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Odontoma-Producing Intraosseous Calcifying Odontogenic Cyst: Case Report

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The present report describes a case of odontoma-producing intraosseous calcifying odontogenic cyst in a 36-year-old Black male in the right mandibular bicuspid region. The lesion involved an unerupted permanent canine, which was displaced to the mandible base and a calcified mass that was later recognized as an odontoma. The lesion was surgically removed.

Key Words: odontogenic cyst, Gorlin cyst.

INTRODUCTION

Calcifying odontogenic cyst was first categorized as a distinct entity by Gorlin et al. (1), and was named after him since then. According to Shear (2), it accounts for 1% of jaw cysts. As the number of reports increased, it was proposed that calcifying odontogenic cyst was indeed a heterogeneous group of entities, with distinct histopathologic findings. We present a case of intraosseous odontoma-producing calcifying odontogenic cyst involving an unerupted permanent canine.

CASE REPORT

A 36-year-old Black male complained of a painless swelling in the right bicuspid region of the mandible. Intraoral examination revealed a firm enlargement in the buccal right bicuspid region extending from the canine to the second bicuspid. The overlying mucosa had a normal aspect. The deciduous right canine was still present, and the permanent one was missing. Detailed examination of the involved teeth revealed no mobility or tenderness to palpation. There were also no signs of caries, pulp pathosis or periodontitis.

A panoramic radiograph (Figure 1) revealed a well-defined unilocular radiolucent lesion involving the anterior region of the mandible, extending from the right second bicuspid to the left lateral incisor. The

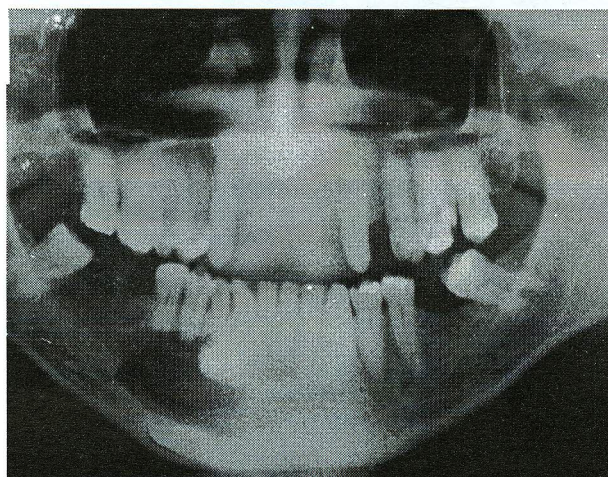


Figure 1. Orthopantomograph showing a well-defined unilocular radiolucent lesion involving the anterior region of the mandible, extending from the right second bicuspid to the left lateral incisor. The permanent canine is seen in the inferior border of the mandible. A radiopaque mass is present in the periapical region of the right deciduous canine and the lateral incisor.

permanent canine was displaced to the inferior border of the mandible. A radiopaque mass was present in the periapical region of the right deciduous canine and the lateral incisor. The right bicuspid exhibited marked root resorption. Radiological differential diagnosis included adenomatoid odontogenic tumor, intraosseous calcifying odontogenic cyst, cystic odontoma, ossifying fibroma, ameloblastic fibro-odontoma and calcifying epithelial odontogenic tumor. Calcifying epithelial odontogenic tumor is most often found in the posterior region of the mandible, and the margins of this lesion are often scalloped (3). Ameloblastic fibro-odontoma is usually encountered in children of 10 years of age, on

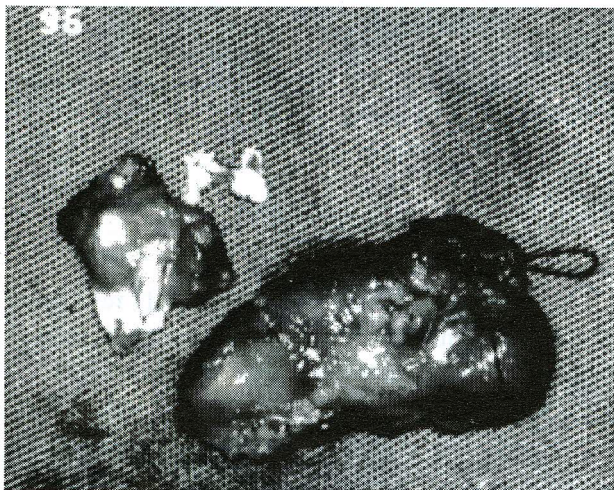


Figure 2. Surgical specimens. Some denticles can be observed.



Figure 3. Microscopically, a basal epithelial layer can be seen, above which various cell layers are present. Close to the basal layer, ghost cells (arrows) are readily observed and a large calcification surrounded by inflammatory cells is in close contact with the connective tissue.

average. Although ossifying fibroma has many features in common with this case, it is seen more often in females. As an additional step, aspiration was performed, and the fluid obtained confirmed the cystic nature of the lesion.

The teeth involved in the lesion received endodontic treatment before the surgical excision of the lesion, during which the involved permanent canine was also removed (Figure 2). Healing proceeded uneventfully.

Microscopically, a cystic lesion was observed, which was lined with a basal layer of columnar cells whose nuclei were located next to the basement membrane (Figure 3). Above this layer, there was a variable number of cell layers. In some places, loosely arranged cells could be seen resembling stellate reticulum cells, and intermixed with them, ghost cells and calcifications. Ghost cells have been regarded as pathognomonic for calcifying odontogenic cyst for a long time, and are recognized by their characteristic eosinophilic appearance. Denticles were also present, leading to the diagnosis of odontoma-producing intraosseous unicystic calcifying odontogenic cyst.

DISCUSSION

Since 1971, calcifying odontogenic cyst has been described by the World Health Organization as a non-neoplastic cystic lesion in which the epithelial lining shows a well-defined basal layer that is often many cells thick and that may resemble the stellate reticulum of an enamel organ, and masses of ghost epithelial cells that may be in the epithelial cyst lining or in the fibrous capsule (4). The ghost cells may become calcified and dysplastic dentin may be laid down next to the basal cell layer of the epithelium. It may be associated with complex odontoma or with tissue resembling an ameloblastic fibro-odontoma.

As the number of reports increased, it was proposed that calcifying odontogenic cyst was indeed a heterogeneous group of entities, with distinct histopathologic findings that included a solid tumor. Praetorius (5) proposed a subclassification for the heterogeneous group of calcifying odontogenic cysts, in which the cystic lesions were separated from the neoplasms (solid lesions). These researchers further divided the cystic entity into three types, the simple unicystic type, the unicystic odontoma-producing type, and the

ameloblastomatous proliferating type. Calcifying odontogenic cyst may be otherwise described as a developmental odontogenic lesion that has its cutaneous counterpart in Malherbe's calcifying epithelioma (1,6).

This case report is in agreement with the literature finding that calcifying odontogenic cysts occur predominantly as an intraosseous lesion. Other authors have reported only 13 to 21 percent of the cysts to be peripheral (extraosseous) lesions (7,8). The area affected in this case has been considered the most commonly affected site: about 65% of the reported cases were found in the incisor-canine area (3,7). The age of the patient is also in agreement with the literature that the mean age when the calcifying odontogenic cyst is diagnosed is 33 years (3). The radiographic findings of this case (unilocular radiolucency with a radiopaque mass and well-circumscribed borders) are encountered in the majority of odontoma-producing intraosseous calcifying odontogenic cysts (9,10).

The radiopaque mass observed in the panoramic radiograph of this case, later diagnosed as odontoma components, are considered to be present in 24-50% of the reported cases (3,7). These cases were classified as a subtype of calcifying odontogenic cyst under the term odontoma-producing type (7), or as a cystic variant associated with odontoma (4,8). Hirschberg et al. (10) proposed that this variant should be classified as a separate entity for which they suggested the name *odontocalcifying odontogenic cyst*. They observed that this variant was more prevalent in females, with a mean age at discovery of 16 years (compared with 34.4 years in the simple calcifying odontogenic cyst group) and that most cases were located in the maxilla. The findings of our case are not in accordance with the above mentioned observations. Hirschberg et al. (10) claimed that the separation of this variant from the heterogeneous group of calcifying odontogenic cyst could lead to a better understanding of its pathogenesis. Although the majority of the cases of this variant indeed have the features pointed out by Hirschberg et al. (10), it may be noteworthy that these characteristics may not be sufficient to classify this variant as a separate entity. In this regard, our case may indicate that this variant may be more common in younger adults, but it may also be discovered in older persons, so that a different pathogenesis might not be associated with this variant. The possibility of occur-

rence of an odontoma associated with calcifying odontogenic cyst may result from factors present at a certain age, but it seems that the same odontogenic epithelium is present in calcifying odontogenic cyst and in this particular variant. Furthermore, it seems that the same characteristic epithelium is involved in the various clinical presentations of this lesion.

In spite of the low frequency of this lesion and the fact that most cases are surgically removed and heal uneventfully, there must be a close follow-up, because there have been reports of association with carcinoma, adenomatoid tumor and ameloblastoma (11-13).

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RESUMO

Pistöia GD, Gerlach RF, dos Santos JCB, Montebelo Filho A. Odontoma produzindo cisto odontogênico calcificante intra-ósseo: relato de caso. *Braz Dent J* 2001;12(1):67-70.

O presente relato descreve um caso de odontoma que produziu um cisto odontogênico calcificante, num homem de 36 anos, da raça negra, na região de premolares inferiores direitos. A lesão envolveu um canino permanente não irrompido, o qual foi deslocado para a base da mandíbula e uma massa calcificada que foi posteriormente reconhecida como um odontoma. A lesão foi removida cirurgicamente.

Unitermos: cisto odontogênico, cisto de Gorlin.

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