Management of Inverted Papilloma of the Temporal Bone

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Abstract

Objective:
1. Understand the clinical presentation, etiology, and histopathology of inverted papilloma of the temporal bone.
2. Discuss current treatment and management of inverted papilloma of the temporal bone as well as considerations for future management.

Methods:
Inverted papilloma (IP), a benign neoplasm typically arising within the sinonasal tract, has rarely been reported to occur outside the sinonasal tract. Only seven cases of temporal bone IP have been reported. We reviewed our two cases of temporal bone IP and discuss their management.

Results:
Our first case was a patient with a history of sinonasal IP who had undergone three previous endoscopic resections when she presented with a subepithelial mass extending from the posterior wall of the ear canal. She was found to have extensive bony destruction and underwent a retrobulbarine approach with tympanomastoid obliteration. The resection was monitored with both baseline and follow-up PET-CT scans. The second case was a primary lesion with an atypical osteoclastic appearance of a granular mass extending from the middle ear through a tympanic membrane perforation. This was managed with a tympanomastoidectomy and clinically followed for recurrence.

Conclusion:
Temporal bone IP is a rare neoplasm with no definitive treatment algorithm. We recommend aggressive surgical resection with close postoperative follow-up. PET-CT may be useful for identifying multicentric disease at presentation or for early identification of recurrent disease. HPV detection may identify IPs at risk for multicentric or recurrent disease.

Introduction

Inverted papilloma is a benign neoplasm arising from ectodermal speech derived sinonasal mucosa of the lateral nasal wall. Although benign, inverted papillomas are often locally aggressive, multicentric, and can be associated with bony destruction and extension into the paranasal sinuses, orbits, and anterior skull base. Rarely, inverted papillomas arise at extra-sinosal tract sites involving the pharynx, larynx, and temporal bone. Only 18 cases involving the temporal bone, including primary and secondary disease, have been reported since 1987.1-14 We present two additional cases of extra-sinosal disease and an overview of their clinical presentation and surgical management. Furthermore, we review the role of HPV screening in extra-sinosal disease and discuss the potential for baseline screening.

Case Presentations

Case #1: A 49-year-old woman with a past history of sinonasal inverted papilloma status post three endoscopic resections presented with an eight month history of progressive right-sided hearing loss. She further reported right-sided otalgia, tinnitus, aural fullness, vertigo, and new onset right-sided facial twitching. Physical examination revealed right-sided facial myoclonus, facial nerve paresis (House Brackmann Scale II/VI) and a bluish, blanching mass protruding from the posterior external auditory canal anteriorly, completely obstructing visualization of the tympanic membrane.

CT Scan of the Temporal Bone and MRI (Figures 1A & B) demonstrated an expansive mass filling the middle ear cavity with bony erosion of the external auditory canal and the intratemporal facial nerve.

The patient underwent a right radical mastoidectomy and decompression of the facial nerve with tympanomastoid obliteration using abdominal fat. Gross total resection was achieved. The patient did well postoperatively with complete return of facial nerve function. Post-operative CT Temporal Bone, MRI and PET/CT were negative for persistent or recurrent disease.

Case #2: A 76-year-old man with a fifteen-year smoking history was referred for evaluation of right-sided chronic otitis media. The patient reported a two year history of decreased hearing and tinnitus in his right ear but denied otalgia, otorhea, vertigo, trauma, or prior otologic surgery.

Microscopic examination of the right ear revealed a small posterior perforation of the tympanic membrane, through which there was extension of polypoid tissue with adjacent neovascularization. An audiogram demonstrated a moderately severe to profound mixed hearing loss with 88% discrimination on the side of the lesion. A CT scan was obtained (Figure 2) demonstrating soft tissue density filling the middle ear cavity. The patient underwent a tympanomastoidectomy and final pathology was consistent with inverted papilloma. A flexible fiberoptic exam failed to reveal intranasal disease or other potential sites of primary disease.

The patient did well post-operatively and was clinically free of disease on physical exam and on repeat CT scan one year post-operatively.

Discussion

Inverted papillomas (IP) are a well-defined group of benign neoplasms typically arising within the sinonasal tract but only 18 cases of temporal bone involvement have been reported.1-14 There are several hypotheses regarding etiology. The first is the transmission of primary disease through the eustachian tube to the middle ear2 however recurrent disease at the primary site or contiguous spread of disease has never been shown. A second hypothesis suggests that possible ectopic foci of Schneiderian mucosa, a result of an error during embryological development, are the root cause.3 A third hypothesis proposes a genetic predisposition to account for the disease’s predilection for multicentricity.4 Another proposal suggests influence of progesterone receptors in rapid growth of IP of the temporal bone5 although in both of our reported cases immunostains for progesterone receptors were negative.

Current literature suggests an association between human papilloma virus (HPV) and IP, however only two cases of the eighteen reported temporal bone cases have shown definite HPV involvement.12,17,20 One hypothesis is a “hit and run” phenomenon in which HPV may be the inciting event of transformation, but is not necessary for maintenance of the neoplastic changes.21 Another author suggested a difference in sensitivity for HPV detection between polymerase chain reaction (PCR) and in situ hybridization (ISH).22 However, there is conflicting data on which technique is superior.22 Neither of the cases we report stained positive for HPV types 6, 11, 16, and 18 by ISH.

The clinical presentation of IP of the temporal bone is varied. While IP of the sinonasal tract predominantly affects men, IP of the temporal bone occur with a female-to-male ratio of 2:1.14 Of the eighteen cases reported, ten were found to be primary disease. In addition to Case #2 above, six of these previously reported cases were associated with chronic otitis media with complaints of hearing loss in the affected ear.5,9 There is an 82% risk of recurrence in the middle ear if adequate margins are not achieved and thus aggressive surgical resection is the preferred method of treatment.1,2,12,13,24 In addition to resection, postoperative radiation therapy has been previously discussed as an option for aggressive lesions of the temporal bone however there is a lack of evidence to support its efficacy.9

Literature regarding post-surgical surveillance for recurrent disease is scant. In a study of four patients with positron emission tomography-computed tomography (PET-CT) scanning, F-18 fluorodeoxyglucose (18FDG) avidity was a reliable indicator of the presence of recurrent disease. Two other studies with small sample sizes showed a potential role for PET-CT in differentiating benign sinonasal inverted papillomas from those with coexistent malignancy.26,27 In one of our reported cases, PET-CT displayed 18FDG avidity at the site of tumor recurrence, suggesting that PET-CT may in fact be an appropriate test for surveillance of extra-sinosal disease, however lack of supporting data prompts further investigation of the subject.

Conclusions

Despite the exceedingly rare presentation of inverted papilloma of the temporal bone, we recommend patients with sinonasal inverted papilloma and otologic complaints undergo work-up for possible temporal bone involvement. We recommend aggressive surgical resection and PET-CT for identifying multicentric disease at presentation as well as for follow-up and early identification of recurrent disease. In regards to HPV detection, the virus’ role remains unclear; however, given the increasing incidence of HPV related head and neck cancer, we recommend both ISH and PCR analysis in order to increase chance of detection.

References


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