CASE REPORT

Maxillary unicystic ameloblastoma – case presentation

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ABSTRACT

BACKGROUND. Approximately one fifth of all neoplasms arising from the embryonic odontogenic apparatus are ameloblastomas. The origin of ameloblastomas is in the epithelial cells involved in the formation of teeth. The mandible is the most affected site, but maxillary ameloblastoma is the most dangerous kind because of its invasive and aggresive evolution.

MATERIAL AND METHODS. A 43-year-old woman was referred to our Department with right nasal obstruction, right sided facial fullness with paraesthesia and chronic frontal headache. The CT scan examination revealed an osseous tumor, with cystic resemblance, arising from the alveolar plate and occupying the right maxillary sinus. Complete removal of the tumor was performed. The histopathological diagnosis was luminal unicystic ameloblastoma.

CONCLUSION. Maxillary ameloblastomas are relatively rare tumors, especially the unicystic type, and their high recurrence rate requires complete surgical removal. In case of luminal unicystic ameloblastoma, complete removal of the tumor offers the best hope for cure without radiotherapy or chemotherapy and reduces the recurrence incidence.

KEYWORDS: unicystic ameloblastoma, maxilla, luminal type, odontogenic tumor

INTRODUCTION

First described by Churchill in 1933, ameloblastomas are the most common odontogenic benign tumors with epithelial origin[1,2]. They represent almost 10% of all tumors of the maxilla and mandible, in 80-85% of the cases involving the mandible and in 15 to 20% the maxilla[3-6]. 50% of the ameloblastomas of the maxilla develop in the molar area, involving the maxillary sinus in 15% of the cases[2,6].

Based on the overall histologic architecture, ameloblastomas can be divided into three types: solid or multicystic, unicystic and peripheral or extraosseous[7]. The unicystic type of ameloblastoma is more frequently encountered asymptomatically in the posterior mandible[7,8].

Because of its invasive natural evolution, the treatment of choice in maxillary ameloblastomas is radical surgery; recurrence can occur in 90-100% of situations, in case of incomplete removal[6].

CASE REPORT

A 43-year-old woman was referred to our Department with right nasal obstruction, right sided facial fullness with paraesthesia and chronic frontal headache. Duration of the symptoms was almost two years. She underwent different treatments for right chronic rhinosinusitis with no clinical improvement.

The ENT clinical examination and nasal endoscopic evaluation revealed the congestion of the right nasal mucosa, with no anterior or posterior discharge and no pathologic lesions in the middle meatus.

The result of the cranio-facial CT-scan showed an oval osseous tumor of 18.1/24.4mm, with cystic resem-

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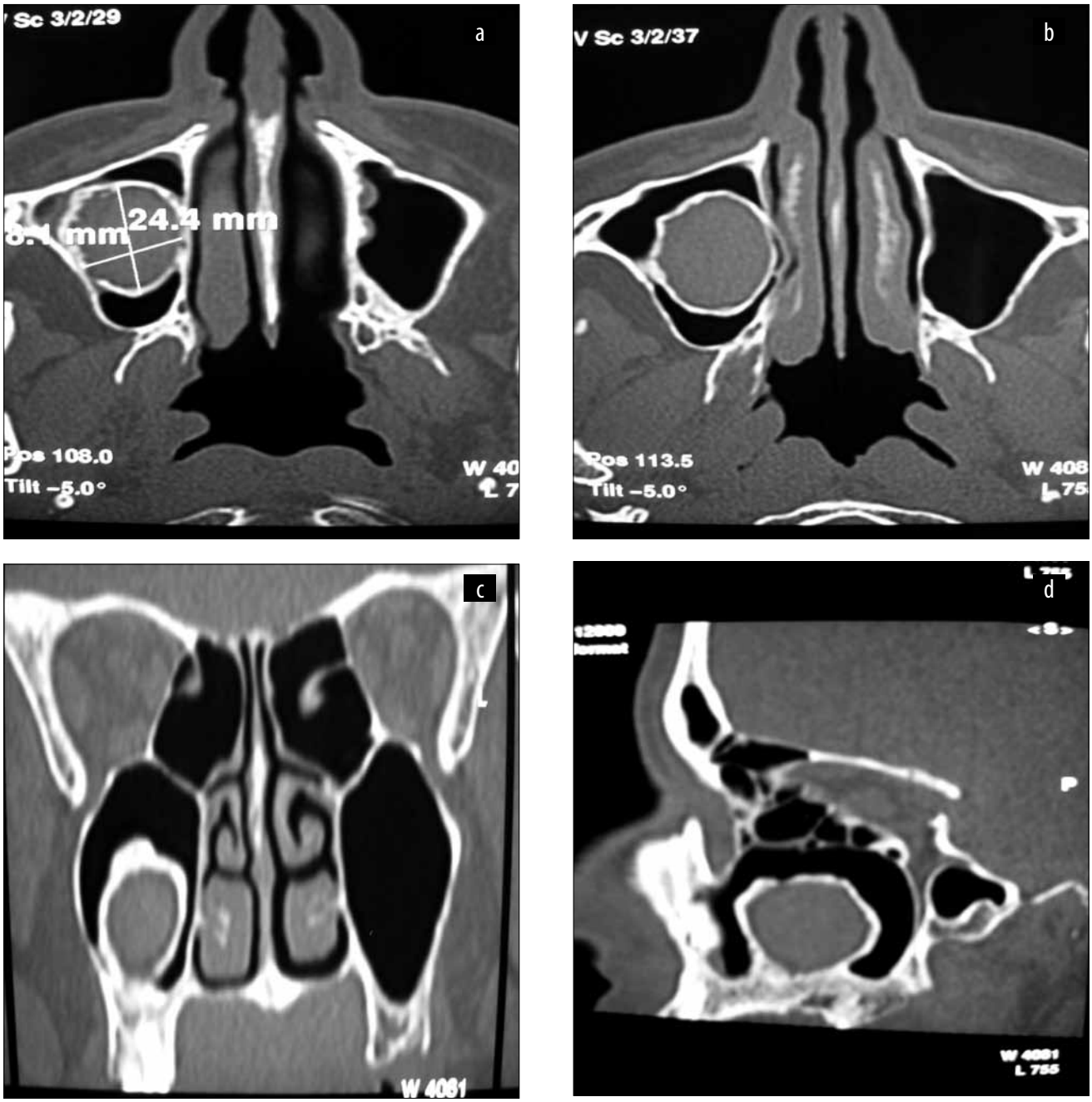

Figure 1 Cranio-facial CT scan – axial **(a,b)**, coronal **(c)**, sagittal **(d)** slices - oval osseous tumor of 18.1/24.4mm, with cystic resemblance, arising from the alveolar plate and occupying almost entirely the right maxillary sinus.

blance, arising from the alveolar plate and occupying almost the entire cavity of the right maxillary sinus (Figure 1a,b,c,d).

The treatment consisted in Caldwell-Luc approach of the right maxillary sinus, performed under general anaesthesia (Figure 2), with complete removal of the tumor. The histopathologic diagnosis was luminal unicystic ameloblastoma, with a hyper-chromatic polarized basal layer and the overlying epithelium being loosely cohesive and resembled stellate reticulum (Figure 3).

The patient's postoperative evolution was within normal limits, with no recurrence at 10 month reassessment.

DISCUSSIONS

Approximately one fifth of all neoplasms arising from the embryonic odontogenic apparatus are ameloblastomas[9]. The origin of ameloblastomas is in the epithelial cells involved in the formation of teeth (including enamel organ, odontogenic rests - cell rest of Malassez, cell rest of Serre, epithelial lining of odontogenic cyst)[10,11].

The most common type of ameloblastoma originates centrally within the bone and tends to be invasive and aggressive. Rarely, an ameloblastoma will

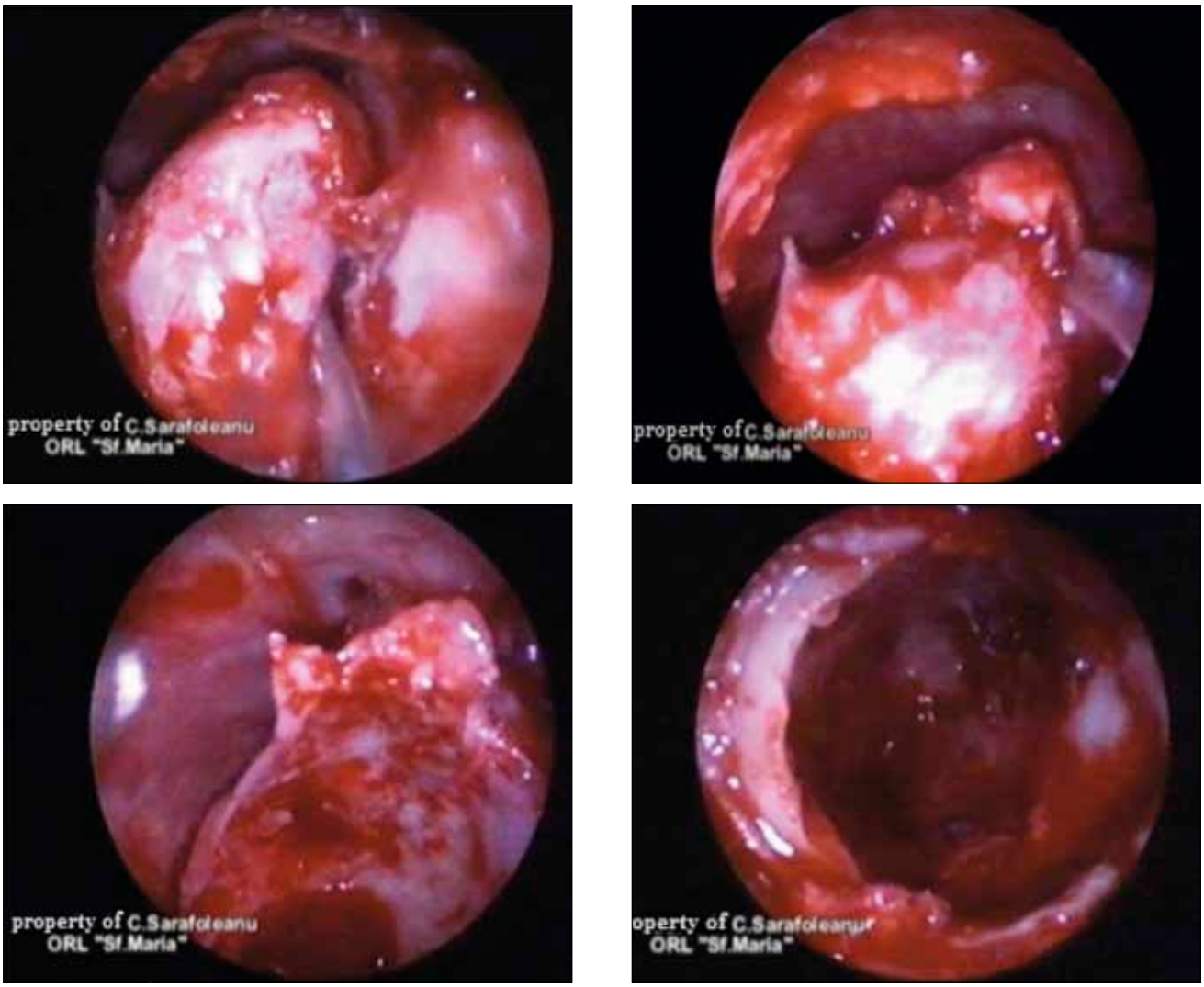

Figure 2 Endoscopic examination – intraoperatory view

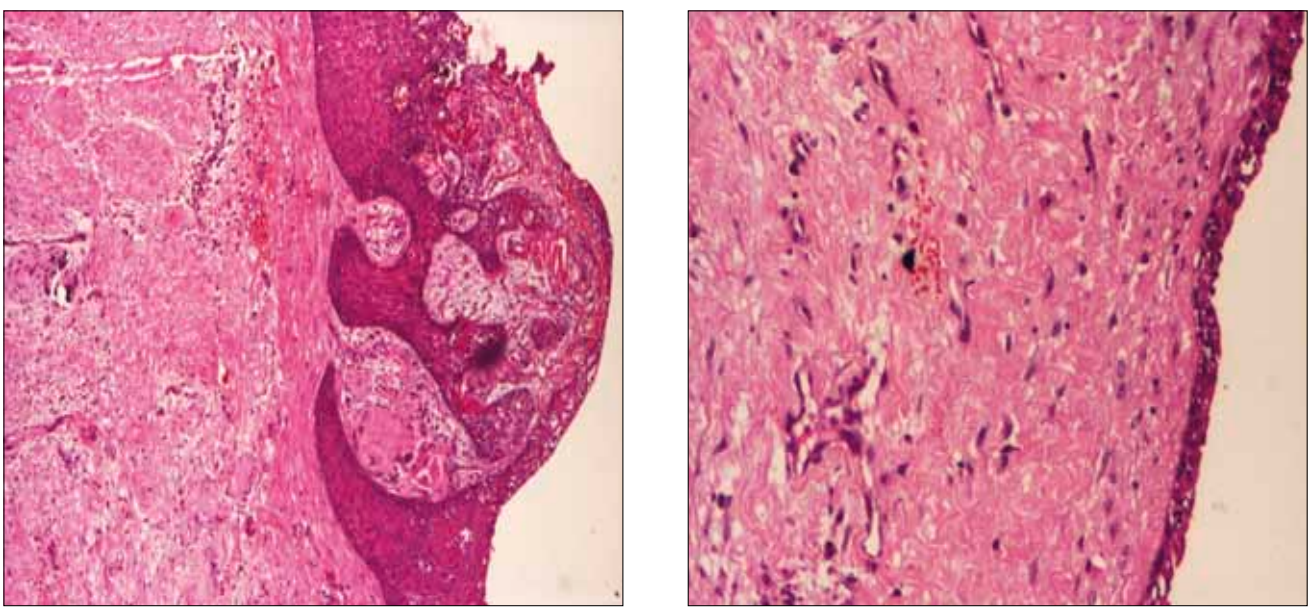

Figure 3 Histopathologic examination - ameloblastic epithelium with hyper-chromatic polarized basal layer; the overlying epithelium was loosely cohesive and resembled stellate reticulum.

arise from the wall of a dentigerous cyst – unicystic ameloblastoma[12].

The unicystic ameloblastoma was first described by Robinson and Martinez in 1977[13]. Later on, Ackermann et al. classified this tumor into three types, according to prognostic and therapeutic aspects[14-16].

The first type, luminal unicystic ameloblastoma, is characterized by a lesion consisting of fibrous cyst wall with a lining of ameloblastic epithelium; the tumor is confined to the luminal surface of the cyst. According to the microscopic evaluation, the overlying epithelium is loosely cohesive and resemble stellate reticulum. The same aspect was found in our case.

Intraluminal or plexiform unicystic ameloblastoma is the second form described and it is characterized by epithelial nodules which arise from the cystic wall lining into the lumen.

The third ameloblastoma type, mural unicystic ameloblastoma, has a fibrous wall infiltrated by typical follicular or plexiform ameloblastoma.

The radiological characteristic aspect of this tumor is a honeycombed or bubble-like radiolucent structure, without distinct borders or calcification in the matrix. Resorption of the roots of adjacent teeth is also a common finding. CT or MRI scans are very useful, but not pathognomonic, the ameloblastoma being often misdiagnosed as a dentigerous cyst or odontogenic keratocyst[17].

Maxillary ameloblastoma is the most dangerous kind and requires "en bloc" resection, because of its high recurrence rate. Surgery in these cases is the treatment of choice and it must be radical from the very beginning[18]; moreover, long-term follow-up is advised because ameloblastomas' recurrence may be long delayed.

CONCLUSIONS

Maxillary ameloblastomas are relatively rare tumors, especially the unicystic type, their high recurrence potential warranting expedient complete removal of the tumor. Although clinical and radiological evaluation aid in the differential diagnosis, the histopathologic examination is the one which confirms the positive diagnosis of ameloblastoma.

In case of luminal unicystic ameloblastoma, complete removal of the tumor offers the best hope for cure without radiotherapy or chemotherapy and reduces the recurrence incidence. Also, it requires long-term follow-up using CT scan examination.

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