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**CASE REPORT**

# Maxillary Plexiform Ameloblastoma with Local Recurrence: A Case Report

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## ABSTRACT

### INTRODUCTION

Maxillary ameloblastoma is a rare and misleading entity that is often misdiagnosed.

### CASE REPORT

Here we report a case of maxillary ameloblastoma revealed by spontaneous dental avulsion in a 62-year-old woman.

The tumor was excised via endoscopic endonasal approach and the histologic diagnosis of ameloblastoma, in its plexiform variant, was established. An MRI was performed at 12 months, showing local recurrence.

### CONCLUSION

Imaging plays an important role in the diagnosis and management of maxillary sinus ameloblastoma despite its low specificity. The diagnosis of certitude remains anatomopathologic.

## KEYWORDS

Ameloblastoma; Maxillary sinus; Plexiform; Local recurrence

## INTRODUCTION

Ameloblastoma is a benign tumor derived from the odontogenic epithelium [1]. It is usually located in the mandible. Ameloblastoma of the maxilla occurs relatively rarely: it represents only 0.1% of sinonasal tumors [2]. It can be locally invasive. It is a rarely malignant or metastatic tumor, and it grows slowly [1].

Clinically, symptoms are nonspecific, posing diagnostic problems with other aggressive tumors in this region, including certain cysts and odontogenic tumors, inverted papilloma's, and malignant tumors [3]. The CT-MRI pair is essential, which allows one to ensure that there is no contraindication to biopsy and establish extension assessment [4]. The final diagnosis is made by histological examination. They have two main histological types: follicular or plexiform.

The invasive nature of these tumors requires surgical resection [3].

We report a rare case of maxillary sinus ameloblastoma in a woman who presented to our hospital with spontaneous dental avulsion and nasal obstruction.

## CASE REPORT

In a study by Zwahlen that included 3677 patients over a 15-years period, 85% of the ameloblastomas were preferentially located in the mandible [5-7]. Localization in the maxillary sinus is relatively rare, accounting for only 1% of tumors in this region [1].

For maxillary ameloblastoma, the distribution of age and sex is evenly distributed [3,8]. In some studies, a peak in frequency is observed between the third and fourth decades [6,9] (Figure 1).

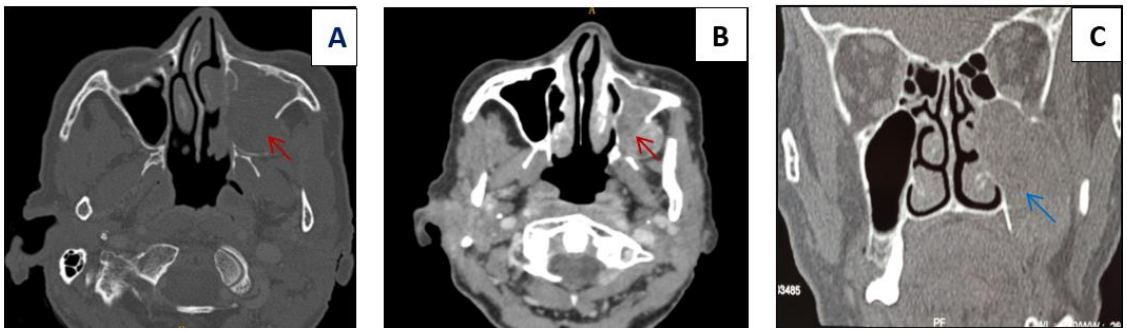


**Figure 1:** Facial CT scan showing total filling of the left maxillary sinus with lysis of the posterior (red arrow) and inferior (blue arrow) walls; axial section with bone fenestration **(A)** and tissue fenestration after PDC injection **(B)** and coronal section **(C)**.

In the maxillary sinus, ameloblastoma is characterized by greater local aggressiveness [2]. This is due to the long onset of symptoms and the spongy nature of the maxillary bone, in contrast to the compact nature of the mandibular bone, which offers less resistance to tumor extension and invasion of neighbouring structures (Figure 2).

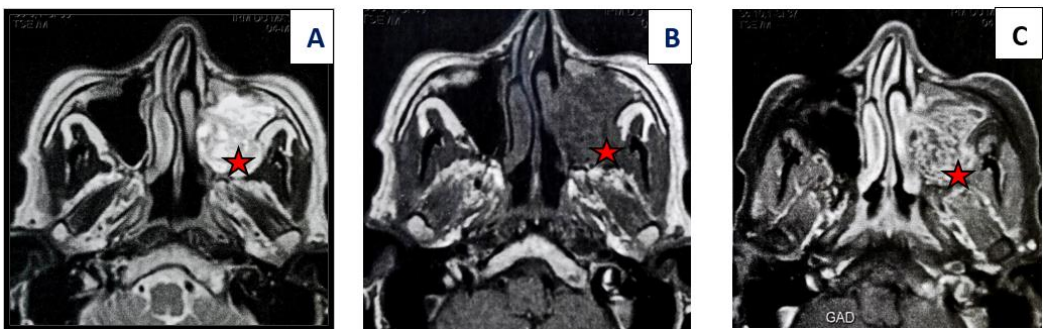


**Figure 2:** MRI of the facial mass: Filling of the left maxillary sinus in T2 hypersignal **(A)**, T1 isosignal **(B)**, with heterogeneous contrast after gadolinium injection **(C)** and extension to the infratemporal fossa (star).

Clinically, ameloblastoma of the maxillary sinus remains asymptomatic for a long time, due to its slow growth. In advanced cases, such as ours, the extension to adjacent structures usually results in rhinological signs, notably nasal obstruction and epistaxis, or facial deformity [3].

There is a wide range of differential diagnosis that causes difficulty in the diagnosis of ameloblastoma. These include odontogenic maxillary tumors, such as odontogenic keratocytes, as well as benign maxillary sinus formations such as inverted papilloma, maxillary sinus mucocele and invasive fungal pseudotumors, or sinus cancers [10].

The World Health Organization (WHO) simplified the classification of ameloblastoma in 2017. This classification, based on clinicopathological criteria, resulted in 3 types [11]; unicystic, extraosseous or peripheral and metastatic. In the case of our patient, it was a peripheral type of ameloblastoma (Figure 3).

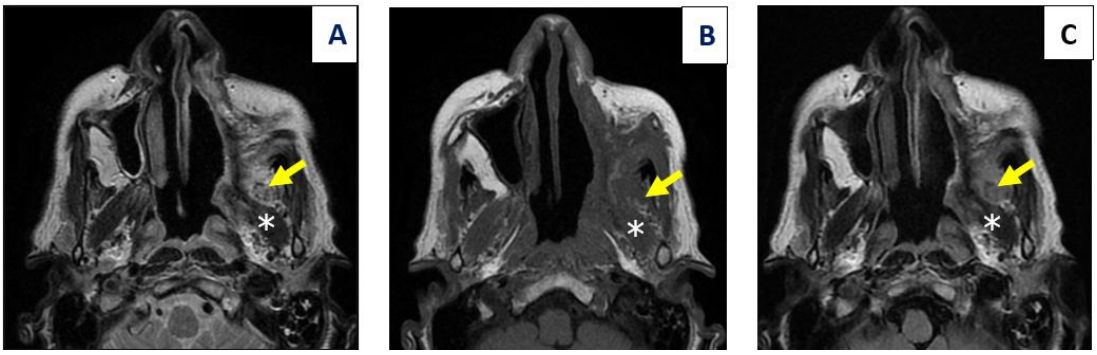


**Figure 3:** MRI facial mass: tumor recurrence (yellow arrow) at the level of the Infratemporal fossa opposite the medial pterygoid muscle (asterisk); In T2 hypersignal **(A)**, T1 isosignal **(B)** and contracting **(C)**.

Imaging has become an indispensable tool in the diagnostic approach and preoperative assessment. High-spatial-resolution CT scans can be used to assess tumor size, bone changes, and anatomical variants at risk for surgery. MRI provides a better tumor tissue analysis of the tumor and evaluates extensions to the deep spaces of the face and endocardium, allowing us to assess the operability of this tumor [12,13].

Radiologically, three groups of ameloblastoma can be distinguished: Solid or multi-cystic, unicystic and peripheral [13]. On computed tomography (CT), ameloblastoma is described as an expansive iso- or hypodense mass with heterogeneous peripheral enhancement. Its multilocular presentation with irregular boundaries gives it a "soap-bubble" appearance [13,14], and is associated with thinning or even destruction of the bone cortex and resorption of tooth roots [14]. The radiological findings in our case were not specific.

The cone Beam is an excellent alternative to CT. It provides an excellent assessment of bone involvement, with a good special resolution to describe extension to the maxillary sinus, nasal cavity, or orbit [12]. It has a sensitivity of 77% and a specificity of 90% for the diagnosis of ameloblastoma [13]. However, its low tissue contrast sometimes poses problems for differential diagnosis with other odontogenic cysts [13,15].

On MRI, the ameloblastoma presents as a homogeneous intermediate T1 signal and T2 hyper signal with enhancement of the nodular and wall [14]. Perfusion and diffusion sequences show a perfusion curve with an

early intense peak and a washout <200 s [15], a high ADC of the cystic portion (2.48 ± 0.20 103 $mm^2/s$)) and for the solid part the ADC is (1.39 ± 0.16 103 $mm^2/s$) [16].

The diagnosis is based on histopathological examination.

The most common histological types seen in maxillary ameloblastomas are the follicular and plexiform types, followed by the acanthomatous s type. Interestingly, the desmoplastic variant, although rare, shows a predilection for the maxilla [3].

Immunohistochemistry can help diagnosis. It excludes entities with histological features similar to ameloblastoma. All ameloblastoma cells express CK19, which is an odontogenic epithelium marker. Ameloblastomas have a very low proliferation index (MıB1/Ki67) [3,17].

Treatment of maxillary ameloblastomas has not yet been established.

Conservative treatment, such as curettage, has up to 100% possibility of local recurrence [3].

Radical surgical resection, with a margin of at least 1 cm to 2 cm, has shown lower percentages of recurrence. A partial or complete maxillectomy has been shown to be extremely effective in preventing recurrence compared to conservative treatment [16,18].

Endoscopic sinus surgery can revolutionize the management of certain AM. It has been shown to be highly successful for a variety of other nasosinusal tumors, such as inverted papillomas and skull base tumors. Endoscopic techniques avoid facial scarring and minimize the appearance of maxillary defects compared to partial or total maxillectomy [16].

## CONCLUSION

Ameloblastomas are odontogenic epithelial tumors that mainly affect the mandible. Maxillary ameloblastoma is a rare entity that poses several diagnostic challenges. Its aggressive, locally expansive nature in imaging may mimic other nasosinus tumors. Imaging has become an indispensable tool in the diagnostic approach and preoperative evaluation.

The diagnosis requires histopathological examination. The endoscopic approach is very safe and effective approach to completely resect these tumors.

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