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Original Article

Postoperative Late Hearing Deterioration in Cholesteatoma with Labyrinthine Fistulas

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BACKGROUND: A labyrinthine fistula is a severe complication of middle ear cholesteatoma that can cause profound sensorineural hearing loss and vertigo. However, there is no consensus regarding the transition to postoperative hearing. Although hearing deteriorates gradually with a delay in some cases of labyrinthine fistula, insufficient consideration has been given to this point. We examined perioperative changes in cases of middle ear cholesteatoma with labyrinthine fistulas.

METHODS: We retrospectively reviewed the medical records of 578 patients with middle ear cholesteatoma who underwent tympanoplasty at our hospital between 2016 and 2021. Patients with labyrinthine fistulas were selected; their perioperative bone-conduction hearing was assessed. Fistula depth was determined following the classification reported by Dornhoffer et al. The hearing was compared preoperatively, early postoperatively (3-6 months), and 1 year postoperatively.

RESULTS: Forty-eight patients (8.3%) had labyrinthine fistulas. Regarding depth, 21 cases were type I, 14 were type IIa, 3 were type IIb, and 10 were type III. Preoperative bone-conduction hearing was significantly poor in invasion type IIb or deeper cases. Cases with type IIb or deeper fistulas, multiple fistulas, or vertigo deteriorated postoperatively. Type III cases or those with multiple fistulas deteriorated further from the early postoperative period to 1 year postoperatively. Concerning frequency, 500 and 2000 Hz showed a delayed deterioration.

CONCLUSION: This is a valuable report of delayed hearing loss after surgery in patients with a labyrinthine fistula. This change is associated with the labyrinthine fistula's depth and multiple fistulas—this is important during preoperative counseling of patients undergoing surgery.

KEYWORDS: Cholesteatoma, labyrinthine fistula, tympanoplasty, hearing loss, vertigo

INTRODUCTION

Labyrinthine fistula is a severe complication of middle ear cholesteatoma that can cause profound sensorineural hearing loss and is found in approximately 2.9%-12.5% of middle ear cholesteatomas.[1,2] There is no consensus on the postoperative hearing transition, as few reports have evaluated postoperative hearing in more than 1 time period. We sometimes encounter cases of labyrinthine fistula in which hearing deterioration is slow and delayed. In addition, labyrinthine fistulas can cause vertigo, which is said to occur in 56% of the cases.[3] However, its relationship with hearing is unclear. Therefore, we aimed to examine the perioperative hearing changes in cases of middle ear cholesteatoma with labyrinthine fistulas.

MATERIALS AND METHODS

We retrospectively reviewed the medical records of 578 patients with middle ear cholesteatoma who underwent tympanoplasty at the Department of Otorhinolaryngology, Jikei University Hospital, Tokyo, between 2016 and 2021. The study was approved by the Ethics Committee of Jikei University (Approval Number: 32-205 10286), and informed consent was obtained by the opt-out method.

Patients with labyrinthine fistulas were selected, and their perioperative bone-conduction (BC) hearing was assessed. A labyrinthine fistula was defined as a defect in the bone labyrinth during surgeries. The fistula depth was classified using the classification

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reported by Dornhoffer et al.[4] In addition, the locations of the fistulas were recorded as lateral semicircular, posterior, superior semicircular canal, or cochlea-vestibule. Patients with recurrent cholesteatomas were included; however, those who underwent previous surgery for labyrinthine fistulas were excluded. Cholesteatoma and labyrinthine fistula were surgically confirmed in all cases. The cholesteatoma diagnosis was based on the presence of intraoperative keratinized squamous epithelium and middle ear debris or histopathological examination of the excised tissue. The cholesteatomas were removed entirely, and the fistulas were treated with autograft materials, such as the temporal muscle fascia, bone pate, and cartilage. In all cases, computed tomography scans obtained approximately 1 year postoperatively confirmed that the labyrinthine fistulas had been repaired properly.

The hearing was compared preoperatively, early postoperatively (3-6 months), and 1 year postoperatively. The guidelines of the Committee on Hearing and Equilibrium of the American Academy of Otolaryngology-Head and Neck Surgery[5] were followed. The pure tone average (PTA) was calculated as the mean of the 0.5-, 1-, 2-, and 3-kHz thresholds. Its association with vertigo was also examined. The results were expressed as means with 95% confidence intervals. All analyses were performed using a statistical software package JMP version 13.0 and JMP Pro version 14.0.0 (SAS Institute Japan, Tokyo, Japan) and included the Wilcoxon rank-sum and signed-rank tests to assess differences between groups. Statistical significance was set at a *P*-value of <.05.

RESULTS

This study included 48 patients (8.3%) with labyrinthine fistulas (Table 1). Overall, 15 patients were female, 33 were male, and their mean age was 48.6 ± 16.9 years. When the fistula depth was assessed according to the Dornhoffer et al classification, type I fistulas were found in 21 patients, type IIa in 14, type IIb in 3, and type III in 10 patients. Fistulas were present in the lateral semicircular canals of 43 patients, superior semicircular canals of 9, posterior semicircular canals of 6, and cochlea-vestibule of 5 patients. Some had multiple fistulas. Specifically, 37 patients had single fistulas, and 11 had multiple fistulas. Twenty-one patients (43.8%) experienced vertigo preoperatively. Audiological examinations (PTA) were performed in all the cases. The overall mean preoperative BC hearing was 25.9 dB, with significant differences between fistula type I and III and IIa and III (I: 17.7 ± 19.0 dB, IIa: 20.6 ± 15.0 dB, IIb: 32.1 ± 30.0 dB, III: 48.5 ± 37.3 dB). It was possible to conduct audiological follow-ups in 46 patients in the early-postoperative period and 45 patients in the 1 year postoperative period. There was one case in which preoperative hearing was scaled out; this was excluded from the study of postoperative changes.

Table 1. Comparison of Demographic and Clinical Characteristics

	Total	Extent Grade of Labyrinth Fistula			
		I	IIa	IIb	III
Characteristics	n = 48	n = 21	n = 14	n = 3	n = 10
Age, years					
Mean	48.6	47.2	48.1	50.2	48.8
Range	11-82	16-80	24-73	37-82	30-68
Sex, n (%)					
Male	33 (68.8)	17 (80.9)	7 (50)	1 (33.3)	8 (80.0)
Female	15 (31.3)	4 (19.0)	7 (50)	2 (66.7)	2 (20.0)
Multiple fistulas, n (%)					
Presence	11 (22.9)	1 (4.8)	2 (14.3)	1 (33.3)	7 (70.0)
Absence	37 (77.1)	20 (95.2)	12 (85.7)	2 (66.7)	3 (30.0)
Preoperative vertigo, n (%)					
Presence	21 (43.8)	8 (38.1)	5 (35.7)	2 (66.7)	6 (60.0)
Absence	27 (56.3)	13 (61.9)	9 (64.3)	1 (33.3)	4 (40.0)
Preoperative bone-conduction hearing (dB)					
Mean	25.9	17.8	20.6	32.1	48.5
Range	0-105	5.0-92.5	0-51.3	1.3-61.3	0-105

The extent grade of the labyrinth fistula is classified according to the classification reported by Dornhoffer et al.[4] Hearing level expressed as median (± SD) dB.

Postoperative hearing changes are shown in Table 2. The mean early postoperative hearing was 29.3 dB, and the mean 1 year postoperative hearing was 32.0 dB, without significant change. As for the types, we assessed the postoperative hearing change separately for IIa and below and IIb and above, as there have been previous reports[6] on hearing deterioration in IIb and above fistulas. The results showed that hearing was worse preoperatively (I and IIa: 18.9 dB, IIb and III: 44.7 dB), early postoperatively (I and IIa: 20.3 dB, IIb and III: 58.2 dB), and 1 year postoperatively (I and IIa: 22.5 dB, IIb and III: 65.5 dB). The difference between hearing 1 year postoperatively and early postoperatively was insignificant (I and IIa: 2.2 dB, IIb and III: 7.2 dB); however, the hearing of types III and above was significantly worse compared with types IIb and below (I, IIa, and IIb: 1.9 dB, III: 10.9 dB) (Figure 1). There was no preoperative difference in the presence or absence of multiple fistulas (single: 24.7 dB, multiple: 30.0 dB). However, the hearing was significantly worse in the early postoperative period (single: 24.2 dB, multiple: 48.0 dB) and 1 year postoperatively (single: 24.9 dB, multiple: 57.0 dB). The difference between 1 year postoperatively and early postoperatively was also significantly poor (single: 1.3 dB, multiple: 11.3 dB). There was no preoperative difference in the presence or absence of vertigo (absence: 21.1 dB, presence: 32.1 dB). However, the hearing was significantly worse in the early postoperative period (absence: 19.0 dB, presence: 42.8 dB) and 1 year postoperatively (absence: 20.9 dB, presence: 47.3 dB). The difference between 1 year postoperatively and early postoperatively was not significantly worse (absence: 1.9 dB, presence: 5.3 dB). Regarding frequency, the

MAIN POINTS

- The relationship between labyrinthine fistulas and hearing loss is unclear.
- We examined perioperative hearing changes in middle ear cholesteatoma with labyrinthine fistulas.
- Preoperative bone-conduction hearing was significantly worse in patients with type IIb invasion or greater, multiple fistulas, or vertigo.
- One-year postoperative hearing was significantly worse than early postoperative hearing at 500 and 2000 Hz in patients with type III invasion or multiple fistulas.
- The possibility of delayed postoperative hearing should be explained to patients undergoing surgery.

Table 2. Hearing Transition with Extent Grade of Labyrinth Fistula, Multiple Fistulas, and Vertigo

Bone-conduction hearing	Total	Extent Grade of Labyrinth Fistula		Fistula		Vertigo	
		I, IIa	IIb, III	Single	Multiple	Absence	Presence
Pre (dB)	25.9 ± 25.6, n = 48	18.9 ± 17.3, n = 35	44.7 ± 35.3*, n = 13	24.7 ± 24.0, n = 37	30.0 ± 32.4, n = 11	21.1 ± 18.0, n = 27	32.1 ± 32.9, n = 21
Early (dB)	29.3 ± 30.3, n = 46	20.3 ± 19.7, n = 35	58.2 ± 40.1*, n = 11	24.2 ± 26.0, n = 36	48.0 ± 38.5*, n = 10	19.0 ± 17.6, n = 26	42.8 ± 37.8*, n = 20
Early minus pre (dB)	5.2 ± 18.4, n = 44	1.4 ± 9.3, n = 34	20.0 ± 34.0*, n = 11	0.9 ± 7.2, n = 35	21.9 ± 34.9*, n = 9	−0.1 ± 7.8, n = 26	12.7 ± 25.9*, n = 18
Post 1 y (dB)	32.0 ± 33.2, n = 45	22.5 ± 21.5, n = 35	65.5 ± 45.3*, n = 10	24.9 ± 27.3, n = 35	57.0 ± 41.2*, n = 10	20.9 ± 18.3, n = 26	47.3 ± 42.6*, n = 19
Post 1 y minus early (dB)	3.2 ± 8.4, n = 42	2.2 ± 6.4, n = 35	7.2 ± 14.0, n = 7	1.3 ± 5.0, n = 34	11.3 ± 14.5*, n = 8	1.9 ± 4.7, n = 26	5.3 ± 12.3, n = 16

The extent of the labyrinth fistula was classified according to the classification reported by Dornhoffer et al. Hearing level expressed as median (± SD) dB.
Pre (dB), preoperative bone-conduction hearing; Early (dB), early postoperative (3-6 months) bone-conduction hearing; Post 1 y (dB), 1 year postoperative bone-conduction hearing.
* *P*-value < .05 is considered statistically significant.

postoperative hearing was significantly worse at all frequencies. One-year postoperative hearing was significantly worse than early postoperative hearing at 500 and 2000 Hz (250 Hz: pre 23.0 dB, early 31.8 dB, 1 year 34.1 dB; 500 Hz: pre 24.2 dB, early 32.3 dB, 1 year 35.5 dB; 1000 Hz: pre 22.9 dB, early 30.5 dB, 1 year 33.0 dB; 2000 Hz: pre 31.8 dB, early 38.0 dB, 1 year 40.0 dB; 3000 Hz: pre 33.6 dB, early 39.9 dB, 1 year 41.1 dB; 4000 Hz: pre 31.8 dB, early 39.5 dB, 1 year 40.8 dB).

DISCUSSION

Labyrinthine fistulas increase the risk of developing sensorineural hearing loss. However, the clinical features vary, and there are no established trends in preoperative and postoperative hearing loss due to labyrinthine fistulas.

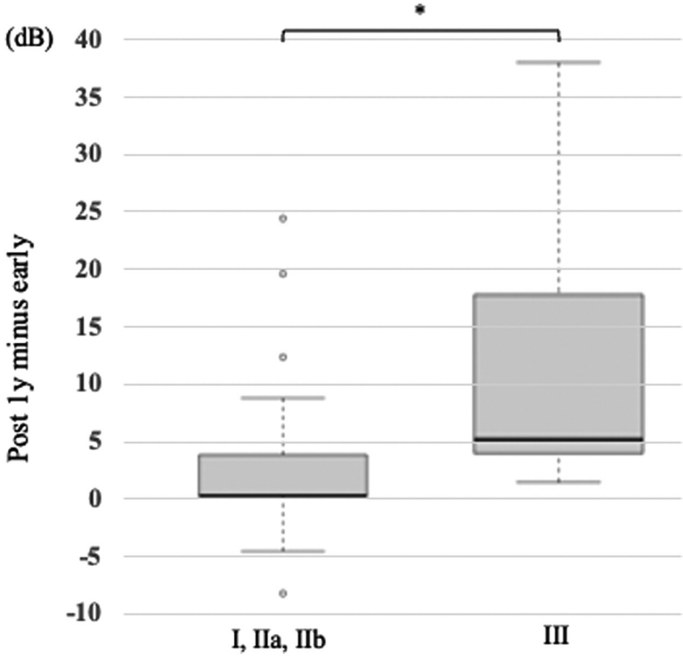

Figure 1. Differences between early postoperative (3-6 months) and 1 year postoperative bone-conduction hearing. The difference between early postoperative and 1 year postoperative bone-conduction hearing in type III was significantly larger than in type IIb or below. **P*-value < .05 is considered statistically significant.

To summarize our study's significant results, the preoperative BC hearing was significantly worse in patients with type IIb invasion or greater. The outcome of patients with type IIb or deeper fistulas, multiple fistulas, or vertigo deteriorated postoperatively. Moreover, the outcome of patients with type III or multiple fistulas deteriorated further from the early postoperative period to 1 year postoperatively. One-year postoperative hearing was significantly worse than early postoperative hearing at 500 and 2000 Hz, respectively.

Our results showed a similar percentage of labyrinthine and lateral semicircular canal fistulas compared to previous reports.[1,2] Some reports have indicated no significant difference in hearing between the types of fistulas[6,7]; however, our results revealed a significant difference in preoperative BC hearing between fistula types I and III and types IIa and III. Previous reports have demonstrated that labyrinthine fistulas of IIb or deeper are prone to irreversible sensorineural hearing loss due to ion balance disruption in the labyrinth caused by extension into the labyrinth[8]; therefore, the present results of poor hearing in patients with IIb or deeper invasion are not surprising. However, there have been cases where hearing was preserved, suggesting that factors such as the site and depth should be considered. In the present study, as in other reports,[9] the lateral semicircular canal was involved in many cases, making it difficult to assess each site separately. Even if the number of cases is large, evaluating cases with multiple fistulas is difficult because we cannot determine which fistula is the main cause.

Many reports[4,6,7] on postoperative hearing evaluation consider only 1 postoperative result. Katsura et al[10] evaluated BC levels in the operated ear 1 and 5 years after surgery. They reported that BC at 1 and 2 kHz was significantly worse at 5 years compared to 1 year after surgery, while BC hearing at 0.5 and 4 kHz was not significantly worse at 5 years compared to 1 year after surgery. This study is similar to our report but with 14 fewer cases. Although the observation period is long, various factors come into play within those 5 years. We believe that the examination period of our study is appropriate when focusing on the effects of fistulas. Although there is concern that the surgical treatment may influence the results, we performed the same technique on the included patients and confirmed the closure of the fistula in all cases.

The study has some limitations. First, the cases do not show a normal distribution and have a large variability. Therefore, the results only apply to the present study in some cases. Second, although the overall number of cases is not small compared to similar studies of labyrinthine fistulas, the sample size is still small, especially for IIb cases in which the cholesteatoma matrix extends to the perilymphatic space and extra-lymphatic drainage occurs; however, the membranous labyrinth is preserved, and this is not easy to determine correctly during surgery. Therefore, we should be aware that BC hearing can worsen with a delay in type II or deeper cases. Further studies on perioperative hearing should be conducted in more cases.

In conclusion, this is a valuable report of delayed hearing loss after surgery in patients with labyrinthine fistulas. It is essential to confirm the depth of the labyrinthine fistula and the presence of multiple fistulas to predict the transition of postoperative hearing. If patients present with these findings even in the early postoperative period when hearing is good, the likelihood of late deterioration should be noted, and this should be explained to the patient undergoing surgery.

Ethics Committee Approval: This study was approved by the Ethics Committee of Jikei University, Tokyo, Japan (Approval Number: 32-205 10286; Date: September 14, 2020).

Informed Consent: Informed consent was obtained from the patients who agreed to take part in the study.

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