference between the two cohorts. The concept that hyperacusis is an increased auditory responsiveness related to auditory neuronal degradation is not novel [10]. LDLs are the most commonly used measurement in the assessment of hyperacusis which can be challenging to quantify. The American Academy of Audiology considers 100 dB HL for the frequencies from 500 Hz to 8000 Hz to be the accepted normal reference level for LDLs. However, some measure of caution must be used in the interpretation of LDL testing. Both the sensitivity and specificity of LDLs as well as the test-retest reliability must be considered when using this test to confirm a diagnosis of hyperacusis. The reliability and consistency of the results are strongly dependent on the consistency and accuracy of the instructions provided by the tester. Despite this, some studies have shown that LDLs are useful for the assessment and follow up of patients with hyperacusis. The opinion of LDL utility remains mixed.

4.5. Limitations

This study has several limitations. First, due to the small sample size, this study lacks statistical power. Next, meaningful interpretation of subject HQ scores is limited using a non-validated questionnaire. A new questionnaire, the Inventory of Hyperacusis Symptoms, has recently been described [22]. This scale assesses hyperacusis symptoms in 5 domains: psychosocial impact, emotional arousal, functional impact, loudness and communication. In future studies we will consider using this questionnaire to more accurately categorize impact of subject's hyperacusis symptoms. Another limitation is the lack of structured, objective measurement of intraoperative stapes mobility. The present study referenced capitulum displacement to the tip width of a 1 mm straight micropick. These results seem to indicate that some subjects have a stapes mobility significantly larger than the range of normal suggested by the literature. However, limited data on the mobility of the stapes is available for review and most sample sizes are quite small. Additionally, some studies are performed in fresh postmortem bodies [13], others on cadaveric specimen [12], and few are performed in vivo [14,15]. As noted above, there are mechanical differences in sound transmission between human and cadaveric specimens, further complicating the acquisition of accurate measurements [20].

5. Conclusions

Consistent with previously reported results [10,11], this study provides evidence that round and oval window reinforcement using either tragal perichondrium or temporalis fascia may reduce sound sensitivity in patients suffering from hyperacusis. Temporalis tissue was found to be easier to use. Although these data are underpowered to show a statistically significant difference between the two cohorts, they seem to suggest that hypermobility of the mobile stapes may play a role in hyperacusis and that results are better when there is excess tissue placed around the stapes. We theorize that these patients may have sustained an injury affecting the micromechanics of the ossicular chain at the level of the oval window and that mechanical reinforcement of these structures explains improvement in their symptoms. Further investigation of stapes mobility both in normal ears and in patients with hyperacusis is necessary to clarify this relationship. Whether there is real hypermobility of the stapes, excess tissue reinforcement of the stapes and oval window appears to help a significant number of patients tolerate loud sounds better.

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