

# Five years follow-up of a keratocyst odontogenic tumor treated by marsupialization and enucleation: A case report and literature review

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## Abstract

Odontogenic cysts are considered as nonneoplastic benign lesions. Among the cysts, keratocyst odontogenic tumor (KCOT) is an intra-osseous tumor characterized by parakeratinized stratified squamous epithelium and a potential for aggressive, infiltrative behavior, and for the possibility to develop carcinomas in the lesion wall. Thus, the aim of this study was to describe a clinical case of KCOT in a young patient and discuss the treatment alternatives to solve this case. A 15-year-old male was referred for treatment of a giant lesion in his left side of the mandible. After the biopsy, a diagnostic of KCOT was made, and the following procedures were planned for KCOT treatment. Marsupialization was performed for lesion decompression and consequent lesion size reduction. Afterward, enucleation for complete KCOT removal was performed followed by third mandibular molar extraction. After 5 years, no signs of recurrence were observed. The treatment proposed was efficient in removing the KCOT with minimal surgical morbidity and optimal healing process, and the first and second mandibular molars were preserved with pulp vitality. In conclusion, this treatment protocol was an effective and conservative approach for the management of the KCOT, enabling the reduction of the initial lesion, the preservation of anatomical structures and teeth, allowing quicker return to function. No signs of recurrence after 5 years were observed.

**Keywords:** Keratocystic tumor, odontogenic cyst, odontogenic tumor, treatment alternative

## Introduction

Keratocyst odontogenic tumor (KCOT) is a benign neoplasm defined by the World Health Organization as a benign unicystic or multicystic, intra-osseous tumor of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium and a potential for aggressive, infiltrative behavior, and high recurrence rate.<sup>[1]</sup> Radiographically, is most often unilocular or multilocular well-circumscribed radiolucent lesion, surrounded by smooth or scalloped margins with sclerotic borders. KCOT has presumably arisen from cell rests of the dental lamina or from offshoots of the basal cell layer of the oral epithelium. The

differential diagnosis involves odontogenic cyst, dentigerous cyst, and ameloblastoma.<sup>[2,3]</sup>

Keratocyst odontogenic tumor frequently occur in the second, third or fourth decades of life in the posterior region of the mandible and ascending ramus, with a high incidence in male patients. Signs and symptoms most frequently found include pain, abscess, swelling, infection and discharge, cellulitis, and trismus,<sup>[4]</sup> but there are also cases with no symptoms. A perceptible number of cases is diagnosed incidentally during regular dental inspections.<sup>[5]</sup> It accounts for approximately 12–14% of all odontogenic cysts of the jaws. Some studies reports recurrence rates for intra-osseous odontogenic keratocysts ranging from 5% to 62%, although several studies examining a large number of cases indicate a recurrence rate of approximately 20–30% up to 10 years after treatment, though it is more common during the first 5–7 years.<sup>[2,6,7]</sup> The potential high risk of recurrence and the long intervals described in the literature explain the necessity for long-term follow-up.

The conservative treatment for this pathology includes marsupialization, decompression, enucleation, and curettage. More aggressive approach is based on osteotomy, lesion resection, use of chemical agents like Carnoy's solution, cryotherapy with liquid nitrogen or peripheral osteotomy.<sup>[8]</sup> The type of treatment is controversial, but depends on innumerable factors including: (i) Localization and size of the lesion; (ii) patient age; (iii) or whether the KCOT is recurrent or primary. However, if the patient is in the first or second decade of life and has still unerupted teeth involving

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KCOTs, aggressive surgery would be not the best choice over conservative treatment.<sup>[9]</sup>

The ultimate goal of surgical procedures is to control and to reduce potential recurrence without morbidity to the patient. Marsupialization is a technique in which the cyst lining is everted and sutured to the adjacent mucosa forming a cavity that remains open to the oral environment, allowing relieve the intracystic pressure and enable the cavity to decrease slowly in size. Afterward, when enough bone has been deposited, and the surrounding vital structures have been saved from damage, enucleation of the lesion can be performed as a second procedure.

Pogrel and Jordan<sup>[10]</sup> consider marsupialization as a definitive treatment for KCOT in a selected number of cases. The benefits of the marsupialization and decompression include: (i) Preservation of oral tissues; (ii) avoidance of surgical damage to important anatomical structures, such as the inferior alveolar nerve; (iii) maintenance of pulp vitality; (iv) prevention of dental extractions and developing teeth; (v) gradual decrease in the cystic cavity; decrease in the incidence of mandibular fracture; (vi) low risk of recurrence, and (vii) minimal surgical morbidity.<sup>[10]</sup> Likewise, there are also disadvantages with this procedure including: (i) The cyst cavity must be kept clean to prevent infection and the patient must irrigate the cavity several times per day; (ii) pathologic tissue is left *in situ* without thorough histologic examination.

Here, we report a clinical case in which KCOT was diagnosed and successfully treated with conservative marsupialization and the subsequent enucleation, with 5 year of follow-up.

## Case Report

A 15-year-old Caucasian male, accompanied by his parents was admitted to the Department of Diagnosis and Surgery at Araraquara Dental School with chief complaints of swelling in his left side of the face [Figure 1]. No systemic alterations and pain were reported. The patient denied use of alcohol and smoke. Panoramic radiography was brought by the patient and dated 1-year before, in the time wherein underwent surgery for his right third inferior molar removal. In this examination, a radiolucent image of 4 mm was associated to his left third mandibular molar. Clinical examination revealed intra and extra oral swelling, good plaque control, and no periodontal disease.

A new panoramic radiography and a computed tomography (CT) scan were requested to the patient. The panoramic image showed a radiolucent area surrounded by a radiopaque halo around the unerupted third molar, blocking their eruption [Figure 2]. The size of the lesion was increased compared to the first radiographic image. CT scan showed a cavity in the left mandibular body with a partial absence of buccal bone plate and part of the lingual bone

wall preserved [Figure 3]. Based on clinical and radiographic features, the diagnosis of a KCOT was suggested, and the treatment possibilities were discussed with the patient and parents.

Initially, an incisional biopsy was performed to confirm the diagnosis. The pathological findings were associated with KCOT and showed fragmented connective tissue, intra-osseous cavity composed mainly by stratified squamous lining with acanthosis and discontinuity of the subjacent connective tissue. In the alveolar bone was observed resorption areas with apposition areas characterized by reversion and/or incremental lines. No signs of malignancy were detected. The patient was informed about the diagnosis, and a minimally invasive surgery to KCOT removal was proposed and accepted by the patient. Written informed consent was obtained prior to initial treatment.

The treatment of the KCOT started with the marsupialization of the lesion by excision of the overlying mucosa and opening of the window of 1.5 cm size into the cystic cavity and suturing the cyst lining to the oral mucosa. The cavities in the anterior part of the mandible were kept open using a syringe by the patient and suturing in place a short piece of drain. This approach allowed freely cyst drain. During the period of lesion decompression the patient and family were instructed to maintain good oral hygiene and to clean the area with saline solution.

At 3 months, after the marsupialization for decompression, a second approach was performed to remove the KCOT. Enucleation followed by the KCOT removal was performed, and the left third mandibular molar was extracted [Figure 4]. After 9 months, the second mandibular molar had erupted, and the lesions size had decreased. Patient returns periodically in order to follow-up the treatment with clinical and radiographic examinations [Figure 5]. After 5 years, CT scan showed no evidence of lesion recurrence, great bone healing, and the second molar showed normal function with pulp vitality [Figure 6].

## Discussion

The term keratocyst odontogenic was first introduced in 1956 by Philipsen.<sup>[11]</sup> The odontogenic keratocystic tumor, formerly known as the KCOT, received its new designation in order to better characterize its neoplastic nature.<sup>[12]</sup> It is a benign developmental odontogenic tumor with many distinguishing clinical and histologic features including: (i) A potential for locally destructive behavior; (ii) a relatively high recurrence rate; and (iii) designation as a consistent finding in the nevoid basal cell carcinoma syndrome, or Gorlin syndrome. This cystic lesion most frequently presented in the second, third, and fourth decades of life at the posterior mandible of male patients, which corroborate our findings. This could be elucidated by the hypothesis that KCOTs originate from the

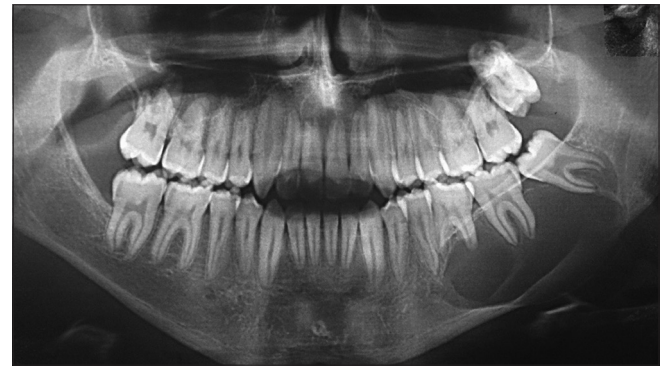
basal layer of oral epithelium, or the remnants of the dental lamina and these epithelial residues may be associated to the formation of a KCOT.<sup>[13]</sup> It accounts for approximately 12–14% of all odontogenic cysts of the jaws. It has a high recurrence rate with reports ranging from 20% to 60%.<sup>[2,3,14,15]</sup>

Radiographically, KCOT can appear the unilocular or multilocular lesion. Small unilocular cysts can be confounded with periapical, dentigerous, lateral periodontal cysts

or gingival cysts, and larger unilocular KCOT can mimic ameloblastoma. A unilocular KCOT appears as a well-defined



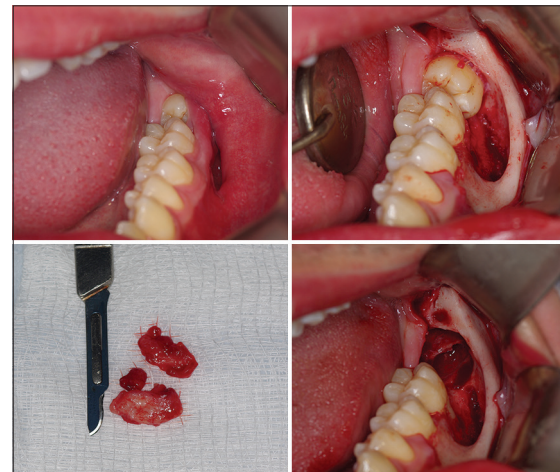
**Figure 1:** Clinical view of the patient showing swelling in his left side of the mandible



**Figure 2:** Second panoramic image showed a radiolucent area surrounded by a radiopaque halo around the unerupted third molar



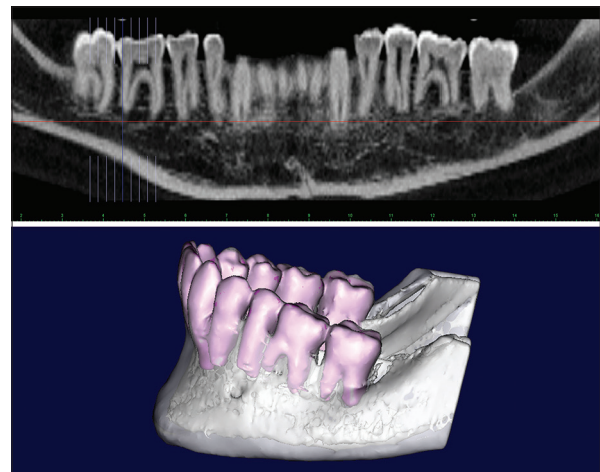
**Figure 3:** Computed tomography scan showed a cavity in the left mandibular body. Part of the lingual bone wall could be seen



**Figure 4:** Enucleation of the lesion followed by third molar tooth removal



**Figure 5:** Five year follow-up intraoral view



**Figure 6:** Panoramic computed tomography and the three-dimensional reconstruction of the mandible after 5 years showing complete healing of the keratocyst odontogenic tumor and vitality of the first and second mandibular molars



radiolucent lesion. Root resorption, extrusion of erupted tooth or displacement of impacted erupted teeth may be evident.

Histologically, the KCOT is characterized by a uniform, usually corrugated parakeratinized epithelium, thick cells presenting a flat basal surface lining, called the fibrous wall. The histology of the KCOT is pathognomonic: The cystic cavity is lined with a thin layer of connective tissue covered by orthokeratinized or parakeratinized stratified squamous epithelium.<sup>[12]</sup>

Pindborg, *et al.*<sup>[16]</sup> established the following histopathologic criteria for this lesion: (i) The epithelium lining is usually very thin and uniform in thickness with little or no evidence of ridges; (ii) there is a well-defined basal cell layer, and the component cells are cuboidal or columnar in shape and often in a palisaded arrangement; (iii) there is a thin spinous cell layer which often shows a direct transition from the basal cell layer; (iv) the cells of the spinous cell layer frequently exhibit intracellular edema; (v) keratinization is predominantly parakeratotic, but it may be orthokeratotic; (vi) the keratin layer is often corrugated; and (vii) the fibrous cyst wall is generally thin and usually uninfamed.

Among the techniques for the treatment of KCOTs, the most described approaches are: Simple surgical tumor enucleation and marsupialization, decompression by marsupialization followed by enucleation, enucleation associated with mucosal excision, peripheral osteotomy, and chemical curettage using solutions based on ferric chloride (FeCl<sub>3</sub>) and ethanol (Carnoy's solution).<sup>[17]</sup>

Large KCOTs are often managed by marsupialization, which relieves the pressure within the cystic cavity and allows new bone to fill the defect with minimal surgical morbidity. Contiguous structures such as tooth, the maxillary sinus, or the inferior alveolar canal can be saved from damage. The procedure is highly successful in decreasing the cyst size before enucleation, and is useful to avoid extensive surgery, and is considered the first option for the treatment of large KCOT.<sup>[18,19]</sup> Here, we choose to use a conservative treatment by means of marsupialization due to the size of the lesion, patient age, teeth involvement and because of the proximity to the mandibular alveolar canal which could be damaged if more aggressive surgical procedure were performed.

Pogrel and Jordan<sup>[10]</sup> advocated marsupialization as a definitive treatment for KCOTs in a certain number of cases. The authors showed that in some cases only the marsupialization for decompression, associated with hygiene and antibiotics were sufficient to promote the reduction of the lesion until its complete disappearance, followed by new bone formation. These findings are in agreement with another study that showed a distinct change in the epithelium after marsupialization that could no longer be classified as KCOT.<sup>[5]</sup> A systematic review reported that the

marsupialization for KCOTs treatment resulted in 40% of tumor recurrence after 5 years of follow-up.<sup>[6]</sup> Our results after 5 years showed no recurrence of the lesion, and all the important structures (teeth and inferior alveolar canal) were preserved. Besides, conservative treatment in this case allowed a quicker return of function without morbidity to the patient.

A recent study<sup>[20]</sup> evaluated 112 mandibular KCOTs in 109 patients who had undergone surgical enucleation. The authors showed a recurrence rate of 28 tumors treated by this technique, where seven had multiple recurrences. However, another study<sup>[17]</sup> sustain that 60% of KCOTs recurrences are from treatments using only simple enucleation, especially due to the difficulty in removing all cystic epithelium, and due to the involvement of some anatomical structures as the maxillary sinus, dental roots and inferior alveolar canal, which could be facilitated by decreasing the preenucleation cystic volume. For these reasons, it is important to emphasize the need for posttreatment follow-up for at least 15 years.

Another treatment option for the KCOTs is using chemical agents. Carnoy's solution is a solution composed by 1 g of FeCl<sub>3</sub> dissolved in 24 mL of absolute ethanol, with 12 mL of chloroform and 4 mL of glacial acetic acid, and is used as irrigating solution for the complementary treatment of KCOTs. Usually used for 5 min after KCOTs enucleation, it denatures the bone cystic cavity eliminating the cystic lesion. However, this solution is a neurotoxic substance that can result in neural commitment if in contact with the inferior alveolar nerve, and besides the Carnoy's solution have carcinogenic components, presenting recurrence rate from 25.6% to 30.3%.<sup>[6,17,21]</sup>

Pogrel *et al.*<sup>[21]</sup> in an attempt to reduce the KCOT recurrence rate, established a protocol including bone osteotomy, 1–2 mm beyond the postenucleation cystic margin with methylene blue staining, aiming to remove any remnants of cystic epithelium. However, this technique is often considered invasive especially if involves large cystic extensions and anatomical structures, leading to paresthesia, and sinus complications.

The use of marsupialization followed by enucleation and/or curettage has been showed optimal results and low recurrence rate, which are in agreement with the results of the present case. Moreover, this approach is considered an effective and less invasive technique in the treatment of KCOTs, reducing the lesion size by drainage and decompression up to 47% of the initial size,<sup>[17]</sup> decreasing the aesthetic and functional damages, allowing the preservation of important anatomical structures.<sup>[22]</sup> An interesting finding of this case is that the first and second mandibular molar was preserved, and the patient did not show any signs of paresthesia related to injury in the mandibular alveolar nerve. The patient does not show any sign of necrotic pulp in his first mandibular molar even

with a small resorption in the root apex, not being necessary additional procedures. Sanchez-Siles *et al.*<sup>[23]</sup> showed a clinical case of KCOT treatment using the marsupialization technique prior to enucleation. This approach allowed the lesion reduction of 50 mm × 25 mm to 13 mm × 13 mm in diameter (radiographic aspect), facilitating the complete removal of the lesion with less-invasive approach and without recurrence within the 1-year of follow-up corroborating the results of this case report.

## Conclusion

The use of the marsupialization technique followed by enucleation for the treatment of KCOTs was an effective and conservative approach for the management of the KCOT, enabling the reduction of the initial lesion, the preservation of anatomical structures and teeth, allowing quicker return to function. No signs of recurrence after 5 years were observed.

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