Surgical treatment of superior semicircular canal dehiscence: a single-centre experience in 63 cases.

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Abstract

Background: Superior semicircular canal dehiscence syndrome may require surgical intervention for disabling symptoms. Various surgical procedures have been described but there is no consensus on any treatment algorithm. Methods: In this single-centre retrospective study, we report the results of the procedures performed between 2006 and 2019 using the three main surgical approaches, middle fossa approach (MFA), transmastoid approach (TMA) and round window reinforcement (RWR). The outcome on cardinal cochlear and vestibular symptoms, audiometric results and changes in cVEMPs were analysed. The patients were also interviewed 12 months to 13 years post-treatment to establish their overall satisfaction following surgery. Results: 63 patients were divided into three groups: 42 MFA; 12 RWR; 9 TMA. Post-surgical control rates exceeded 80% for the majority of symptoms in the MFA and TMA groups, and ranged from 11.1 to 83.3% for the RWR group. Over 90% of MFA or TMA patients and 60% of the RWR cohort were satisfied overall with their treatment. Hearing thresholds were intact following surgery in the MFA and TMA groups. There was one case of profound post-operative deafness in the RWR group. Discussion: MFA and TMA are both safe and effective techniques in the treatment of disabling SSCD. Since MFA is the more invasive technique, this study suggests that TMA should be proposed as first-line treatment, temporal bone anatomy permitting. RWR outcomes are more random and this option could be offered to patients at risk under general anaesthesia. Key words: Minor syndrome, superior canal dehiscence, hearing loss, vertigo.

MAIN DOCUMENT

Key points:

- Both the middle fossa and transmastoid approaches are safe and effective in the treatment of disabling superior semicircular canal dehiscence.

- Cochlear symptoms (tinnitus, autophony and aural fullness) improved in almost 85% of MFA patients and 80% of TMA patients.

- On average, hearing thresholds did not change significantly after TMA and improved slightly after MFA.

- Vestibular symptoms (dizziness, oscillopsia, Tullio, effort-induced vertigo) improved in 80% of MFA patients compared to almost 90% of TMA patients.

- The RWR outcomes are inconsistent. This option should therefore be offered to patients at risk under general anaesthesia.

Introduction:

Superior semicircular canal dehiscence was described for the first time by Lloyd Minor in 1998, correlating hypersensitivity phenomena in the inner ear to defective coverage of the superior canal[1]. This dehiscence of the otic capsule creates a direct interface between the perilymph, the membranous canal and the overlying dura-mater, which acts as third window (in addition to the physiological oval and round windows). The presence of this third window increases the difference in pressure between the two normal windows creating a low impedance pathway in the direction of the labyrinth via which acoustic energy is dissipated. The resulting loss of energy is illustrated by an increase in hearing thresholds in terms of air conduction. However, this mechanism also increases the transmission of bone vibrations to the perilymph fluids via the labyrinth, generating bone conduction hyperacusis.

The symptoms triggered are heterogeneous but can be extremely disabling, combining cochlear signs such as autophony or pulsatile tinnitus, and vestibular signs including Tullio's phenomenon or oscillopsia[2]. Surgical treatment is proposed in the event of disabling symptoms [3]. The first surgical procedure was described by Minor and comprised plugging the canal via a middle fossa approach (MFA) [4]. This approach was subsequently widely used with different techniques for treating dehiscence (capping, resurfacing [5]). In 2008, Agrawal and Parnes suggested using the transmastoid approach (TMA) to access the superior canal – a slightly less invasive approach and one more familiar to otologists[6]. Several series of TMA patients have reported similar results to those obtained with the MFA but critics condemn it for not providing a direct view of the dehiscence. In 2002, Kartush *et al.* introduced the window reinforcement technique via the canal to suppress the effect of the third window, described in Silverstein *et al.* [7] . Their results were deemed satisfactory in 4 out of 6 cases treated, but subsequent studies carried out with this technique yielded variable results [8].

Objective The purpose of this study is to present the results obtained with each of the three techniques in a single-centre cohort of patients who underwent surgery due to symptomatic superior semicircular canal dehiscence.

Method:

Ethical considerations The local institutional Review Board approved this retrospective study and waived the need for written informed consent (RnIPH 2021-03).

Study Design, settings The data of all patients who underwent surgery for disabling superior semicircular canal dehiscence (SSCCD) between 2006 and 2019 in our tertiary referral centre were analysed retrospectively.

Participants SSCCD was confirmed in all patients by dedicated computed tomography of the temporal bone, with at least one disabling, related cochlear or vestibular symptom [9] and at least one objective test indicating the presence of a third window [videonystagmography (VNG) with pressure tests; cervical vestibular-evoked myogenic potentials (cVEMP) with threshold study]. Each patient in the cohort was assigned to the group corresponding to the surgical strategy selected (MFA, TMA or RWR). Patients who were inadequately controlled and subsequently reassessed using another approach were analysed a second time in the group for the second approach used. Patients with associated dehiscence of the posterior canal were not selected.

Outcome measures, analysis Symptomatic control was assessed between 6 and 12 months post-surgery for the cardinal cochlear and vestibular symptoms of SSCCD reported by participants prior to surgery (sensation of deafness, fullness, tinnitus, autophony, instability, sound- (Tullio) or effort-induced vertigo and oscillopsia). Hearing thresholds in air-conduction (AC) and bone-conduction (BC), cervical Vestibular Evoked Myogenic Potentials (cVEMPs) and Video Head Impulse Testing (VHIT) were assessed pre-operatively, and between 6 to 12 months after surgery. Pure-tone average (PTA) was calculated on four frequencies (0.5, 1, 2 and 4 kHz).

All subjects were also interviewed 6 months to 13 years post-treatment to establish their overall satisfaction. A descriptive statistical analysis of mean values and 95% confidence intervals was performed to study quantitative variables. Percentages were used to describe qualitative variables. Mean audiometric thresholds were compared between the pre- and postoperative setting in each group using a Wilcoxon rank-sum test

(XLStat for Excel) with a level of significance of $\alpha = 0.05$.

Results:

Baseline characteristics During the study period, 54 patients underwent surgery (31 women and 23 men) including 7 bilateral procedures, and 2 patients underwent another surgery using a second approach. In case of bilateral dehiscence, the most affected ear, basing on symptoms and/or cVEMPs threshold, was chosen for the first procedure. We therefore examined the results recorded in 63 ears. The median age was 50 years [standard deviation (SD)=12.38], and the right/left ratio 1.03. The distribution of symptoms reported by patients on diagnosis is listed in Table 1. The most common symptom was instability (92.1%), followed by tinnitus (82.5%) and a sensation of deafness (81.0%). The hearing thresholds are presented in Table 2. Pressure VNG was positive (abnormal eye movement on applying Valsalva's manoeuvre and/or intense sound stimulation) in 82% of cases when performed (n=56/63). The mean cVEMP threshold was 74.7 dB (n=58/63; SD=11.9), 8/58 did not present any threshold anomaly and increased anomaly was reported in 43/58 cases.

Post-surgical clinical course MFA was performed on 42 ears, RWR on 12 and TMA on 9. The average hospital stays were 5.12 days (SD=1.29), 1.58 days (SD=0.52) and 3.67 days (SD=0.71), respectively. Results relating to cardinal symptom control are presented in Table 1. The post-operative control rates exceeded 80% for all symptoms in the MFA and TMA groups except for instability (78.4% in the MFA group) and sensation of deafness (27/36, i.e. 75.0% in the MFA group, 3/6 in the TMA group). With regard to instability, 4 MFA patients whose condition did not improve presented bilateral dehiscence and underwent secondary surgery in the opposite ear. In the RWR group, symptomatic control was 47.06% for cochlear symptoms and 74.07% for vestibular symptoms including 58% for instability control. The patients were contacted at the time of this study and were asked to comment on overall improvement following surgery. 38/42 MFA patients (90.5%) reported a (partial or complete) improvement compared to 9/9 TMA patients with 3 patients reporting a partial improvement in terms of symptoms. Conversely, 41% of RWR patients reported no improvement (Figure 1). Regarding the patients who underwent bilateral surgery, 7/7 initially underwent a MFA procedure. Three of these patients reported a complete improvement after this initial procedure and 4 a partial improvement. All of the uncontrolled symptoms were vestibular in nature (instability 4/7; Tullio's phenomenon 2/7; oscillopsia and effort-induced vertigo 1/7). One MFA patient whose condition had not improved presented contralateral dehiscence but did not wish to undergo repeat surgery.

Regarding hearing, the mean PTAs in AC and BC are presented in Table 2. There was no significant difference between mean pre- and post-operative values for the MFA (BC: + 0.83 dB, p=0.693) and TMA (BC: +1.94 dB, p=0.120; AC: +2.79 dB, p=0.192) groups, except for the statistical improvement in the mean AC thresholds in the MFA group (AC: -3.39 dB, p=0.02). One patient in the RWR group developed cochleovestibular syndrome 10 days after surgery, which led to profound hearing loss. A significant deterioration in the mean thresholds was therefore noted in this group following surgery (BC: +14.06 dB, p=0.008; AC: +12.40 dB, p=0.049). We assessed the number of patients whose hearing thresholds increased by more than 10 dB at 4000Hz in both BC and AC. This corresponded to 4 out of 12 patients who underwent RWR surgery and approximately 10% of the MFA and TMA patients.

The cVEMP evaluation (n = 51/63) highlighted a mean post-operative threshold of 86.47 dB (SD=11). On an individual level, 11 patients maintained thresholds below 80 dB. In the MFA group, 6/42 maintained abnormal thresholds (14.29%), but 5 of these 6 patients reported an overall improvement; 3/12 RWR patients maintained abnormal thresholds (37.5%) with no improvement in 2 of these patients. As regards the TMA group, the threshold did not revert to normal in 2/9 patients (22.22%) but clinical improvements were noted in both cases.

Discussion:

The treatment of SSCCD has gradually changed since it was first described by Minor in the early 2000s [4]. Many studies have validated surgery as standard treatment for symptomatic patients and report an overall success rate (defined as complete or partial disappearance of symptoms) of over 90% [10].

The MFA is most suited to the anatomy since it provides a direct view on the dehiscence and facilitates treatment of the bony defect. Further, it facilitates the concomitant treatment of large tegmen defects when associated. Access nevertheless remains difficult when the dehiscence is located in the medial portion of the canal, behind or even in contact with the superior petrous sinus [11]. This approach also poses a potential, albeit limited, risk of neurological complications[12]. Several teams have shown that the use of an endoscope can limit temporal retraction and improve the view of the dehiscence particularly when the arcuate eminence defect is along a low-lying tegmen [13]. No approach-related complications were observed in our series of MFA patients. Efficacy in terms of symptom control was established with an improvement in all symptoms, except subjective deafness, in almost 80% of cases[14]. Our results are similar to those found in the literature with a control rate ranging from 75% for sensation of deafness to 96% for fullness. In terms of maintained labyrinth function, some studies have not shown any significant worsening in cochlear and vestibular functions [15] while others reported significant worsening of up to 36% in high-frequency hearing thresholds [14]. Mean hearing thresholds were unchanged in our series but approximately 10% displayed sensorineural hearing loss at 4000 Hz.

The development of TMA in this indication has extended the treatment of SSCCD to more otolaryngology surgeons familiar with this approach. Several teams plug the canal on both the ampullated and non-ampullated sides [6,16]. In theory, this technique poses a greater risk of labyrinth complications compared to resurfacing. This is partly due to the risk of damaging the membranous labyrinth and due to the drilling close to the labyrinth [17]. Moreover, it does not provide a direct view of the dehiscence and may be insufficient to treat multiple or large tegmen defects when associated. However, satisfactory results are still being recorded with TMA with control rates for hearing symptoms and induced vertigo above 80% with hearing thresholds maintained [16]. These observations are consistent with the results recorded in our work. Despite having a small series (n=9), symptom control exceeded 80% for all symptoms except sensation of deafness. No complications were noted in this group and a shorter hospital stay was recorded compared to MFA.

Round window reinforcement via the transcanal approach heralds a return to a physiological model with 2 mobile windows [7]. In their first patient series, post-operative sensorineural hearing loss was observed in 2 out of 3 patients with complete round window obturation. This technique was thus abandoned in favour of round window reinforcement. Significant improvements in symptoms in 4 out of 6 patients in the initial series and in all cochlear and vestibular symptoms, apart from the sensation of deafness, in a multicentre study involving 19 subjects were documented with this technique[7]. Among the studies investigating this alternative, Succar *et al* . [18] highlighted a subjective improvement in 64% of patients (9/14) and stable BC thresholds. In contrast, 50% of patients presented a decline of more than 10 dB in AC thresholds. In our series, 12 patients underwent RWR surgery but this alternative was abandoned in 2016 given the random nature of the results obtained (overall improvement of less than 60%). In addition, 1 patient presented cophosis secondary to post-surgical labyrinthitis confirmed by a labyrinthine hypersignal on the MRI scan. Profound deafness persisted despite repeat surgery to remove the obturation material and apply dexamethasone. RWR remains a minimally invasive alternative[8] and may be considered in patients at high anaesthetic or surgical risk.

No meta-analysis could be carried out to effect a robust comparison of these different approaches[19] since the studies include mostly retrospective cases with few participants and variable endpoints from one study to the next. In their systematic review, Nguyen *et al.* found that auditory symptoms were more often improved after MFA compared to TMA (72% versus 59%) without any difference for vestibular symptoms [14]. However, fewer complications appear to have occurred with TMA and scenarios were less severe compared to MFA[19]. Depending on the anatomy, the authors advocate the transmastoid approach when the dehiscence is facing the superior petrous sinus. Conversely, in the event of a poorly pneumatized mastoid with low-lying tegmen, this technique is less comfortable than the MFA[5,11]. On our site, TMA has gradually replaced MFA as first-line therapy subject to favourable anatomical conditions.

The study of VEMPs has largely demonstrated its interest in the diagnosis of SCCSSD to investigate the third window effect. Abnormally low thresholds are generally found together with an abnormally increased amplitude. This is indicative of dehiscence-induced vestibular hypersensitivity [20]. These anomalies may be linked to the size and location of the dehiscence [21]. In most cases, thresholds revert to normal following surgery but this normalisation does not always correlate with clinical improvement. In our series as in other studies [22], normalisation of cVEMPs is not strictly associated with clinical improvement, and vice-versa. However, 80% of patients had both pre and post-operative recording of cVEMPs, and the retrospective nature of our study limited the data completeness.

VHIT is another potentially useful technique for the diagnosis and post-surgical control of these patients. It is used to analyse individual semicircular canal function. According to a recent study, a 20% decrease in superior canal function is immediately observed following surgery before reverting to normal [23]. This selective, transient hypofunction could reflect the elimination of the pathological flow of perilymph in the canal, or an inflammatory reaction associated with the handling of the membranous labyrinth in the said canal. However, plugging rarely reaches the ampulla, the level at which recorded VHIT responses are detected. This may explain the normalised gain away from surgery since an endoluminal flow may persist at this level. In our series, some patients experienced a significant change in terms of gain in the treated canal whilst normal function and no correlation with symptom changes were documented in others.

Conclusion This retrospective study confirms that excellent results are obtained with both MFA and TMA in terms of symptom control and labyrinthine preservation. TMA remains a less invasive technique and may be used as first-line therapy under favourable anatomical conditions. Conversely, RWR does not facilitate satisfactory symptom control. It can be considered in fragile patients to avoid general anaesthesia but the risk of failure should be pointed out. Many grey areas remain in terms of the link between pathophysiology, clinical signs and objective electrophysiological results. A better understanding of how these different parameters are interlinked might help to identify those patients who would not benefit from this type of procedure.

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Tables & figures with captions:

		MFA	RWR	TMA
Subjective deafness	n preoperative	36/42	9/12	6/9
	n improvement	27/36	1/9	3/6
	% improvement [CI95]	75.0% [61.9 - 88.1]	11.1% [0 - 28.7]	50.0% [17.3 – 82.7]
Aural fullness	n preoperative	25/42	7/12	5/9
	n improvement	24/25	3/7	4/5
	% improvement [CI95]	96.0% [90.1 - 100]	42.9% [14.9 - 70.9]	80.0% [53.9 - 100]
Tinnitus	n preoperative	34/42	12/12	6/9
	n improvement	29/34	8/12	5/6
	% improvement [CI95]	85.3% [74.6 - 96.0]	66.7% [40.0-93.3]	83.3% [59.0 - 100]
Autophony	n preoperative	20/42	6/6	7/9
	n improvement	17/20	4/6	7/7
	% improvement [CI95]	85.0% [74.2 - 95.8]	66.7% [40.0-93.3]	100%
Cochlear symptoms	% improvement	**84.35%** [73.36 – 95.34]	**47.06%** [18.82 – 75.30]	**79.17%** [52.64 – 100]
Instability	n preoperative	37/42	12/12	9/9
	n improvement	29/37	7/12	8/9
	% improvement [CI95]	78.4% [65.9 - 90.8]	58.3% [30.4 - 86.2]	88.9% [68.4 - 100]
Tullio's phenomenon	n preoperative	17/42	6/12	0/9
	n improvement	14/17	5/6	0/0
	% improvement [CI95]	82.35% [70.8 - 93.9]	83.3% [62.2 - 100]	-
Effort-induced vertigo	n preoperative	20/42	6/12	4/9
	n improvement	16/20	5/6	4/4
	% improvement [CI95]	80.0% [67.9 - 92.1]	83.3% [62.2 - 100]	100.0%
Oscillopsia	n preoperative	16/42	3/12	6/9
	n improvement	13/16	3/3	5/6
	% improvement [CI95]	81.3% [69.5 - 93.1]	100%	83.3% [59.0 - 100]
Vestibular symptoms	% improvement	**80.00%** [67.90 – 92,10]	**74.07%** [49.27 – 98.87]	**89.47%** [69.42 – 100]

Table 1: Pre-operative symptom distribution and post-operative improvement. Abbreviations: MFA, middle fossa approach; n, number of participants; RWR, round window reinforcement; TMA, transmastoid approach; [CI95], 95% confidence interval.

		MFA	RWR	TMA
Bone Conduction (BC)				
Preoperative PTA	m (sd)	17.86 dB (+/- 21.89)	19.2 dB (+/- 15.7)	15.0 dB (+/- 13.4)
	[CI95]	[11.24 – 24.48]	[10.32 – 28.03]	[6.25 – 23.75]
Postoperative PTA	m (sd)	18.7 dB (+/- 21.6)	33.2 dB (+/- 32.3) *	16.9 dB (+/- 12.6)
	[CI95]	[12.17 – 25.23]	[14.92 – 51.48]	[8.67 – 25.13]
Air Conduction (AC)				
Preoperative PTA	m (sd)	32.7 dB (+/- 25.3)	24.8 dB (+/- 14.5)	23.6 dB (+/- 15.7)
	[CI95]	[25.05 – 40.35]	[16.60 – 33.00]	[13.34 – 33.86]
Postoperative PTA	m (sd)	29.3 dB (+/- 25.6) *	37.2 dB (+/- 30.9) *	26.4 dB (+/- 14.0)
	[CI95]	[21.56 – 37.04]	[19.72 – 54.68]	[17.25 – 35.55]
Evolution of High-frequency thresholds				
BC & AC 4 kHz worsening > 10 dB	n	4/41	4/12	1/9
	% [CI95]	9.76% [0.67 – 18.84]	33.33% [6.66 – 60.01]	11.11% [0 – 31.64]
AC 8 kHz worsening > 10 dB	n	7/41	4/12	2/9
	% [CI95]	17.07% [5.56 – 28.59]	33.33% [6.66 – 60.01]	22.22% [0 – 49.38]

Table 2: Changes in hearing thresholds. Abbreviations: dB, decibel; m, mean; MFA, middle fossa approach; PTA, pure-tone average; RWR, round window reinforcement; sd, standard deviation; TMA, transmastoid approach; [CI95], 95% confidence interval. Note: Significant differences between preoperative and postoperative values are indicated by * ($p < 0.05$, Wilcoxon Test)

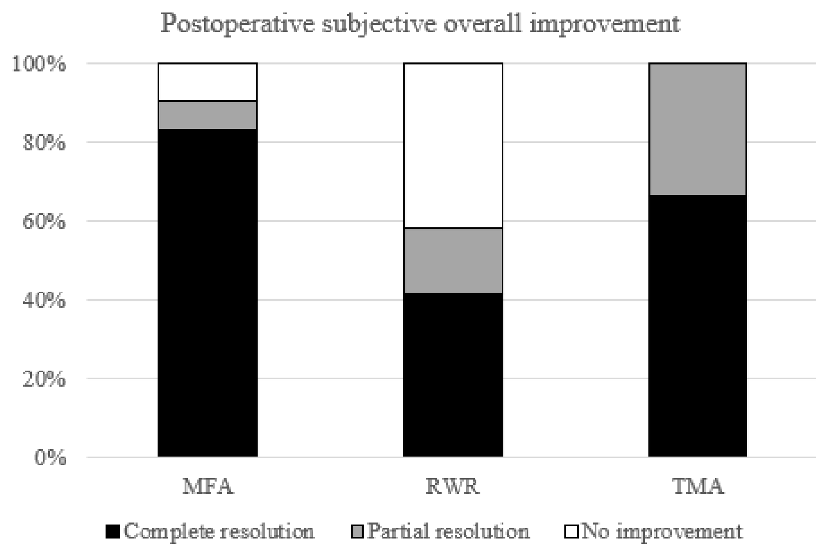

Figure 1: Long-term subjective overall improvement. Abbreviations: MFA, middle fossa approach; RWR, round window reinforcement; TMA, transmastoid approach.

		MFA	RWR	TMA
Subjective deafness	n preoperative	36/42	9/12	6/9
	n improvement	27/36	1/9	3/6
	% improvement [CI95]	75.0% [61.9 - 88.1]	11.1% [0 - 28.7]	50.0% [17.3 – 82.7]
Aural fullness	n preoperative	25/42	7/12	5/9
	n improvement	24/25	3/7	4/5
	% improvement [CI95]	96.0% [90.1 - 100]	42.9% [14.9 - 70.9]	80.0% [53.9 - 100]
Tinnitus	n preoperative	34/42	12/12	6/9
	n improvement	29/34	8/12	5/6
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Autophony	n preoperative	20/42	6/6	7/9
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	% improvement [CI95]	85.0% [74.2 - 95.8]	66.7% [40.0-93.3]	100%
Cochlear symptoms	% improvement	**84.35%** [73.36 – 95.34]	**47.06%** [18.82 – 75.30]	**79.17%** [52.64 – 100]
Instability	n preoperative	37/42	12/12	9/9
	n improvement	29/37	7/12	8/9
	% improvement [CI95]	78.4% [65.9 - 90.8]	58.3% [30.4 - 86.2]	88.9% [68.4 - 100]
Tullio's phenomenon	n preoperative	17/42	6/12	0/9
	n improvement	14/17	5/6	0/0
	% improvement [CI95]	82.35% [70.8 - 93.9]	83.3% [62.2 - 100]	-
Effort-induced vertigo	n preoperative	20/42	6/12	4/9
	n improvement	16/20	5/6	4/4
	% improvement [CI95]	80.0% [67.9 - 92.1]	83.3% [62.2 - 100]	100.0%
Oscillopsia	n preoperative	16/42	3/12	6/9
	n improvement	13/16	3/3	5/6
	% improvement [CI95]	81.3% [69.5 - 93.1]	100%	83.3% [59.0 - 100]
Vestibular symptoms	% improvement	**80.00%** [67.90 – 92,10]	**74.07%** [49.27 – 98.87]	**89.47%** [69.42 – 100]

		MFA	RWR	TMA
Bone Conduction (BC)				
Preoperative PTA	m (sd)	17.86 dB (+/- 21.89)	19.2 dB (+/- 15.7)	15.0 dB (+/- 13.4)
	[CI95]	[11.24 – 24.48]	[10.32 – 28.03]	[6.25 – 23.75]
Postoperative PTA	m (sd)	18.7 dB (+/- 21.6)	33.2 dB (+/- 32.3) *	16.9 dB (+/- 12.6)
	[CI95]	[12.17 – 25.23]	[14.92 – 51.48]	[8.67 – 25.13]
Air Conduction (AC)				
Preoperative PTA	m (sd)	32.7 dB (+/- 25.3)	24.8 dB (+/- 14.5)	23.6 dB (+/- 15.7)
	[CI95]	[25.05 – 40.35]	[16.60 – 33.00]	[13.34 – 33.86]
Postoperative PTA	m (sd)	29.3 dB (+/- 25.6) *	37.2 dB (+/- 30.9) *	26.4 dB (+/- 14.0)
	[CI95]	[21.56 – 37.04]	[19.72 – 54.68]	[17.25 – 35.55]
Evolution of High-frequency thresholds				
BC & AC 4 kHz worsening > 10 dB	n	4/41	4/12	1/9
	% [CI95]	9.76% [0.67 – 18.84]	33.33% [6.66 – 60.01]	11.11% [0 – 31.64]
AC 8 kHz worsening > 10 dB	n	7/41	4/12	2/9
	% [CI95]	17.07% [5.56 – 28.59]	33.33% [6.66 – 60.01]	22.22% [0 – 49.38]

TABLE 2: Changes in hearing thresholds. Abbreviations: dB, decibel; m, mean; MFA, middle fossa approach; PTA, pure-tone average; RWR, round window reinforcement; sd, standard deviation; TMA, transmastoid approach; [CI95], 95% confidence interval. Note: Significant differences between preoperative and postoperative values are indicated by ***** ($p < 0.05$, Wilcoxon Test)

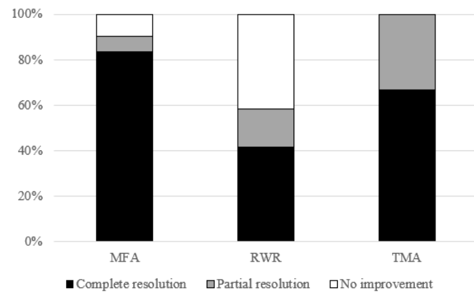

FIGURE 1: Long-term subjective overall improvement. Abbreviations: MFA, middle fossa approach; RWR, round window reinforcement; TMA, transmastoid approach.