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Original Article

Correlation between Surgical Outcome and Stage of Acquired Middle Ear Cholesteatoma: Revalidation of the EAONO/JOS Staging System

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OBJECTIVES: This study aimed to evaluate the intraoperative findings, recurrence rate, and hearing outcome of cholesteatoma surgery and correlate them with the newly proposed EAONO/JOS Joint Consensus Statement.

MATERIALS and METHODS: The records of 407 patients diagnosed with chronic otitis media and cholesteatoma between 2009 and 2017 were reviewed. After the exclusion of records with unsatisfactory surgical notes and anamnesis, 353 patients were included in the study. The 290 patients who had undergone primary surgery and 63 who had undergone revision surgery were evaluated separately.

RESULTS: Total 162 of 290 (56%) patients had retraction pocket cholesteatoma and 128 of 290 (44%) patients had non-retraction pocket cholesteatoma. Eighty (28%) patients had stage I, 114 (39%) had stage II, 91 (31%) had stage III, and 5 (2%) had stage VI disease. The recurrence rate was 6.9% (20/290). The average age of these patients at the time of the second operation was 23.31±10.3 years. Twelve patients had (60%) recurrent cholesteatoma, and eight (40%) had residual cholesteatoma. Hearing outcome and surgical technique were significantly associated with the disease stage; however, the recurrence rate showed no such association.

CONCLUSION: We concluded that the EAONO/JOS staging system is beneficial for estimating the postoperative hearing results and planning the surgical technique. However, there was no significant relationship between the recurrence rate and the EAONO/JOS staging system. We believe that additional factors, such as infection, ossicles, and invasion, predict the recurrence. Widespread use of the EAONO/JOS staging system will enable better evaluation of surgical outcomes and prognosis.

KEYWORDS: Cholesteatoma, hearing, classification, staging

INTRODUCTION

Cholesteatoma is a cystic lesion formed from keratinizing squamous epithelium in the temporal bone. It is a local invasive disease that causes chronic infection, hearing loss, and life-threating complications. Surgical treatment is the only treatment option. The primary objective of surgical treatment is to first remove the cholesteatoma completely and to create a dry, safe, and disease-free ear and then restore hearing. Recurrence rate and residual disease are the most important problems in cholesteatoma surgery. The critical decision during the operation is to find a acceptable balance on teeterboard between favourable hearing result and recurrence rate. Based on the surgical technique, up to 25% subjects exhibit residual disease [1]. Thus, before the operation, the surgeon should gather all the available evidence and speak to the patient about the expected outcome and risks. Microscopic examination and radiologic evaluation of the patient are beneficial. Another helpful tool is the staging system. These systems have been developed by international experts, as per their long-time experiences. These systems are not only important for estimating the outcome, but also play a role in sharing and comparing patient information. Several staging systems have been proposed for this purpose. Tos, Lien, Sanna are well-known researchers on this subject [2-5]. Further, institutional committees have proposed systems for cholesteatoma staging [6-8].

This study was presented at the The 9th EAONO Instructional Workshop and Consensus on Auditory Implants, 18-23 June 2018, Copenhagen, Denmark.

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The recent publication on this subject is the European Academy of Otology and Neurotology/Japan Otological Society (EAONO/JOS) Joint Consensus Statement about middle ear cholesteatoma classification [9]. James et al. studied the validity of this system with prospective data from nine centers around the world. They found that the inter- and intra-rater reliability were high [10]. They also recorded that the recurrence rate rises with increase in the disease stage; however, the increase was not statistically significant. They mentioned the difficulties experienced while conducting multicenter retrospective studies and proposed prospective data acquisition. Further, they did not mention about the hearing outcome. Fukuda et al. focused on hearing in 34 pars flaccida cholesteatoma patients and found a significant correlation between the outcome and the EAONO/JOS stage [11]. Several studies have described cholesteatoma definitions and classifications. There are different strategies for classification, such as "presumed etiology and pathophysiology", "pathophysiology, location, ossicular defects, and presence of complications", "extension of the disease", "location of origin on the tympanic membrane, "direction of extension of disease", "origin and location of disease", "extent of involvement", and "inflammation status". However, few clinical trials have evaluated the evidence level or compared them with the other systems [12]. We planned a single-center retrospective study to overcome the linguistic discrepancies of data among different clinics. This study aimed to evaluate the intraoperative findings, recurrence rate, and hearing outcome of cholesteatoma surgery and correlate them with the new proposed EAONO/JOS Joint Consensus Statement.

MATERIALS AND METHODS

The records of patients diagnosed with chronic otitis media and cholesteatoma between 2009 and 2017 were reviewed. Four hundred and seven patients with pathologically proven cholesteatoma were included. Fifty-three patients with unsatisfactory anamnesis and surgical notes were excluded. Total 353 patients were enrolled. The 290 patients who had undergone primary surgery and 63 who had undergone revision surgery were evaluated separately. Ethical approval was obtained from the University Ethics Committee (60116787-020/20944).

All the patients were classified as per the classification and staging system proposed by the EAONO/JOS Joint Committee (Tables 1, 2). STAM system divides middle ear into 6 parts as; the tympanic cavity (T), the attic (A) and the mastoid (M), the difficult access sites (S) includes S1, the supratubal recess (also called the anterior epitympanum or protympanum) and S2, the sinus tympani[9].

Hearing outcomes were reported according to the Committee on Hearing and Equilibrium of the American Academy of Otolaryngology- Head, and Neck surgery guidelines. Pure tone average air-bone gap (PTA-ABG) of four frequencies (0.5, 1, 2, and 3 kHz) at least one year after the surgery was used [13]. The patients who did not undergo follow-up audiometry in our hospital or did not complete the one-year follow-up were not included in the comparison. We set two levels of success as PTA-ABG ≤ 10 and ≤ 20 dB.

Surgical procedures [Transcanal atticotomy (TCA), canal wall-up mastoidectomy (CWU), canal wall-down mastoidectomy (CWD), and revision mastoidectomy (RM)], graft materials (facia, cartilage, and both), ossiculoplasty techniques (incus, PORP, TORP, and cortical bone), bone erosions, and dehiscence were noted in detail.

The patients who had undergone revision surgery were evaluated separately. If the patients had the first surgery in our department, we compared the surgical records and classified them as having residual or recurrent cholesteatoma.

Table 1. The EAONO/JOS classification for acquired middle air cholesteatoma

The EAONO/JOS classification for acquired middle ear cholesteatoma
1. Retraction pocket cholesteatoma
a) Pars flaccida (attic) cholesteatoma
b) Pars tensa cholesteatoma
c) Combination of pars flaccida and pars tensa cholesteatoma
2. Non-retraction pocket cholesteatoma
a) Cholesteatoma secondary to tympanic membrane perforation
b) Cholesteatoma following trauma/otologic procedures

Table 2. The EAONO/JOS staging system for acquired middle air cholesteatoma

The EAONO/JOS staging system for acquired middle ear cholesteatoma
1Stage I: Cholesteatoma localized in the primary site
Stage II: Cholesteatoma involving two or more sites
Stage III: Cholesteatoma with extracranial complications or pathologic conditions including:
Facial palsy
Labyrinthine fistula with conditions at risk of membranous labyrinth
Labyrinthitis
Postauricular abscess or fistula
Zygomatic abscess
Neck abscess
Canal wall destruction more than half the length of the bony ear canal
Destruction of the tegmen with a defect that requires surgical
Adhesive otitis; total adhesion of the pars tensa
Stage IV: Cholesteatoma with intracranial complications including
Purulent meningitis
Epidural abscess
Subdural abscess
Brain abscess
Sinus thrombosis
Brain herniation into the mastoid cavity

MAIN POINTS

- EAONO/JOS staging system is beneficial in estimating the postoperative hearing results.
- There is no significant relationship between the recurrence rate and EAONO/JOS staging system.
- If classification and staging are used together, it will be a more effective prediction tool(Ex: stage I and pars flaccida RPC).

Statistical Analysis

All statistical analyses were performed using the Statistical Package for the Social Sciences (SPSS) version 25 for Windows (IBM Corp.; Armonk, NY, USA). Continuous variables are presented as mean±-standard deviation, minimum, and maximum values. Categorical variables are presented as numbers and percentages. We used the Pearson Chi-Square test and Fisher's Exact Test for comparing the groups; $p<0.05$ were considered statistically significant.

RESULTS

Patient Population

The number of patients who had undergone primary surgery in our department was 290. One hundred seventy-three (60%) patients were men, and 117 patients (40%) were women. The mean age of the patients was 35.53±17.1 (3-77) years. The mean follow-up duration was 4.14±2.54 years.

Total 63 patients had undergone a revision mastoidectomy for cholesteatoma between 2009 and 2017. The mean age of these patients at the time of the second operation was 23.31±10.3 (10-64) years and the average time between the two operations was 27.3±20.3 months.

Cholesteatoma Classification and Staging

Total 162 of 290 (56%) patients had retraction pocket cholesteatoma (RPC), and 128 (44%) of 290 patients had non-retraction pocket cholesteatoma (non-RPC). We found that 80 (28%) patients had stage I, 114 (39%) had stage II, 91 (31%) had stage III, and 5 (2%) had stage VI dis-

Table 3. The number of primary cases according to the EAONO/JOS classification and staging systems

The number of patients according to cholesteatoma classification and stage	Total	Stage I	Stage II	Stage III	Stage IV
Retraction Pocket Cholesteatoma					
Pars Flaccida	69	22 (32%)	29 (42%)	17 (25%)	1 (1%)
Pars tensa	37	8 (22%)	16 (43%)	12 (32%)	1 (3%)
Combination	56	-	15 (27%)	39 (70%)	2 (3%)
Non- Retraction Pocket Cholesteatoma					
Secondary to perforation	123	49 (40%)	51 (41%)	22 (18%)	1 (1%)
Following trauma/otologic procedures	5	1 (20%)	3 (60%)	1 (20%)	0 (0%)
Total	290	80 (28%)	114 (39%)	91 (31%)	5 (2%)

Table 4. Surgical technique, recurrence rate, and hearing outcomes after one year according to the stage of the cholesteatoma were shown. The type of the surgical technique was ignored while calculating hearing outcomes

	Stage I	Stage II	Stage III	Stage IV
Surgical technique				
TCA	13 (16%)	2 (2%)	0 (0%)	0 (0%)
CWU	45 (56%)	32 (28%)	11 (12%)	0 (0%)
CWD	22 (28%)	80 (70%)	80 (88%)	5 (100%)
Residual and recurrent cholesteatoma				
Residual	2 (2.5%)	1 (0.8%)	5 (5.5%)	0 (0%)
Recurrent	1 (1.2%)	6 (5.2%)	5 (5.5%)	0 (0%)
Total	3 (3.7%)	7 (6.0%)	10 (11.0%)	0 (0%)
Bone erosions and dehiscence over the critical areas				
Facial Nerve	5 (6%)	26 (23%)	40 (44%)	3 (60%)
Dural plate	1 (1%)	7 (6%)	13 (14%)	4 (80%)
LSC	0 (0%)	0 (0%)	23 (25%)	2 (40%)
Carotid artery	0 (0%)	0 (0%)	2 (2%)	0 (0%)
Jugular vein	0 (0%)	0 (0%)	3 (3%)	3 (60%)
Hearing outcome				
Air-bone gap ≤ 10 dB	16 (24%)	6 (7%)	7 (9%)	0 (0%)
Air-bone gap ≤ 20 dB	39 (58%)	35 (40%)	20 (27%)	0 (0%)
Air-bone gap ≤ 30 dB	62 (92%)	52 (60%)	38 (51%)	1 (50%)

TCA: transcanal atticotomy; CWU: canal wall-up; CWD: canal wall-down; LSC: Lateral Semicircular canal

Table 5. Surgical technique, recurrence rate, and hearing outcomes after one year according to the classification of the cholesteatoma. The type of surgical technique was ignored while calculating the hearing outcomes

	Pars Flaccida RPC	Pars Tense RPC	Combined RPC	Secondary to perforation	Following trauma/ otologic procedures
Surgical technique					
TCA	8 (12%)	2 (5%)	0 (0%)	5 (4%)	0 (0%)
CWU	15 (21%)	13 (35%)	11 (20%)	46 (37%)	3 (60%)
CWD	46 (66%)	22 (60%)	45 (80%)	72 (59%)	2 (40%)
Residual and recurrent cholesteatoma					
Residual	3 (4.3%)	1 (2.7%)	2 (3.6%)	2 (1.6%)	0 (0%)
Recurrent	1 (1.4%)	2 (5.4%)	3 (5.4%)	6 (4.9%)	0 (0%)
Total	4 (5.7%)	3 (8.1%)	5 (9%)	8 (6.5%)	0 (0%)
Bone erosions and dehiscence over the critical areas					
Facial Nerve	15 (22%)	6 (16%)	25 (45%)	27 (22%)	1 (20%)
Dural plate	3 (4%)	4 (10%)	11 (20%)	7 (6%)	0 (0%)
LSC	6 (9%)	2 (5%)	13 (23%)	4 (3%)	0 (0%)
Carotid artery	0 (0%)	0 (0%)	2 (3.5%)	0 (0%)	0 (0%)
Jugular vein	1 (1.5%)	1 (3%)	3 (5%)	3 (2%)	0 (0%)
Hearing outcome					
Air-bone gap ≤ 10 dB	10 (19%)	3 (11%)	3 (7%)	13 (12%)	-
Air-bone gap ≤ 20 dB	29 (55%)	9 (33%)	10 (24%)	46 (42%)	-
Air-bone gap ≤ 30 dB	39 (74%)	15 (55%)	21 (50%)	69 (69%)	-

TCA: transcanal atticotomy; CWU: canal wall-up; CWD: canal wall-down; Retraction Pocket Cholesteatoma; LSC: Lateral Semicircular canal

ease. The detailed classification and staging of the patients are shown in Table 3. In stage IV patients, we diagnosed two epidural abscesses, one subdural abscess, one sinus thrombosis, and one brain herniation.

Hearing Outcome

The hearing results were compared with pure tone audiograms that were taken at least one year after the surgery. The patients who did not undergo follow-up audiometry in our hospital or did not complete one year of follow-up were excluded. The PTA-ABGs of 231 patients were calculated (Tables 4, 5). The best hearing results were achieved in patients with pars flaccida RPC (PTA-ABG ≤ 20 dB: 55%). In all the groups, the hearing results were worse for higher cholesteatoma stage (p=0.001). There was a significant relationship between the classification and hearing outcome (p=0.01). The detailed analysis of hearing outcomes according to the stage and classification together is shown in Table 6.

Surgical Technique

More than 50% of the patients had CWD surgery regardless of their stage. However, with increase in the disease stage, the ratio of CWD surgery was increasing (p<0.001). There was also a significant relation between the surgical technique and classification (p=0.03). The detailed analysis of surgery is shown in Tables 4, 5. More stage I patients underwent CWU surgery.

Reconstruction of the tympanic membrane was performed in 216 (74%) patients. Temporal facia (49%), tragal cartilage (5%), and both (46%) were used as graft materials. Ossicular reconstruction was performed for 135 (47%) patients. The cortical bone (42%), incus (26%), PORP (19%), and TORP (13%) were preferred for reconstruction.

Bone Erosions and Dehiscence over the Critical Areas

We recorded the erosions and dehiscence over critical areas (Tables 4, 5). In particular, the combined RBC group had a high rate of erosions.

Analysis of Cholesteatoma Recurrence

Total 63 patients underwent revision mastoidectomy for cholesteatoma. Only 20 of 63 patients had their first operations in our clinic; therefore, we could not evaluate the majority of recurrences if they had residue or recurrent disease. The recurrence rate was 6.9% (20/290). The average age of these patients at the time of the second operation was 23.31±10.3 years. Twelve patients had (60%) recurrent cholesteatoma, and eight (40%) had residual cholesteatoma. The mean revision time was 14.8 months for residual cholesteatoma and 41.6 months for recurrent cholesteatoma. The classification and staging of recurrences is shown in Tables 4, 5. There was no correlation between the stage or classification of the cholesteatoma and the recurrence rate (p>0.05).

DISCUSSION

Cholesteatoma is histologically benign but locally destructive; it can cause result in very different clinical pictures, changing from a limited lesion without hearing loss to a life-threating intracranial abscess. The spread of cholesteatoma and complications are effective in surgical

Table 6. Detailed hearing outcome as per the classification and staging

		Air-bone gap (dB)		
	n of patients	0-10	0-20	0-30
Pars flaccida RPC				
Stage I	19	6 (32%)	14 (74%)	17 (89%)
Stage II	19	2 (11%)	9 (47%)	13 (68%)
Stage III	14	2 (14%)	6 (42%)	9 (64%)
Stage IV	1	0 (0%)	0 (0%)	0 (0%)
Subtotal	53	10 (19%)	29 (55%)	39 (74%)
Pars Tensa RPC				
Stage I	6	2 (33%)	3 (50%)	4 (66%)
Stage II	11	0 (0%)	4 (36%)	5 (45%)
Stage III	10	1 (10%)	2 (20%)	6 (60%)
Subtotal	27	3 (11%)	9 (33%)	15 (55%)
Combined RPC				
Stage II	13	1 (8%)	4 (31%)	9 (69%)
Stage III	29	2 (7%)	6 (21%)	12 (41%)
Subtotal	42	3 (7%)	10 (24%)	21 (50%)
Secondary to perforation				
Stage I	42	8 (19%)	22 (52%)	32 (71%)
Stage II	45	3 (7%)	18 (40%)	25 (55%)
Stage III	21	2 (9%)	6 (28%)	11 (52%)
Stage IV	1	0 (0%)	0 (0%)	1 (100%)
Subtotal	109	13 (12%)	46 (42%)	69 (63%)
Grand Total*	231	29 (13%)	94 (41%)	144 (62%)

RPC: retraction pocket cholesteatoma

*Grand Total: The n of patients who had follow-up audiometry in our hospital one year after the surgery.

planning, hearing outcome, and operational risks. We used our retrospective data to revalidate the EAONO/JOS classification and staging systems in order to investigate whether they correlated with the surgical planning, operational findings, and outcome. We found that 56% of the cholesteatoma cases in our series originated from retraction pockets. Pars flaccida was the most frequent side (43%) followed by combined (34%) and pars tensa (23%). Most patients were classified as having stage II disease (39%), followed by those with stage III disease (31%), and those with stage I disease (28%). The hearing outcome worsened significantly and the need for CWD surgery increased significantly as the disease stage progressed. Although the recurrence rate was rising with the increasing stage, it was not statistically significant.

Matsuda et al. evaluated only RPC according to the JOS system [14]. They found that with the increasing stage, the hearing outcome, and recurrence rates worsened significantly. Moreover, in the EAONO/JOS system, the hearing outcome worsened and the recurrence rate increased with an increase in the stage of cholesteatoma in our series; however, we did not find any significant relation with the recurrence rate. Non-RPC cholesteatoma secondary to tympanic perforation was found in 42% of the patients, and non-RPC following trauma/otologic procedures were found in only 2% of the patients.

To our knowledge, only one study has evaluated the correlation between the EAONO/JOS staging system and hearing outcome. Fukuda et al. evaluated 34 patients with pars flaccida RPC and found that in the early stages, the hearing outcome was better. They also concluded that the EAONO/JOS staging system could be used as a prognostic indicator for hearing outcomes [11]. Our findings support these findings. We found a strong relationship between the stage and the hearing outcome. We also recognized that hearing outcome was best in the pars flaccida group, while it was worst in the combined group. Therefore, combined use of the staging and classification would enable more accurate estimation of the hearing outcome.

There are several surgical techniques used for cholesteatoma treatment. Different surgical approaches are preferred by different surgeons, as per the cholesteatoma stage, expected hearing outcome, and patient compliance. We found that the disease stage had a stronger association with the choice of surgical technique than the classification. The need for CWD surgery was higher in those with higher disease stage. This relationship was also reported by Matsuda et al. [14].

One of the important problems in cholesteatoma surgery is recurrence. It has been believed that the technique is closely related to the recurrence rate. In a large review study, Kelly et al. concluded that CWU is mostly related to residual disease, while CWD is related to recurrent disease [15]. In our series, we found that the prevalence of residual and recurrent diseases progressively increased with the increase in the disease stage. The ratio of recurrences in stages I, II, and III was 3%, 6%, and 11%, respectively; however, this increase was not statistically significant. While our results are in agreement with those of James et al., Matsuda et al. found a statistically significant difference between pars flaccida and pars tensa cholesteatoma [10, 14]. They also reported a significantly higher recurrence rate in stage II and III than in stage I patients [14]. When we considered the classification of cholesteatoma, the highest recurrence rate was found in the combined RPC group (9%), followed by that in those with pars tensa RPC (8.1%), secondary to perforation (6.5%) and pars flaccida RPC (5.7%). These results suggest that recurrence is more closely related to the stage of the cholesteatoma than the surgical technique.

Cholesteatoma can destroy the bony cover of the critical areas, such as the facial nerve and dural plate. It was reported that the intraoperative facial dehiscence ratio changed from 11%-33%, and the labyrinth fistula ratio changed from 3%-13% [16-18]. In our series, we found that 25% patients had facial nerve dehiscence, 9% had labyrinth fistula, and 9% had dural plate destruction. The combined RPC group was prone to complications more than the other groups.

One of the main limitations of the study is the retrospective study design because it reduces the variety of the available data. Prospective clinical trials that compare different systems will provide a better understanding of the usefulness.

CONCLUSION

Cholesteatoma is a very important locally destructive clinical situation. Treatment success is closely related to the surgical technique,

localization of the disease, and invaded part of the temporal bones. Therefore, detailed diagnostic investigation and provision of informing to the patient about the expectations are crucial before the operation. We found that postoperative hearing results were worse when the disease was diagnosed in more advanced stages. Total 82% of the patients with stage I had favorable (< 20 dB) hearing outcomes. Dehiscence on the critical areas was more frequent in combined RPC. CWD was the main type of operation preferred in patients with disease stage II or more. We concluded that EAONO/JOS staging system is beneficial in estimating the postoperative hearing results and planning the surgical technique. However, there is no significant relationship between the recurrence rate and the EAONO/JOS staging system. Additional factors are involved in the prediction of recurrence, such as infection, ossicles, and invasion. The combined use of classification and staging will be a more effective prediction tool (e.g., stage I and pars flaccida RPC). Widespread use of the EAONO/JOS staging system will help us perform better evaluation of the surgical outcomes and prognosis.

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