



## Recurrent maxillary ameloblastoma: A case report

Mahdi Azadi, Farnoosh Mohammadi, Narges Hajiani \*

Department of Oral and Maxillofacial Surgery, School of Dentistry, Tehran University of Medical Sciences, Tehran, Iran.

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\*Corresponding author:

Narges Hajiani

Department of Oral and Maxillofacial Surgery,  
School of Dentistry, Tehran University of Medical  
Sciences, Tehran, Iran.

Tel: +98-21-66581544

Fax: +98-21-66428655

Email: na72hajiani@gmail.com

### ABSTRACT

Ameloblastoma is one of the most common types of oral odontogenic tumors. As per literature, ameloblastoma mostly occurs in the mandible but the maxillary ameloblastoma has a more aggressive behavior due to anatomical features. Also, unicystic ameloblastoma may have lower recurrent rate. In this case report, we present a 60-year-old male patient with a history of unicystic ameloblastoma, which the intraluminal adenomatoid odontogenic tumor excisional biopsy surgery was performed but the patient didn't follow the treatment completely, and after two years he came back with swelling of the right upper alveolar ridge. After the second surgery, the histopathologic report revealed a mixed plexiform-follicular ameloblastoma recurrence and it seemed that previous surgery was not sufficient and more radical treatment is needed for the lesion.

**Keywords:** Ameloblastoma; Maxilla; Adenomatoid odontogenic tumor.

## Introduction

One of the most common tumors of the jaw is ameloblastoma. This tumor was first recognized by Cusack in 1827 and explained by Broca in 1868 [1]. Ameloblastoma occupies 30% of all benign odontogenic originated tumors of the jaw [2]. It is a benign tumor that could be locally invasive but has a high tendency to recur. Most of the diagnosed cases are in the third to fifth decades of life and there is a rare occurrence in children [3]. Ameloblastoma has a variable geographic prevalence. It is the most common odontogenic tumor in China and Africa and also stands as the second common type of the

odontogenic tumor in the America and Canada [4]. At the beginning, the clinical features present as a swelling without any symptom, gradually followed by tooth mobility or displacement and root resorption. Consequently, cortical bone involvement and expansion may lead to functional compromise [5]. Ameloblastomas have been categorized into three biologic subtypes based on the behavioral pattern, clinical and radiographic features and prognostic factors. The three categories are cystic, solid, and peripheral. There are several histopathologic subtypes, but the most common are follicular, plexiform, acanthomatous, granu-

lar and desmoplastic [6]. When an ameloblastoma grows up to a considerable size, it can threaten the airway and the gastrointestinal system and can even increase the mortality risk, therefore it can be symptomatic. Specially in the maxilla due to its spongy structure, ameloblastomas can reach a significant size and spread through the sinuses, orbits, nasal cavity or cranium [7]. Oncogenic promoters for transformation of odontogenic epithelium into ameloblastoma are some molecules and genetic factors that are linked to dysregulation of the variant genes strongly associated with sonic hedgehog, mitogen-activated protein kinase, and WNT/b-catenin signaling pathways [8].

There are variable types of treatment for this tumor, but recurrence rate is related to the treatment approach, which can be as high as 15% to 25% and 75% to 90%, after radical and conservative treatment respectively [9]. Maxillary ameloblastomas are considered rare and because of the anatomic consideration and adjacent structures are treated more severely to decrease the risk of reoccurrence [10]. In this case, almost 33% of maxillary ameloblastomas involve the maxillary antrum and the nasal floor [11] and tend to grow with buccal expansion and invade the nasal floor [12]. Unicystic ameloblastoma is presented as a cystic lesion in clinicoradiographic features like a mandibular cyst, but histopathologically, it involves some attributes of typical ameloblastomatous epithelium [13].

In unicystic ameloblastoma, the recurrence rate is as low as 10% to 25% with the conservative treatment. However, in some studies it is expressed that this rate is influenced by the degree of ameloblastic epithelial invasion so this variety should be considered in the treatment planning [14]. Adenomatoid odontogenic tumor (AOT) is another odontogenic epithelial tumor. AOT requires only surgical excision. But presenting with unicystic ameloblastoma or another hybrid tumor, this may change the treatment plan in a more radical way and a long term follow up may be required [15].

So ameloblastoma is considered as a major maxillofacial related problem, due to functional and cosmetic impressions especially in the maxilla. unicystic ameloblastoma can cause similar consequences by incorrect treatment. Here, we are reporting a case with the ameloblastoma of the upper jaw, arising from a unicystic ameloblastoma with atypical presentation of intraluminal proliferation, resembling adenomatoid odontogenic tumor that may often be misdiagnosed.

## Case Report

A 60-year-old male patient with past medical history of ischemic heart disease, type 2 diabetes mellitus, hypertension and long-term opioid addiction presented to the department of the maxillofacial surgery of Shariati Hospital in Tehran, Iran. The patient had a history of previous exact site excisional biopsy surgery two years ago with histopathologic diagnosis of unicystic ameloblastoma, with a focus of intraluminal proliferation resembling adenomatoid odontogenic tumor. In that excisional biopsy, microscopic examination was as follows according to the histopathologic report. A cystic lesion lined by stratified squamous epithelium demonstrating palisading and prominent reverse polarity at some areas in close association with stellate-like reticulum cells. Also, the epithelium showed areas of swirling pattern. A focus of dentinoid material aggregation was seen in close association with supporting connective tissue (Figure-1 & 2).

The patient didn't complete his treatment and after two years he came back with swelling of the posterior right maxillary alveolar ridge, which occurred over the past year. General clinical examination revealed complete edentulous ridge of both jaw, normal mouth opening, normal mucosa, swelling of right part of the palate and alveolar ridge that was partially bony and partially soft. Normal orofacial sense was declared by the patient and muscle movement was normal. On examination, no lymph nodes were tender and palpable (Figure-3).

The patient revealed that he lost his last teeth around 10 years ago and had been wearing complete dentures. Radiographic Evaluation of spiral face CT scan (axial, coronal, sagittal, 3D) without contrast, OPG and CBCT revealed a lytic expansion lesion in the right maxillary bone and its alveolar process compressing right maxillary sinus and bulging into right oral cavity. Perforation was observed in some regions of buccal and palatal bone (Figure-3). Based on the clinicoradiographic examination, a provisional diagnosis of recurrent unicystic ameloblastoma was made.

Then an incisional biopsy was carried out and the hematoxylin and eosin stained tissue section revealed epithelial odontogenic tumor composed of bilayered strands, small cystic spaces and follicles lined by tall columnar peripheral ameloblast-like cells with reverse polarity, palisaded nuclei and loose fibrovascular stroma representing a plexiform ameloblastoma. In the next step, surgical resection of the solid mass compos-

ing of bone and soft tissue was carried out with safe margin and the right total maxillectomy, according to Weber Fergusson's approach. The orbit was preserved, but due to involvement, inferior nasal turbinate was resected too (Figure-4). Histological sections from the surgical specimen revealed a solid, homogenous tan surface lesion lined by irregular strands of epithelium, bordered by columnar palisading cells that surround an island of cells resembling stellate reticulum. The lesion was diagnosed as mixed plexiform-follicular ameloblastoma.

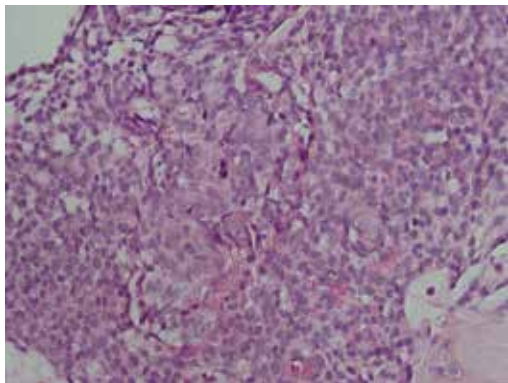


Fig. 1. AOT formation in odontogenic epithelium.

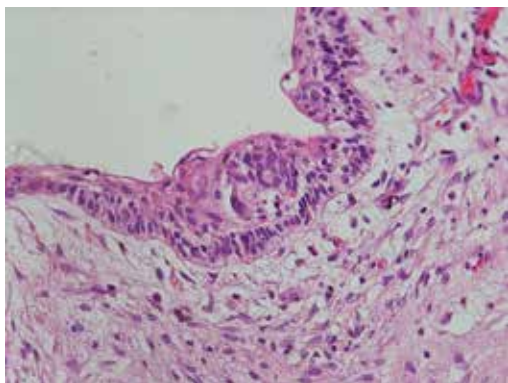


Fig. 2. Varying histologic patterns of ameloblastoma are appreciated at low power.



Fig. 3. Clinical and radio graphical appearance of the lesion.

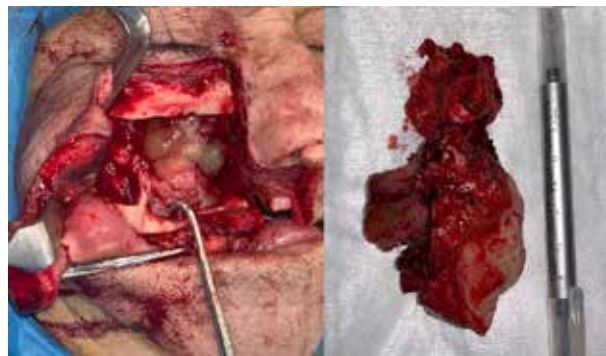


Fig. 4. A representative macroscopic specimen of the ameloblastoma and surgery approach.

## Discussion

Ameloblastoma is considered as one of the most common benign odontogenic tumors of the oral cavity. Posterior mandibular region is the most common site, but the maxillary ameloblastomas behave more aggressively and are often diagnosed when invasion into adjacent anatomic structures occurs [4,10]. Sinonasal ameloblastomas are radiographically different from their peers in the other regions of the jaw. Sinonasal lesions are frequently radiopaque, solid lesions that fill the nasal cavity or sinus. In contrast ameloblastomas within the jaw appear to be more radiolucent and commonly are “honeycomb-like” or “bubble-like” [16]. Some of the ameloblastomas are accompanied by other odontogenic tumors such as AOT. Solitary AOT has a striking tendency to occur in younger patients, especially in the anterior part of their jaws [17].

Occurrence of AOT with ameloblastoma in the posterior of mandible, especially in older patients may indicate that such lesions represent unicystic ameloblastomas with secondary AOT alteration. So, it is important to examine the full tissue specimens to find any ameloblastomatous changes [15]. Recurrence of ameloblastoma can be related to variable factors such as surgical approach, region and histopathologic type. Conservative management has a higher incidence of recurrence than radical excision. Some studies suggest that conservative treatments, by limiting resection (enucleation), curettage, partial maxillectomy without recurrence are also successful [13,18].

## Conclusion

According to our study, it is suggested that a unicystic ameloblastoma especially in the maxilla can behave like a solid ameloblastoma or convert to it. Also accompanying with AOT does not show a better prognosis and it seems that more severe treatments like block resection is better than conservative surgeries in

ameloblastomatous lesions, due to propensity for regional invasion and risk of recurrence.

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### Conflict of Interest

There is no conflict of interest to declare.

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