Ameloblastoma Presenting as a Sinonasal Tumor: A Rare Entity

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Abstract

A case of ameloblastoma of the left maxilla presenting as a sinonasal tumor in a 60-year-old man is reported. Ameloblastoma is a benign, locally invasive odontogenic epithelial tumor, originates from dental epithelium of enamel itself or its epithelial tissues. Usually it restricts itself to the oral cavity in most of the cases. However, rarely it can present as sinonasal mass, exhibit a locally aggressive behaviour with a high level of recurrence. In our case, the tumor was presented as a radiographically soft tissue mass filling the left sinonasal cavity. After Functional Endoscopic Sinus Surgery (FESS), the patient has pursued a uneventful clinical course upto 1.5 years of follow-up then presented again with local recurrence of tumor.

Keywords: Ameloblastoma, Odontogenic Epithelium, Sinonasal Tract

Introduction:

Ameloblastoma is a benign locally invasive odontogenic epitelial 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.[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)extraosseous/peripheral type.[4] Solid ameloblastomas affect the mandible preferably, especially the posterior region.[5] The literature showed that solid ameloblastoma occurred as the least frequent in maxillary bone.[3,5,6] Most of the cases involving the sinonasal cavity are tumors that originated in the maxilla and secondarily extend to the sinonasal cavities. There are only few cases of primary sinonasal ameloblastomas, reported in literature without connection with gnathic areas.[7]

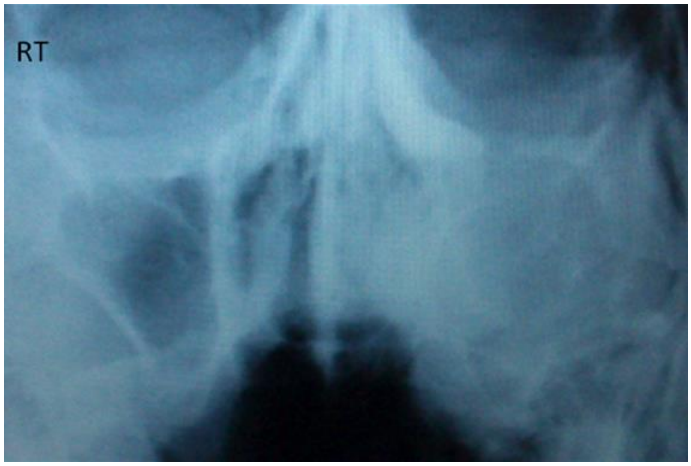
Case Report:

We report a case of ameloblastoma in the maxillary sinus in a 60-year-old man, presented with clinical complaints of recurrent sinusitis i.e. headache, nasal stuffiness and obstruction. Patient undergone through general and local examination as well as paranasal sinuses radiograph and CT examination were carried out. Diagnostic nasal endoscopy was also performed.

On X ray -Water`s view of paranasal sinuses, there was presence of soft tissue density mass lesion filling the left sinonasal cavity with loss of borders of maxillary sinus and lateral wall of nasal fossa on the left side. Haziness and opacification of the left antrum

and relative haziness of left nasal fossa was compared with the contralateral side (Figure No. 1).

Figure No. 1: Water's view of paranasal sinuses shows haziness and opacification of the left maxillary antrum along with loss of borders of maxillary sinus and lateral wall of nasal fossa on left side.



CECT demonstrated a massive expansile soft tissue density lesion, with no significant contrast enhancement, occupying the entire left maxillary sinus and extension in to left nasal fossa and choana, causing thinning and rarefection of sinus walls. The lateral nasal walls as well as ethmoidal sinuses were involved. Nasal septum is displaced to right side. Osteomeatal complex, inferior and lateral wall of antrum on the left side were not distinguishable (Figure No. 2, 3).

Considering absolute benign features, a provisional diagnosis of benign sinonasal polypoidal mass was made, and endoscopic surgical excision was planned. Patient underwent FESS(Functional endoscopic sinus surgery) with uncinectomy, MMA, anterior ethmoidectomy, partial excision of left middle turbinate and left polypectomy under general anaesthesia. Tissue sample was sent for histopathological diagnosis.

Histopathological report revealed, benign lesion showing plexiform and follicular arrangement of epithelial tissue with cells showing reverse polarity, stellate epithelium was present in between the epithelial tissue. On the basis of these findings diagnosis of ameloblastoma was made.

After FESS(Functional endoscopic sinus surgery), the patient has pursued a uneventful clinical course upto 1.5 years of follow-up then presented again with local recurrence of tumor. CT examination for paranasal sinuses was repeated and a radical left maxillectomy was performed.

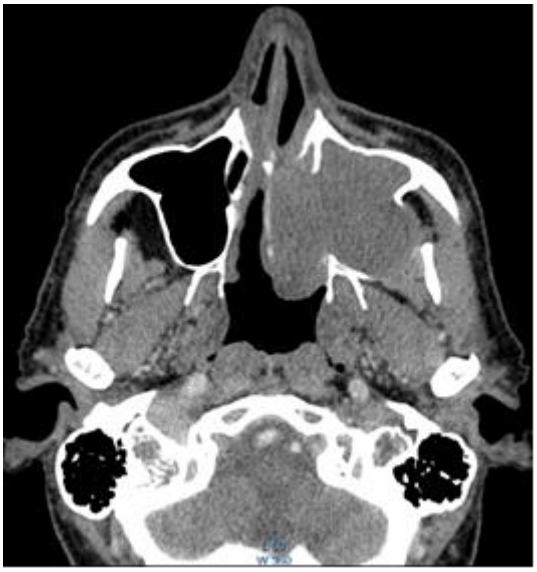
Discussion:

Ameloblastomas are benign, locally invasive epithelium-derived odontogenic tumors that typically originated in mandible and less often in the maxilla. The presence of ameloblastomas in the sinonasal region is usually secondary to an extension of a tumor of gnathic origin into this area.[7] However, primary sinonasal ameloblastomas, without extension from the gnathic region, have also been reported.[3]

On the basis of imaging modalities, diagnosis of benign soft tissue sinonasal mass lesion was made with differentials of anterochoanal polyp, sinonasal polyposis, antral mucocele and other benign aggressive tumoral lesion were considered. Although malignant lesions such as SCC were not completely ruled out in differential diagnosis of maxillary sinus tumoral lesions, but there was no evidence of any obvious bony destruction to support the diagnosis. In addition, significant expansion, thinning and rarefection of sinus walls by the mass lesion, lowered the possibility of malignancy.

Surgical resection is usually the treatment of choice. Recently FESS(Functional endoscopic sinus surgery) of benign sinonasal tumors has resulted in less radical surgical approach, decreased morbidity and better tumor control.[8] Yearly follow up for atleast 5 years after radical surgery has been proposed in cases of solid ameloblastomas.[3] In cases of maxillary involvement, at least 10 years follow up has been recommended as high recurrence rate is reported. These lesions are locally aggressive and become dangerous clinically as they can invade adjacent sinus and vital structures.[3, 9] Treatment decisions for ameloblastoma should based on the individual patient situation and the best judgement of the surgeon. [10]

Figure No. (2a, 2b): Axial CECT scan shows soft tissue density lesion affecting left sinonasal region (figure 2a), causing thinning and rarefaction of sinus walls on bone window (figure 2b).



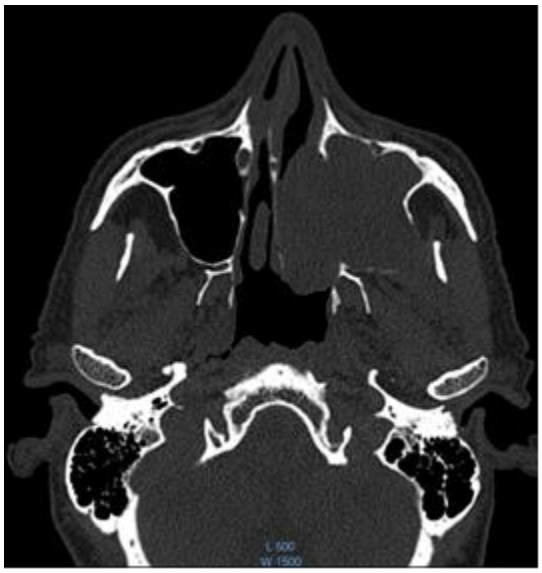
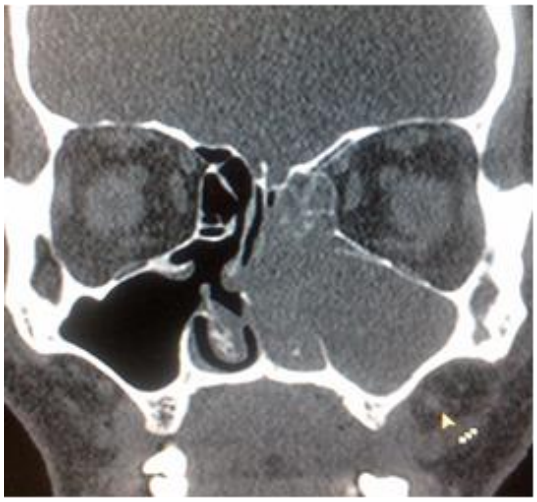


Figure No. (3a, 3b): Coronal(Figure3a) & Sagittal(Figure3b) CT sections show the extent of the large expansile soft tissue lesion located in left maxillary sinus and nasal fossa, displacing medial and lateral walls.



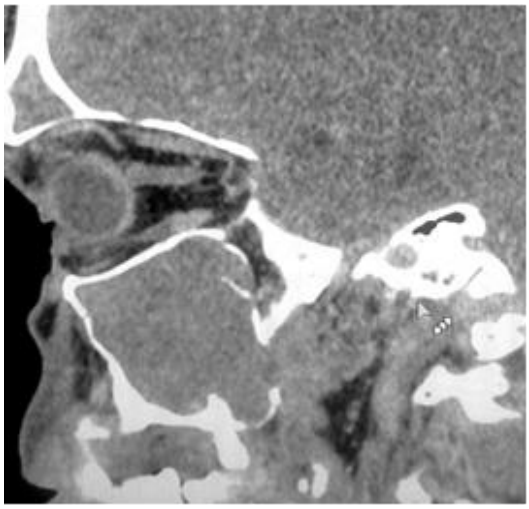
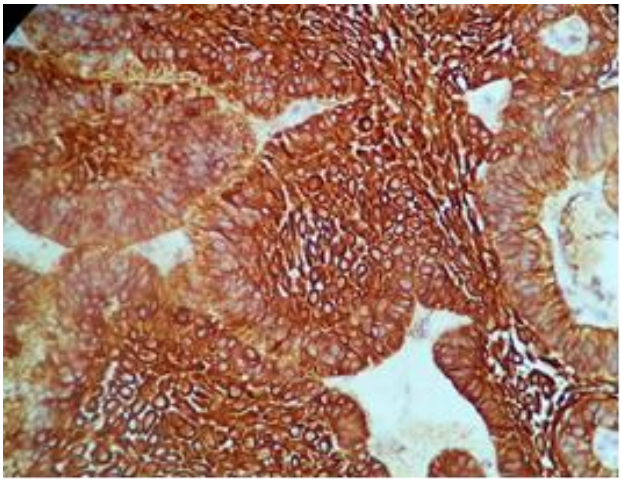
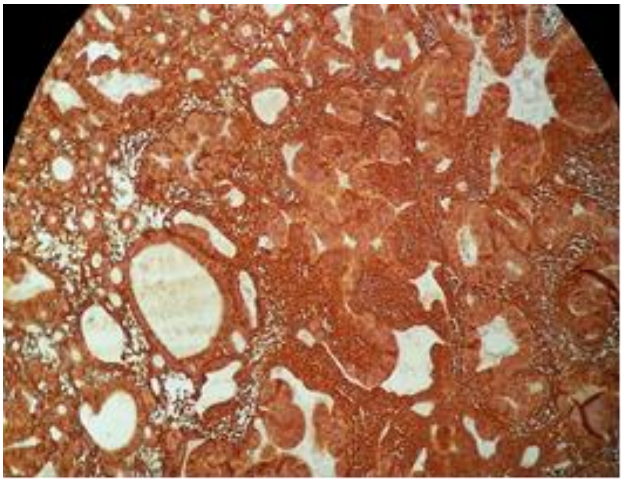


Figure No. (4a, 4b): Follicular islands of odontogenic epithelium presenting ameloblastoma





Conclusion:

Generally, ameloblastomas are originated in jaw bones. Primary Sinonasal ameloblastomas are rare tumors without any extension from the gnathic region. The prognosis of surgical treatment depends upon the extent of the lesion and adjacent structures involvement at the time of presentation instead of origin of lesion.

References:

1. Ledesma-Montes C, Mosqueda-Taylor A, Carlos-Bregni R, et al. Ameloblastomas: a regional Latin-America multicentric study. Oral Dis. 2007;13:303–7.
2. Sun ZJ, Wu YR, Cheng N, et al. Desmoplastic ameloblastoma - A review. Oral Oncol. 2009;45:752–9.
3. Hertog D, van der Waal I. Ameloblastoma of the jaws: A critical reappraisal based on a 40-years single institution experience. Oral Oncology. 2010;46:61–4.
4. Thompson L. World Health Organization classification of tumours: pathology and genetics of head and neck tumours. Ear Nose Throat J. 2006;85:74.
5. Fulco GM, Nonaka CF, Souza LB, et al. Solid ameloblastomas - Retrospective clinical and histopathologic study of 54 cases. Braz J Otorhinolaryngol. 2010;76:172–7.
6. Pitak-Arnnop P, Chaine A, Dhanuthai K, et al. Unicystic ameloblastoma of the maxillary sinus: Pitfalls of diagnosis and management. Hippokratia . 2010;14:217–20.
7. Ereno C, Etxegarai L, Corral M, et al. Primary sinonasal ameloblastoma. APMIS. 2005;113:148–50.
8. London SD, Schlosser RJ, Gross CW. Endoscopic management of benign sinonasal tumors: a decade of experience. Am J Rhinol. 2002;16:221–7.
9. Rastogi R, Jain H. Case report: desmoplastic ameloblastoma. Indian J Radiol Imaging. 2008;18:53–5.
10. Kishore M, Panat SR, Kishore A, Joshi A. Follicular ameloblastoma: A case report. IJSS Case Reports & Reviews. 2014;1(1):1-3.

How to Cite: Kumar A, Sarswat S, Sharma S. Ameloblastoma Presenting as a Sinonasal Tumor: Rare Entity. Int J Adv Health Sci 2014; 1(2): 28-31.