Calcifying Cystic Odontogenic Tumor: Case Report

Corresponding Author:
Prof. Sergio E Cury,
DDS PhD, Oral Pathology - UniFOA - University of Volta Redonda, 27.310-060 - Brazil

Submitting Author:
Prof. Sergio E Cury,
DDS PhD, Oral Pathology - UniFOA - University of Volta Redonda, 27.310-060 - Brazil

Article ID: WMC002583
Article Type: Case Report
Submitted on: 07-Dec-2011, 10:49:01 AM GMT   Published on: 07-Dec-2011, 05:12:17 PM GMT
Article URL: http://www.webmedcentral.com/article_view/2583
Subject Categories: ORAL MEDICINE
Keywords: Calcifying odontogenic cyst, Gorlin\'s cyst, Odontogenic cysts, Odontogenic tumors, Calcifying odontogenic cystic tumor

How to cite the article: Cury S E, Cury S N, Cury M , Calderoni A , Fajardo V D, Carvalho M R, Luderer L A. Calcifying Cystic Odontogenic Tumor: Case Report . WebmedCentral ORAL MEDICINE 2011;2(12):WMC002583

Copyright: This is an open-access article distributed under the terms of the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original author and source are credited.

Source(s) of Funding:
None

Competing Interests:
In exchange for publication of the Article, Author represents and warrants to the Journal and the Publisher, together with their officers and directors, that the Article delivered for publication is original and does not infringe the patent, trademark, copyright, trade secret rights or other proprietary rights of third parties ("IP Rights"). Author also represents that has no financial interest or arrangement with any entity which interest or arrangement might be perceived to bear on the objectivity of the Article.
Calcifying Cystic Odontogenic Tumor: Case Report

Author(s): Cury S E, Cury S N, Cury M , Calderoni A , Fajardo V D, Carvalho M R, Luderer L A

Abstract

The following is a report of a 16-year-old boy with history of pain in the maxillary left central incisor and in the maxillary sinus area. Radiographs showed a well-demarcated, unilocular mixed radiolucent-radiodense lesion.

Introduction

The calcifying cystic odontogenic tumor (CCOT) is an uncommon benign cystic neoplasm of odontogenic origin, characterized by an ameloblastoma-like epithelium with ghost cells that may calcify, first described by Gorlin et al in 1962(1,2,3). It has been shown to have extensive diversity in its clinical and histopathological features, as well as in its biological behavior, and most cases present cystic characteristics, few are of the solid type (15%), and its rare malignant transformation is well documented(3-5). It represents about 2% of all odontogenic cysts and tumors and 3.5% in the Brazilian population(4,6). In 1992, the World Health Organization (WHO) classified CCOT within the groups of neoplasms and tumors originated from odontogenic tissues, yet confirmed that most cases are non-neoplastic(2,5). A great variety of clinical and histological features have been reported and several classifications have been proposed(7-9).

Radiographically, the CCOT is usually a mixed lesion, with uni or multilocular radiolucent area, containing different amounts of radiopaque material(3,8). The association with the apices of teeth reveals an incidence of root resorption in 75% to 77% of cases(10,11).

The most notable features of this pathologic entity are the ghost epithelial cells with a tendency to calcify and the occasional association of this finding with certain odontogenic tumors, including the odontoma and ameloblastoma(1-3).

Case Report(s)

A 16-year-old white boy with history of pain in the maxillary left central incisor and in the maxillary sinus area was referred by his local dentist to the Department of Oral Surgery at the Dental School of University of Volta Redonda, Brazil. Intraoral inspection revealed that the mucosa was normal, and a light tumefaction was observed at the region between the maxillary left central and lateral incisors. Occlusal radiograph showed a well-demarcated, unilocular mixed radiolucent-radiodense lesion extending from the maxillary right central incisor to the maxillary left first molar region. The radiographic image also presented reactive sclerotic lines and expanded cortical bone, thereby creating direct contact between the cyst wall and the oral mucosa. Displacement of the central and lateral incisors and root resorption of the central incisor were also observed (Figure1 A).

Surgical enucleation had been performed under local anesthesia. The pathologic report showed a cystic lesion with4.5 cm in its greatest diameter, soft consistence and brownish color. During the procedure, the lower aspect of the left lateral wall of the maxillary sinus near the first molar was observed and revealed no erosion caused by the cystic lesion.

Histological examination was performed at the Department of Oral Pathology at this University. Microscopic findings showed a cystic cavity lined by a remarkable and well-defined polarized basal layer of prominent palisade and mildly hyperchromatic cuboidal and columnar cells that focally resembled the ameloblastic epithelium. Overlying this layer, there were epithelial cells exhibiting similar arrangement as the central stellate reticulum of the tooth bud. Between them, the typical ghost cells were observed. The ghost cells were large and eosinophilic, with aberrant keratinization. Foreign body giant cells were found in close relationship with the aberrant keratin, and calcified focal bodies were observed. The cyst wall was composed of a hypercellular immature fibrous tissue, and a focal area of inflammatory cells was also present (Figure 1 B, C, D, E and F).

No recurrence was recorded after 18-month follow-up.

Discussion

CCOT is an uncommon benign odontogenic lesion that was first distinguished as a separate entity by Gorlin et al in 1962(1,3). Although named and defined as a cyst, there is no agreement in the literature regarding its classification as a cyst or a neoplasm, since some examples of CCOT show areas suggestive of neoplasia(7,12). In addition, several
classifications have been suggested in the literature, each of them trying to separate its cystic from solid variants, but none has been universally accepted (2,7,13).

The CCOT normally appears as a painless, slow-growing tumor, equally affecting the maxilla and mandible, with predilection for the anterior segment (incisor/canine area). It generally affects young adults in the third to fourth decade of life, without gender predilection(5). It is usually composed of a cystic cavity with fibrous capsule, lined by an odontogenic epithelium with typical microscopic characteristics (presence of variable amounts of aberrant epithelial cells without nuclei, which are named ghost cells)(2,13).

The development of intraosseous or extraosseous varieties of CCOT is related in the literature, but it seems to depend on the location of odontogenic epithelium, which constitutes the source of the lesion. However, the site does not seem to have any relation with the behavior or histological features of the cyst(2,14).

Radiographically, the CCOT is usually a mixed lesion, with radiolucent area, uni or multilocular, that contains different amounts of radiopaque material(3,8). McGowan and Browne(15), in 1982, found that the presence of mineralization was approximately twice as frequent in microscopic examination compared to radiographic analysis. The present case seems to support this conclusion, since it had no or very low radiographically detectable calcified bodies in the lesion.

In a study by Tanimoto et al(11), in 1988, the presence of root resorption was detected in approximately 75% to 77% of cases. In the present case, root resorption was found and is in accordance with this affirmation, even though lida et al(1), in 2006, reported that the occurrence of root resorption is not common. The present case also exhibited expanded cortical bone and direct contact between the cyst wall and the oral mucosa, in agreement with the study of Praetorius et al(9) 1981.

All histological findings support the histological descriptions in the literature(2,3,16,17). The CCOT can be found alone, as in the present case, or associated with other odontogenic tumors, as odontoma, ameloblastoma, ameloblastic fibroodontoma, odontoameloblastic tumor, calcifying epithelial odontogenic tumor and adenomatoid odontogenic tumor(3,9). The malignant transformation of a preexisting benign CCOT could occur, yet is extremely uncommon(3,12).

Finally, treatment of the cystic variety of CCOT is usually conservative and consists of enucleation with curettage for intraosseous lesions and local excision for peripheral lesions. The prognosis is good and only occasional recurrences have been reported(16,17).

References

Well-demarcated, unilocular mixed radiolucent-radiodense lesion (occlusal radiograph) (A); prominent and well-defined polarized basal layer of prominent palisade and mildly hyperchromatic cuboidal and columnar cells that focally resemble ameloblastic epithelium-low magnification 100X H&E (B and C); eosinophilic ghost cells with aberrant keratinization, calcified focal bodies and focal area of inflammatory cells-high magnification 400X (D); ghost cells, calcified bodies and epithelial cells exhibiting similar arrangement as the central stellate reticulum of the tooth bud-high magnification 1000X (E and F).
Disclaimer

This article has been downloaded from WebmedCentral. With our unique author driven post publication peer review, contents posted on this web portal do not undergo any prepublication peer or editorial review. It is completely the responsibility of the authors to ensure not only scientific and ethical standards of the manuscript but also its grammatical accuracy. Authors must ensure that they obtain all the necessary permissions before submitting any information that requires obtaining a consent or approval from a third party. Authors should also ensure not to submit any information which they do not have the copyright of or of which they have transferred the copyrights to a third party.

Contents on WebmedCentral are purely for biomedical researchers and scientists. They are not meant to cater to the needs of an individual patient. The web portal or any content(s) therein is neither designed to support, nor replace, the relationship that exists between a patient/site visitor and his/her physician. Your use of the WebmedCentral site and its contents is entirely at your own risk. We do not take any responsibility for any harm that you may suffer or inflict on a third person by following the contents of this website.