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Original Article

Clinical Characteristics of Pars Tensa Cholesteatoma: A Comparative Study of Area-Based Classification Systems Proposed by the Japanese Otological Society and the European Academy of Otology & Neuro-Otology

Masaomi Motegi, Yutaka Yamamoto, Takeshi Tada, Masahiro Takahashi, Sayaka Sampei, Hiromi Sano, Tsunetaro Morino, Manabu Komori, Masahiro Miura, Kazuhisa Yamamoto, Yuichiro Yaguchi, Yuika Sakurai, Hiromi Kojima

Department of Otorhinolaryngology, Jikei University School of Medicine, Tokyo, Japan (MM, YY, TT, MT, SS, HS, TM, MK, M.Miura, KY, YS, HK)
Department of Otorhinolaryngology, St. Marianna University School of Medicine, Kanagawa, Japan (Y.Yaguchi)

ORCID IDs of the authors: M.M. 0000-0003-0947-6526; Y.Y. 0000-0002-4577-1234; T.T. 0000-0003-3774-6239; M.T.0000-0002-7286-2326; S.S. 0000-0002-7305-3205; H.S. 0000-0002-5145-2942; T.M. 0000-0003-1299-8659; M.K. 0000-0001-8223-9186; M.Miura 0000-0003-3708-1089; K.Y. 0000-0001-6509-929X; Y.Yaguchi 0000-0002-6313-0780; Y.S. 0000-0002-9768-3850; H.K. 0000-0003-2967-2110.

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OBJECTIVES: To assess the clinical characteristics of extent patterns in pars tensa cholesteatoma.

MATERIALS and METHODS: This was a retrospective chart review. Forty-four patients with pars tensa cholesteatoma who underwent primary surgery at a tertiary academic medical center were included. The main outcomes measured were sex, age, clinical background, and stage classification of pars tensa cholesteatoma (including the extent of cholesteatoma and involvement of the sinus tympani) according to two staging classifications: criteria advocated by the Japanese Otological Society (JOS) and those advocated by the European Academy of Otology and Neuro-Otology (EAONO)/JOS joint consensus statements.

RESULTS: The mean patient age ± standard deviation was 38.4±19.6 years. The patients comprised 19 men and 25 women. According to the JOS classification, 18 ears (40.9%) were classified as stage I, 22 (50.0%) as stage II, and 4 (9.1%) as stage III. According to the EAONO/JOS joint consensus statements, 14 ears (31.8%) were classified as stage I, 26 (59.1%) as stage II, and 4 (9.1%) as stage III. Fourteen ears (31.8%) demonstrated involvement of the sinus tympani. Four ears (9.1%) that were originally categorized as stage I cholesteatoma by the JOS criteria showed sinus tympani invasion and were subsequently categorized as stage II according to the EAONO/JOS criteria.

CONCLUSION: We determined the clinical characteristics of pars tensa cholesteatoma based on the novel and well-defined classification criteria. Further studies including long-term outcomes are necessary to demonstrate the clinical relevance of the discrepancy between the two criteria with respect to involvement of the sinus tympani.

KEYWORDS: Cholesteatoma, classification, sinus tympani, etiology, staging

INTRODUCTION

Acquired cholesteatoma has a varying pathophysiology highlighted by clinically different characteristics. Surgery for cholesteatoma remains challenging because it requires complete exenteration and hearing improvement regardless of the varying extent of pathophysiology. Thus, surgical findings have been revised over time, and many surgeons consider these findings to correlate with postoperative results, including recidivism, hearing improvement, and surgical complications [1-5]. However, the extent to which middle ear cholesteatoma is linked with postoperative outcomes remains unclear.

Manifestations of acquired cholesteatoma have been classified into three types: pars tensa cholesteatoma, pars flaccida cholesteatoma, and cholesteatoma secondary to tympanic perforation. Because of only a few reports [1-3, 6] clarifying the definition of pars ten-

Corresponding Address: Masaomi Motegi E-mail: m_motegi@hotmail.co.jp

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sa cholesteatoma, it is unclear whether the grade of extent, including invasion of the tympanic sinus, affects postoperative results. Recently, the Japanese Otological Society (JOS) [7] and European Academy of Otology and Neuro-Otology (EAONO) [8-10] proposed classification criteria for middle ear cholesteatoma. For the first time, both groups clearly defined each type of cholesteatoma and firmly categorized the range of extent areas depending on each type of acquired cholesteatoma. Both criteria are similar in their classification approaches, which depend on the number of sub-sites of the middle ear cavity involved; however, there the involvement of the sinus tympani is disputable.

Here we present evidence regarding the extent of pars tensa cholesteatoma based on these novel staging criteria. The objective of the current study was to assess the clinical characteristics of pars tensa cholesteatoma as a preliminary step before evaluating how the preoperative severity of disease extension correlates with the postoperative prognosis.

MATERIALS AND METHODS

We retrospectively reviewed the medical records, including surgery videos, of 292 patients who underwent primary surgery for cholesteatoma at a tertiary academic medical center between January 2009 and December 2015. A total of 292 ears of patients with cholesteatoma comprising 183 pars flaccida (62.7%), 55 congenital (18.8%), 44 pars tensa (15.1%), 8 secondary to tympanic perforation (2.7%), and 2 unclassifiable (0.7%) ears were assessed. Forty-four ears from patients meeting a previously published definition of pars tensa cholesteatoma [7-10] were included. The study protocol was approved by the institutional review board of our center, with a waiver of informed consent for the retrospective medical records review (approval number: 29-208) (8824)).

In patients with pars tensa cholesteatoma, the male-to-female ratio, age distribution, clinical background, stage classification of cholesteatoma extent, and involvement of the sinus tympani were evaluated. We classified the extent of cholesteatoma according to the Classification and Staging of Cholesteatoma proposed by the JOS in 2015 [7] and used the EAONO/JOS joint consensus statements for the definitions, classification, and staging of middle ear cholesteatoma [8-10]. Based on both criteria, the extent of cholesteatoma was categorized into stages I, II, III, and IV. In stage I, the lesion is limited to the tympanic cavity; in stage II, the lesion extends beyond the tympanic cavity; in stage III, intratemporal complications and/or pathological conditions are present, including facial nerve palsy, labyrinthine fistula, total adhesion of the pars tensa, and neck abscess; and in stage IV, intracranial complications are present, including brain abscess and sinus thrombosis. Using the JOS criteria, stages II and III were further subclassified according to the extent of the epithelium within the following regions: protympanum (P), tympanic cavity (T), attic or epitympanum (A), and mastoid cavity (M). According to the EAONO/JOS criteria, the extent of cholesteatoma was subclassified into the following regions: difficult access sites (S), T, A, and M. The S sites include the supratubal recess (also termed the anterior epitympanum or protympanum) (S1) and sinus tympani (S2). Thus, there is a discrepancy between the criteria because the latter designates the sinus tympani as one sub-site, which is covered by T in the former classification. For instance, if the epithelium invades the T, A, or M without involvement of the sinus tympani or any complication, it would be classified as "Stage II TAM."

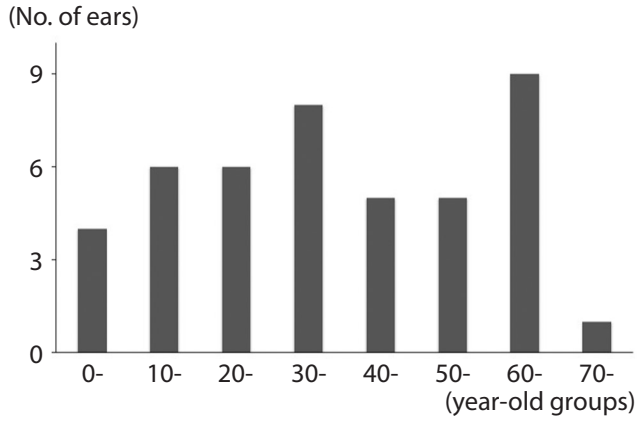

Figure 1. Age distribution of patients with pars tensa cholesteatoma. There was a wide age distribution; two peaks corresponding to patients in their 30s and 60s were observed.

RESULTS

Patient Age, Sex, and Background

Pars tensa cholesteatoma was observed in 44 ears of 44 patients, including 25 ears of 25 women and 19 ears of 19 men. The age of patients at the time of surgery was 4-72 years, with a mean ± standard deviation of 38.4±19.6 years. There was a wide distribution of age, and two peaks were observed among patients in their 30s and 60s (Figure 1). Some abnormalities were detected in the contralateral tympanic membrane of 10 ears (22.7%). Specifically, five ears (11.4%) exhibited cholesteatoma, four (9.1%) showed retraction or adhesion of the tympanic membrane, and one (2.3%) showed otitis media with effusion.

Type and Stage Classification of Pars Tensa Cholesteatoma

Eighteen ears (40.9%) were classified as stage I, 22 (50.0%) as stage II, and 4 (9.1%) as stage III according to the JOS staging criteria (Table 1). The extent patterns in ears with stage II pars tensa cholesteatoma were as follows: TA, 5; TAM, 16; and PTAM, 1. Patterns in ears with stage III pars tensa cholesteatoma were as follows: TAM, 2; PTAM, 1; and T, 1. In the four ears with stage III pars tensa cholesteatoma, three exhibited total adhesion of the pars tensa, and two of these ears exhibited a labyrinthine fistula. Conversely, 14 ears (31.8%) were classified as stage I, 26 (59.1%) as stage II, and 4 (9.1%) as stage III according to the EAONO/JOS criteria (Table 2). The patterns in ears with stage II pars tensa cholesteatoma were as follows: ST, 4; TA, 3; STA, 2; TAM, 8; and STAM, 9. The patterns in ears with stage III were: T, 1; TAM, 2; and STAM, 1. Of the four ears with stage III pars tensa cholesteatoma, three exhibited total adhesion of the pars tensa, and two of these ears exhibited a labyrinthine fistula. None of the ears were classified as stage IV pars tensa cholesteatoma based on either set of criteria.

Involvement of the Sinus Tympani

Fundamentally, both the JOS and EAONO/JOS criteria categorize the staging of the cholesteatoma based on the number of sub-sites involved. However, the difference between these criteria largely depends on the classification of the sinus tympani as one sub-site. Thus, if we classify a ear that exhibits sinus tympani involvement using the EA-

Table 1. Stage classification and extent patterns for pars tensa cholesteatoma based on JOS classification criteria (n=44)

Stages	n	Protympanum (P) (%)	Tympanum (T) (%)	Attic (A) (%)	Mastoid (M) (%)
Stage I	18	0 (0.0)	18 (100.0)	0 (0.0)	0 (0.0)
Stage II	22	1 (4.5)	22 (100.0)	22 (100.0)	17 (77.3)
Stage III	4	1 (25.0)	1 (25.0)	3 (75.0)	3 (75.0)
Stage IV	0	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)
Total	44	2 (4.5)	44 (100.0)	44 (100.0)	20 (45.5)

JOS: Japanese Otological Society

Table 2. Stage classification and extent patterns for pars tensa cholesteatoma based on EAONO/JOS consensus statements (n=44)

Stages	n	Difficult access site (S) (%)	Tympanum (T) (%)	Attic (A) (%)	Mastoid (M) (%)
Stage I	14	0 (0.0)	14 (100.0)	0 (0.0)	0 (0.0)
Stage II	26	15 (57.7)	26 (100.0)	22 (84.6)	17 (65.4)
Stage III	4	1 (25.0)	4 (25.0)	3 (75.0)	3 (75.0)
Stage IV	0	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)
Total	44	16 (36.3)	44 (100.0)	25 (56.8)	20 (45.5)

EAONO: European Academy of Otology and Neuro-Otology

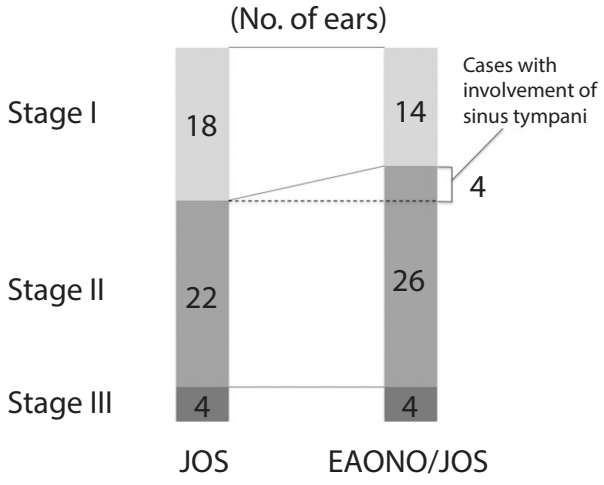

Figure 2. Difference in stage distributions of pars tensa cholesteatoma according to the JOS and EAONO/JOS criteria. Four ears (9.1%) that were classified as stage I according to JOS criteria were classified as stage II according to EAONO/JOS criteria.
JOS: Japanese Otological Society; EAONO: European Academy of Otology and Neuro-Otology

ONO/JOS criteria, we count an additional sub-site than when using the JOS criteria. There were 14 ears (31.8%) with cholesteatoma exhibiting involvement of the sinus tympani. Specifically, 4 (22.2%), 10 (45.5%), and 0 ears (0.0%) showed involvement of the sinus tympani in stages I, II, and III, respectively, according to the JOS criteria. Therefore, four ears with cholesteatoma and sinus tympani involvement were categorized as stage I according to JOS criteria; however, they were categorized as stage II when the EAONO/JOS criteria were used (Figure 2).

DISCUSSION

Middle ear cholesteatoma is empirically considered to exhibit varying clinical manifestations depending on the type of cholesteatoma. According to the JOS [7], pars tensa cholesteatoma is defined as a combination of sinus and tensa cholesteatoma. However, sinus cholesteatoma is described as a sequela of posterosuperior retraction or perforation of the pars tensa extending to the tympanic sinus and posterior portion of the tympanic membrane, and tensa cholesteatoma is described as a sequela that arises from within the whole tympanic membrane retraction or perforation, including the inferior and anterior segments, according to the EAONO [8-10]. Previous studies supported the assertion that cholesteatoma progression could influence postoperative outcomes [2, 11, 12]. With respect to pars tensa cholesteatoma, the ossicles are likely to be destroyed at an early stage [11]; therefore, hearing loss becomes more noticeable in patients with pars tensa cholesteatoma, even at early stages. Furthermore, the rate of recurrence is higher in pars tensa cholesteatoma, and the proportion of postoperative hearing improvement is lower than in other types of cholesteatomas [1]. However, minimal evidence based on analysis of the etiological correlation between the extent of cholesteatoma and prognosis has been reported [4, 6, 13]. Recent staging criteria from the JOS and EAONO/JOS criteria were advocated as the first national or international standards for middle ear cholesteatoma. To our knowledge, this is the first study to include well-defined clinical evidence that reflects more detailed and accurate pathogenesis of pars tensa cholesteatoma.

In the present study, the incidence of pars tensa cholesteatoma was found to be marginally higher among women. This result is consistent with previously reported sex ratios of pars tensa cholesteatoma [14]; in contrast, the incidence rate of all types of cholesteatoma was higher among men [15, 16]. In agreement with other reports [15, 16], the number of patients who were elderly at the time of surgery was higher in pars tensa cholesteatoma than in other types of cholesteatoma. This may be because of a higher age of onset associated with pars tensa cholesteatoma or because of the longer time course between onset and surgery due to its slow progression.

The proportion of ears classified as stage I according to the JOS and EAONO/JOS criteria at the time of surgery were 40.9% and 31.8%, respectively. In a study conducted by Matsuda et al. [17], 32.0% of ears were classified as stage I, which is similar to the percentage of ears classified as stage I in our study. Furthermore, when Matsuda et al. [17] compared the types of cholesteatomas at the time of surgery, the ears that were classified as stage I in patients with pars tensa cholesteatoma comprised a larger proportion than the ears classified as stage I in patients with pars flaccida cholesteatoma [17]. Therefore, pars tensa cholesteatoma may be more prone to remain in the primary site than pars flaccida cholesteatoma.

In our study, no correlation was observed between the extent of cholesteatoma and involvement of the sinus tympani. Previous studies reported that sinus tympani involvement in cholesteatoma was detected intraoperatively in 9.6%-37.5% of patients [3, 13]. To our knowledge, the present study provides the first evidence for the involvement of the sinus tympani in pars tensa cholesteatoma (31.8%). Particularly, there is a difference between the JOS and EAONO/JOS criteria when classifying the staging in pars tensa cholesteatoma. According to the EAONO/JOS criteria, ears are classified as higher than stage II when the pathology of pars tensa cholesteatoma involves the sinus tympani. In the present study, four ears (9.1%) that were classified as stage I according to the JOS criteria were categorized as stage II when using the EAONO/JOS criteria (Figure 2). Future studies evaluating postoperative outcomes should exemplify the importance of this discrepancy.

Here we provided evidence regarding the clinical characteristics of pars tensa cholesteatoma based on new and well-established classification criteria. Thus, our data, including detailed characteristics of the extent patterns in pars tensa cholesteatoma, may contribute to a surgical strategy tailored according to the degree of extension. Recently, microscopic surgery with endoscopic assistance and totally endoscopic ear surgery have become popular options for cholesteatoma treatment. Endoscopic assistance is useful for complete surgical resection of the pathological tissue, particularly when it includes regions exhibiting invasion of the tympanic sinus, in which the endoscopy allows better visualization. Further, in the case of more localized cholesteatoma, detailed preoperative knowledge may be very useful for determining whether the entire surgery should be performed under endoscopic guidance. Regarding prognostic factors, preoperative complications could influence the surgical strategy. For instance, in cases where total adhesion of the tympanic membrane occurs, which is classified as stage III, a low proportion of postoperative hearing improvement can be predicted. Additionally, preoperative information regarding the labyrinth fistula may lead to a preference for the canal wall down technique.

This study has some limitations. First, our results were subject to selection bias because the intraoperative findings were strongly influenced by the surgical indications. Despite potential bias in the data, our findings essentially focused on the pathogenesis of pars tensa cholesteatoma. Second, we did not evaluate postoperative hearing levels or cholesteatoma recidivism. Further studies including long-term outcomes may demonstrate the clinical relevance of the difference between the JOS and EAONO/JOS criteria with respect to the involvement of the sinus tympani.

CONCLUSION

Our findings indicate that the staging criteria advocated by the JOS and EAONO/JOS appropriately reflect the detailed pathogenesis of pars tensa cholesteatoma. The involvement of the sinus tympani in pars tensa cholesteatoma was assessed, and a difference in stage classification between the JOS and EAONO/JOS criteria regarding that area was found. Analysis of a homogeneous population based on a more detailed extension classification by the JOS and EAONO/JOS criteria aids better understanding of the clinical characteristics of pars tensa cholesteatoma. Further studies, including assessments of postoperative outcomes, are necessary to determine the clinical importance of detailed information on extension in pars tensa cholesteatoma, including the involvement of the sinus tympani.

Ethics Committee Approval: Ethics committee approval was received for this study from the Institutional Review Board of the Jikei University School of Medicine (No. 29-208(8824)).

Informed Consent: Informed consent is not necessary due to the retrospective nature of this study.

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Conflict of Interest: The authors have no conflict of interest to declare.

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