Primary middle ear papilloma: A case report

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We report a case of primary middle ear papilloma in a 66-year-old male with underlying chronic suppurative otitis media, presenting with right sided otorrhea with progressive aural mass formation. He underwent right mastoidectomy and tumour excision. Diagnosis of right middle ear papilloma with no malignant features was confirmed with histopathological examination. The patient recovered well with no recurrence up to 12 months.

Keywords: Papilloma, otitis media, otorrhea, aural mass.

INTRODUCTION

Inverted papilloma (IP) is a benign tumour of sinonasal region which constitutes 0.6–1.5 in 100,000 population per year, tend to recur and capable of malignant transformation.[1] IP is a type of Schneiderian papilloma, commonly found at sinonasal territory, and less commonly identified at extranasal sites such as pharynx, lacrimal sac, and middle ear.[2] Involvement of the middle ear and mastoid is less frequent, widely reported as the concurrent of sinonasal inverted papilloma, migrating through the Eustachian tube and also as ectopic migration of Schneiderian membrane embryologically.[2] Etiology are multifactorial, including chronic inflammation, viruses, environment, and allergy.[3] It affects adults to the elderly, and the most frequent symptoms are chronic otorrhoea with hearing loss.[4] Examination usually revealed polypoid-looking middle ear mass with discharge through a perforated tympanic membrane.[5] Here, we present a report of primary middle ear papilloma.

CASE PRESENTATION

A 66-year-old male with a history of underlying left temporal bone squamous cell carcinoma underwent left subtotal temporal bone resection with superficial parotidectomy and radiotherapy in the year 2006. After completed treatment, repeated CT scan showed no evidence of tumour recurrence. He complained of intermittent right-sided otorrhea during routine follow-up, which was already present for more than 20 years and treated as chronic otitis media without cholesteatoma. Apart from that, there was hearing loss and occasional tinnitus on the same side. No otalgia, vertigo, aural fullness, facial asymmetry nor constitutional symptom was present.

Examination of the right ear showed subtotal tympanic membrane perforation with purulent discharge. Soft tissue mass was seen arising from the middle ear at the superior region of perforated tympanum. Nasal and oropharynx examinations were normal. Pure tone audiometry revealed moderate to severe mixed hearing loss level, while there was profound hearing loss in the left ear.

Multiple doses of topical eardrops were prescribed but the symptoms continued. The soft tissue mass was enlarging and protruding laterally beyond the perforated edge of the tympanic membrane. Imaging with HRCT scan revealed a soft tissue density lesion within the anterior epitympanum, while the rest of the middle ear cavity including ossicles were intact, and no evidence of chronic mastoiditis.

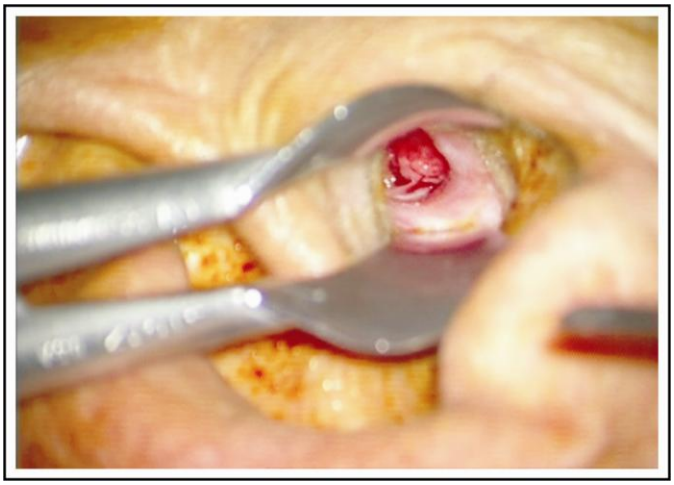

Fig. 1: Polypoidal mass.

The patient underwent right-sided modified radical mastoidectomy and tympanoplasty type IV. Polypoidal mass was found filling up the mesotympanum and epitympanum (Fig. 1). There was an erosion of the short

process of the incus. Incus and head of malleus were nibbled and removed to facilitate surgery. He had an uneventful surgery and was discharged.

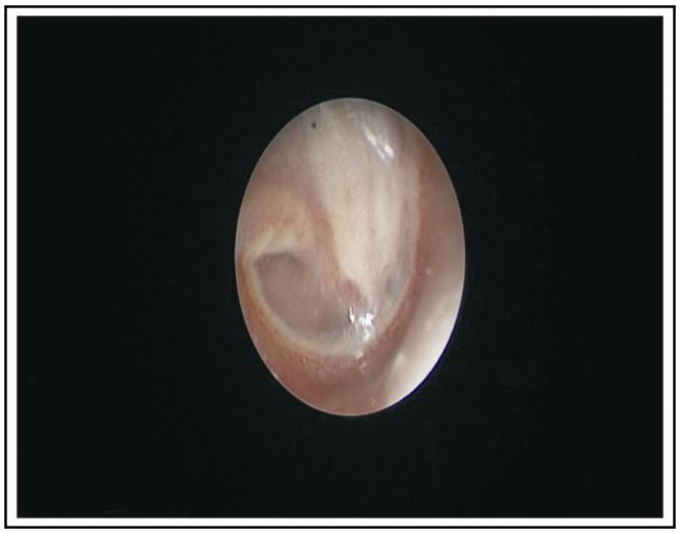

Fig. 2: Mastoid cavity.

Histopathological examination of the mass revealed fragments of tissue forming papillary structure lined by hyperplastic stratified squamous epithelium with central fibrovascular core with no evidence of malignancy. The adjacent antrum, middle ear showed granulation tissue formations. During postoperative review, the ear was dry and the neo-tympanic membrane appeared healthy. The mastoid cavity was well epithelialized (Fig. 2). The patient recovered well with no recurrence up to 12 months of follow-up.

DISCUSSION

Schneiderian papilloma is a benign disease of the nose and para nasal sinus mucosa.[6] IP is the most commonly reported type of Schneiderian papilloma.[7] IP mostly originate from the lateral wall of the nose or maxillary sinus, with a reported incidence of 0.2 to 1.5 per 100,000 people per year.[3] The occurrence of papilloma in the middle ear was first documented by Stone et al in 1987[8] and up to 57 cases were reported at the time being excluding current report.[2]

Several mechanisms are proposed. Stone et al hypothesized that the occurrence of middle ear papilloma was due to direct extension from eustachian tube, however it was found that many of the cases had normal eustachian tubes.[5] The second mechanism involves an embryonic migration of an ectopic Schneiderian epithelium into the middle ear.[5] The third mechanism was the inflammatory process related to chronic suppurative otitis media, which in turn stimulated the development of Schneiderian type epithelium which give rise to IP in the middle ear.[4] In present case, there was chronic suppurative otitis media as well as chronic mastoiditis on HRCT scan, which supports the third mechanism.

Hearing loss, followed by otorrhea are the most common symptoms of middle ear papilloma.[4,9] In the current case, there was long-standing suppurative otitis media with persistent discharging ear. Other symptoms reported in the literature are aural fullness, otalgia, and facial nerve paralysis.[9] Papilloma arising from the sinonasal region including middle ear are previously known as Schneiderian Papilloma as it is lined with Schneiderian epithelium, ectodermal derived respiratory mucosa.[2,5]

Inverted, oncocytic, and exophytic papillomas are three histologically distinct forms of papillomas that arise from this distinctive epithelium. The most frequent type of sinonasal papilloma is inverted papilloma. IP is lined with squamous, respiratory, or transitional epithelium with an endophytic architectural pattern on histology. Exophytic papilloma is lined by similar epitheliums of IP, but with an exophytic architectural pattern, whilst oncocytic papilloma is lined by oncocytic epithelium and possibly show either endophytic or exophytic architectural pattern.[7]

In current case, it is difficult to distinguish between the types of papilloma due to the fragmented nature of the specimen, and Human Papilloma Virus (HPV) in-situ hybridization or polymerase chain reaction (PCR) test was not done as it is not available in our center. IP is associated with low-risk HPV 6 and 11 as well as high-risk HPV (HPV 16 and 18) whereas exophytic papilloma is usually associated with low-risk HPV and oncocytic papilloma does not associate with HPV infection. IP and oncocytic papilloma have the risk of malignant transformation; 2.8 to 24.6%, 4 to 17%, respectively.[7]

Association of sinonasal papilloma with middle ear papilloma though uncommon but well known, especially the inverted type.[2] Therefore, examination and exclusion of nasal symptoms and sinonasal disease are essential to delineate the possible origin of the middle ear pathology, which was negative in the current case.

There are no standard guidelines on the management of middle ear papilloma due to the limitation of cases and reports. But the mainstay of treatment modalities for middle ear IP reported are surgical excision and mastoidectomy as the preferred method of choice.[10] The recurrence is high, with reported about 11.5-78% of recurrence of lesion after adequate treatment.[1,2,10] Thus long-term follow-up is required to detect recurrence and malignant transformation[5]. Malignant transformation to

carcinoma is possible especially for IP of the middle ear, accounts from 1 to 53%.[10]

In summary, Schneiderian papilloma of the middle ear is a rare benign primary pathology, whereby long-standing inflammation of the ear such as chronic discharging ear is a risk factor. The mainstay of treatment is complete surgical resection with a good prognosis. Due to the high risk of recurrence and tendency for malignant transformation, long-term follow-up is mandatory.

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