Inverted papilloma of the nasal cavity is a benign neoplasm, although it can be locally invasive and has the potential for malignant degeneration. Inverted papilloma of the temporal bone is extremely rare. We describe a case of a 44-year-old woman who was treated for nasal inverted papilloma and was later found to have inverted papilloma of her temporal bone. The patient required several procedures to remove the inverted papilloma from the nasal cavity and temporal bone, and she is currently free of recurrence.

Inverted papilloma (IP) is the second most common benign neoplasm of the nasal cavity, though it is relatively rare with a reported frequency of 0.6 to 1.5 per 100,000 per year. It is more common in men than women, and it usually occurs in the fifth to sixth decades of life (1). Nasal IP originates from the Schneiderian mucosa of the sinonasal tract and is most commonly attached on the lateral nasal wall (2). At the temporal bone, IP is exceedingly rare and much less is known about this entity than nasal IP. Only 32 cases of IP involvement of the temporal bone have been reported in the literature (3–6).

**CASE DESCRIPTION**

A 44-year-old woman presented with nasal obstruction, and a right nasal mass consistent with IP was found on exam. The patient underwent endoscopic resection of this lesion in 2009 and was found to have multifocal IP located on the right middle turbinate, the right posterior nasal septum, and the left torus tubarius. The patient was reported to have persistent disease on the left torus tubarius; she was subsequently lost to follow-up. She presented 21 months later with new right-sided conductive hearing loss and underwent myringotomy with tympanostomy tube placement. At the time of surgery, a fleshy mass was biopsied from the middle ear that was consistent with IP with severe dysplasia. Nasal endoscopy at that time indicated recurrent disease on the right side, without involvement of the eustachian tube orifice intranasally.

The patient underwent right middle ear exploration and was found to have tumor encroaching into the eustachian tube orifice. She also underwent directed biopsies endoscopically of suspicious nasal lesions. Specimens from the middle ear and nasal cavity revealed IP and no malignancy. Definitive endoscopic resection was performed on the patient’s nasal lesions, at which point the margins were clear of papilloma by final pathology. Four months after this resection, additional middle ear exploration was performed, and disease extending into the eustachian tube orifice with focal high-grade dysplasia was found (Figures 1 and 2). The IP in the middle ear was completely grossly removed with forceps and potassium titanyl phosphate laser. The patient had a 30 pack-year history of smoking, and at this time, she stopped smoking and switched to electronic cigarettes. She currently has an anterior tympanic membrane perforation allowing for office surveillance. Genotyping for human papillomavirus (HPV) was negative in the nasal tissue but positive for HPV 11 in the middle ear space.

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**Figure 1.** T1 MRI post-contrast demonstrating mild enhancement of the lesion in the right middle ear encroaching on the eustachian tube (white arrow) adjacent to the carotid artery (black arrowhead).
At 3-year follow-up from her last procedure, the patient showed no evidence of tumor persistence or recurrence in the nose or the ear. Follow-up magnetic resonance imaging (MRI) at that time was also negative for disease. The current plan of treatment involves close observation with serial MRI and endoscopic surveillance of the nasal cavity and middle ear. The patient has been discussed at the multispecialty tumor board for possible radiation in the future should her disease develop malignant transformation.

**DISCUSSION**

Middle ear involvement with IP is an unusual finding that is rarely reported in the literature. There are three theories for the development of temporal bone IP: direct spread of tumor through the eustachian tube, conversion of ectopic rests of Schneiderian membrane in the temporal bone, and embolic seeding of tumor cells outside the sinonasal tract. The most common presenting symptoms of temporal bone IP are hearing loss and otorrhea. Middle ear IP is associated with an increased risk of malignancy, particularly if the lesions are recurrent. A recent series of 32 patients with temporal bone IP reported invasive carcinoma in 28% of patients and carcinoma in situ in 16%.

The middle ear lesion in this case demonstrated HPV 11 positivity. Nasal IPs have been associated with this and other HPV isotypes. However, HPV 16 and 18 have been more classically associated with malignancy. A recent meta-analysis of 31 case-controlled studies showed statistically significant association of high-risk HPV subtypes 16 and 18 with malignant sinonasal IP—when type 18 having a stronger association with malignancy than type 16. The role of HPV in the development of IP has yet to be elucidated. Additionally, the clinical significance of isolating HPV in the middle ear specimen but not in the nasal specimen is unclear. Roh et al performed a retrospective review of 54 patients and found no significant difference in HPV status and rate of recurrence; in the same review, there was an association between smoking and IP recurrence, although this finding was not statistically significant. Since our patient has stopped smoking, she has experienced no additional recurrence of her IP.

The primary treatment modality for this benign disease is surgical. Mitchell et al reported a combined lateral and anterior skull base approach for extirpation of the eustachian tube in order to definitively manage malignant transformation of IP in this area. Complete excision of the tumor is important to reduce the risk of recurrence. Recurrence rates with mucosal stripping have been measured at 52.2%, whereas drilling the tumor base, cautering the tumor base, and complete excision have recurrence rates of 4.9%, 4.7%, and 0%, respectively. Recurrence at the tumor base is thought to be caused by rests of abnormal epithelium or tumor within the bone. Radiation, though not used routinely in most cases of IP, has been used as adjuvant therapy in certain cases. To date, there are no definitive recommendations regarding the role of radiation in treating this rare entity.

In this case, the authors will continue close observation and serial imaging. They have had thorough discussions with the patient regarding adjuvant radiation due to the recurrent nature of her tumor with severely dysplastic features. This case underscores the need for further data on this topic.


**Figure 2.** Hematoxylin and eosin stains demonstrating (a) middle ear inverted papilloma with moderate to severe dysplasia (100×) and (b) numerous mitotic figures within middle ear inverted papilloma (400×).