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CASE REPORT

A Case of Middle Ear Papilloma Originating from the Eustachian Tube

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Papilloma in middle ear is extremely rare in otolaryngology field. Inverted papilloma involving the middle ear has a high recurrence rate and a possibility of malignant change. As these tumors seldom involve the middle ear, there are no estimates on the incidence of these lesions in this particular site and only a few reports in the literature. We experienced a case of papilloma of the middle ear originating from the Eustachian tube accompanied by cholesteatoma. Thirty-year-old woman visited our clinic and was treated with open canal wall tympanomastoidectomy, There was a pink papillomatous mass located at tympanic orifice of the Eustachian tube. That mass was idenfified as papilloma. There was no recurrence of cholesteatoma and papilloma 2 years after the operation. This paper aims to report a case of primary middle ear papilloma in a patient with no account of previous nasal disease with literature.

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Introduction

In otolaryngology, a papilloma has been reported in the nasopharynx, lacrimal gland, larynx, and temporal bone. However, its origin from the inside of the middle ear cavity in the temporal bone is a rare case especially with otitis media that accompanies cholesteatoma; this rare case has not yet been reported. The pathogenesis and relationship with cholesteatoma of papilloma originated from the inside of middle ear cavity has not fully understood. A papilloma is histologically a benign tumor but it tends to invade locally. Since incomplete removal leads to recurrence, the removal range must aim eradication of the tumor. Initially, this case was thought to be otitis media with cholesteatoma or simple chronic otitis media which we planned to treat surgically but accompanying papilloma was found later. We report this rare case with literature reviewing.

Case Report

A 30- year old female visited our clinic complaining of partial hearing loss in left ear and yellowish purulent otorrhea which started 7-8 years ago. The patient had no history of significant prior diseases. Hearing in the left ear decreased to the extent that she was unable to perform a proper conversation on the phone. She had neither autophonia nor hyperacusis. During systemic examination, dizziness, otalgia, and tinnitus were not found. In physical examination, right tympanic membrane was normal, and light-pink-colored soft-tissue was found which partially filled the left external auditory meatus. This soft-tissue was connected to the middle ear cavity through superior part of tympanic cavity, the otorrhea was draining through the defect of tympanic membrane, and the *Providencia stuartii* was isolated by culture examination. The inferior part of the tympanic membrane was totally adhered to middle

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ear showing no movement on air otoscopy examination and Valsalva maneuver. Whitish keratin material appeared in the retraction pocket and the malleus was not observed (Fig. 1A). The nasal cavity and pharyngeal cavity were normal according to physical examination. Conductive hearing loss (65/15dB) was found in pure tone audiometry before the operation (Fig. 1B). On computed tomographic scans of the temporal bone after otorrhea was decreased by pharmacological treatment, in the superior lateral area, the mastoid was sclerotic and epitympanum, middle ear cavity, and aditus ad antrum were filled with soft tissue density. These soft tissue density was also found in the external acoustic meatus and the Eustachian tube connecting both structures. The ossicles were eroded so that only their part was found. In coronal scan image, bone destruction around epitympanic space was observed and soft tissue was found in the mastoid antrum, facial recess and hypotympanum (Fig. 2 A, B). Based on both the physical findings and computerized axial tomography, the soft tissue was suspected to be inflammatory tissue due to otitis media with cholesteatoma, adenoma, or squamous cell carcinoma. On the taken magnetic resonance imaging to identify invasion to surrounding tissues, a mass which extends from external acoustic meatus to the Eustachian tube had definite boundary. The mass was observed with the isosignal intensity on T1 weighted image and high signal intensity of distal end on T2 weighted view (Fig. 2 C, D). Although biopsy of the soft tissue from the external acoustic meatus confirmed granulomatous inflammatory tissue, operation was indicated for differential diagnosis and radical treatment; thus, the surgery was performed under general anesthesia. We found the white tumor in the middle ear cavity which looked like papilloma during the operation. The stem of the mass was in the Eustachian tube, the cabbage-shaped body was directed to the middle ear cavity, and it was 1.5 x 1 cm in size. Since there was no adhesion with surrounding tissue, the body and stem were removed simultaneously by en-bloc dissection. Whitish keratin materials separated from the mass had accumulated in the epitympanum and the auditus ad antrum. We removed the cholesteatoma completely. The incudostapedial joint was separated already. The malleus and incus were removed because they appeared to be eroded mostly. Open cavity mastoidectomy was performed on the sclerotic mastoid for easy recurrence observation. Ossiculoplasty with PORP and tympanoplasty with temporalis muscle fascia were performed afterwards. Due to the possibility of remnant cholesteatoma, we decided to consider second-look operation according to the follow-up result and pathological opinion of the tumor. The surgery was ended without placing ventilation tube since the condition of the Eustachian tube was good. The biopsy result reported that the mass, in H&E stain, was cholesteatoma and papilloma characterized by growth on the epithelium and

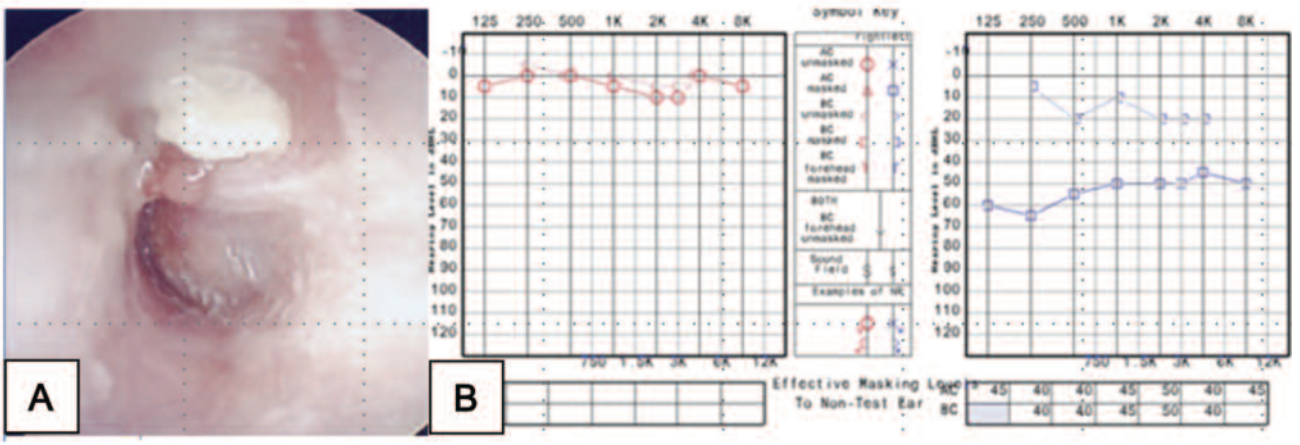

Figure 1. A: The inferior part of the tympanic membrane was totally adhered to middle ear and whitish keratin material appeared in the retraction pocket. **B:** Pure tone audiometry showed that air bone gap was 50dB, conductive hearing loss in left ear.

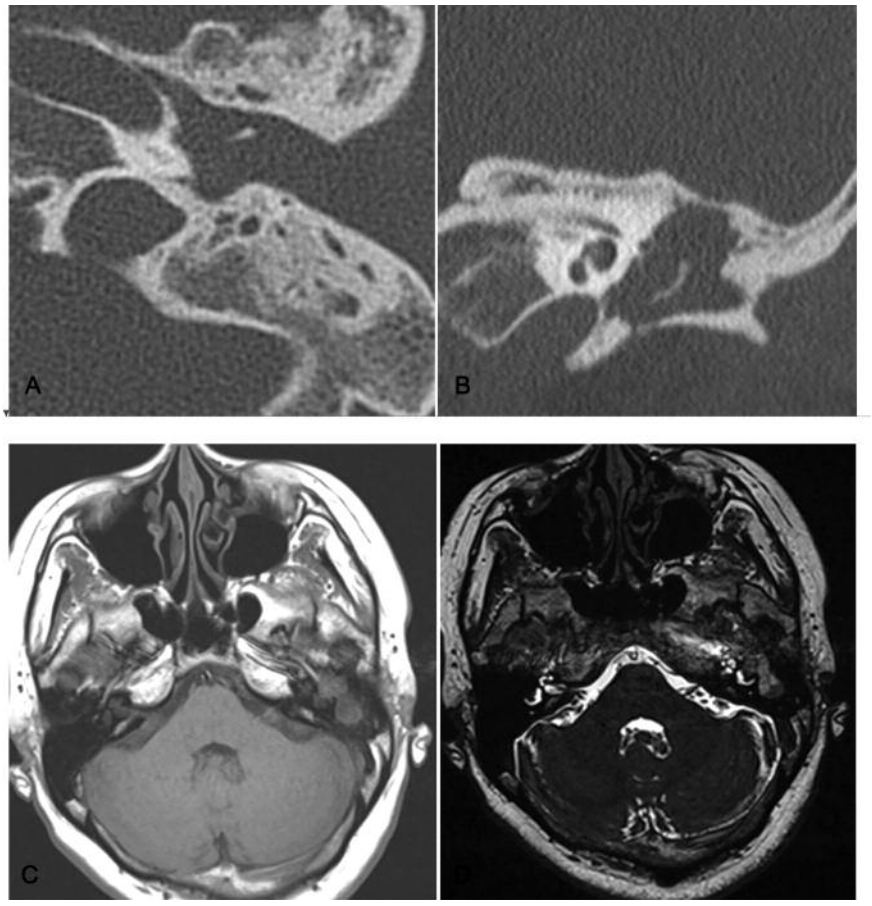

Figure 2. A and B Temporal CT. A(Axial view) A: The white arrow shows E-tube was obstructed by soft tissue density. Middle ear was filled with total soft tissue density. B(Coronal view): Ossicle was eroded and scutum was blunt but tegmen was intact.
C and D Temporal MRI. C(T1 weighted image): The white arrow shows that low signal intensity with definite boundary lesion which extends from left external auditory canal to the Eustachian tube, D (T2 weighted image): The white arrow shows that heterogenous signal intensity with definite boundary lesion.

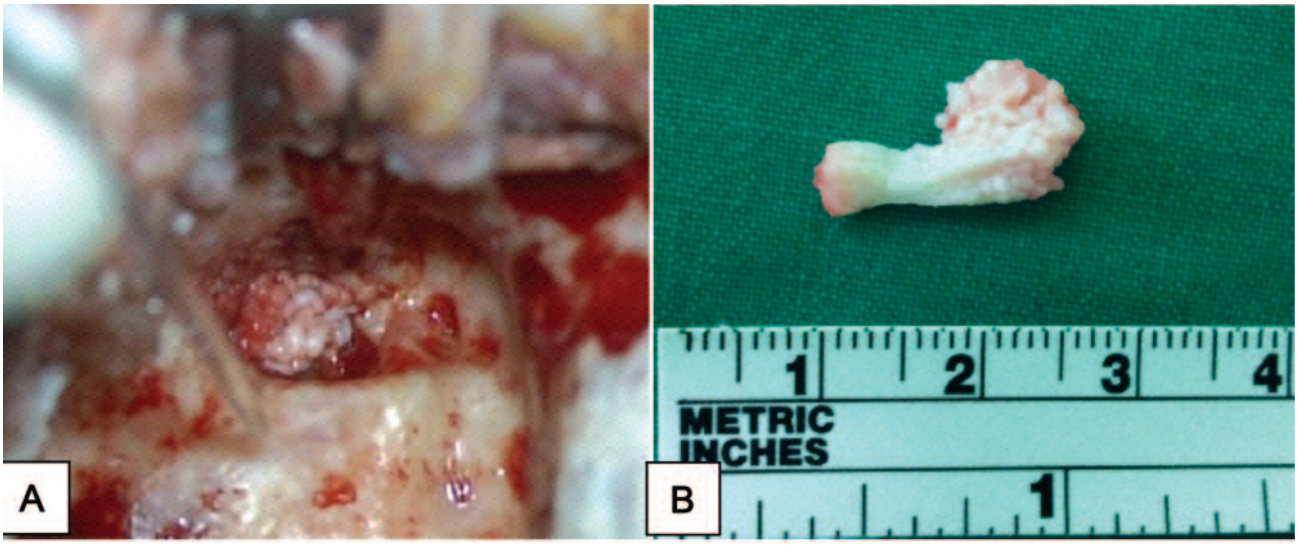

Figure 3. A: The surgical view shows that middle ear papilloma originated from the Eustachian tube. **B:** The papilloma length is about 1.5 x1.0 cm size.

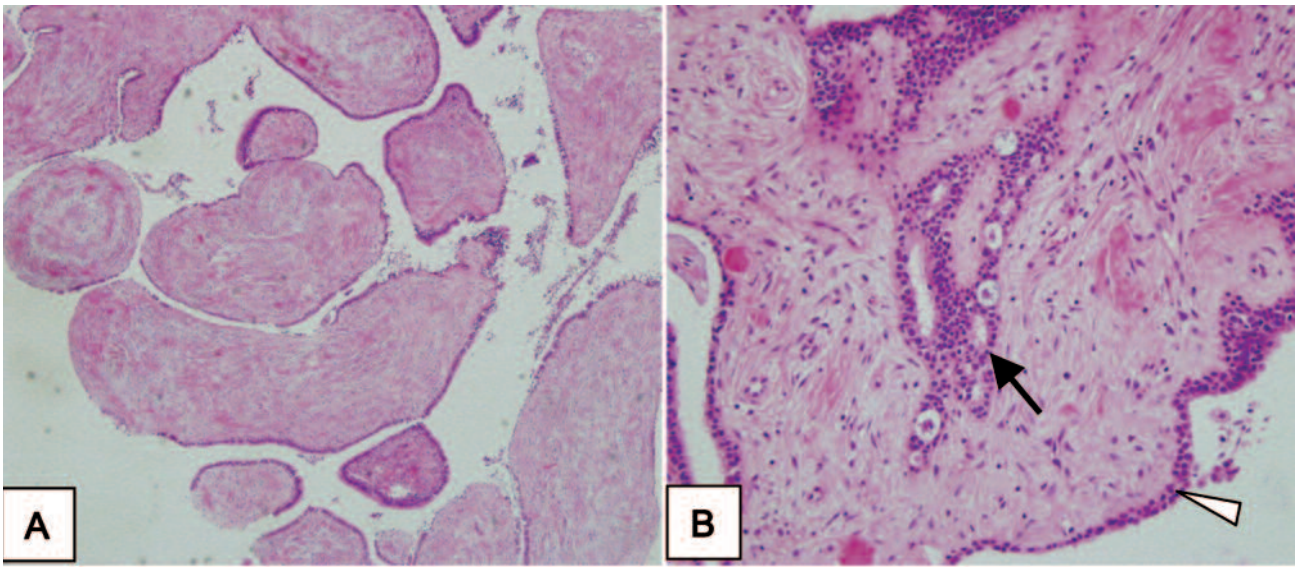

Figure 4. Histological finding A: The epithelial hyperplasia with abundant stroma is noted (H & E stain, 40). B: The black arrow showed fibrous epithelial gland growth surrounded by fibrous stroma. The arrow head showed that epithelial lining (H & E stain, 100).

inclusion of fibroepithelial glands. Currently, the patient has been observed for 24 months at outpatient clinic after the surgery. Pure tone audiometry hearing test result showed air/bone gap is approximately 25dB which is much better than before the surgery. There was no recurrence of mass, graft tympanic membrane was well regenerated, and ventilation of middle ear was good by now.

Discussion

A papilloma is originated from squamous epithelium or schneiderian epithelium. In the case of direct involvement of the mastoid or middle ear cavity has been explained with direct invasion and metaplasia theories. The first theory is direct involvement from the nasal sinus to the Eustachian tube and second theory is metaplasia of the middle ear mucosa caused by chronic inflammation [1]. For this case, there was no significant finding at the nasal cavity or nasopharynx on physical examination but papilloma in the middle ear of which the stem was originated from the Eustachian tube and was transformed into cabbage shape only in the middle ear cavity. Since the mass was single, and was accompanied by otitis media with cholesteatoma, mucosal degeneration in the Eustachian tube should have been checked for more accurate investigation. According to the literature, differential diagnosis of papilloma in the middle ear cavity can be adenoma, jugulotympanic ganglioma, acoustic neuroma, meningioma, and so on [2]. A papilloma is classified into 1. fungiform papilloma, 2. inverted papilloma, and 3. cylinderical papilloma [2]. In this case, pathology department reported the tumor as a papilloma without further classification since it did not fall into neither of the three mentioned above.

As deciding surgical treatment of papilloma, radical eradication is required for it tends to frequently recur if it is removed incompletely. However in this case, open cavity mastoidectomy with tympanoplasty was performed in the consideration of second-look operation and the advantage over intact canal wall mastoidectomy such as better visual access, and more complete removal. The patient has been followed up for 24 months after the surgery, and no retraction of the grafted tympanic membrane due to recurred or survived tumor so far.

The relationship between papilloma and cholesteatoma in this case can be explained by invagination theory [3,4]. A papilloma blocked the entrance of the Eustachian tube in the middle ear cavity causing malfunction which arises negative pressure in middle ear cavity and eventually causes retraction of the tympanic membrane. In this way, the movement of epithelial cell impaired, and exfoliated epithelium cannot escape causing the accumulation of keratin; thus, the cholesteatoma must have formed from it. Although both papilloma and cholesteatoma have high local recurrence rate, open mastoidectomy was chosen

instead of radical mastoidectomy which was typically indicated. Since compromised surgical method was performed in the consideration of hearing, age, and various other factors, careful follow-up at outpatient clinic is required. In addition, ventilation tube insertion must be performed immediately if worsening of ventilation is observed in the future.

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