Journal of Craniomaxillofacial Research

Vol. 7, No. 2 Spring 2020

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.

ARTICLE INFO

Article Type:
Case Report

Received: 2 Sep. 2019
Revised: 8 Nov. 2019
Accepted: 1 Feb. 2020

**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 butthe 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 seemedthat 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% ofall benign odontogenic originated tumors of the jaw [2]. It is a benign tumor that could be locally invasive but has a high tendency to reoccur. Most ofthe 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. Itis the most common odontogenic tumor in China and Africaand also stands as the second common typeof 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 expansionmay lead to functional compromise [5]. Ameloblastomas has been categorized into three biologic subtypes based on the behavioral pattern, clinical and radiographic features and prognostic factors. The three categories arecystic, 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 threatentheairway and the gastrointestinal system and caneven increase the mortality risk, thereforeit 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 thevariant 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 severelyto 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 clinico radiographic featuressolike a mandibular cyst, but histopathologically, it involvessome attributes of typical ameloblastomatous epithelium [13].

In unicystic ameloblastoma, the recurrence rate is as low as 10% to 25% with the conservative treatment. Howeverin 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 planin 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 acase with theameloblastoma of the upper jaw, arising from a unicystic ameloblastoma withatypical presentation of intraluminal proliferation, resembling adenomatoid odontogenic tumor that mayoften be misdiagnosed.

Case Report

A 60-year-old male patient with past medial 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 hada 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 follow 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 occurredover thepast year. General clinical examination revealed complete edentulous ridge of both jaw, normal mouthopening, normal mucosa, swelling of right part ofthe 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 andpalpable (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 expiation 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 fibro vascular stroma representing a plexiform ameloblastoma. In the next step, surgical resection of the solid mass compos-

ing of bone and soft tissue was carriedout with safe margin andthe 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 surgicalspecimen revealed a solid, homogenous tan surface lesion lined by irregular strandsof epithelium, bordered by columnar palisading cells thatsurround an island of cells resembling stellate reticulum. The lesion was diagnosed as mixed plexiform-follicular ameloblastoma.

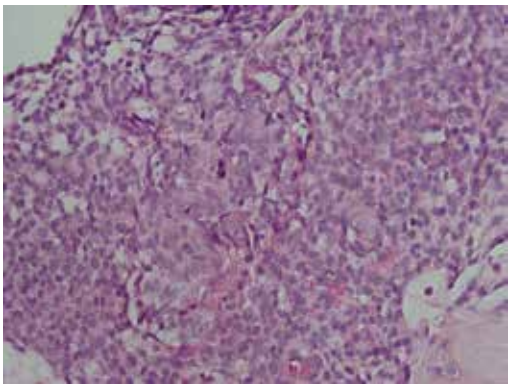

Fig. 1. AOT formation in odontogenic epithelium.

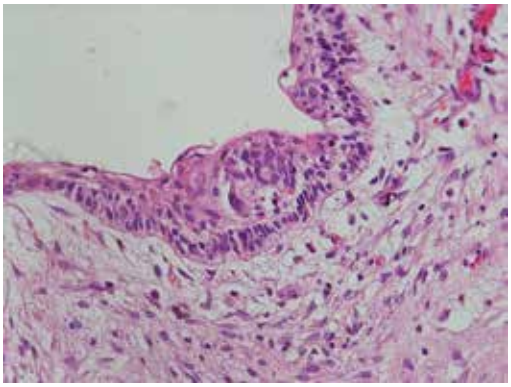

Fig. 2. Varying histologic patterns of ameloblastoma are appreciated at low power.

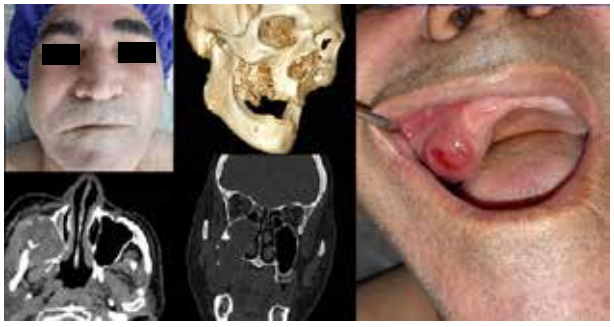

Fig. 3. Clinical and radio graphical appearance of the lesion.

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 arefrequently radiopaque, solid lesions that fill the nasal cavity or sinus. In contrast ameloblastomas within the jaw appear to bemore 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 theirjaws [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 AOTalteration. 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 dos not show a better prognosis and it seems that more severet reatments like block resection is better than conservative surgeries in

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

Acknowledgment

We would like to thank the Department of Oral and Maxillofacial Pathology of the Dental Faculty of University of Tehran, for all of their collaboration and technical support.

Conflict of Interest

There is no conflict of interest to declare.

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Please cite this paper as:
Azadi M, Mohammadi F, Hajiani N; Recurrent maxillary ameloblastoma: A case report. J Craniomax Res 2020; 7(2): 101-104