



## Odontogenic Keratocyst Mimicking Periapical Cyst: Case Report

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### Abstract

Keratocyst is an odontogenic cyst that, due to its aggressiveness, growth potential, and high recurrence rate was once classified as an odontogenic tumor by the World Health Organization in 2005. However, it was reclassified as an odontogenic cyst in the 2017 classification. Keratocyst is most often diagnosed on routine radiographic examinations. It can be uni or multilocular. It has a higher incidence in the posterior region and ramus of the mandible. In the maxilla, it is more frequently found in the canine region. The purpose of this paper is to present a case of a unilocular keratocyst that affected the maxillary premolar region, mimicking a periapical cyst. The lesion was surgically removed by enucleation, one of the recommended techniques for the treatment of keratocysts. Besides enucleation, marsupialization and bone resection, or a combination of these techniques, are also indicated. Keratocysts have a high recurrence rate, requiring a long period of clinical and radiographic follow-up.

**Keywords:** *Odontogenic Keratocyst; Odontogenic Cysts; Odontogenic Tumors; Oral Diagnosis; Oral Surgery*

### Introduction

The odontogenic keratocyst is an intraosseous lesion, originally classified as an odontogenic cyst. In the World Health Organization classification in 2005, in view of its characteristics such as growth potential, aggressiveness of the lesion, genetic and histopathological aspects, the keratocyst was considered as a benign cystic neoplasm, being called keratocystic odontogenic tumor [1-9]. Later, in the 2017 classification, the World Health Organization redefined the nomenclature to keratocyst [10,11].

Keratocysts are slow growing. However, they are very aggressive and locally invasive, and can reach large proportions and cause bone resorption [2-4,6-8,10-15].

The diagnosis occurs most often accidentally, in routine radiographic examinations [4]. They can be found in patients at any age. However, approximately 60% of cases are diagnosed in the 10 to 40 years of age range. The mandible is most affected, ranging from 60 to 80% of cases, with higher incidence in the posterior region and the ascending ramus. The keratocyst affects the maxilla from 20 to 30%, with higher frequency in the canine region [1,4,6,8,10,11-17]. Keratocysts have no gender predilection [6,15].

Radiographically, the osteolytic lesion is characterized by a unilocular radiolucent image, which can also be multilocular, but less frequently [1,10]. It is generally well delimited and surrounded by bone sclerosis [12]. In the maxilla, one should be aware of misinter-

pretation, leading the dental surgeon to suspect a periapical lesion or lateral periodontal cyst [13,15]. The association of multiple keratocysts with other manifestations may indicate basal cell nevoid carcinoma syndrome [1,3,6,7,18-20]. Imaging examinations should be employed to exclude this condition.

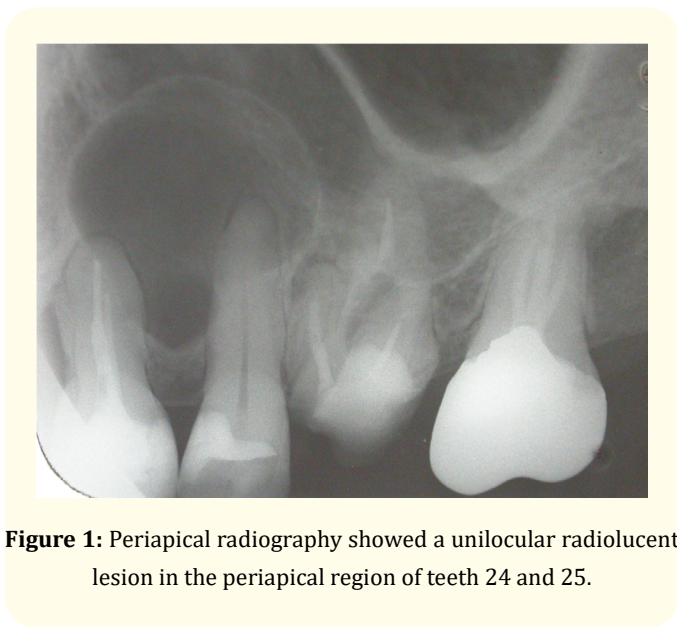
The indicated treatment is surgical removal, by means of enucleation, marsupialization, bone resection or a combination of these techniques [1,6,10,18,21]. Keratocysts have a high recurrence rate, ranging from 22 to 60% [2,3,4,6-8,12-15,21].

The purpose of this article is to present a case of an unilocular keratocyst that affected the upper premolar region, mimicking a periapical cyst, and was surgically removed by the enucleation technique.

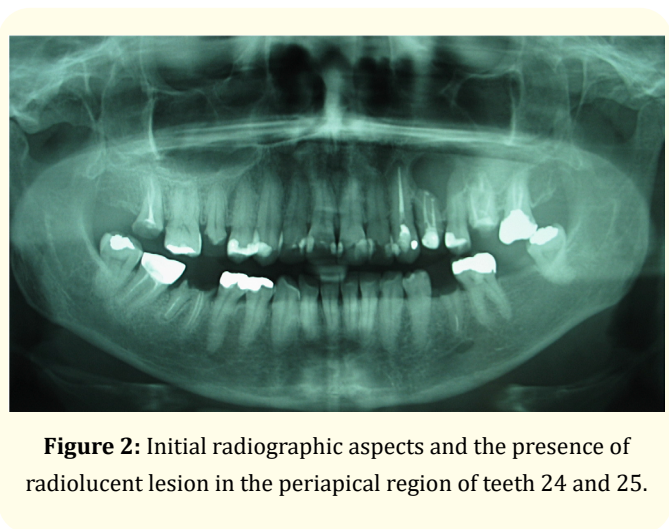
### Case Report

A Caucasian female patient, 54-years-old, came to a private clinic for dental treatment.

Routine periapical and panoramic radiography (Figure 1 and 2, respectively) showed a unilocular, circumscribed radiolucent lesion in the periapical region of teeth 24 and 25. Computed tomography was requested for better detailing and surgical planning.

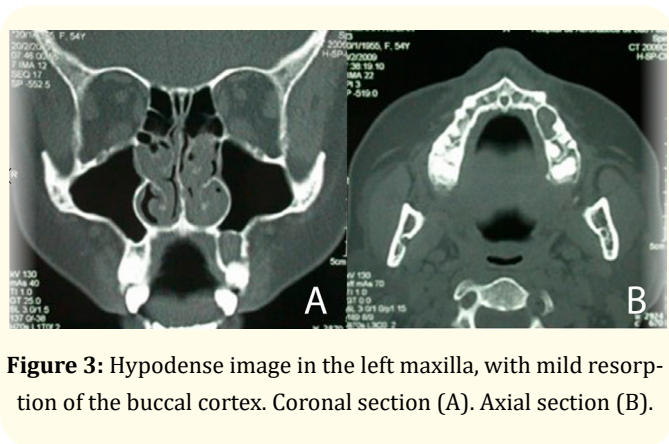


**Figure 1:** Periapical radiography showed a unilocular radiolucent lesion in the periapical region of teeth 24 and 25.



**Figure 2:** Initial radiographic aspects and the presence of radiolucent lesion in the periapical region of teeth 24 and 25.

Computed tomography revealed a hypodense image in the left maxilla, with mild resorption of the buccal cortical bone (Figure 3).



**Figure 3:** Hypodense image in the left maxilla, with mild resorption of the buccal cortex. Coronal section (A). Axial section (B).

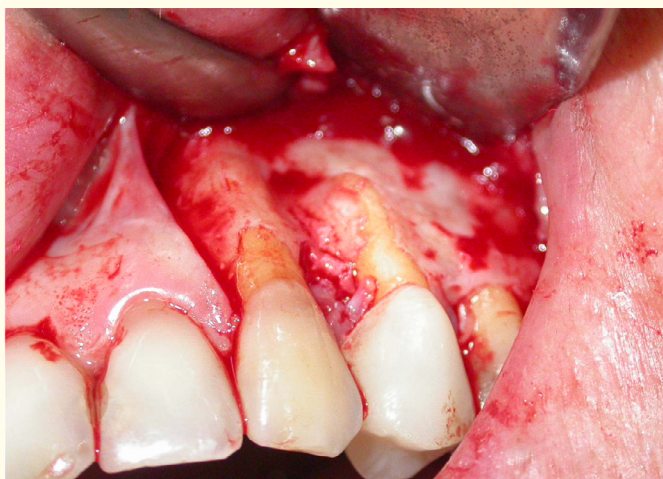
Clinically, no bulging of the buccal cortex was observed. The diagnostic hypothesis was periapical cyst.

Surgical removal was indicated to the patient, discussing all the risks and care related to the procedure. After consent for the procedure, surgery was scheduled. No systemic diseases or conditions that would compromise the surgical procedure were reported.

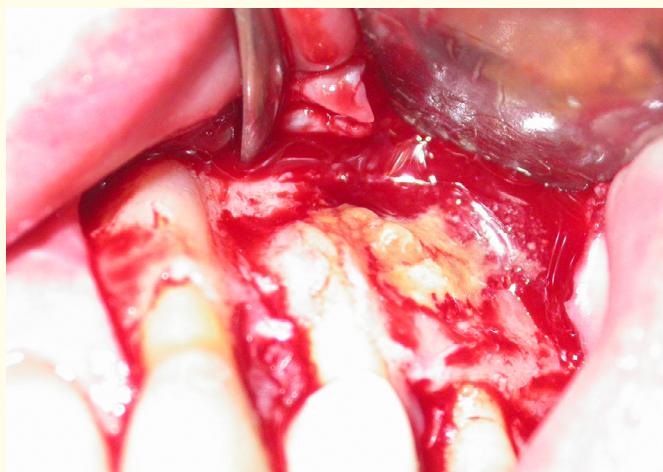
Under local anesthesia, an intrasulcular incision was made from tooth 23 to tooth 26, with two relaxers, for flap detachment (Figure 4). The buccal cortical was slightly curetted until the lesion



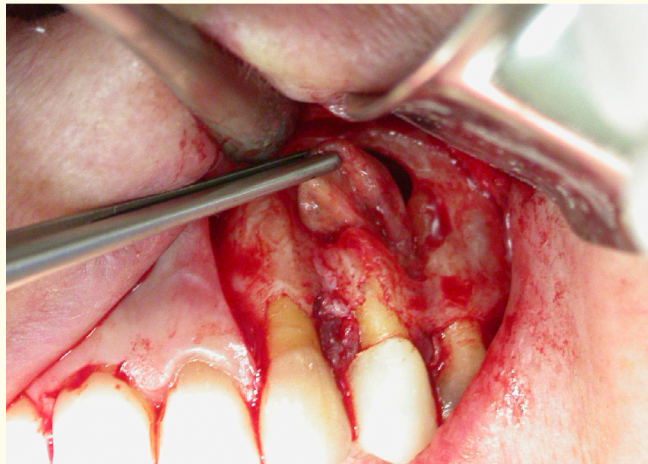
was exposed, since there was cortical resorption (Figure 5). Subsequently, the lesion was curetted and completely removed (Figure 6). An apicectomy was performed on tooth 24 and filled with calcium hydroxide cement (Figure 7). The bone cavity was washed thoroughly and the region was sutured (Figure 8). Analgesic, anti-inflammatory and antibiotic drugs were prescribed to the patient.



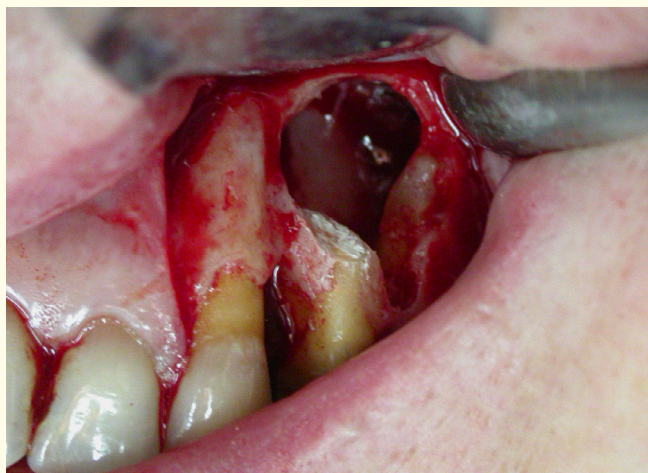
**Figure 4:** Gingival flap after incision between teeth 23 and 26.



**Figure 5:** Buccal cortical slightly curetted until the lesion.

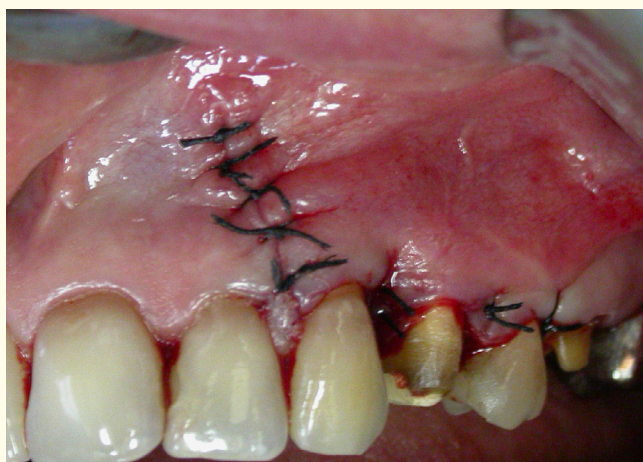


**Figure 6:** Lesion curetted and completely removed.



**Figure 7:** Apicectomy on tooth 24 and filled with calcium hydroxide cement.

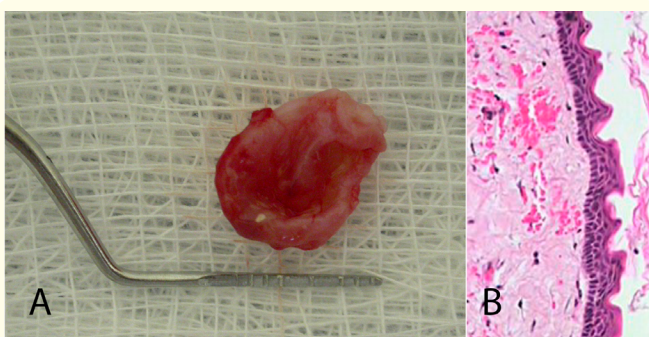
The fragment of the lesion (Figure 9A) was fixed in 10% formalin and sent to the Laboratory of Surgical Pathology of the School of Dentistry, University of São Paulo. Histopathological examination revealed squamous, stratified epithelium with hyperchromatic cell nuclei arranged in palisade. The stroma showed a sparse inflammatory infiltrate composed of loose fibrous connective tissue (Figure 9B). The diagnosis was odontogenic keratocyst.



**Figure 8:** Region sutured.



**Figure 10:** Evaluation after 15 days and removal of the sutures.



**Figure 9:** Fragment of the lesion (A). Histopathological aspects of the odontogenic keratocyst (magnification: 40X) (B).

The patient was evaluated after 15 days, and the remaining sutures were removed (Figure 10). No complaints and/or complications were reported. The patient was evaluated again 30 days after the surgical procedure (Figure 11).

The patient has been followed for 18 months with no signs of recurrence.

### Discussion

Odontogenic keratocyst is relatively frequent in stomatologic practice, surpassed only by root cyst and dentigerous cyst [1,2,10,11].



**Figure 11:** Evaluation postoperative (30 days): satisfactory healing.

Clinically, both cortical bone expansion and bone resorption can be observed [1,6,10,11,15].

Radiographically, the osteolytic lesion is characterized by uni or multilocular radiolucent image [1,10]. It is usually well delimited and surrounded by bone sclerosis [12]. In the posterior maxilla, it is possible for the lesion to involve the maxillary sinus, destroying its floor [14]. As observed in the present case, odontogenic kerato-



cyst can assume radiographic features that may induce the diagnostic hypothesis of periapical cyst. The clinical, radiographic, and trans-surgical features of keratocyst commonly induce the diagnostic hypothesis of ameloblastoma (when the pattern is multilocular) and dentigerous cyst (unilocular), when associated with the included tooth [1,10]. Additionally, adenomatoid odontogenic tumor, calcifying odontogenic cyst and ameloblastic fibroma are included in the differential diagnosis [11].

It is lined by uniform stratified pavementous epithelium, most often parakeratinized, and basal layer composed of columnar or cuboidal cells arranged in palisade [1,2,10,12,18]. Some characteristics of the neoplastic nature of the lesion supported the World Health Organization classification in 2005, changing the nomenclature from odontogenic keratocyst to keratocystic odontogenic tumor [1,2,5-8]. Marked expression of p53 protein, perlecan deposits, and intense expression of heparanase were correlated with the neoplastic properties of the tumor [7,9]. Currently, keratocyst is classified as an odontogenic cyst by the latest World Health Organization classification in 2017 [1,10].

Surgical removal is the indicated treatment, by means of enucleation (with or without Carnoy's solution), marsupialization, bone resection, or a combination of the techniques [1,2,6,10,11,18,21]. It has been established that the more the aggressive nature of the keratocyst, the more invasive the treatment should be, thus reducing the recurrence rate [1,2,8,10,11,18].

Keratocysts have a high recurrence rate, ranging from 22 to 60%, usually associated with the remaining tooth or the surgical technique [2,3-8,10-15,18,21]. Recurrence can occur even 10 years after surgery [3]. From this perspective, prolonged clinical-radiographic follow-up of the patient after removal of the keratocyst is recommended [11].

## Conclusion

Odontogenic keratocysts are classified as cystic, slow-growing lesions, although they are more aggressive in comparison to radicular and dentigerous cysts. It presents radiographically as a uni or multilocular radiolucent lesion, and can reach large dimensions. In the maxilla, they can mimic periapical cysts. Treatment is surgical, with variations in techniques (enucleation, marsupialization, or resection). It has a high recurrence rate, and long-term clinical and radiographic follow-up is required.

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