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Ameloblastomatous Calcifying Odontogenic Cyst: A Rare Lesion

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Introduction: Calcifying odontogenic cyst (COC) is a unique and uncommon odontogenic cyst classified into four groups of cystic, odontoma producing, ameloblastomatous proliferating and neoplastic ones.
Case Presentation: A 34-year-old Iranian man complaining of a painless facial and palatal swelling of the left side of the maxilla persisted for approximately three years was referred to the department of oral and maxillofacial surgery, Hamadan University, Iran. Panoramic film revealed a well-defined multilocular mixed radiolucent and radioopaque lesion of the maxilla at the left side. An incisional biopsy was obtained. Based on the histopathologic findings, ameloblastomatous COC was diagnosed.
Discussion: We reported a rare case of COC. According to Praetorius et al. classification, this patient comes under the category of type 1C (ameloblastomatous proliferating). Many patients with ameloblastomatous COC should be reported to understand its biological behavior as possible.

*Keywords:*Cyst; Calcifying Odontogenic Cyst; Radiolucent

1. Introduction

Calcifying odontogenic cyst (COC) is a unique and uncommon odontogenic lesion first described by Gorline et al. in 1962. In 1971, WHO defined COC as a cystic lesion (1, 2). COC is a developmental odontogenic cyst. It is considered as a rare lesion and accounts for 1% of jaw cysts (3). There are heterogeneous groups of COC that demonstrate variable clinical and histopathological features. In total, 86-98% of COCs are cystic and others are neoplastic (1, 4). In 2005, WHO reclassified COC as calcifying cystic odontogenic tumor (4). Ameloblastomatous COC is a rare variant of COC, so we presented a patient with this type of COC to understand its clinical and biological behaviors.

2. Case Presentation

A 34-year-old Iranian man was referred to the department of oral and maxillofacial surgery, Beasat Hospital, Hamadan University of Medical Sciences, Iran in March 2013. The patient's chief compliant was left-sided painless facial and palatal swelling persisted for approximately three years. Swelling started gradually and increased to gain the current size. His medical history was not-contributory. Head and neck clinical examination revealed mild left midfacial enlargement, so that the ipsilateral nasolabial fold was obliterated partially. No associated pain or paresthesia was present. Furthermore, there was no evidence of nasal obstruction or cervical lymphadenopathy. Intraoral, the patient was edentulous and there was a buccal and palatal expansion of the left maxilla extending from almost the lateral incisor region to the first molar area. This bony hard swelling was not tender to palpation and the overlying mucosa was normal. Panoramic film revealed a well-defined multilocular mixed radiolucent and radio-opaque lesion of the maxilla at left side (Figure 1).

Computed tomography disclosed a large corticated lytic lesion, which caused medial displacement of the lateral wall of the nasal cavity and upward movement of maxillary sinus (Figure 2).

Based on clinical and radiological findings, a provisional differential diagnosis of calcifying odontogenic cyst (COC), Cemento ossifying fibroma and ameloblastoma were considered. Adenomatoid odontogenic tumor was not included in differential diagnosis because of the lack of teeth impacted teeth. An incisional biopsy was obtained

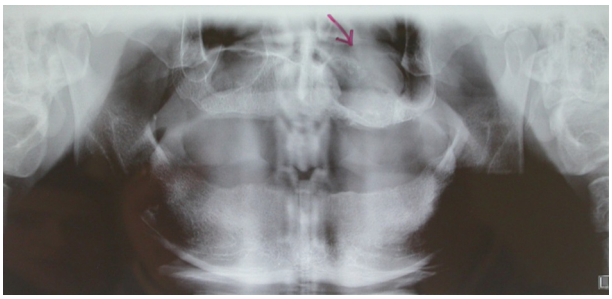


Figure 1. Panoramic Film, a Well-Defined Multilocular Mixed Radiolucent and Radio-Opaque Lesion of the Maxilla at Left Side

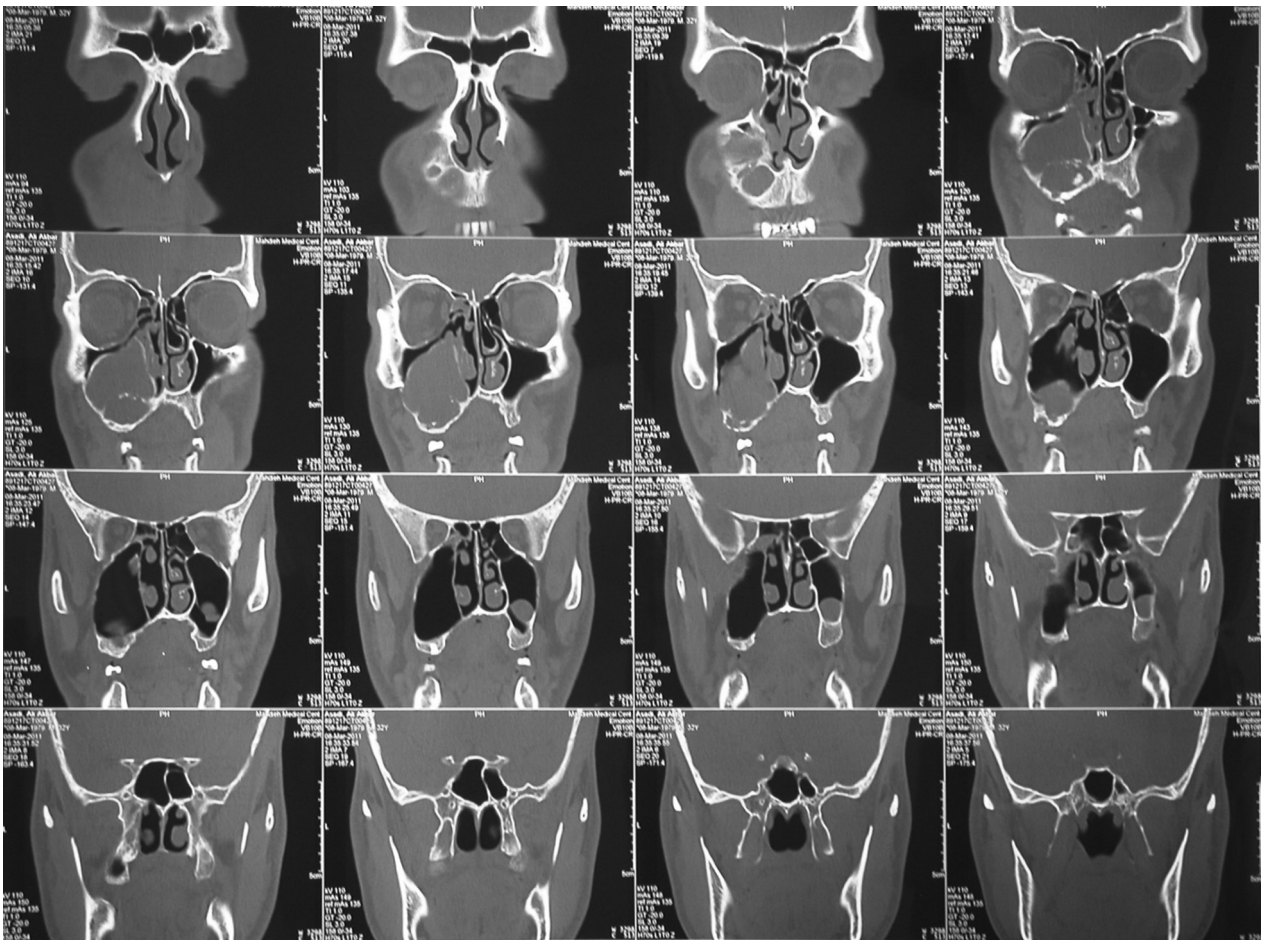
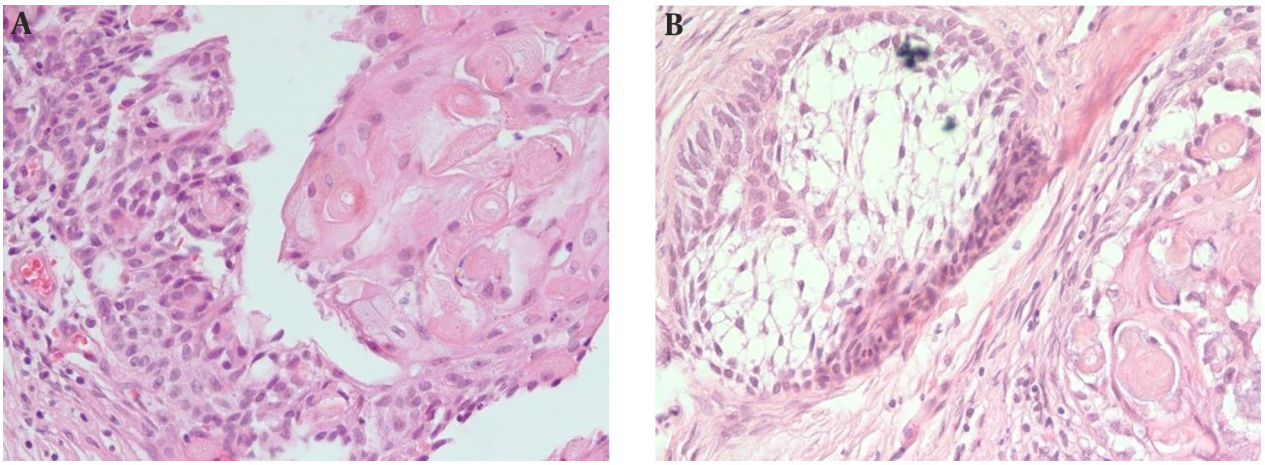


Figure 2. Computed Tomography, a Large Corticated Lytic Lesion, Which Caused Medial Displacement of the Lateral Wall of the Nasal Cavity

Figure 3. Photomicrograph of the Cystic Lesion



A) Cystic lesion lining by odontogenic epithelium with ghost cells, B) Ameloblastomatous proliferations with mainly follicular pattern were seen in the connective tissue wall of the cyst (Haematoxylin and Eosin Stain; ×100).

from the anterior region of the lesion to establish final diagnosis. During the biopsy procedure, a cystic lesion filled with purulent fluid was observed. Histopathologic examination revealed cystic lesion lined with basal cell layer resembling the ameloblast cells. Cells above the basal cell layer resembled stellate reticulum and had loosely

arrangement. Many masses of ghost cells and spherical calcifications were seen within the epithelium and connective tissue wall of the cyst. Ameloblastomatous proliferations with mainly follicular pattern were seen in the connective tissue wall of the cyst (Figure 3 A, B). Based on these histopathologic findings, ameloblastomatous COC was diagnosed.

The patient was operated under general anesthesia with nasotracheal intubation. During the operation, cystic lesion was enucleated through intraoral approach. The cyst was easily removed from the bone and curettage of the bony cystic wall was performed by a surgical bur after enucleating the cyst. Microscopic examination of excisional biopsy reconfirmed the previous diagnosis of incisional biopsy. The postoperative course was uneventful. No evidence of recurrence was noted after one year since writing the manuscript (Figures 1 - 3).

3. Discussion

COC is an unusual lesion with variable clinical and histopathological features. Some of them have cystic features but others show characteristics of a solid neoplasm (3, 5). Therefore, there are some classifications of COC. In 1981, Praetorius first described the classification of COC as type 1A (cystic), type 1B (odontoma producing), type1C (ameloblastomatous proliferating), and type 2 (neoplastic). Toida and et al. classified COC as cyst and neoplasm (4, 6-8). The cystic variant accounts for 80-98% of lesions and neoplasm variant 11.5%. Buchner classified this lesion as peripheral and central. The frequency of COC is estimated as less than 1% (1). Of 766 cases diagnosed by the oral and maxillofacial pathology department of Hamadan University of Medical Sciences, 7 (0.9%) cases of COC were reported, which only 2 (0.26%) cases had ameloblastomatous COC. In 2006, Jones et al. reported 21 (0.3%) cases of COC of 7121 odontogenic cyst from the files of department of oral pathology in Sheffield (England) (9). Of 14106 cases diagnosed by the oral and maxillofacial pathology department of Tehran University of Medical Sciences, only 56 (0.4%) cases of COC were reported (5). Ameloblastomatous COC is a rare variant type of COC (1). COC affects maxilla and mandible equally with predilection to the anterior side. It presents with a painless slowly growing lesion affecting young adults in the second decade of life without gender predilection (4). Kamboj, Aithal, and Ide et al. reported cases of ameloblastomatous COC with swelling of mandible, which is inconsistent with our study (1, 3, 10). In the present study, ameloblastomatous COC occurred in a man in fourth decade of life. Aithal and Singh presented female cases in the third decade of life and Menal reported a case of in a man in the third decade (3, 8). COC is generally a unilocular radiolucent lesion; some of them are well-defined and others may be ill-defined or poorly defined in radiology (1, 2). As they mature, appear as a mixed radiolucent/radiopaque lesion (2). In this case, radiographic examination showed a well-defined and corticated multilocular mixed lesion in the left side of maxilla extending from lateral incisor to the first molar, which caused medially displacement of lateral wall of the nasal cavity and upward displacement of maxillary sinus. This was similar to Sonone et al. study in which the expansile lytic lesion extended into the right maxillary antrum and anterior portion of the right nasal cavity (11). Unlike the study of Balaji et al. which reported loss of lamina dura of the lateral incisor and the study of Menat et al. which inferior displacement of third molar and root resorption of second molar were reported; there was no evidence of root resorption and tooth displacement in our study (4, 12). Ameloblastomatous COC is the same as unicystic ameloblastoma histologically, but there are calcifications and ghost cells within the epithelium and connective tissue in the COC (3). Our case had histopathological criteria of ameloblastomatous COC mentioned above, so we classified this lesion as ameloblastomatous COC as a rare lesion. We reported a rare case of COC. According to Praetorius classification, this case belonged to type 1C category (ameloblastomatous proliferating). Many cases with ameloblastomatous COC should be reported to understand its biological behavior.

Authors' Contributions

Study concept, design and acquisition of data: Setareh Shojaei, Reza Jamalpour, Shirin Modabbernia and Shokoofeh Jamshidi; analysis and interpretation of data: Setareh Shojaei, Reza Jamalpour, Shirin Modabbernia and Shokoofeh Jamshidi; drafting of the manuscript: Setareh Shojaei, Reza Jamalpour, Shirin Modabbernia and Shokoofeh Jamshidi; critical revision of the manuscript for important intellectual content: Reza Jamalpour; study supervision: Shokoofeh Jamshidi.

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