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Journal of Oral Medicine, Oral Surgery, Oral Pathology and Oral Radiology

Journal homepage: [www.joooo.org](http://www.joooo.org)**Case Report****Odontogenic keratocyst with extensive dystrophic calcification in anterior maxilla: A case report**

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**Abstract**

**Background:** The odontogenic keratocyst, usually located in the posterior mandible, exhibits a unilocular or multilocular radiolucent area and well-defined histopathological characteristics.

**Case Presentation:** A 41-year-old woman with an asymptomatic swelling in the anterior maxilla that progressed over two months. Radiographic examination using a panoramic radiograph revealed a mixed image with radiopaque foci. Differential diagnoses included desmoplastic ameloblastoma and ossifying fibroma. A histological analysis revealed an epithelium lining a capsule characterized by a basal layer of columnar palisaded cells and a wrinkled parakeratin layer. In some areas, the epithelium was detached from the capsule, and many areas of dystrophic calcification were observed in the capsule.

**Conclusion:** Although some studies mention calcifications in the capsule of the keratocyst, there is no conclusive study on the mechanism of development of these calcifications. This keratocyst is notable because exhibits extensive areas of calcification that, to the best of our knowledge, have not previously been reported.

**Keywords:** Calcification; Keratocyst odontogenic tumor; Maxilla; Dystrophic; Oral medicine.

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**1. Introduction**

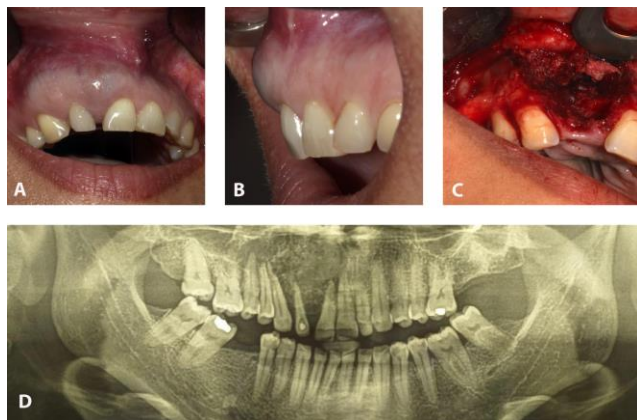
Odontogenic keratocysts (OKC) account for 19% of all jaw cysts. Because of their high recurrence rates and bone destruction, they are often considered to be locally aggressive developmental lesions.<sup>1</sup> Most commonly, it appears as a unilocular or multilocular radiolucency in the posterior mandibular region. The term is defined as "a benign uni- or multicystic intraosseous tumor of odontogenic origin with pseudostratified squamous epithelium lining and potential aggressive infiltrative characteristics." Single or multiple cysts can occur, with multiple OKCs being common in syndromic cases.<sup>2</sup>

**2. Case Presentation**

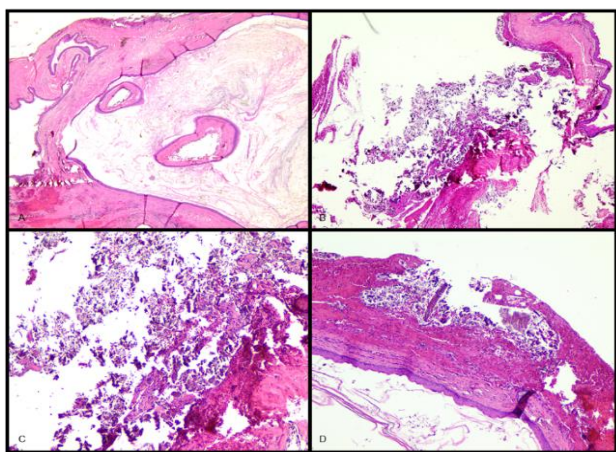
Female patient, 41 years old, was referred to the Dentistry Service at João de Barros Barreto University Hospital with asymptomatic swelling with fibrous consistency, measuring 4 x 1 cm in size in the anterior maxilla, with a two-month progression (**Figure 1 A,B**). The panoramic radiograph revealed a mixed image with radiopaque foci, causing divergence of the dental roots of teeth 11, 12, and 13 (D). Systemic conditions and medication history was not relevant for the continuity of the case.

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Based on the patient's history, clinical examination, and radiographic findings, two diagnostic hypotheses were considered: desmoplastic ameloblastoma and ossifying fibroma. After preoperative exams, an incisional biopsy (**Figure 1 C**) was performed for histological analysis. The histopathological analysis showed an epithelium lining a capsule. The epithelium displayed a palisaded layer of columnar basal cells and a wrinkled parakeratin layer. In some areas, the epithelium detached from the capsule. Many areas of dystrophic calcification were observed within the capsule (**Figure 2**).



**Figure 1:** A,B): Clinical appearance of the swelling on the vestibular view, shown in lateral view. C): Surgical procedure by which a linear incision was made with a scalpel. D): Panoramic radiograph showing a mixed radiopaque/radiolucent (arrows) lesion of the anterior maxilla, along with divergence and external root resorption of the área



**Figure 2:** Histopathological features of the lesion. A, B and C): Fragment of cystic capsule of odontogenic origin, partially coated by a layers of parakeratinized epithelium. The capsule was compose of loose connective tissue, blood vassel and moderate mononuclear inflammatory infiltrate  $\times 10$ , Hematoxylin-eosin (HE). A high-resolution version of this slide for use with the Virtual Microscope is available as eSlide: VM07240. D): An epithelium lining a capsule characterized by a basal layer of columnar palisaded cells and a wrinkled parakeratin layer (original magnification  $\times 40$ ,

HE). A high-resolution version of this slide for use with the Virtual Microscope is available as eSlide: VM07237

### 3. Discussion

The Odontogenic Keratocyst represents a cyst with an alteration in the PTCH gene, with a preference for the posterior mandible region in patients aged 10 to 40 years.<sup>3-5</sup> It exhibits a unilocular or multilocular radiolucent area with well-defined, regular radiopaque margins and has high recurrence rates.<sup>5</sup> Histopathological characteristics are highly distinctive and include a wrinkled epithelial surface layer, a consistent epithelial thickness (between 6 and 8 cell layers), a "picket fence" appearance of the basal cells, separation of the epithelium from the connective tissue stroma, and the presence of satellite or daughter cysts.<sup>6</sup> Some reports in the literature show that areas of calcification can occur in the OKC capsule.<sup>6-8</sup>

The literature suggests that dystrophic calcification may occur by degeneration as a result of necrobiosis or a foreign body reaction. Inductive changes with mesenchymal cells, the abundance of crystalline calcium phosphates, hydroxyapatite, whitlockite, and inorganic phosphates in the fluid of OKC, as well as odontogenic epithelial remnants may be mechanisms by which these calcifications occur in the lesion wall and in similar lesions.<sup>7,8</sup>

### 4. Conclusion

All ages are affected, with a peak in the first or fourth decade and a second smaller peak in the elderly, a slight male predominance can be observed. The risk of recurrence following traditional enucleation is between 20-30%, although a wide range has been reported.<sup>5</sup>

The present case, however, differs from those previously reported, because it extensive dystrophic calcification of several areas within the capsule, which is a unique phenomenon that have never been reported to the best our knowledge.

### 5. Source of Funding

None.

### 6. Conflict of Interest

None.

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