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## Inverted Papilloma of the Middle Ear Presenting as an Aural Polyp

### ABSTRACT

**Objective:** To present a rare case of inverted papilloma of the middle ear in a 77-year-old man presenting with an external auditory canal polyp of the right ear.

**Methods:**

**Design:** Case Report  
**Setting:** Tertiary Private Hospital  
**Patient:** One

**Results:** A 77-year-old man presenting with external auditory canal mass underwent tympanoplasty with canal wall down mastoidectomy. Histopathologic examination revealed inverted papilloma.

**Conclusion:** With only 30 cases reported in the literature, inverted papilloma of the middle ear is a rare disease entity that may mimic other benign conditions such as cholesteatoma. It requires further investigation to devise a rational approach to diagnosis and management. Regular post-operative monitoring is essential due to high recurrence and malignant transformation rate while post-operative radiotherapy remains controversial and requires further investigation.

**Keywords:** *Inverted papilloma, cholesteatoma, middle ear*

**Middle ear infection** and its symptoms are quite common in our setting such that consultation and management (mostly surgical) are often delayed. Mastoidectomy with or without tympanoplasty usually suffices for straightforward aural polyps and cholesteatoma but what if a presumed common condition turns out to be something unexpected and rare? We report one such case.

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**CASE REPORT**

A 77-year-old hypertensive man consulted due to progressive hearing loss in his right ear. Since childhood, he experienced recurrent episodes of right aural discharge that persisted until adulthood. There were no consults or medications. Four years prior, he noticed decrease in hearing acuity in the right ear with otorrhea and ear pain. His hearing progressively worsened prompting consult in our institution.

On physical examination, there was no gross deformity of the auricle and no tragal tenderness noted on both ears. Video otoscopy showed a pinkish, fleshy, mass with smooth surface and irregular borders in the right ear, characteristic of an aural polyp. (Figure 1) The mass obstructed 70% of the external auditory canal limiting the view of the tympanic membrane. Minimal whitish discharge was also noted. The left middle ear appeared normal.

Plain computed tomography scan of the temporal bone showed a soft tissue density in the antrum and epitympanum extending to the external auditory canal of the right ear on axial view and blunting of the right scutum on coronal view. The middle ear ossicles were not identified in the right ear. (Figure 2)

Pure tone audiometry revealed mild sensorineural hearing loss on the left and profound mixed hearing loss on the right ear. Tympanometry revealed a Type A tympanogram on the left and a Type C tympanogram on the right ear.

With an impression of chronic otitis media AS, active with cholesteatoma formation, a canal wall down mastoidectomy with tympanoplasty via post auricular approach with temporalis fascia underlay graft, ossiculoplasty and meatoplasty was performed on the

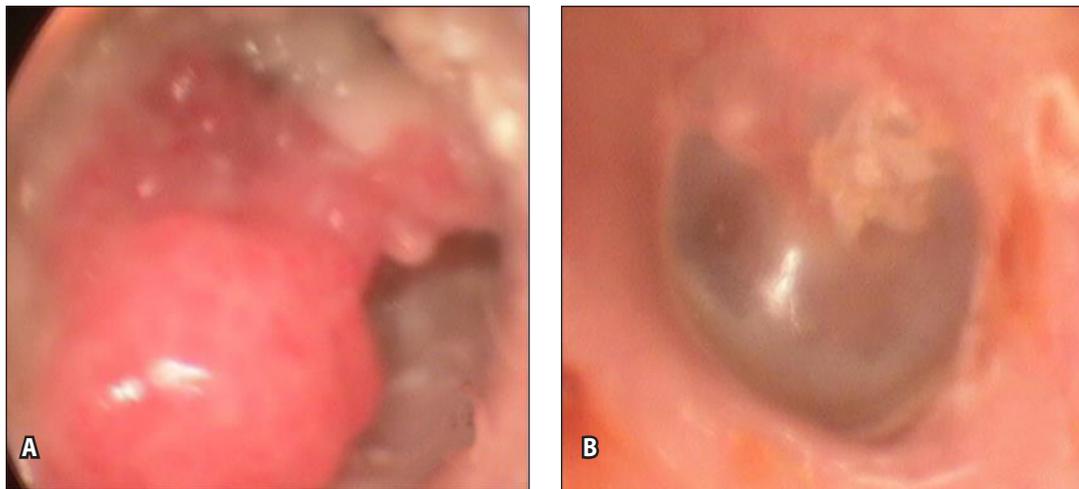


Figure 1. A. Video otoscopy (still image) shows the polypoid mass occupying the right external auditory canal; B. Normal otoscopy of the left ear

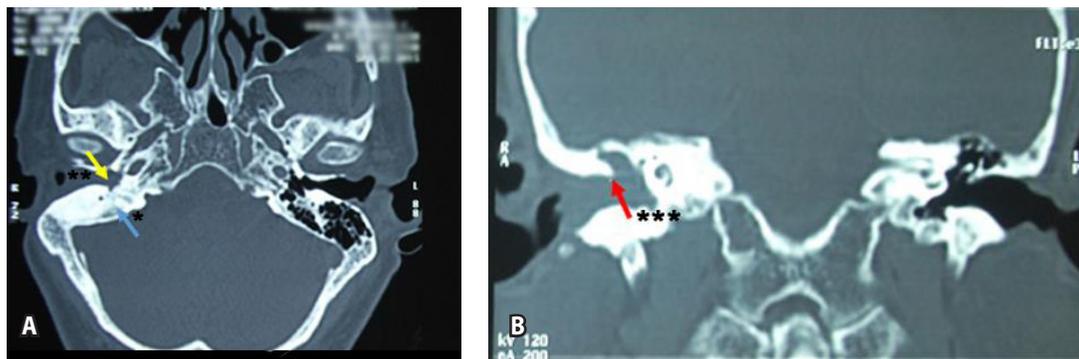
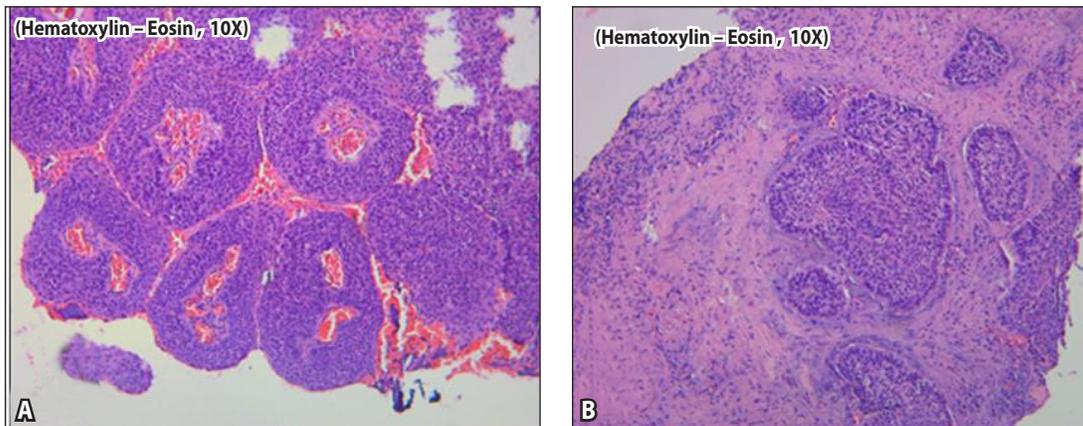
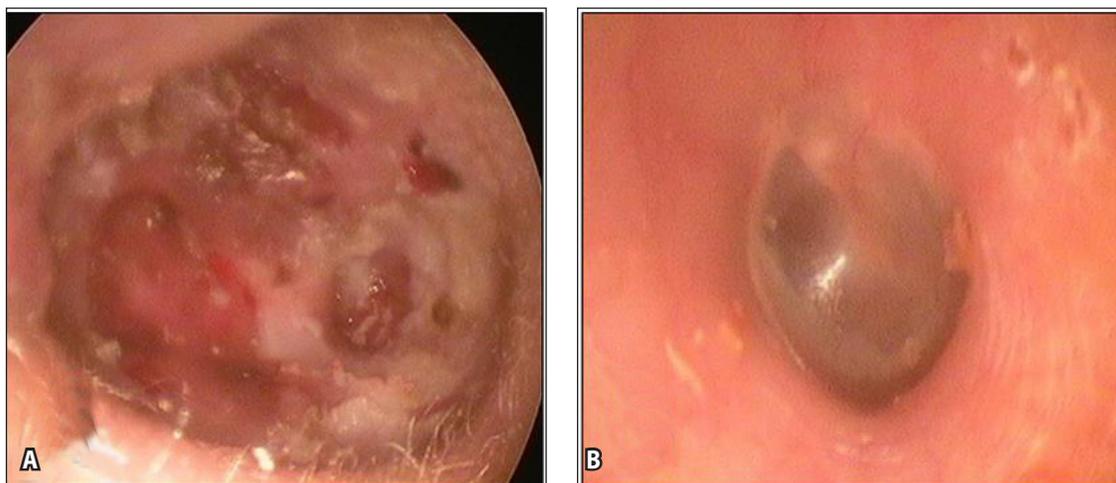


Figure 2. Plain CT Scan, A. Axial view showing a soft tissue density occupying the antrum (\*) extending to the external auditory canal (\*\*); B. Coronal view shows blunting of the scutum (\*\*\*)



**Figure 3.** Histopathologic sections (Hematoxylin – Eosin), **A.** Scanning view showing tissue fragments lined by hyperplastic respiratory epithelium; and **B.** Low power view [10x magnification] showing epithelial surface inversion inside the stroma characteristic of inverted papilloma



**Figure 4. A.** Video otoscopy (still image) 3 months post-operatively showing no recurrence of otorrhea and aural mass in the right external auditory canal; **B.** Normal otoscopy of the left ear

right. Intraoperatively, a firm, fleshy, polypoid mass in the middle ear cavity extending to the mastoid antrum was completely removed. Whitish epithelial debris was also noted in the middle ear cavity. Significantly, there was no mass observed in the eustachian tube opening. Surgical specimens submitted for histopathology consisted of the aural polyp, cholesteatoma and middle ear mass. The cholesteatoma was confirmed as such but the aural polyp and middle ear tissue were diagnosed as consistent with inverted papilloma. (Figure 3)

The post-operative course was uneventful with unremarkable two-week and three-month follow ups. There was no ear canal mass and the tympanoplasty graft was intact. (Figure 4) Repeat pure tone audiometry showed profound mixed hearing loss in the right ear.

### DISCUSSION

“The presence of an aural polyp in a chronically infected ear should be considered to be a cholesteatoma until proven otherwise.”<sup>1</sup> Our patient presented with a polypoid external auditory canal mass on the background of a 20-year history of chronic otorrhea with imaging findings suggestive of cholesteatoma. Hence, the patient was managed as an aural polyp. Biopsy of the ear canal mass was not done pre-operatively and we proceeded directly with mastoidectomy as it is the definitive management for cholesteatoma. However, an unexpected result of inverted papilloma was the final histopathologic finding of the middle ear mass.

Aural polyps are granulation tissues usually found at the junction



between an eroding cholesteatoma and bone.<sup>1</sup> Like the tip of an iceberg, an aural polyp may contain significant clues to a serious underlying infection or benign lesions such as cholesteatoma. Grossly, an aural polyp may be described as a solitary, polypoid, hyperemic mass, often friable, obstructing the view of the tympanic membrane as seen in our patient.<sup>2</sup>

Other benign lesions such as inverted papilloma of the middle ear may also present as an external auditory canal mass. However, it is not surprising that inverted papilloma was not initially considered due to the extreme rarity of the case.<sup>3</sup> Ear canal malignancies may also present as such. However, clinical course and radiologic evidences may differ from a simple condition of cholesteatoma. Shuang *et al.* described the radiologic findings of malignant ear canal and middle ear conditions. Both squamous cell carcinoma and adenocarcinoma present radiologically as a hypodensity on CT imaging signifying necrosis and involvement of the deep structures such as subcutaneous tissues, petrous bone and carotid artery wall in both conditions on MRI.<sup>4</sup> These findings were not present in our patient.

Inverted papilloma, also called “Schneiderian papilloma” is a common tumor arising from Schneiderian epithelium of the nasal cavity and paranasal sinuses.<sup>5</sup> It is the second most common benign tumor of the nose and paranasal sinuses and is the most common surgical indication for benign sinonasal tumor.<sup>5</sup> Inverted papilloma can also be found in other areas of the body such as the urinary tract specifically the urinary bladder and in extremely rare conditions, it could also arise in the middle ear. As of 2012, Rubin *et al.*<sup>3</sup> reported there were only less than 20 cases of inverted papilloma in the literature. However, our search of the English literature on MEDLINE (PubMed), EMBASE and Google Scholar using the keywords “inverted papilloma,” “inverting papilloma,” “ear canal,” “middle ear,” “aural,” yielded a total of 31 cases as of May 2018.

Inverted papilloma of the nose and paranasal sinuses may arise due to several factors. Occupational hazards such as outdoor and industrial occupations and exposure to different smokes, dust and aerosols, may all be considered potential risk factors.<sup>6,7</sup> However, for inverted papilloma in the middle ear, no research has been published to our knowledge that associates occupational hazards with middle ear papilloma. While it seems interesting that this patient worked as a traffic enforcer for five years exposing himself to various pollutants encountered in the streets, we can only conjecture that the background underlying chronic otitis media with a persistent tympanic membrane perforation may have exposed the middle ear to environmental pollutants that may have predisposed to developing a middle ear papilloma.

Another etiology of sinonasal papillomas include infection with

human papilloma virus (HPV) specifically strains 6 and 11. However, this concept remains controversial for the middle ear.<sup>4,8</sup> Of the three hypotheses proposed by Rubin *et al.*, the first is migration of paranasal sinus inverted papilloma cells via the Eustachian tube, as suggested by the considerable percentage of patients with an ipsilateral paranasal sinus inverted papilloma.<sup>4</sup> In our patient, imaging showed no signs of nose and paranasal sinus involvement. Post-operative nasal endoscopy showed no signs of an intranasal mass either. Thus, development of papilloma in our patient may be explained by the second (an abnormality of embryonic migration of ectopic Schneiderian membrane into the mucosa of the middle ear) or third hypothesis (chronic otitis media stimulating the development of Schneiderian mucosa).<sup>4</sup>

Otorrhea and hearing loss are the main symptoms associated with inverted papilloma of the middle ear.<sup>3,8</sup> Additionally, Pou *et al.* reported its frequent association with persistent middle ear effusion and ipsilateral sinonasal tumors.<sup>11</sup> Although middle ear papilloma may be different from sinonasal papilloma in terms of epidemiology and pathology, both are locally aggressive, have a high tendency to recur and are associated with malignancy.<sup>4,9,10</sup> The malignant transformation rate for middle ear papilloma is 35.3%, higher than that observed in paranasal sinus inverted papilloma and the recurrence rate is 56.25%.<sup>4</sup> The difference may be attributed to the difficulty of complete resection of inverted papilloma in the complex middle ear and mastoid cavity compared to the large paranasal sinuses.<sup>4</sup> It is also worth noting that recurrence and malignant transformation is usually observed in cases with which sinonasal inverted papilloma was initially diagnosed and managed prior to middle ear involvement. In the summary of case reports by Kainuma *et al.*, recurrence is almost always noted in cases of simultaneous sinonasal and middle ear papilloma.<sup>11</sup> On the other hand, primary inverted papilloma of the middle ear showed no recurrence except for two cases wherein the surgeon did conservative surgeries prior to radical mastoidectomy.<sup>11</sup> Hence, we can only assume that middle ear papilloma with sinonasal involvement has more aggressive features compared to primary middle ear inverted papilloma and requires more meticulous follow-up surveillance. Still, its similarities to cholesteatoma in terms of presentation, clinical course, local aggressiveness and morphology makes the diagnosis difficult and is only confirmed on final histopathology after the surgery.

One dilemma in middle ear papilloma is the management. Although all would agree that surgery is the mainstay of treatment, it is difficult to define a standard surgical strategy based on limited data available in the literature.<sup>4</sup> Current recommendations include complete surgical excision followed by long term surveillance for newly diagnosed cases, while post-operative radiotherapy is reserved for recurrent disease



and tumors with malignant transformation.<sup>4,8,10,11</sup> However, it is still necessary to conduct further investigations to come up with a rational and standard approach in the management of this rare condition.

In summary, middle ear inverted papilloma is a rare disease entity. Awareness of this rare disease makes us more careful in the management of benign conditions such as cholesteatoma of the middle ear that may present similarly. Further investigations are required to identify the risk factors and its etiology and to come up with a rational approach to its management. Complete excision is necessary but performing extensive surgery such as temporal bone resection or post-operative radiotherapy are still controversial. Post-operatively, its high recurrence rate and possibility of malignant transformation require close monitoring and should include repeated otoscopic examination and imaging procedures such as CT scan or MRI.

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