

A Rare Case of Sinonasal Ameloblastoma with Immunohistochemistry

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ABSTRACT

Introduction: Ameloblastoma is benign locally aggressive dentine epithelial tumor. Mandible and maxilla are common sites of involvement. Calretinin is specific marker and useful to differentiate ameloblastoma from Keratocystic odontogenic tumor. The rarity of site and presentation of lesion makes us report this case.

Case report: 46 year old female had recurrent nasal blockage. Polyps in right nostrils were seen on Rhinoscopy. Polypectomy was done for suspected antrochoanal polyps. Ameloblastoma was diagnosed on histopathology and confirmed by Immunohistochemistry with Cytokeratin, Calretinin and WT1.

Conclusion: Preoperative diagnosis by biopsy in unilateral nasal masses should be carried out to expect the unexpected. Immunohistochemistry should be performed in cases having rare site to confirm the diagnosis.

Keywords: Sinonasal Ameloblastoma, Calretinin, WT1, Cytokeratin

INTRODUCTION

Ameloblastoma is a benign neoplasm of epithelial origin with invasive and destructive growth characteristics.¹ The recurrence rate is high in cases of incomplete surgical resection.²

The estimated incidence of ameloblastomas is approximately 0.5 per million populations per year. There is no significant gender predilection. Most cases are between 30 and 60 yrs of age groups.³ According to the recent Classification of Odontogenic Tumors, by World Health Organization (WHO), four subtypes of benign ameloblastomas are recognized: the 1) Solid / multicystic, 2) Desmoplastic, 3) Unicystic and the 4) Extrasosseous / peripheral type.⁴

Solid ameloblastomas affect the mandible preferably, especially the posterior region. The literature showed that solid ameloblastoma occurred as the least frequent in maxillary bone.^{5,6} Most of the cases involving the sinonasal cavity are tumors that originated in the maxilla and secondarily extend to the sinonasal cavities.

CASE REPORT

46 year old female came with recurrent nasal blockage and right sided purulent nasal discharge since 4 months. Rhinoscopy showed polyps in right nostril. MRI showed lobulated non-enhancing soft tissue in right maxillary sinus extending into right nasal cavity. It extends posteriorly up to the right posterior choana, protruding into the nasopharynx. There is thinning of the medial wall. Significant mucosal thickening in right ethmoid sinus. Right middle and inferior turbinate appear partly obscured by soft tissue within the

right nasal cavity. Findings suggestive of antrochoanal polyp on radiological investigation. Clinically diagnosed as right antrochoanal polyp. Hence polypectomy was done.

We received multiple polypoidal bits aggregating to 4cc largest measuring 1.8cm x 1cm. On histopathological examination the polypoidal bits showed dentine epithelium arranged in trabeculae, follicles and in plexiform pattern. Reverse polarization in epithelium seen. Stroma shows stellate reticulum cells and scanty lymphocytic infiltrate.

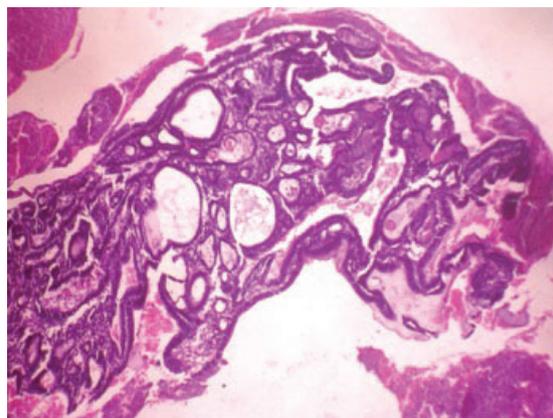


Figure-1A:

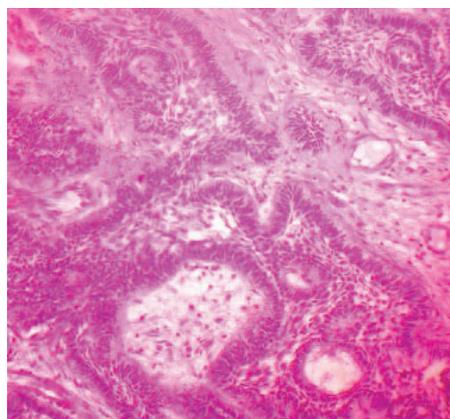


Figure-1B:

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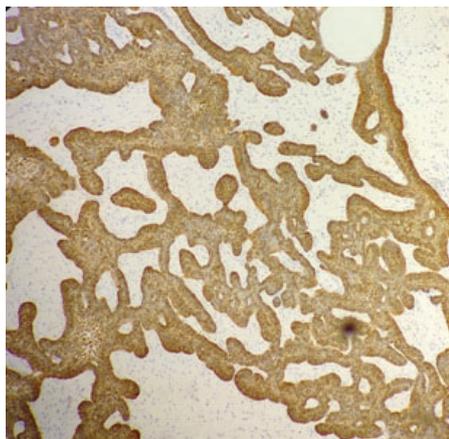


Figure-2:

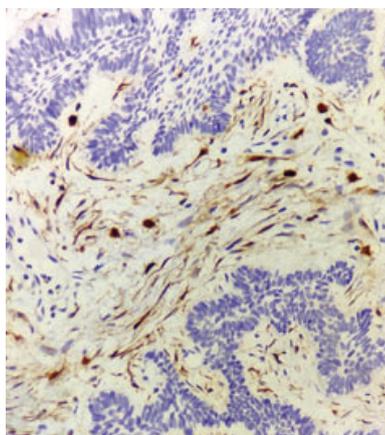


Figure-3:

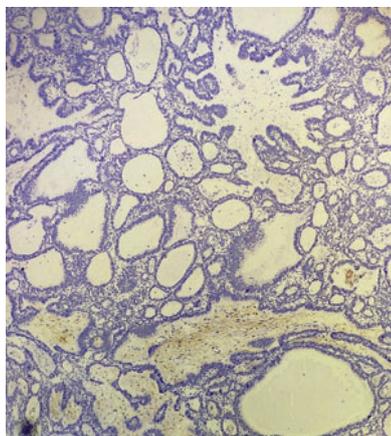


Figure-4:

Follicles and plexiform cords and peripheral palisading (Figure-1A)

Epithelium with reverse polarization and stroma shows stellate like reticular cells.(Figure-1B)

CK14: strongly positive in dentine epithelium, Nuclear and Cytoplasmic. (Figure2)

CK19: weak focal positivity in dentine epithelium.

Calretinin: positive in stellate like reticulum cells only. Nuclear and cytoplasmic (Figure-3)

WT1: Negative (Figure-4)

Diagnosis of solid Ameloblastoma was confirmed with Immunohistochemistry.

DISCUSSION

The rarity of origin of tumor, rarity of presentation of tumor, rarity of histological variant at that site makes this case extraordinary.

Ameloblastoma is a benign locally invasive odontogenic epithelial tumor with slow growth.¹ It originates from dental epithelium of enamel itself or its epithelial tissues or from the cells of the basal layer of the oral mucosa.⁷ Generally these tumor restricts itself to oral cavity or jaw. Mandible is most common site. There are only few cases of primary sinonasal ameloblastomas, reported in literature without connection to gnathic areas.⁸

The solid/multicystic ameloblastoma can histopathologically be divided into a follicular and a plexiform type. The follicular type can be further subdivided into a spindle cell type, an acanthomatous type, a granular type and a basal cell type. The plexiform type contains basal cells arranged in anastomosing strands with an inconspicuous stellate reticulum. The stroma is usually delicate, often with cyst like degeneration.⁹ Unicystic ameloblastomas are lined by a variable epithelium. It could be any combination of one with typical ameloblastic characteristics, nonkeratinizing squamous cells and metaplastic. In such cases, the differentiation of odontogenic cysts from the unicystic ameloblastoma can be problematic.¹⁰

This case was clinically diagnosed as antrochoanal polyp may be because of extent of lesion and radiological features of the mass. MRI in this case showed extension into choana and nasopharynx, and thickening in sinus walls without bony destruction or rarefaction. Only mild thinning of medial wall of maxillary sinus was seen.

An antrochoanal polyp presents as a single polyp arising from the maxillary sinus passing through the sinus ostia and extending backward towards the choana.¹¹ Intranasal lesions of neoplastic conditions may present as typical firm proliferative lesions with destruction of surrounding structures or may mimic inflammatory conditions in their early stages with the presence of polyps on endoscopy. Radiologically all malignant lesions are characterized by soft tissue mass with aggressive bone destruction of the adjacent walls.¹²

Immunohistochemical evaluation of ameloblastomas using epithelial and connective tissue specific markers help in studying the histogenesis and assessing the biological behaviour. In the human enamel organ CK5, 7, 8, 13, 14, 17 and 19 are present. The dental lamina and the enamel organ express CK14, except in the secretory ameloblasts and in portions of the stellate reticulum/stratum intermedium. Antibody against CK19 labels the preameloblasts and the secretory ameloblasts, and part of the external enamel epithelium/ dental lamina.¹³ The intense expression of keratin 14 in ameloblastoma by all tumor cells suggests that they may retain basal cell characteristics with a potential for proliferation.¹⁴

Calretinin may be used as a specific immunohistochemical marker for neoplastic ameloblastic epithelium. It serves as an

important diagnostic adjunct in the differential diagnosis of ameloblastoma and keratocystic odontogenic tumor.¹⁵ This case showed calretinin positivity in stellate-like reticulum cells.

The Wilms' tumor 1 gene (WT1) was originally isolated and described as the gene responsible for Wilms' tumor. WT1 plays a remarkable role as an oncogene rather than a suppressor gene, in tumorigenesis of ameloblastoma. Staining of WT1 is variable. In our case there was no staining of WT1.¹⁶ Immunohistochemistry is not usually employed to assist diagnosis and classification of odontogenic tumors. It may be helpful to reach a correct diagnosis in doubtful situations.¹⁷ In this case the immunohistochemical markers confirmed diagnosis of ameloblastoma.

In all cases of unilateral nasal mass, high index of suspicion of neoplastic etiology is must. The difficulty in early diagnosis is because lesions of paranasal sinus develop in a closed anatomic space not causing symptoms in the early stages and symptoms become evident only in late stages when they expand toward the nasal cavity or nasopharynx with bony destruction and complications. Favorable outcomes depend on an early diagnosis and appropriate management. Nasal endoscopy with biopsy remains the gold standard for diagnosis.¹⁸

CONCLUSION

Neoplastic lesions of nose and paranasal sinus are one of the most challenging conditions to diagnose and treat. Unilateral sinonasal neoplastic conditions during their early stages often mimic an inflammatory pathology. Preoperative biopsy in unilateral sinonasal lesions is gold standard for diagnosis of odontogenic tumors.

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