CASE REPORT

Varied Presentations of Inverted Papilloma

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Abstract

Inverted papillomas form about 0.4 to 5% of nasal tumors. They usually present as unilateral firm, bulky, red and vascular masses. We present three cases which were initially diagnosed as a maxillary mucocele, pansinusitis and antrochoanal polyp based on clinical and radiological findings. It is important to keep the diagnosis of inverted papilloma in mind in unilateral diseases of the paranasal sinuses as a more radical approach is required.

Keywords: Inverted papilloma, maxillary mucocele, pansinusitis, antrochoanal polyp.

INTRODUCTION

Sinonasal papillomas are benign neoplasms of the pseudostratified ciliated columnar epithelium that lines the nasal cavity and paranasal sinuses originating from the schneiderian membrane. The first case was described by Ward in 1854. However, it was Ringertz in 1938 who described the histologic characteristic of inversion of this lesion through the surface epithelium. Although uncommon in the general population, papillomas comprise between 0.4% and 5% of primary nasal tumors. The peak age of presentation is the 5th and 6th decades, and there is a strong male preponderance.[1] Histologically, inverted papillomas are characterized by an invagination of neoplastic epithelium into the underlying stroma, without transgression of the basement membrane. The prototypical sites of occurrence are along the lateral nasal wall, particularly in the middle meatus. The etiology of inverted papillomas however, remains poorly understood.[2] Proposed causes include allergies, chronic sinusitis, airborne pollutants, and viral infection. Human Papilloma Virus (HPV) is an epitheliotropic virus that has recently been found to be associated with the occurrence of inverted papillomas.[3,4] Despite being benign, *Inverted papillomas* may demonstrate aggressive local invasion typically presenting with unilateral nasal obstruction, rhinorrhea, and facial pain or pressure.[5,6] The other important characteristic of inverted papillomas is their association with squamous-cell carcinoma (SCC). The incidence of malignant transformation, variably reported as being between 5% and 21%.[1]

CASE REPORTS

Case 1

A 35 years old Malay gentleman presented to the ENT Department with complaints of persistent right sided nasal block associated with mucopurulent discharge of 2 months duration. On endoscopic examination, the right middle turbinate was flushed to the lateral wall with pus oozing from the middle meatus. We proceeded with a computed tomographic scan which showed that the entire right maxillary cavity was filled with polyps and that its medial wall was thinned out and pushed medially as indicated by the alphabet 'A' in the scan and was reported as a right maxillary mucocele (Fig. 1).

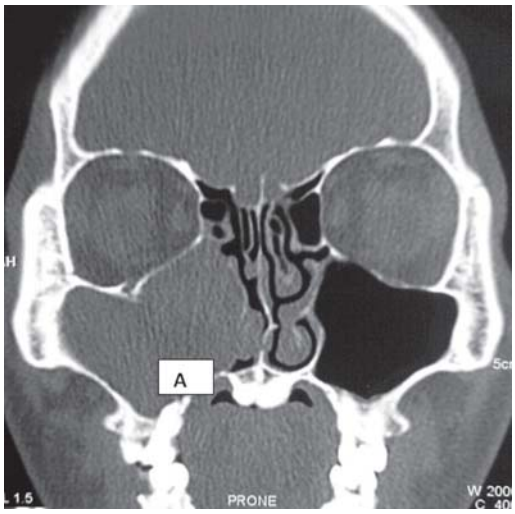

Figure 1: Right maxillary mucocele

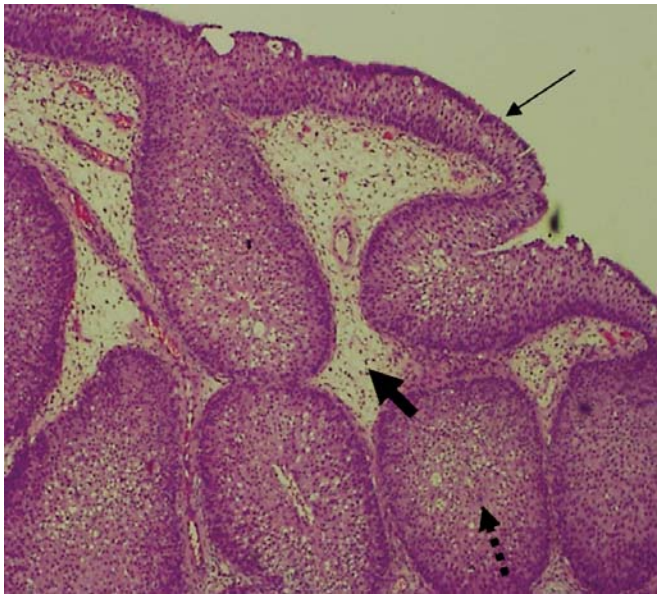

Figure 2: Hematoxylin and eosin stain (magnification X10). Thin arrow showing the surface epithelium, large arrow showing the stroma and dotted arrow showing the nest of squamous epithelium

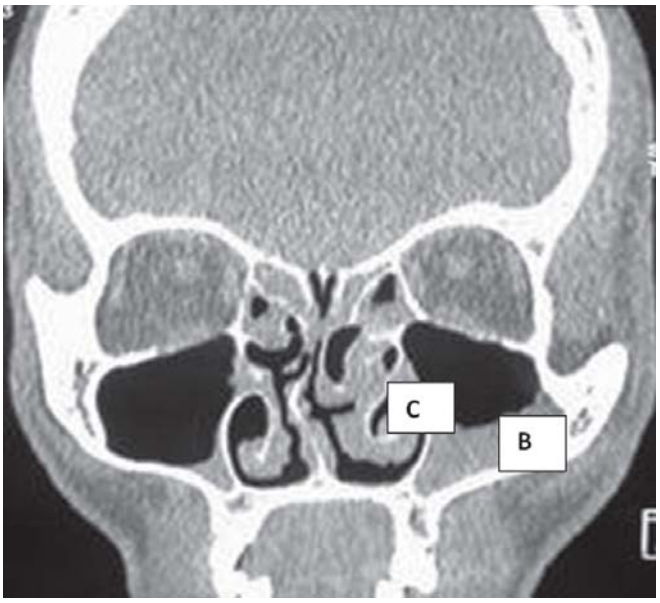

Figure 3: Pansinusitis with left concha bullosa

He was planned for endoscopic sinus surgery. Intraoperatively we found that the entire right maxillary cavity was filled with friable polypoidal mucosa. Histological section showed invaginating nests of benign squamous epithelium in the stroma, hence it was interpreted as features suggestive of *Inverted papilloma* (Fig. 2). Therefore, he was planned for a second surgery which was a medial maxillectomy at which tumor clearance was achieved**.** He has since then been on regular follow-up and free of disease.

Case 2

A 42 years old Malay gentleman presented to our clinic with history of a right sided submandibular swelling with odynophagia of one month duration. He however, had no nasal symptoms. On endoscopic examination it was noted that he had minimal polyps in the left middle meatus. We proceeded with a CT scan which revealed mucosal thickening in both maxillary, ethmoidal, frontal and sphenoidal sinuses with blocked osteomeatal complexes indicated by the alphabet 'B', also seen was pneumatization of the left middle turbinate marked by the alphabet 'C' hence it was reported as Pansinusitis with a Left Concha Bullosa (Fig. 3).

He was then planned for functional endoscopic sinus surgery with right submandibular gland excision. Intraoperatively we noted polypoidal mucosa over the left bulla ethmoidalis and maxillary ostium which was cleared totally. The intranasal histopathological section was interpreted as *Inverted papilloma* and the right submandibular gland showed inflammatory changes. On follow-up, the endoscopic examination revealed no evidence of residual disease and since then he is disease free.

Case 3

A 59 years old Indian gentleman came to see us with complaints of persistent left sided nasal block not resolving with medical treatment. Endoscopic examination revealed extensive polyps within the left nasal cavity. We proceeded with a CT scan which showed that the left maxillary sinus was completely filled with polyps with extension into the left nasal cavity was reported as Left Antrochoanal Polyp as indicated by the alphabet 'D' (Fig. 4).

He underwent left endoscopic medial maxillectomy due to the intraoperative findings where the left middle meatus was occluded by polyps but the left maxillary cavity was filled with friable tissue which was adherent to the sinus walls. Histopathological section was interpreted as *Inverted papilloma.* His follow-up over the last one year has been regular with no evidence of recurrence.

DISCUSSION

Inverted papillomas despite their benign histological appearance, often demonstrate aggressive local invasion. Due to the aggressive nature of these lesions, their tendency to erode bone and the possibility of extranasal extension, complete wide local resection is recommended either through

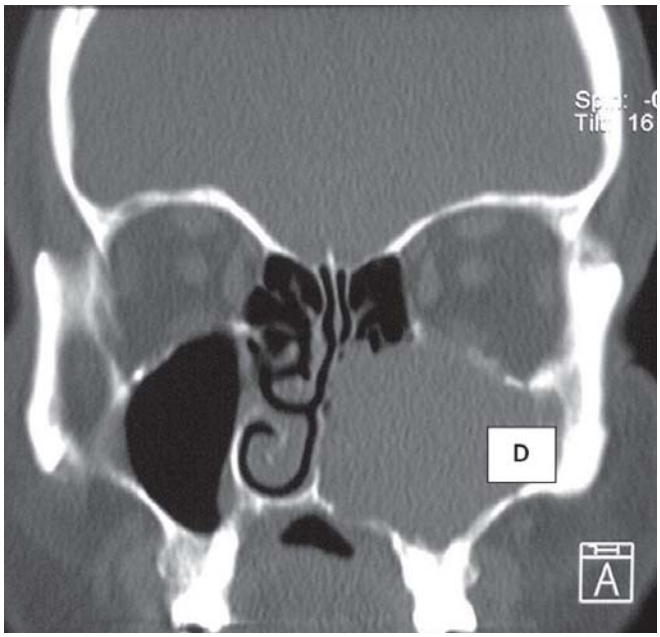

Figure 4: Left antrochoanal polyp

a lateral rhinotomy or an endoscopic approach. A recent meta-analysis and another systematic literature review support endoscopic approach as a favorable treatment option compared with open approaches.[7,8]

Radiography is important in the evaluation of patients with inverted papilloma as it aids in the planning of type of surgery required. On computed tomography, an inverted papilloma appears to have soft-tissue density and enhances heterogeneously with contrast.[9] Thinning or bowing of the bony walls of the sinus is common, and sometimes entrapped or remodeled bone within the tumor can appear as calcifications. These radiographic features are indistinguishable from inflammatory polyps with entrapped debris, making it difficult to accurately diagnose or define the tumor extent as seen in the above three cases. It allows one to assess the extent of disease and the presence of bone erosion or invasion into adjacent structures such as the base of skull or the orbit. These changes may suggest the presence of an associated malignancy, thus altering the management. Although CT scanning has improved the ability to plan surgical resection of *inverted papilloma*, it has a sensitivity of only 69% and specificity of 20%.[10] MRI is an alternative study that is superior to CT scanning in distinguishing papillomas from inflammation and for providing better delineation of the lesions in contrast to surrounding soft tissue. Hence it should be kept in mind that a differential diagnosis of inverted papilloma is lurking behind that conspicuous looking polyp. Conventionally, all unilateral polyps and suspicious-looking lesions need histologic evaluation, especially since the unexpected diagnosis of *inverted papilloma* may occur in apparently normal bilateral polyps. The frequency of this event has been shown to be varying between 0.00% and 0.92%.[11]

Recurrence following surgery for inverted papilloma depends upon tumor location, tumor extent, histology, method of removal, follow-up, demographic and social factors. However, the main reason still remains the method of removal. This is directly related to the completeness of tumor excision, with many recurrent cases actually representing residual disease. This often results from failure to obtain a biopsy sample before surgery, biopsy of the accompanying polypoid disease and not the tumor proper or incorrect diagnosis of the biopsy specimen as inflammatory disease, which leads to limited and subtotal removal. In these cases a second stage surgery should be carefully planned with the aim of complete resection.

CONCLUSION

Inverted papilloma is a benign sinonasal lesion that most commonly arises on the lateral nasal wall with a high incidence for recurrence and local aggressiveness. Appropriate preoperative assessment includes clinical and radiological evaluations, which provide an accurate picture of the lesion. Complete removal of the lesion offers the best chance of minimizing recurrence; therefore cases should be carefully selected for either endoscopic removal or open approaches. Due to the chance for delayed recurrences and the 5 to 21% incidence of malignant transformation, these patients require careful, long-term follow-up. The key to success is locating the origin of the tumor, defining its extent, and completely removing all diseased mucosa with a surrounding rim of normal mucosa.

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