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 SE, Cury SN, Cury M, Calderoni A, Fajardo VD, Carvalho MR, Luderer LA. 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 (CC-BY), 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 SE, Cury SN, Cury M, Calderoni A, Fajardo VD, Carvalho MR, Luderer LA

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 Vota Redonda, Brazil. Intraoral inspection revealed that the mucosa was normal, and a light tumoraction 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 with 4.5 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).
Reviews

Review 1

**Review Title:** Calcifying Cystic Odontogenic Tumor

Posted by Dr. Constantino Ledesma-Montes on 26 Jun 2012 08:36:00 PM GMT

**What are the main claims of the paper and how important are they?:**
To report a case of Calcifying Cystic Odontogenic Tumor.

The claim is not new, but it is valid.

Yes.

Yes.

If a protocol is provided, for example for a randomized controlled trial, are there any important deviations from it? If so, have the authors explained adequately why the deviations occurred?

Not apply.

Yes.

Not apply.

Not apply.

**Rating:** 8

**Comment:**
It is a well done manuscript and the case report is well presented.

**Competing interests:** None.

**Invited by the author to make a review on this article?** : No

**Have you previously published on this or a similar topic?**: Yes

**References:**
One of the references is my chapter in the WHO Blue Book.

**How to cite:** Ledesma-Montes C. Calcifying Cystic Odontogenic Tumor [Review of the article 'Calcifying Cystic Odontogenic Tumor: Case Report ' by ]. WebmedCentral 1970;3(6):WMCRW001988
Review 2

Review Title: "Calcifying Cystic Odontogenic tumor: case report" by Sergio E Cury

Posted by Prof. Omar F Molina on 25 Jan 2012 07:18:08 AM GMT

Rating: 0

Comment:
This study and report of a case is a comprehensive description about periapical granuloma and differential diagnosis of similar lesions occurring in the jaws. This study will expand and enlighten current knowledge about such lesions. The authors should be congratulated for sharing such material with us. However, for the sake of clarity for the reader, some minor changes are recommended:

1. In the introduction:
   Please write: Periapical lesions resulting from pulp necrosis are among the most frequent pathologies occurring in the alveolar bone (Garcia et al, 2007).

2. Introduction in the second paragraph:
   Please write: Thus, such lesion is a consequence of an infection.....

3. In the introduction, fourth paragraph:
   Please write: The latter develop instead of the latter develops (the author should write in the plural as they are talking about periapical abscesses).

4. In the case report, third paragraph:
   Please write: in the region of the basal and mandibular angle....

5. In the case report, fourth paragraph:
   Please write: "expansive to the lingual cortical and the mandibular base areas"...

6. In the case report, fifth paragraph:
   please write: "Because the lesion was located lingually, the surgeon decided to use an extraoral approach, via the neck, until the affected region was reached".

7. In the case report, seventh paragraph:
   Please write: Such evaluation revealed fragments of connective......
   Please write: Additionally, numerous blood vessels, some of which engorged, were found. Areas of erythrocyte extravasation and bacteria colonies completed the panel.....
   Please write: "The pathologist further added that, since the histologic panel was strongly associated with the endo-periodontal impairment of the mandibular right first molar, elimination of the infection area in the region and radiographic follow-up was highly recommended".
   One paragraph before the discussion:
   please write: The examination revealed that the lesion had regressed and the expansion had decreased, nevertheless it was not possible to evaluate if the bone had been repaired. Another computed tomography was taken following eight months and bone formation and repair were observed.

8. In the discussion
   Please write 4 short paragraphs per page for the sake of clarity
   In the third paragraph, please write especial instead of special
   In the third paragraph, please write: "Traditionally, it has been suggested that periapical cysts can be differentiated in periapical radiographs".
   In the same paragraph, please write: Conversely, a granuloma is supposed to present a diffuse, .......
   In the fourth paragraph, please write: "The clinical use of cone beam technology can assist the differential diagnosis.....
   In the same paragraph, please write: "Using computed tomography, a radicular cyst can be differentiated from a periapical granuloma, by a markedly lower density of the cystic cavity when compared with the granulomatous tissue"
   In the same paragraph, please write: "This allows the clinician to decide whether or not the surgery is necessary..."
without waiting for the follow up period to assess. "However, Estrela et al (2009) do not agree and they argue that....."

Invited by the author to make a review on this article? : Yes  
Have you previously published on this or a similar topic?: No  
How to cite: Molina O."Calcifying Cystic Odontogenic tumor: case report" by Sergio E Cury[Review of the article 'Calcifying Cystic Odontogenic Tumor: Case Report ' by ].WebmedCentral 1970;3(1):WMCRW001428
## Review 3

**Review Title:** Calcifying Cystic Odontogenic Tumor: Case Report

**Posted by:** Prof. Luiz Roberto C Manhães Jr on 24 Jan 2012 12:24:21 PM GMT

<table>
<thead>
<tr>
<th></th>
<th>Question</th>
<th>Answer</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Is the subject of the article within the scope of the subject category?</td>
<td>Yes</td>
</tr>
<tr>
<td>2</td>
<td>Are the interpretations / conclusions sound and justified by the data?</td>
<td>Yes</td>
</tr>
<tr>
<td>3</td>
<td>Is this a new and original contribution?</td>
<td>No</td>
</tr>
<tr>
<td>4</td>
<td>Does this paper exemplify an awareness of other research on the topic?</td>
<td>Yes</td>
</tr>
<tr>
<td>5</td>
<td>Are structure and length satisfactory?</td>
<td>Yes</td>
</tr>
<tr>
<td>6</td>
<td>Can you suggest brief additions or amendments or an introductory statement that will increase the value of this paper for an international audience?</td>
<td>No</td>
</tr>
<tr>
<td>7</td>
<td>Can you suggest any reductions in the paper, or deletions of parts?</td>
<td>No</td>
</tr>
<tr>
<td>8</td>
<td>Is the quality of the diction satisfactory?</td>
<td>Yes</td>
</tr>
<tr>
<td>9</td>
<td>Are the illustrations and tables necessary and acceptable?</td>
<td>Yes</td>
</tr>
<tr>
<td>10</td>
<td>Are the references adequate and are they all necessary?</td>
<td>Yes</td>
</tr>
<tr>
<td>11</td>
<td>Are the keywords and abstract or summary informative?</td>
<td>Yes</td>
</tr>
</tbody>
</table>

**Rating:** 7

**Comment:**
This article is interesting due to the controversy in the literature specifies to this lesion.

**Invited by the author to make a review on this article?:** Yes

**Have you previously published on this or a similar topic?:** No

**Experience and credentials in the specific area of science:**
Phd Professor in Radiology in two University.

**How to cite:** Manhães Jr L. Calcifying Cystic Odontogenic Tumor: Case Report [Review of the article 'Calcifying Cystic Odontogenic Tumor: Case Report' by ]. WebmedCentral 1970;3(1):WMCRW001420
Review 4

**Review Title:** Calcifying cystic odontogenic tumor: case report

Posted by **Prof. Brunno S Freitas** on 12 Jan 2012 11:55:15 AM GMT

<table>
<thead>
<tr>
<th></th>
<th>Question</th>
<th>Answer</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Is the subject of the article within the scope of the subject category?</td>
<td>Yes</td>
</tr>
<tr>
<td>2</td>
<td>Are the interpretations / conclusions sound and justified by the data?</td>
<td>Yes</td>
</tr>
<tr>
<td>3</td>
<td>Is this a new and original contribution?</td>
<td>No</td>
</tr>
<tr>
<td>4</td>
<td>Does this paper exemplify an awareness of other research on the topic?</td>
<td>Yes</td>
</tr>
<tr>
<td>5</td>
<td>Are structure and length satisfactory?</td>
<td>Yes</td>
</tr>
<tr>
<td>6</td>
<td>Can you suggest brief additions or amendments or an introductory statement that will increase the value of this paper for an international audience?</td>
<td>Yes</td>
</tr>
<tr>
<td>7</td>
<td>Can you suggest any reductions in the paper, or deletions of parts?</td>
<td>No</td>
</tr>
<tr>
<td>8</td>
<td>Is the quality of the diction satisfactory?</td>
<td>Yes</td>
</tr>
<tr>
<td>9</td>
<td>Are the illustrations and tables necessary and acceptable?</td>
<td>Yes</td>
</tr>
<tr>
<td>10</td>
<td>Are the references adequate and are they all necessary?</td>
<td>Yes</td>
</tr>
<tr>
<td>11</td>
<td>Are the keywords and abstract or summary informative?</td>
<td>Yes</td>
</tr>
</tbody>
</table>

**Rating:** 7

**Comment:**
This is an interesting case report.

Despite the controversy about the classification of CCOT the authors should put the World Health Organization (2005) position

**Invited by the author to make a review on this article?** : Yes

**Have you previously published on this or a similar topic?** : No

**Experience and credentials in the specific area of science:**
PhD and MSc in oral pathology

**How to cite:** Freitas B.Calcifying cystic odontogenic tumor: case report[Review of the article 'Calcifying Cystic Odontogenic Tumor: Case Report ' by ].WebmedCentral 1970;3(1):WMCRW001366
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.