Ameloblastomatous Calcifying Odontogenic Cyst: A Rare Lesion

Setareh Shojaei 1; Reza Jamalpour 1; Shirin Modabbernia 1; Shokoofeh Jamshidi 2,*

1Department of Oral and Maxillofacial Pathology, Hamadan University of Medical Sciences, Hamadan, IR Iran
2Dental Research Center, Department of Oral and Maxillofacial Pathology, Hamadan University of Medical Sciences, Hamadan, IR Iran
*Corresponding author: Shokoofeh Jamshidi, Dental Research Center, Department of Oral and Maxillofacial Pathology, Hamadan University of Medical Sciences, Hamadan, IR Iran. Tel: +98-8118354140, Fax: +98-8118381085, E-mail: dr.jamshidi19@gmail.com

Received: June 17, 2014; Revised: June 20, 2014; Accepted: June 23, 2014

1. 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 radio-opaque 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 Ic (ameloblastomatous proliferating). Many patients with ameloblastomatous COC should be reported to understand its biological behavior as possible.

Keywords: Cyst; Calcifying Odontogenic Cyst; Radiolucent

Copyright © 2014, Hamadan University of Medical Sciences. This is an open-access article distributed under the terms of the Creative Commons Attribution-Non-Commercial 4.0 International License (http://creativecommons.org/licenses/by-nc/4.0/) which permits copy and redistribute the material just in noncommercial usages, provided the original work is properly cited.
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), type 1C (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 1406 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 mediastial 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.

Funding/Support

Hamadan University of Medical Sciences was administrative, technical, and material support.

References


