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Main Articles

Osteoplastic changes in attic cholesteatoma

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Abstract
Eighty-nine cases of attic type cholesteatoma were operated on during a three and a half-year period. Of these, eight cases were characterized by bony tissue proliferation at the aditus ad antrum or mastoid antrum. Sex, age, and hearing levels were not significant in these cases. Bony fixation of the incus and the malleus was seen in six cases. Bony tissue blocked further expansion of attic cholesteatoma at the aditus in four cases, narrowed the epithelial tract to the antrum in two cases, and completely separated the cholesteatoma into two cholesteatomas in two cases. Infectious stimuli, at an early stage of the disease, might stimulate such osteoplastic activity at the aditus. Axial CT scans give useful information regarding structure before surgery.

Key words: Middle ear; Cholesteatoma; Tomography, X-ray computed; Bone regeneration

Introduction

One characteristic of middle ear cholesteatoma is its osteolytic activity (Chole, 1986). The hallmarks of cholesteatoma on computerized tomography (CT) scans i.e. smooth bony expansion of the attic and mastoid antrum, scalloping of the mastoid, and erosion of the ossicles, lateral wall of the attic and anterior tympanic spine (Mafee *et al.*, 1988) are all attributed to the lytic activity of the disease.

Among the cases of middle ear cholesteatoma examined by us, however, we discovered an atypical type, characterized by osteoplastic, rather than osteolytic activity, seen mostly around the aditus ad antrum. The present paper reports eight such cases of atypical attic cholesteatoma.

Materials and methods

In the period between February 1991 (when the first atypical case was recorded) and August 1994, a total of 89 cases of pars flaccida cholesteatoma were primarily treated surgically at Hamamatsu University Hospital or its affiliated hospitals. Of these, eight cases (five male and three female) were categorized as atypical after surgery. Patients' ages ranged from eight to 62 years (average 43 years). Past histories of ear pathology and hearing disorder were less varied among these patients than among patients with ordinary attic cholesteatoma.

Tympanic membrane perforation was found in the pars flaccida of seven patients, but no perforation was seen in the one case in which congenital

TABLE I
PATIENT CHARACTERISTICS AND DISEASE FEATURES

Case no.	Age	Sex	Side	Air hearing level and type	Ossicular fixation	Cholesteatoma location	Fistula of labyrinth	Bone proliferation	Pneumatization in opposite ear
1	8	M	R	38 dB, C	M, I	Attic	+	Aditus	Good
2	26	M	L	32 dB, C	I	Attic–Mastoid	–	Aditus	Poor
3	29	M	L	37 dB, MI	M, I	Attic	–	Aditus	Poor
4	52	M	L	82 dB, MI	M, I	Attic	–	Aditus	Good
5	53	F	R	45 dB, MI	I	Attic	–	Aditus	Fair
6	57	M	R	73 dB, MI	None	Attic/Mastoid	+	Aditus	Good
7	60	F	R	52 dB, C	M, I	Attic–Mastoid	–	Aditus	Fair
8	62	F	L	38 dB, MI	None	Attic/Mastoid	–	Antrum	Post-operative

C, conductive; MI, mixed; M, malleus; I, incus.
Hearing level is the pure tone averages of 0.5, 1, and 2 KHz.

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ossicular fixation was the tentative diagnosis. Surgery confirmed attic cholesteatoma in every cases. In two cases (Table I:Cases 1 and 7), the postero–superior part of the tympanic membrane was adhered to the long process of the incus. No adhesion of the eardrum to the ossicles or promonotory were seen in any of the remaining cases. Various features of these eight cases are summarized in Table I.

Results

In all cases, bony tissue proliferation, mostly plate-form, was found blocking the aditus ad antrum (seven cases), or the mastoid antrum (one case) (Figure 1). Bony plates were incomplete in five cases, and completely sealed the central mastoid cell tract in three, as far as could be determined under an operating microscope. In four cases (Nos 1, 3, 4 and 5) attic cholesteatoma terminated at the plate, with no further cholesteatoma. Mastoid antrum and air cells, however, were found peripheral to the plate though underdeveloped in these cases. These mastoid cavities were lined with thick mucous membrane, or filled by cholesterol granuloma. In two other cases (Nos 2 and 7), a thin epithelial tract, narrowed by the bone at the aditus, proceeded peripherally from the attic cholesteatoma, to form an enlarged cholesteatoma in the mastoid. In one of these (No. 2), the opening of the tract in the attic was found at the middle of the medial attic wall, and in the other (No. 7), at the medial side of the short process of the incus. In the remaining two cases (Nos 6 and 8), independent cholesteatomas were seen in the attic and mastoid, separated by a bone at the aditus or the mastoid. No inflammatory changes were seen in or around the cholesteatoma. In one case (No. 6), a thin epithelial tract from the mastoid cholesteatoma ended blindly in the bony tissue at the aditus.

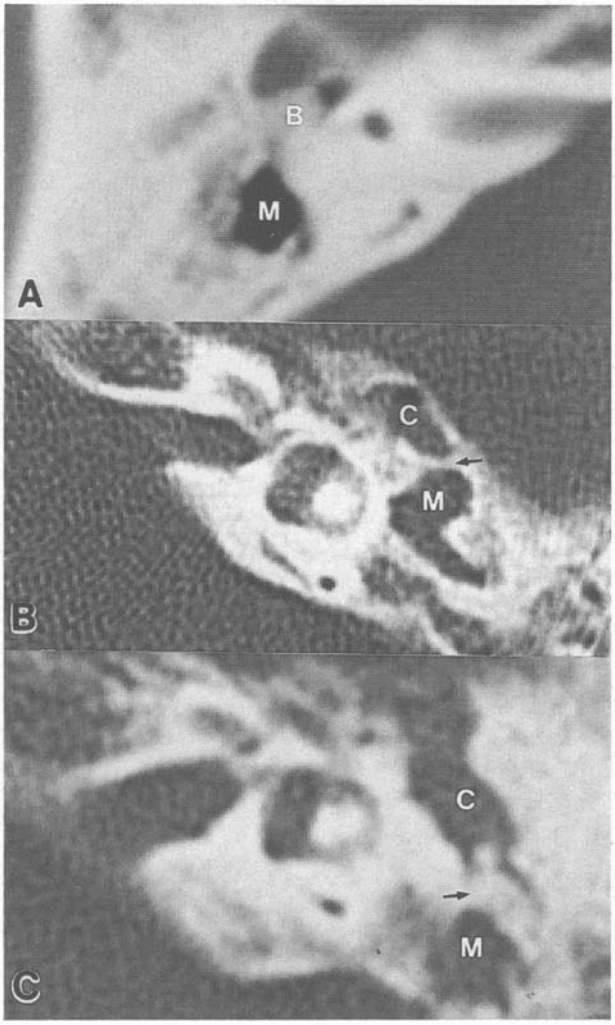

FIG. 1

Axial CT views showing bony proliferation. (A) Bone tissue (white B) fused with the incus blocks the aditus. No cholesteatoma is found in an isolated mastoid antrum (white M) (*Case* 5: Table I). (B) A bony plate (arrowed) separates the cholesteatoma in the attic (white C) and mucous membrane-covered mastoid antrum (white M) (*Case* 3). (C) A bony plate (arrowed) in the mastoid antrum completely separates the cholesteatoma from one in the attic (white C) and one in the mastoid (white M): ossicles were not fixed in this case (*Case* 8).

Ossicular fixation was absent in two cases (Nos 6 and 8), and present in six. In three cases (Nos 1, 3 and 7), both malleus and incus were fixed to the medial attic wall without continuity to the bony plate blocking the aditus. In the remaining three cases, the blocking bone was continuous to both incus and malleus (No. 4), or to the incus body alone (Nos 2 and 5).

Erosion of the dural plates of the middle and posterior cranial fossae was seen in two cases (Nos 6 and 7).

A small piece of proliferated bone was harvested for histological examination from one of the patients. The histological section was found to be composed of lamellar bone with interlacing canaliculi with connective tissue inside (Figure 2).

Temporal bone CT scans were performed along an axial plane in all cases, with direct coronal views also taken for some patients. Pneumatization was poor in all cases. These scans revealed either clearly defined bone shadows (mass- or plate-form), or hazy, hard, tissue shadows in the attic and aditus. Some

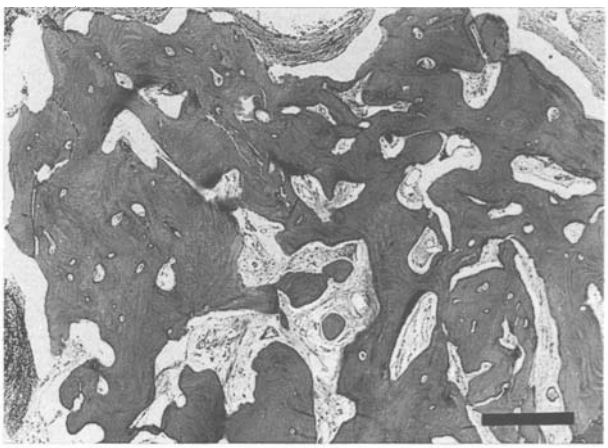

FIG. 2

Sectional view of bone blocking the aditus (Case 6). A bony tissue acquired during surgery shows lamellar bone with interlacing canaliculi, containing connective tissue and capillaries. Bar = 0.5 mm.

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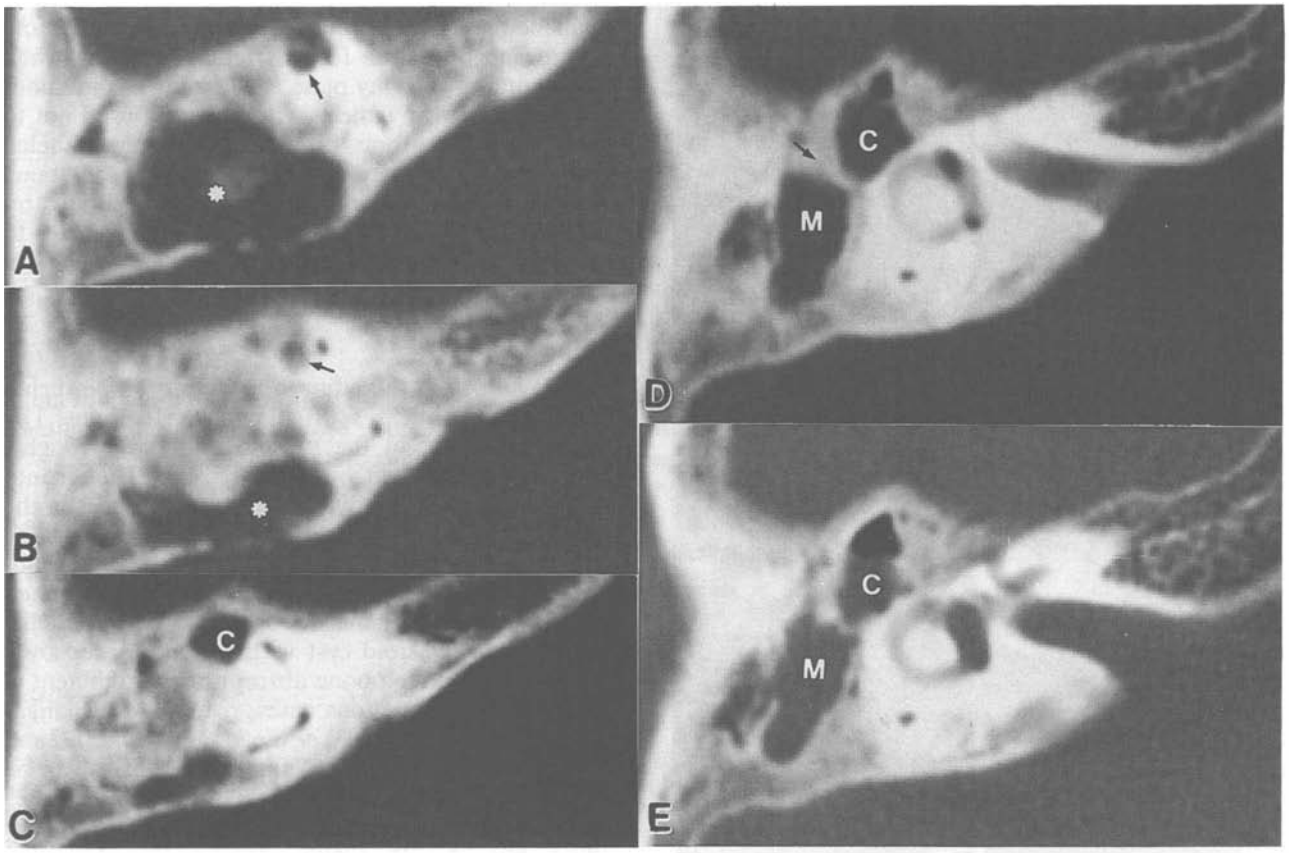

FIG. 3

Axial CT views [five sequential slices of 2 mm thickness from superior (A) to inferior (E)] of Case 7 (right ear). Cholesteatoma in an atypical location (asterisk in A and B) is connected to the attic cholesteatoma (white C, in C, D and E) through a thin epithelial duct (small arrow in A and B). A bony plate (arrowed in D) separates the attic and antrum (white M), which contained no cholesteatoma.

representative findings from continuous slices are shown in Figures 3 and 4. Figure 3 shows exceptional extension of cholesteatoma off the central mastoid cell tract through a narrow bony channel, together with erosion of the dural plate of the posterior cranial fosa. Presence of the erosion of the dural plate (Cases 6 and 7) and fistula of the labyrinth (Cases 1 and 6) indicate coexistence of osteolytic and osteoplastic changes in the same middle ear.

Discussion

Osteoplastic change in the mastoid cavity is frequently seen as a sequel of middle ear infection. Osteoid tissue deposited on the existing bone is then transformed into lamellar bone by osteoblastic activity (Schuknecht, 1993). Such osseous sclerosis narrows the mastoid cavity, but the central mastoid cell tract usually remains open. Another type of osteoplastic change is callus formation occurring post-operatively in the mastoid or middle ear, where there has been trauma to bone or periosteum, and where there is a continuing inflammatory process (Schuknecht, 1993).

The bone proliferation observed in the present cases is different from either the osseous sclerosis or the post-operative callus formation mentioned above. Histologically, however, the bony tissue acquired in *Case* 6 proved to be very similar to callus formation, and thus it is possible to surmise that the bone proliferation in the present cases may be the product of inflammatory stimuli.

Inflammation might have stimulated bone formation at an early stage of the disease as the cholesteatoma was reaching the aditus. Newly-forming bone in the aditus might then have blocked further expansion of the cholesteatoma, or narrowed and made tortuous a still expanding cholesteatoma duct. At times, it might have completely separated the epithelial duct, leaving a separated cholesteatoma in the mastoid. If inflammation occurred at a later stage after the cholesteatoma matrix already occupied the antrum, then osseous sclerosis with a narrowed central mastoid tract would remain.

The reason why bone proliferation occurred at the aditus, and why this type of change has been seen only in the pars flaccida type of cholesteatoma, is not known.

Another possible cause of bone proliferation is an anomaly of the middle ear structure. The fixed ossicles and presence of cholesteatoma without tympanic perforation found once in *Case* 1 suggest this possibility.However, bony tissue proliferation of the type observed in the present cases has not been histologically proven in the congenital cases so far. Reported congenital ossicle fixation is often caused

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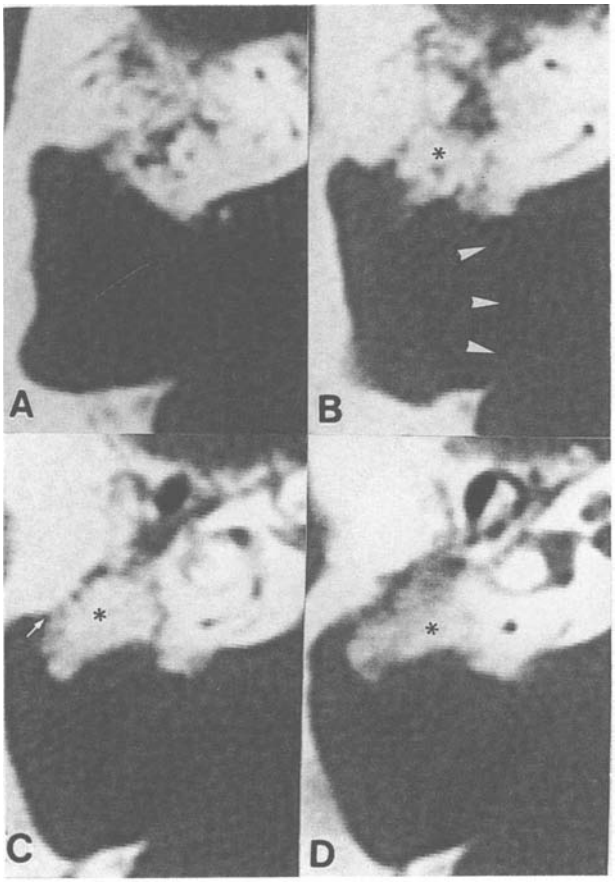

FIG. 4

Axial CT views [four sequential slices of 2 mm thickness from superior (A) to inferior (D)] of Case 6 (right ear). The attic and mastoid cavity, both containing cholesteatoma, are separated by a bone mass (asterisks in B, C and D). Note that a cross-section of this bone is shown in Figure 2. A thin epithelial duct from the mastoid cholesteatoma grows through a narrow bony channel (arrowed in C), but ends up blindly at the aditus. The dural plate of the posterior cranial fossa is vastly eroded (arrowheads in B).

by bony spurs around the malleus head (Schuknecht and Gulya, 1986) whereas the ossicular fixation found in the present cases always occurred around the incus. Bony ankylosis of the incus body is rare in the ossicular anomalies reported so far (House, 1956). We consider absence of the tympanic perforation in *Case* 1 to indicate a healed perforation after cessation of the inflammation.

Cases of bony fixation of the malleus and incus occurring in the attic were discussed by Tos (1970). The majority of his cases were presumed to be caused by infection and the illustrations of fixation provided by him bear little resemblance to our cases. Cholesteatoma was found in only one case out of 12.

There are few reports of bony blockage at the aditus ad antrum in the literature. The aditus blocks reported by Richardson (1963) were made up of soft tissue in most cases, and were thought to be caused by bacterial infection. No relation to middle ear cholesteatoma was mentioned in his paper.

Audiometry results are not characteristic for atypical cholesteatoma; scrutiny of the axial CT scan being the best way to observe this condition. During an operation for attic cholesteatoma, when confronted by a bony plate at the aditus, contiguous to, and fixing the ossicles, care must be used not to miss a cholesteatoma or thin connecting epithelial tract, as they are often hidden by the bone proliferation. Every epithelial tract should be carefully followed. Removal of the bony tissue helps to ensure total exposure.

Conclusions

A bony block at the aditus appears not to be such a rare finding in cases of attic cholesteatoma. The occurrence rate in the present study was roughly nine per cent (eight out of 89 cases). Though not mentioned in this study, various bony changes in the aditus or the attic were also found e.g. an isolated antral cholesteatoma separated by the mucous membrane of the attic, which contained fixed ossicles and epithelial debris but no epithelial matrix, or a mastoid cyst with aditus blocked by a bony plate. Though bone absorption is prominent in most cholesteatomatous cases, osteoplastic changes also frequently occur at some stage of the disease. The cause of the bone formation seems to be inflammatory in most cases.

Acknowledgement

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References

Chole, R. A. (1986) Acute and chronic infection of the temporal bone including otitis media with effusion. In *Otolaryngology, Head and Neck Surgery.* (Cummings, C. W., Harker, L. A. eds.). C. V. Mosby Co., St Louis/ Toronto, pp 2963–2988.

House, H. P. (1956) Diagnostic aspects of congenital ossicular fixation. *Transactions of the American Academy of Ophthalmology and Otolaryngology* **60:** 787–790.

Mafee, M. F., Levin, B. C., Applebaum, E. L., Campos, M., James, C. F. (1988) Cholesteatoma of the middle ear and mastoid. A comparison of CT scan and operative findings. *Otolaryngologic Clinics of North America* **21:** 265–293.

Richardson, G. S. (1963) Aditus block. *Annals of Otology, Rhinology and Laryngology* **72:** 223–236.

Schuknecht, H. F. (1993) *Pathology of the Ear,* 2nd Edition, Lea and Febiger, Philadelpha, pp 555–597.

Schuknecht, H. F., Gulya, A. J. (1986) *Anatomy of the Temporal Bone with Surgical Implications,* Lea and Febiger, Philadelphia, pp 59–63.

Tos, M. (1970) Bony fixation of the malleus and incus. *Acta Otolaryngologica* **70:** 95–104.

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