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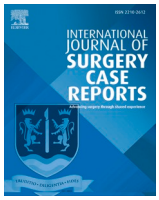

Case report

Asymptomatic ameloblastoma of the maxilla with infratemporal fossa involvement: A case report

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ABSTRACT

Introduction and importance: Diagnosis of odontogenic tumors can be challenging due to their rarity and diverse morphology. The clinical diagnosis could be suspected when it had raised near the tooth. But, when their location is not typical, like inside the paranasal sinuses, the diagnosis is less easy. Maxillary ameloblastomas are rare with only sparse information on their epidemiological, histological and effective management.

Case presentation: A 54-year-old woman presented with ameloblastoma of the left maxillary sinus. Intraoral examination revealed partially edentulous with a 2 cm painless ulceration of the left alveolar process of the maxilla. A biopsy was performed which showed a follicular ameloblastoma. The treatment was surgical. She had a combined *endoscopic and transoral resection of the tumor. The* patient *was diagnosed with* recurrence *within* 3 months from surgery.

Discussion: although rarely reported, clinicians should still suspect and know how to manage such as rare and locally invasive tumor as a maxillary ameloblastoma.

Conclusions: As odontogenic tumors are rare, some entities are infrequently encountered, making the diagnosis more difficult. Clinicians, oral and maxillofacial surgeons, and oral pathologists should be familiar with the ameloblastoma and its differential diagnosis for accurate diagnosis and management.

1. Introduction

Ameloblastoma is a benign aggressive odontogenic tumor that constitute a heterogeneous group of neoplasms with a variety of histological types and clinical behaviors (1).

Ameloblastomas have as origin the odontogenic epithelial. They may arise from any of the following: Rest cells of the dental lamina, developing enamel organ, epithelial lining of odontogenic cysts, basal cells of oral mucosa, heterotopic epithelium in other parts of the body (e.g., pituitary gland) (2).

According to the newly updated classification of the World Health Organization (WHO) in 2017, ameloblastoma is classified into four subtypes: conventional ameloblastoma, unicystic ameloblastoma, peripheral/extraosseous ameloblastoma and metastasizing ameloblastoma (3).

The most common types of ameloblastomas are the follicular and plexiform types also called the“classic/conventional types,”followed by acanthomatous and granularcell types (4).

Ameloblastomas are characterized by an aggressive potential for local invasion and a high recurrent potential requiring accurate histological diagnosis and appropriate treatment (5).

The purpose of this article is to present a rare case of ameloblastoma in the left maxillary sinus with infratemporal fossa involvement in an asymptomatic patient.

This article has been reported in line with SCARE criteria (6).

2. Case presentation

A 54-year-old Tunisian woman was referred to the ENT department following the accidental discovery of an ulceration in front of alveolar processes of the left maxilla. There were no other significant symptoms. In addition, the medical history was clear, and no relevant medical event was documented.

The patient was medically fit and well.

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Nasal endoscopy showed a medial bulging of the left intersinusonasal septum.

There was no mass or polyposis in the nasal cavity. There were no cervical lymph nodes.

As presented in Fig. 1, Intraoral examination revealed partially edentulous with painless ulceration of 2 cm of the left alveolar process of the maxilla.

A computed tomography scan of the paranasal sinus was performed, it sowed a completely obliterated left maxillary sinus by a tumor process. The mass eroded the dorsal wall of the maxillary sinus into the infratemporal fossa and expanded through the medial and inferior wall of the maxillary sinus to the nasal cavity (Fig. 2).

A biopsy performed from the ulceration showed a follicular ameloblastoma without signs of malignancy transformations (Fig. 3).

The tumor limits suggested a transvestibular maxillary sinus surgery. in our case, we discussed the therapeutic course with our patient, who is a well-informed person, and decided to minimize the radicality.

Transnasal functional endoscopic sinus surgery combined with a trans vestibular approach was performed under general anesthesia. We additionally resected parts of the posterior wall of the maxillary sinus. A targeted portion of the inferior portion of the maxilla was removed as a part of the tumor resection. A histological clear margin was being achieved. No primary reconstruction of the buccco sinus defect was performed. The Analysis of the surgical specimen revealed the same morphological features of the incisional tissue.

There were no intraoperative or postoperative complications disturbed the patient's recovery.

A control MRI done after 3 months showed a 14 mm tumor residue in the left pterygopalatine region.

The patient was reoperated by a team made of otorhinolaryngology and maxillofacial surgeons. A trans vestibular approach was performed, and the tumor was resected with clear surgical margins. The reconstruction of the bucco-sinus defect was done.

The patient showed no signs of clinical or radiologic recurrence within a follow-up period of 6 months.

3. Discussion

Ameloblastoma is a rare odontogenic epithelium neoplasm, it accounts for only 1 % of all maxillary tumors and 11 % of odontogenic tumors (7,8). The geographical distribution varies according to country of origin and ethnicity, ameloblastoma has no link to the region of residence or climate, but the frequency is significantly higher in the black African population (9,10).

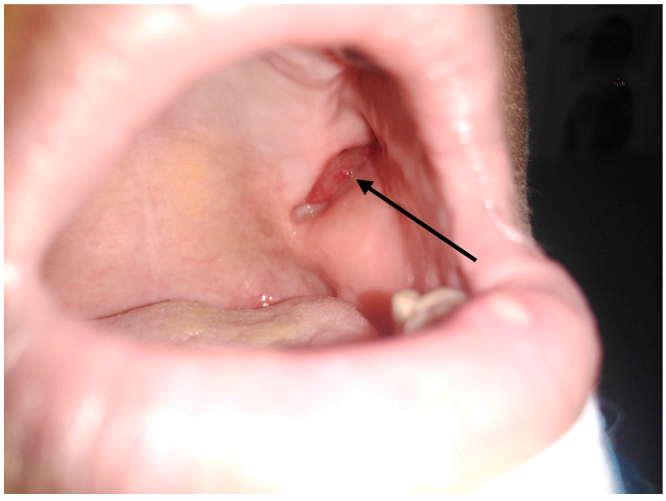

Fig. 1. Ulceration of 2 cm long axis of the left alveolar process of the maxilla (black arrow).

According to the published series, the average age of the patients with Classical ameloblastoma is generally between 40 and 50 years, with an average age of discovery of 40 years (5), Our patient is 54 years old.

Histological examination: The typical morphology of ameloblastoma is that of odontogenic epithelial islets simulating the stellar reticulum, peripherally palisaded by cylindrical cells with inverted nuclear polarity, filled with angular cells loosely arranged (3).

The follicular pattern is the most common response found in the series. as is the case with our patient (11,12). It is characterized by a composition of neoplastic cell nests with central descement of short fusiform epithelial cells resembling the star reticulum of the developing dental germ, surrounded by palisade cylindrical cells with inverted nuclear polarity (3).

For the maxillary location of ameloblastoma, it is rare compared to that occurring at the mandible (11).

Unlike the compact mandible bone, the maxillary is a spongy bone, which allows the tumor to more easily invade and spread to adjacent structures such as the nasal cavity, para nasal sinuses, orbits, para pharyngeal tissues and base of the skull (13), and in these cases the ameloblastoma may manifest with different symptomatology such as facial deformation, usually unilateral, toothache, headache, nasal obstruction, nasal epistaxis and visual disturbances (11). In our case, the patient was asymptomatic with a fortuitous discovery despite the extension of the maxillary ameloblastoma to the nasal cavity, the maxillary sinus and the homolateral infra temporal fossa.

For descriptive reasons, maxillary ameloblastomas are also divided into posterior or anterior tumors. Ameloblastomas have a preference for the posterior maxillary (9).

The radiological aspect is not specific. It varies depending on the type of ameblastoma. However, MRI has shown important impact in the differential diagnosis and therapeutic planning for providing information about soft tissue (15) .

Radical surgery represents current standard treatment option for ameloblastomas and this will depend on the histological subtype, and surgical technique to be performed in order to obtain surgical margins free of neoplasia, which could reduce local recurrences (16,17).

The current treatment of choice is wide local excision with clear margins and immediate reconstruction. An open partial maxillectomy with a 10–15 mm safety margin of healthy bone including the alveolar ridge, the hard palate, the mucosa of the maxillary sinus and the lateral nasal wall is often practiced (10). further radiation can be a feasible therapy option (7).

The risk of recurrence is in the range of 50 % to 72 % of cases (7).

To reduce the recurrence of ameloblastoma it is important to identify the risk factors associated with recurrence. Rong et al. showed that recurrence was associated with several clinical factors such as: character of the site, root resorption and presence of invasion of the maxillary sinus regardless of the surgical method used [14].

4. Conclusion

Ameloblastoma is a rare tumor, benign but locally very aggressive, they can be found anywhere within the maxillary region.

The anatomical particularities of the maxillary sinus make the diagnosis made commonly at a late stage. The most observed histological patterns were the follicular and plexiform types. Its treatment is mainly surgical.

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Ethical approval

This study is exempt from ethical approval at our institution.

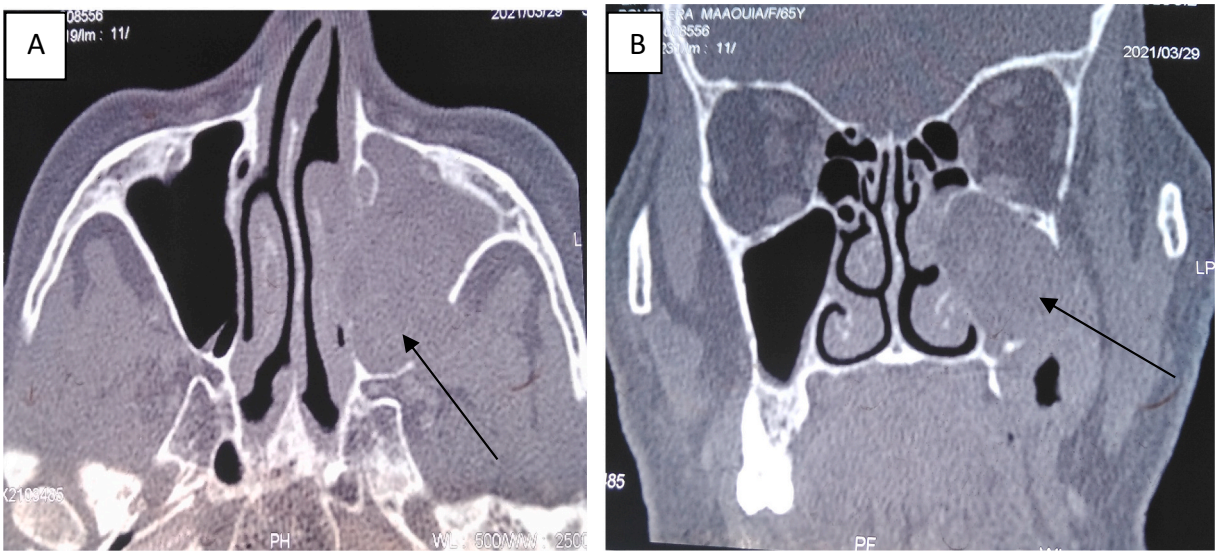

Fig. 2. Preoperative axial (A) and coronary(B) CT scan with a tumorous mass in the left maxillary sinus spreading to infratemporal fossa.

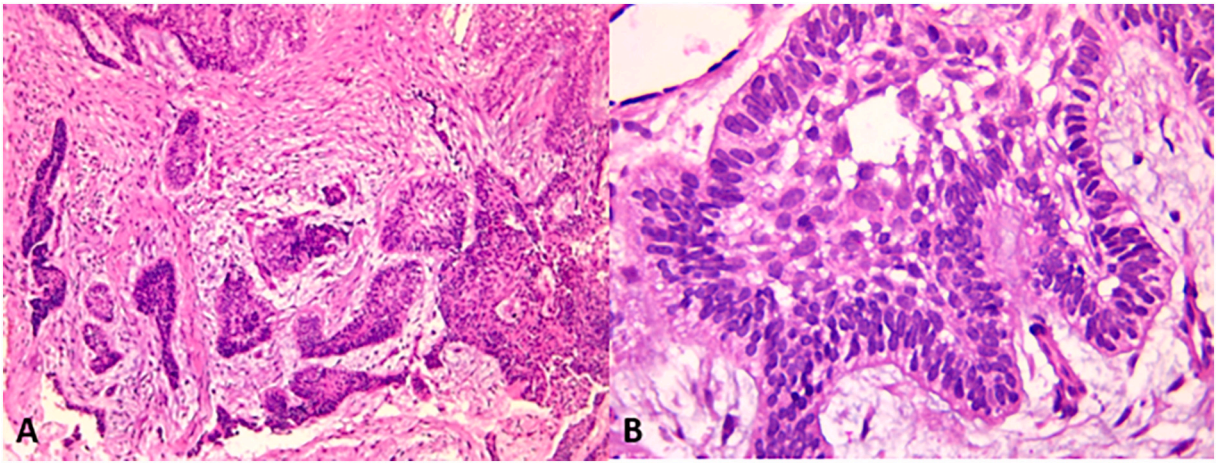

Fig. 3. Follicular ameloblastoma: A/ Anastomosing strands of epithelial cells surrounded by hypocellular stroma (HE × 100). B/ Peripheral palissading tumor cells with more loosely arranged cells in the center (HE × 400).

Consent

Written informed consent was obtained from the patient's parents for publication of this case report and accompanying image. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Registration of research studies

Not applicable.

Provenance and peer review

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Guarantor

Azer Chebil

CRediT authorship contribution statement

Azer CHEBIL: Writing - Original Draft,**Mahdi Hasnaoui:** Writing - Review & Editing,

Salma Bhar: Writing - Original Draft, **Mohamed Masmoudi**: Writing - Review & Editing.

Ahlem Bellalah: Resources, **Kalifa Mighri:** supervision.

Declaration of competing interest

The authors report no declarations of interest.

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