Inverted Papilloma with Focal Respiratory Epithelial Adenomatoid Hamartoma: A Case Report of Interesting Histopathologic Findings

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INTRODUCTION

Inverted papilloma (IP), first described in 1854, is a rare sinonasal tumor mainly affecting adults in their 5th decade, characterized by local aggression, high recurrence rates, and potential association with carcinoma. In 1995, Wenig and Heffner described respiratory epithelial adenomatoid hamartoma (REAH), a benign sinonasal neoplasm with glandular proliferation lined by ciliated epithelium.

IP and REAH share many non-specific sinonasal symptoms such as nasal obstruction, rhinorrhea, epistaxis, facial pain, and headache, but REAH is more commonly associated with olfactory dysfunction (hyposmia or anosmia).

The coexistence of IP and REAH poses a diagnostic challenge given their overlapping clinical and histological features. This report describes a rare simultaneous occurrence, offering unique diagnostic and clinical insights.

CASE PRESENTATION

A 52-year-old male, presented with a greater than 5-year history of left-sided nasal congestion, mouth breathing, intermittent left-sided rhinorrhea, and postnasal drainage. He reported persistent pressure over the left maxillary sinus and intermittent waxy, white nasal discharge from the left side. He denied any changes in taste or smell.

EXAMINATION AND IMAGING FINDINGS

- Nasal endoscopy revealed a grade 3 polyp extending from the left maxillary ostium with fingerlike mucosal projections.
- Pre-operative CT findings show moderate mucosal thickening in the left maxillary sinus with narrowing of the accessory ostium, a polyp in the left posterior nasal cavity extending into the nasopharynx, mild mucosal thickening in the right maxillary sinus, and a leftward deviated nasal septum with a spur abutting the left inferior turbinate (Figure 1 and 2).

DIFFERENTIAL DIAGNOSIS

- Inflammatory polyps, antrochoanal polyps, IP, juvenile nasopharyngeal angiofibroma, fibrous dysplasia, sinonasal squamous cell carcinoma and other malignancies.

OPERATIVE MANAGEMENT

- The patient underwent septoplasty and left endoscopic medial maxillectomy with maxillary sinus lesion resection. The tumor's attachment site was identified, and the lesion was completely excised with intraoperative frozen section confirming IP. No evidence of hyperostotic bone on intraoperative assessment, concordant with preoperative CT imaging.

HISTOPATHOLOGY

- Fragments of IP with focal REAH in the left maxillary sinus, while the left inferior turbinate was negative for tumor.

FIVE MONTH FOLLOWUP

- The patient demonstrates clinical improvement with no evidence of disease recurrence on endoscopic examination, with planned surveillance in three months.

CASE PRESENTATION

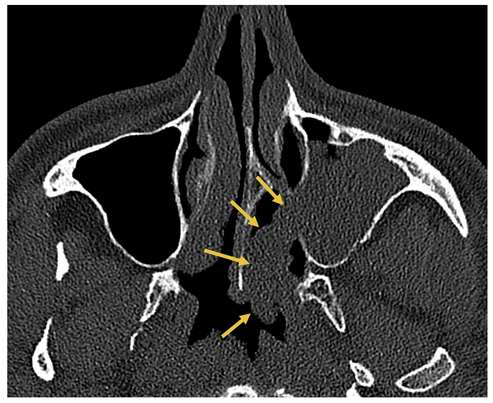

Figure 1. Axial CT in the bone window shows a polypoid mass in the left maxillary sinus and nasal cavity extending into the nasopharynx (arrows); with no aggressive osseous erosion, suggesting a benign etiology.

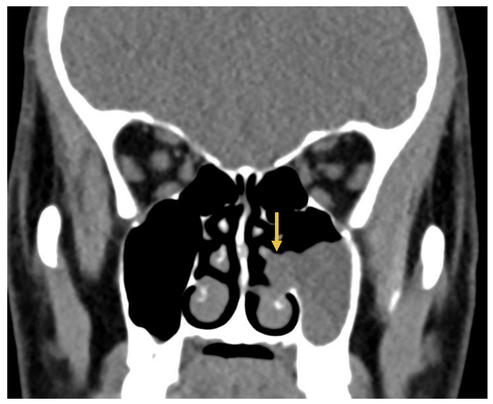

Figure 2. Coronal CT in the soft tissue window confirms the same polypoid mass involving the left maxillary sinus and nasal cavity (arrow), with no osseous stalk identified.

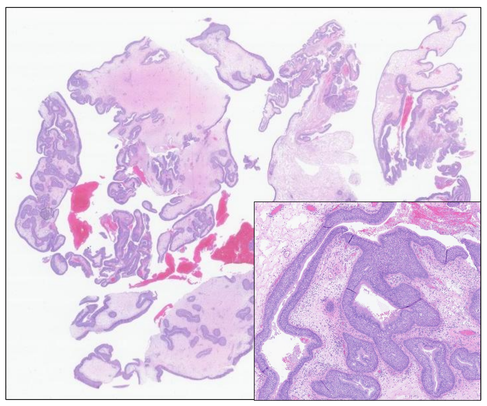

Figure 3. H&E-Stained Section of Left Maxillary Sinus (Inset shows IP component).

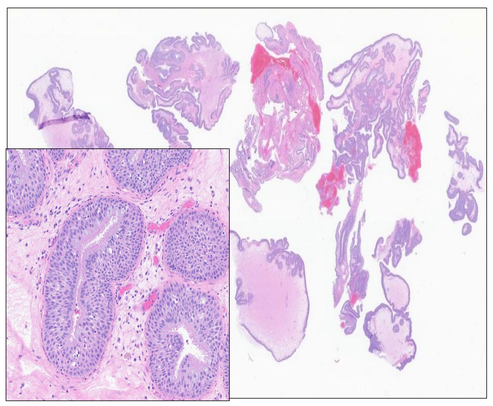

Figure 4. H&E-Stained Section of Left Maxillary Sinus (Inset shows REAH component with thickened basement membrane and ciliated epithelium)

DISCUSSION

IP typically appear endoscopically as exophytic, polypoid masses, and more vascular than an inflammatory polyp. IP arises from invaginated squamous epithelium, most commonly on the lateral nasal wall, and has malignant potential, whereas REAH originates from the olfactory cleft, with benign single-layer respiratory epithelium, and no reported malignant transformation.
While both may share surface respiratory epithelium, histologic features such as proliferating epithelial thickening with mucocytes, mucous cysts, and inflammation favor IP [1]. CT imaging reveals bone remodeling and focal hyperostosis for IP [2] and olfactory cleft opacification for REAH.

Sarioglu (2007) reviewed 38 specimens from 26 cases and found 3 with overlapping IP and REAH features [3], challenging traditional classification and complicating diagnosis. Recent meta-analysis reported that endoscopic resection of IP has a 12.8% recurrence rate, lower than external approaches (16.6%) [4], whereas REAH shows excellent prognosis after endoscopic resection, with no recurrences over 27.2 months follow-up [5]. Regular post-operative endoscopic exams are essential for detecting recurrence, which typically occurs within the first two years.

CONCLUSION

This case represents one of the few reported instances of IP and REAH coexisting in a single paranasal sinus within our retrieval capability. It underscores the need for careful pathological review to prevent misdiagnosis, as IP requires aggressive treatment and long-term surveillance, while REAH needs simple excision with rare recurrence. Accurate detection and differentiation of sinonasal pathologies is crucial for proper management.

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