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Multifocal inverting papilloma of the sinonasal cavity and temporal bone

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ABSTRACT

Introduction: Inverting papillomas (IPs) represent the most common benign neoplasm of the sinonasal cavity and are known for local invasion, proclivity for recurrence, and risk of malignant transformation. IP of the temporal bone (TBIP) is exceptionally rare, with 32 reported cases. We present a new case of multifocal IP of the sinonasal cavity and temporal bone.
Methods: Case report and review of the literature.
Results: A 45-year-old man presented with a left sided biopsy proven IP and associated left sided hearing loss. Imaging demonstrated a left sided nasal mass and a separate non-contiguous soft tissue mass filling the left middle ear without involvement of the eustachian tube. He underwent an endonasal endoscopic gross total resection of the sinonasal lesion and biopsy of the middle ear mass with pathology showing IP. He subsequently underwent a left sided transtemporal resection of the TBIP. Review of the literature, revealed 32 TBIP cases, with 59% having history of associated sinonasal IP and 41% with isolated temporal bone disease. Over half of the patients demonstrated recurrence. In comparison to patients with history of sinonasal IP, isolated TBIP occurred in younger patients, was more common in females, and had less association with HPV and malignant transformation.
Conclusion: TBIP is extraordinarily rare and usually presents with a history of sinonasal IP. Isolated TBIP may be a distinctly different disease process. Disease recurrence is common and risk of malignant transformation is present, so aggressive initial surgical treatment with gross total resection is advocated.

1. Introduction

Inverting papilloma within the temporal bone (TBIP) is extremely rare with only 32 reported cases in the literature [1]. Sinonasal inverted papilloma (IP) is a benign, but locally aggressive neoplasm, which most often originates from the lateral nasal wall and represents between 0.5% and 4% of all sinonasal tumors [2]. There is a risk for transformation into squamous cell carcinoma and recurrence is common [3–7]. TBIP has a higher rate of transformation to SCC at 36% compared to IP within the nasal cavity which has been estimated to have rates between 5% and 21% [8].

Involvement of the temporal bone is hypothesized to occur via several mechanisms including transmission of cells via the eustachian tube, direct extension through the eustachian tube, iatrogenic implantation or seeding, or from stimulation or conversion of residual Schneiderian mucosa within the middle ear by such triggers as chronic otitis media [1,9]. Of the 32 cases previously reported, 13 had isolated TBIP without sinus involvement [1]. The mean age of presentation was 52 and most commonly patients presented with hearing loss and otorrhea [1]. We present a case of TBIP in a patient with concurrent IP of the sinonasal cavity.

2. Case report

A 45-year-old man presented with two-year history of left-sided nasal obstruction, anosmia and diminished hearing. The referring

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community ENT had obtained a biopsy of a left-sided nasal mass, which was consistent with IP. A computed tomography (CT) scan demonstrated a large mass filling the entire left nasal cavity, ethmoid, and sphenoid sinuses, extension into nasopharynx with thinning of the medial orbital wall and cribiform plate. Magnetic resonance imaging (MRI) demonstrated characteristic findings of IP including frond-like heterogeneous enhancement of the lesion (Fig. 1A and B). Additionally, a similar soft tissue mass was seen in the left middle ear surrounding the ossicles (Fig. 1C, E, F). The nasal mass appeared separate from the soft tissue mass filling the left middle ear, and there was no involvement of the Eustachian tube (Fig. 1D).

The patient underwent an endonasal endoscopic gross total resection of the sinonasal tumor. The lesion was pedicled off of the posterior superior septum and did not extend into the Eustachian tube (Fig. 2). The mass abutted but did not erode through the cribiform plate or involve the mucosa of the anterior skull base, and final pathology was consistent with inverted papilloma having high-grade dysplasia (Fig. 4B). At the end of the endonasal endoscopic surgical resection, the left ear was examined and myringotomy performed revealing a polypoid mass within the middle ear space. A biopsy of this mass was obtained and demonstrated IP. The patient subsequently underwent left transotic resection of the extradural TBIP approximately two months after his sinonasal resection, allowing him time to heal from his endonasal operation and come to terms with the multifocal nature of his disease (Fig. 3). Unfortunately, in these intervening two-months, the patient had progression of disease in his temporal bone with new symptoms of worsening hearing and vertigo/disequilibrium. To remove the entire lesion, which was found to extend into the vestibule and cochlea, the ear canal was closed, a fallopian bridge technique was utilized, and complete labyrinthectomy was performed. The Eustachian tube was also dissected 6 mm from its protympanic orifice prior to obliteration; here biopsies obtained that were negative for IP. Final pathology of the main tumor mass demonstrated inverted papilloma with high-grade dysplasia (Fig. 4A), similar to the specimens previously obtained from the nose. The patient has healed well from his operation and has been disease free for 9-months.

3. Discussion

Ward first described sinonasal inverting papilloma in 1854. The pathogenesis of the disease, however, remains largely unclear. Historically, risk factors for IP were smoking, allergy, occupational exposures, or viral infection, particularly HPV [10–12]. Although extensively studied, HPV infection as an etiology is still controversial. A recent study by Roh and colleagues argues against its involvement. This study utilized PCR to target HPV DNA from sinonasal IP specimens, which was then genotyped. They found only 14.8% of their study participants tumors contained HPV DNA, and none of the HPV positive patients were found to have recurrence with mean follow-up of 34.1 months [13]. Their study group population contained 13% smokers. Interestingly, of the patients with recurrent IP, 42.9% of them were smokers – suggesting that smoking may be a greater risk factor than HPV [13]. A recent meta-analysis examined HPV infection and risk for malignant transformation in sinonasal IP and found a significant association between the two, especially with those infected with HPV-18 types [14].

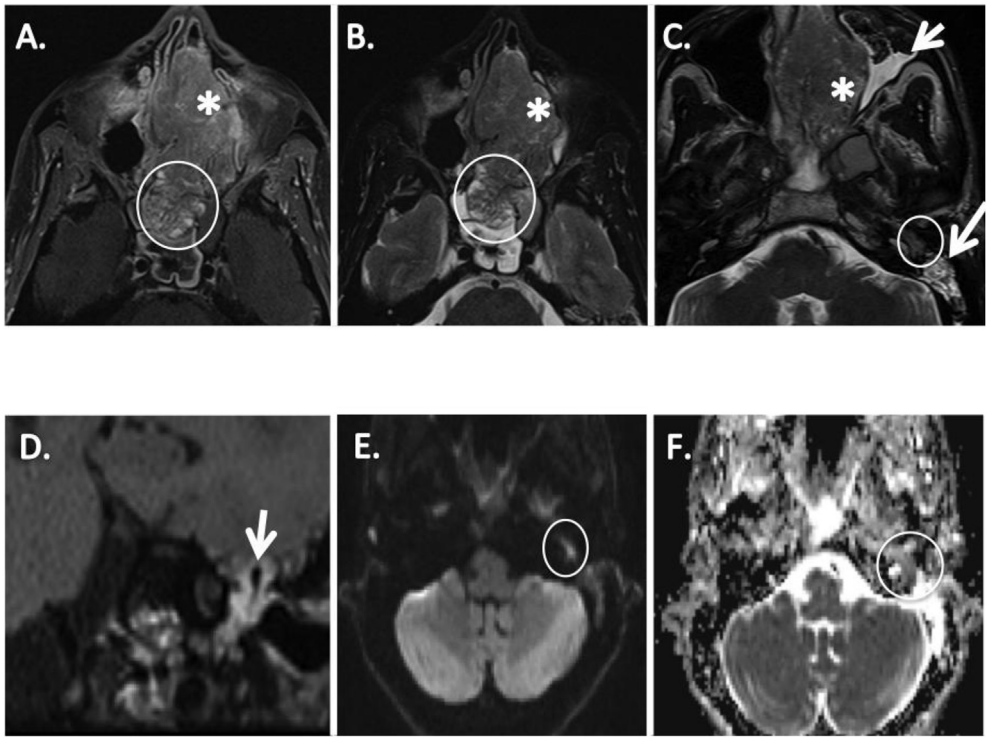

Fig. 1. A. Axial T1 MRI with contrast and B. Axial T2 MRI demonstrate a large intranasal mass (asterisk). A portion of the mass at the sphenoid sinuses (circle) demonstrates frond like heterogeneous enhancement with a "mini-brain" appearance characteristic of inverted papilloma. More anteriorly the heterogeneity is lost consistent with pathologic findings of degeneration to high grade dysplasia. C. Axial T2 MRI demonstrating the large intranasal mass (asterisk) and trapped secretions in the left maxillary sinus (short arrow) as well as effusion of the left mastoid (long arrow), inverted papilloma (circle) with a more dark appearance. D. Coronal T1 MRI with contrast enhancing inverted papilloma throughout the middle ear cavity, surrounding the ossicles (tip of arrow). E. DWI shows bright signal (circle) and ADC map (F.) Show dark signal (circle) indicative of restricted diffusion from tumor cellularity in the middle ear.