

## Case Report

# A rare case of plexiform ameloblastoma of right maxillary sinus

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### ABSTRACT

Plexiform ameloblastoma is a benign odontogenic neoplasm characterized by locally invasive growth and high recurrence rate with incomplete resection. Plexiform ameloblastomas in the sinonasal tract, particularly the maxillary sinus, are exceptionally rare, comprising about 0.11% of sinonasal tumors, with a male predilection and later presentation age (mean 59.7 years) compared to jaw variants. A 40-year-old male presented with recurrent epistaxis and on examination revealed a reddish nasal mass. Initial CT suggested a right maxillary mucocele or low-grade neoplasm. FESS with total excision of the friable mass was done. Histopathology reported plexiform ameloblastoma. At 2-year follow-up, MRI revealed a recurrent 3.9×3.2×3.6 cm lesion with bony erosion, for which right infrastructural maxillectomy was done followed by obturator placement. The patient remains disease-free on subsequent follow-up. While conservative FESS may suffice initially, complete resection via maxillectomy is crucial to prevent early recurrence (typically 1-2 years post-surgery). Complete surgical excision offers excellent prognosis for sinonasal Plexiform Ameloblastoma, with long-term follow up necessary due to local aggressiveness. This case underscores the value of histopathology and serial imaging in management.

**Keywords:** Plexiform ameloblastoma, Epistaxis, Odontogenic tumours, Sinonasal tumours, Maxillary sinus

### INTRODUCTION

Plexiform Ameloblastoma is a benign neoplasm of epithelial origin with invasive and destructive growth characteristics.<sup>1</sup> The recurrence rate is high in cases of incomplete surgical resection.<sup>2</sup> 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 years of age groups.<sup>3</sup> According to the recent Classification of Odontogenic Tumors, by World Health Organization (WHO), four subtypes of benign ameloblastomas are recognized: solid/multicystic, desmoplastic, unicystic and extraosseous/peripheral type.<sup>4</sup> Solid ameloblastomas affect the mandible preferably, especially the posterior region. The literature showed that solid ameloblastoma occurred as the least frequent in maxillary bone.<sup>5,6</sup> Most of the cases involving the sinonasal cavity are tumors that

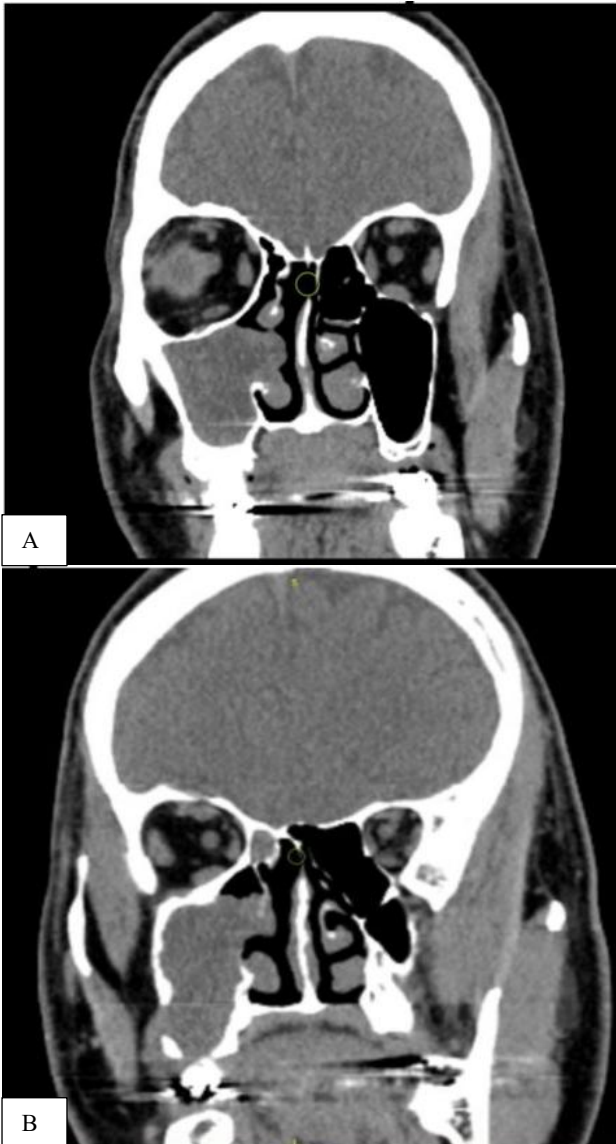
originated in the maxilla and secondarily extend to the sinonasal cavities.

### CASE REPORT

A 40-year-old male presented with complaints of bleeding from the nose for 5 days, sudden onset, 2 episodes, scanty amount which stopped spontaneously. On examination there was a reddish mass on the right side of the nasal septum.

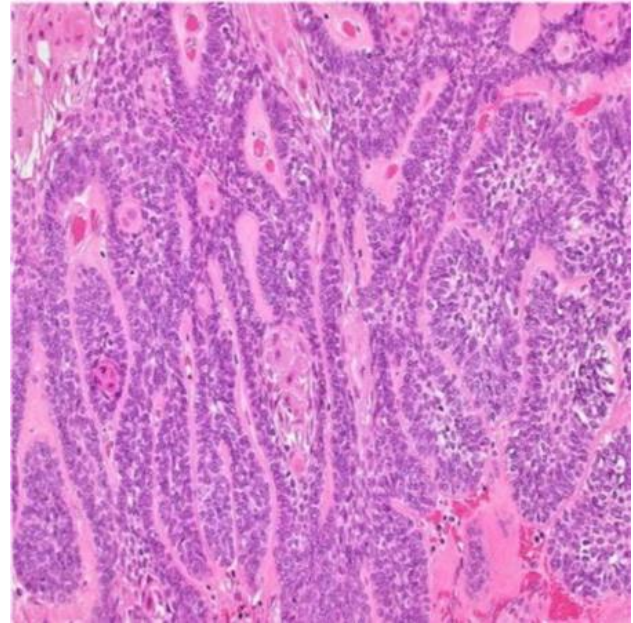
CT OMC showed soft tissue density lesion in the right maxillary sinus extending through the right maxillary ostium, mildly protruding into the right nasal cavity and causing bony erosion of inferior wall of the right maxillary sinus, features suggestive of right maxillary mucocele/ low grade neoplasm. (Figure 1A and B). Patient underwent right limited functional endoscopic sinus surgery (FESS). Intra-operatively, there was a

reddish friable mass filling the entire right maxillary sinus which was removed in toto and sent for histopathological examination (HPE). HPE was reported as plexiform ameloblastoma (Figure 2).

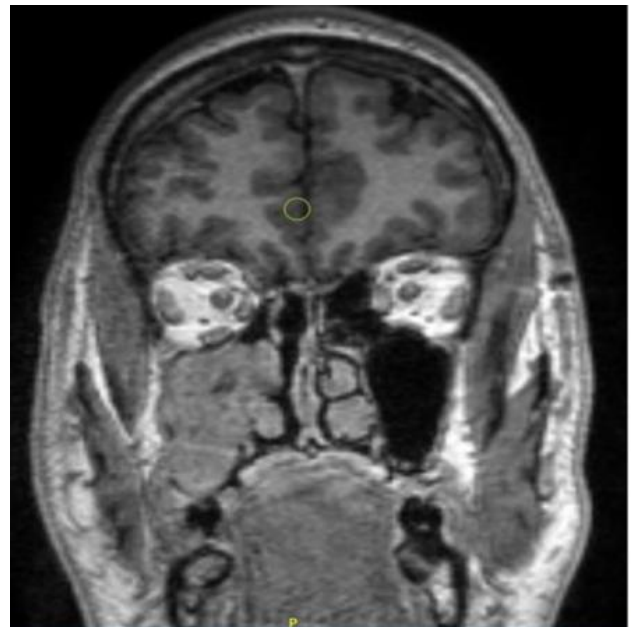


**Figure 1: CT OMC showing soft tissue density lesion in the right maxillary sinus: (A) extending through the right maxillary ostium and mildly protruding into the right nasal cavity and (B) causing bony erosion of inferior wall of the right maxillary sinus.**

On 2 years follow up, patient presented with bleeding from the nose on and off. MRI head and neck showed T1 hypointense, T2/STIR hyperintense lesion (3.9×3.2×3.6 cm) in the right maxillary sinus extending through the maxillary ostium and mildly protruding into right nasal cavity with bony erosion of inferior wall of right maxillary sinus suggestive of residual or recurrent lesion (Figure 3). Patient underwent right infrastructural maxillectomy. Intraoperative and postoperative period was uneventful. This was followed by obturator placement.



**Figure 2: HPE showing features of plexiform ameloblastoma.**



**Figure 3: MRI head and neck showing T1 hypointense, T2 /STIR hyperintense lesion in the right maxillary sinus causing bony erosion of inferior wall of right maxillary sinus and also extending through the maxillary ostium and mildly protruding into right nasal cavity.**

At 9 months follow-up after maxillectomy, the cavity was healthy (Figure 4). Also, follow up MRI head and neck showed no lesion (Figure 5). Patient was doing well at that time. At 2 years follow-up after maxillectomy, patient remains disease free.



**Figure 4: Cavity of right infrastructural maxillectomy at 9 months follow-up.**



**Figure 5: MRI head and neck at 9 months follow-up after maxillectomy.**

## DISCUSSION

Sinonasal ameloblastomas are rare tumors of the sinonasal tract that arise from sinonasal epithelium. A large review found ameloblastomas to comprise approximately 0.11% (n=19,658) of all sinonasal tract tumors.<sup>7</sup> Sinonasal ameloblastomas show a predilection for the male gender of 3.8:1.7 They present approximately 15–25 years later than those occurring in the jaw, at an overall mean age of 59.7 versus 35–45 years of age respectively.<sup>7,8</sup> Clinical presentation commonly includes nasal obstruction, sinusitis, and/or

epistaxis.<sup>7-10</sup> Less common presenting signs and symptoms include facial swelling, headaches, and tearing.<sup>7,9,10</sup> Sinonasal lesions are most frequently described as radiopaque, solid lesions that fill the nasal cavity or sinus.<sup>7</sup>

On gross examination, these primary sinonasal lesions are predominantly solid, grey-white, and rubbery to granular.<sup>7</sup> Histologically, sinonasal ameloblastoma resemble those lesions arising within the bones of jaw. The plexiform pattern is the most common pattern in sinonasal ameloblastomas, but the follicular pattern may also be predominant.<sup>7</sup> The plexiform pattern of ameloblastoma is characterized by back-to-back anastomosing cords of epithelium, with peripheral columnar cells that demonstrate reverse polarity of their nuclei. Between the epithelial cords there are loosely arranged, angulated cells that comprise the stellate reticulum-like component of the tumor. Finally, transition from normal sinonasal tract epithelium to ameloblastomatous proliferation may be demonstrated microscopically and helps confirm the neoplasm is in fact primary to the sinonasal tract. Surgical resection of primary sinonasal ameloblastomas is the treatment of choice.<sup>7-10</sup> Originally, radical surgery was performed on most patients given that treatment success heavily correlates with complete surgical removal of the lesion.<sup>8,9</sup> Conservative surgery or functional endoscopic sinus surgery (FESS) may be attempted in proper clinical setting, however complete removal of the tumor is the most important factor in determining the appropriate procedure.<sup>7-10</sup>

The overall prognosis of sinonasal ameloblastomas is quite good with local recurrence being the most common long-term sequelae. Recurrence tends to occur within 1–2 years following surgery; however, cases have been reported with recurrence occurring many years following surgery.<sup>8</sup> There has been no documented malignant transformation, metastasis or deaths attributed to sinonasal ameloblastomas.<sup>7-9</sup>

## CONCLUSION

Sinonasal Plexiform Ameloblastoma is a benign but locally aggressive variant of Ameloblastoma involving the nasal cavity and/or paranasal sinuses which often mimics malignant tumors. Diagnosis is primarily based on histopathology but radiologic and intra-operative findings help distinguish it from differentials. The treatment of choice is complete surgical excision. Good follow up is very necessary.

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