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Respiratory Epithelial Adenomatoid Hamartoma- A Case Report

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Abstract: ***Background: Respiratory Epithelial Adenomatoid Hamartoma is a rare benign neoplasm which often clinically mimics a malignant lesion. Accurate histopathological interpretation, supported by close clinicopathological correlation, is essential to minimize misdiagnosis, prevent overtreatment, and enhance overall patient outcomes. Objective-To present the case of 66-year-old male patient who presented with nasal obstruction highlighting clinical, radiological and histopathological information. Result - Radiological and clinical features were suggestive of a malignant lesion. Histopathology together with supportive IHC for CK 5/6 and p63 lead to the diagnosis of REAH. The lesion was completely excised, and the patient remained asymptomatic on follow up. Conclusion: The close resemblance of REAH to low-grade sinonasal adenocarcinoma can lead to misdiagnosis. Recognizing this lesion is necessary to avoid unnecessary aggressive interventions.***

Keywords: Respiratory epithelial adenomatoid hamartoma, Nasal obstruction case, Histopathology diagnosis, benign sinonasal lesion, misdiagnosis risk.

1. Introduction

Respiratory epithelial adenomatoid hamartoma (REAH) is an uncommon, benign glandular lesion of the sinonasal tract characterized by proliferation of ciliated respiratory epithelium forming gland-like structures.[1,2] First described as a distinct entity in the late 20th century, REAH is increasingly recognized in routine surgical pathology due to improved awareness and its inclusion in major head and neck pathology classifications.[3] REAH most commonly arises in the posterior nasal septum, although it may also involve the nasal cavity, paranasal sinuses, and nasopharynx.[1,2]

2. Case History

A 66-year-old male patient presented with complaints of nasal obstruction for 2 years which progressed to difficulty in breathing. On examination-firm fleshy mass was seen arising from upper part of nasal cavity (olfactory region) on both sides. The clinical diagnosis was olfactory neuroblastoma/olfactory lymphoma.

3. Investigations

CT scan PNS plain and contrast revealed a polypoidal mass with likely superadded infection in the left maxillary, left ethmoidal and left frontal sinuses nearly completely occluding involved sinuses with extension and widening of middle meatus.

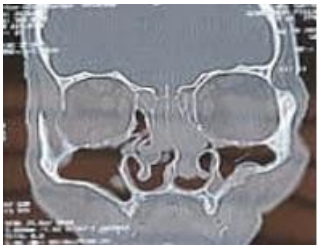

Figure 1

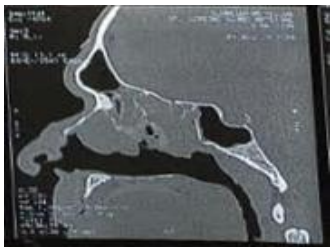

Figure 2

Figure 1 and 2: CT PNS showing polypoidal mass involving left maxillary left ethmoidal and frontal sinuses nearly completely occluding involved sinuses with extension and widening of middle meatus.

Histopathology

Histopathological examination revealed a polypoidal mass lined by respiratory epithelium, proliferated glands devoid of atypia, mixed inflammatory infiltrate and congested vessels in the subepithelial edematous stroma.

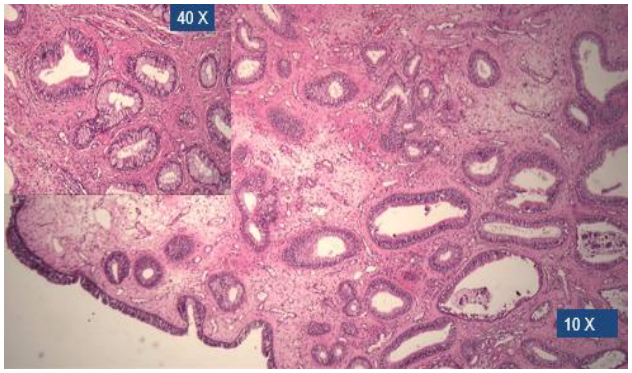

Figure 3: Biopsy revealed a polypoidal mass lined by respiratory epithelium, subepithelial edematous stroma shows proliferated glands, mixed inflammatory infiltrate and areas of infarction.

Immunohistochemistry

CK5/6 and P63 was positive in myoepithelial cells of all glands.

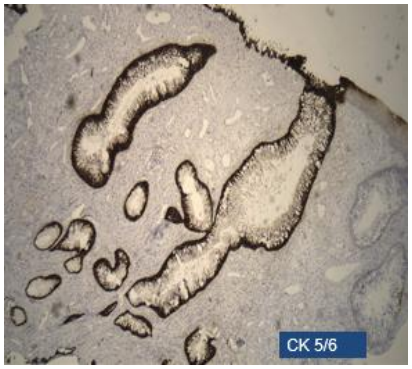

Figure 4: CK 5/6 showing positivity in myoepithelial cells of all glands

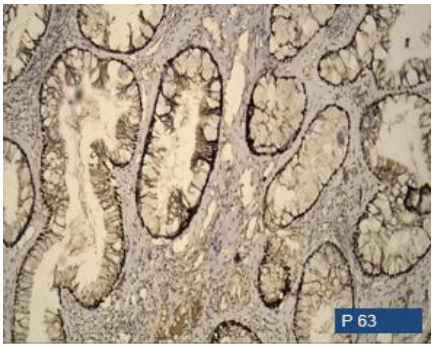

Figure 5: P 63 showing positivity in myoepithelial cells of all glands

4. Discussion

Respiratory epithelial adenomatoid hamartoma (REAH) is an uncommon but increasingly recognized benign sinonasal lesion that can closely mimic malignant tumours on clinical and radiological evaluation.[1] Its pathogenesis is likely multifactorial, with contributions from chronic inflammation and possible neoplastic mechanisms.[2]

REAH predominantly affects adults with a male preponderance and typically arises from the posterior nasal septum and olfactory cleft, although other sinonasal sites may be involved; bilateral presentation is not uncommon.[4] Clinically, patients present with nonspecific symptoms such as nasal obstruction, rhinorrhoea, hyposmia, facial pain, and occasional epistaxis, often resembling chronic rhinosinusitis.[4,5,7]

Histologically, REAH is characterized by submucosal proliferation of well-formed, round to oval glands lined by ciliated respiratory epithelium within an edematous stroma and surrounded by a thickened basement membrane.[1,5] The absence of cytologic atypia, mitotic activity, and invasive growth helps distinguish it from sinonasal adenocarcinoma and other malignancies.[10]Immunohistochemically, the glands are CK7 positive with p63-positive basal cells, while CK20, CDX2, and S100 are typically negative.[10]

Radiologically, REAH appears as a slow-growing, non-invasive mass, often showing olfactory cleft widening (>10 mm), imaging findings are not specific and histopathology remains the gold standard for diagnosis.[2]

The differential diagnosis includes inflammatory nasal polyps, inverted papilloma, and sinonasal adenocarcinoma; unlike REAH, malignant lesions show cytologic atypia and destructive growth.[8]

Management is conservative, with complete endoscopic excision being curative and associated with an excellent prognosis. [9]Accurate recognition is essential to avoid misdiagnosis and unnecessary aggressive treatment.[10]

5. Conclusion

Respiratory epithelial adenomatoid hamartoma (REAH) is a benign lesion of the sinonasal tract that often mimics malignant tumours on clinical and radiological evaluation. Precise histopathological identification is essential, and awareness of its distinctive features helps prevent diagnostic errors and avoid unwarranted aggressive therapy, thereby ensuring appropriate patient care and favourable outcomes.

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