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# Diagnostic and treatment features of keratocystic odontogenic tumors

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## ABSTRACT

Keratocystic odontogenic tumors (KCOT) comprise a unique pathological entity characterized by aggressive/destructive behavior and propensity to recurrence. This study describes the diagnostic and treatment features of a KCOT lesion. A 22-year old man was referred for surgical treatment of pericoronitis on tooth no. 37. Panoramic radiography revealed a unilocular, large radiolucent area extending from tooth no. 36 to the left mandibular ramus. Aspiration and incisional biopsy were performed, and the tissue sample was sent for microscopic evaluation. Microscopically, a cystic lesion was observed, lined by keratinized squamous epithelium and filled with keratin lamellae, confirming the diagnosis of KCOT. Surgery was performed in an outpatient setting and involved osteotomy, detachment of the cystic lesion, and removal of teeth no. 36, 37, and 38. The patient was clinically and radiographically followed for 12 months, and no evidence of recurrence was observed. KCOTs should be considered in the differential diagnosis of lesions affecting the posterior region of the mandible. Accurate clinical, radiographic, and microscopic examinations are essential to establish the definitive diagnosis and choose the most effective therapy.

**Keywords:** Keratocystic odontogenic tumor, odontogenic keratocyst, oral neoplasms.

## Particularidades do diagnóstico e tratamento do tumor odontogênico queratocístico

## RESUMO

O tumor odontogênico queratocístico (TOQ) é uma entidade patológica singular, devido ao seu comportamento agressivo/destrutivo e à sua propensão a recorrências. O presente

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trabalho descreve as particularidades de diagnóstico e tratamento de um TOQ. Um paciente do sexo masculino, com 22 anos de idade, foi encaminhado para tratamento cirúrgico de pericoronarite no dente 37. O exame radiográfico panorâmico revelou uma área ampla, unilocular, estendendo-se do dente 36 até o ramo esquerdo da mandíbula. Punção óssea aspirativa e biópsia incisional foram realizadas, e a amostra de tecido foi encaminhada para análise microscópica. Microscopicamente, observou-se lesão cística, revestida por epitélio escamoso queratinizado e preenchida por lamelas de queratina, confirmando o diagnóstico de TOQ. O procedimento cirúrgico foi realizado em ambiente ambulatorial e envolveu osteotomia, descolamento da luz da lesão e exodontia dos dentes 36, 37 e 38. O paciente foi acompanhado clínica e radiograficamente por um período de 12 meses, e não foi observada recorrência da lesão. O TOQ deve ser considerado no diagnóstico diferencial de alterações da região posterior da mandíbula. Exames clínicos, radiográficos e microscópicos precisos são essenciais no estabelecimento do diagnóstico e na escolha da modalidade terapêutica mais eficaz.

**Palavras-chave:** Tumor odontogênico queratocístico, queratocisto odontogênico, neoplasias orais.

## INTRODUCTION

Odontogenic keratocysts, recently reclassified as keratocystic odontogenic tumors (KCOTs) (1), are among the most controversial and frequent pathological entities affecting the maxillofacial region (2-4). Their aggressive/destructive behavior and proneness to recurrence have led the condition to be classified as a benign neoplasm, in spite of actually being a simple cystic lesion (5).

KCOTs are twice more frequent in the mandible than in the maxilla (6,7). Their main clinical manifestations are swelling and/or pain, even though some asymptomatic cases have also been reported (8-10). Radiographically, KCOTs present non-pathognomonic features (2,9,10): they may appear as small or large, round or ovoid radiolucent lesions, often with scalloped, multilobulated, distinct margins. Involvement of an impacted tooth has been reported in 25 to 40% of cases (6,7).

Histopathologically, KCOTs present some distinguishing features compared with other odontogenic tumors. The epithelium may show budding of the basal layer into underlying connective tissue, with formation of detached microcysts, termed daughter cysts (2). Treatment remains controversial, and different approaches have been reported in the literature (11). As a conservative method, simple enucleation with or without curettage and marsupialization can be performed. More aggressive methods include peripheral osteotomy, chemical curettage with Carnoy's solution, and resection (8,12).

The present article describes the case of a 22-year old man who presented with a KCOT involving teeth no. 36, 37, and 38. Diagnosis and treatment features are discussed.

## CASE REPORT

A 22-year old male patient of African descent was referred to the Department of Oral and Maxillofacial Surgery of the Municipal Health Department of Goiânia, state of Goiás, Brazil, in December 2008, for surgical treatment of pericoronitis on tooth no. 37. Patient medical history was negative for other conditions, except for allergy to sodium dipyrone. Oral examination revealed unilateral swelling of the left cheek region, of hard consistency, with no point of fluctuation, presence of purulent discharge, citric yellow in color, on the distal surface of tooth no. 37, and no eruption of tooth no. 38. There were no spontaneous complaints (Figure 1A).

Panoramic radiography revealed a unilocular, large radiolucent area extending from the mesial surface of the distal root of tooth no. 36 to the left mandibular ramus. Tooth no. 38 was completely involved in the radiolucent lesion and displaced into the left mandibular ramus. No root resorption was noted (Figure 1B). The patient underwent aspiration, incisional biopsy, and histological examination. The latter revealed a cystic lesion, lined by keratinized squamous epithelium and filled with keratin lamellae (Figure 1C-E), confirming the diagnostic hypothesis of KCOT.

The surgical procedure was performed in an outpatient setting under local anesthesia with no sedation. Preventive drug therapy with 875 mg amoxicillin/clavulanic acid combined with 750 mg paracetamol and 100 mg nimesulide was administered 1 hour before surgery. An incision was made along the left mandibular ramus, and a mucoperiosteal flap was raised, with intrasulcular extension to teeth no. 36 and 37. Osteotomy was performed using a 701 bur attached to a high-speed handpiece, with subsequent exposure of the cystic lesion, which was then detached until the region of the coronoid process and left mandibular condyle. Thereafter, teeth no. 36, 37, and 38 were removed to avoid condylar base fractures (Figure 1F-H). Occlusion sutures were made using 4.0 nylon monofilament, and the patient was instructed to keep the medication in the postoperative period (10 days). The patient was followed for 12 months and underwent clinical and radiographic examinations every visit. The healing process was satisfactory, with no evidence of recurrence throughout the follow-up period (Figure 1 I).

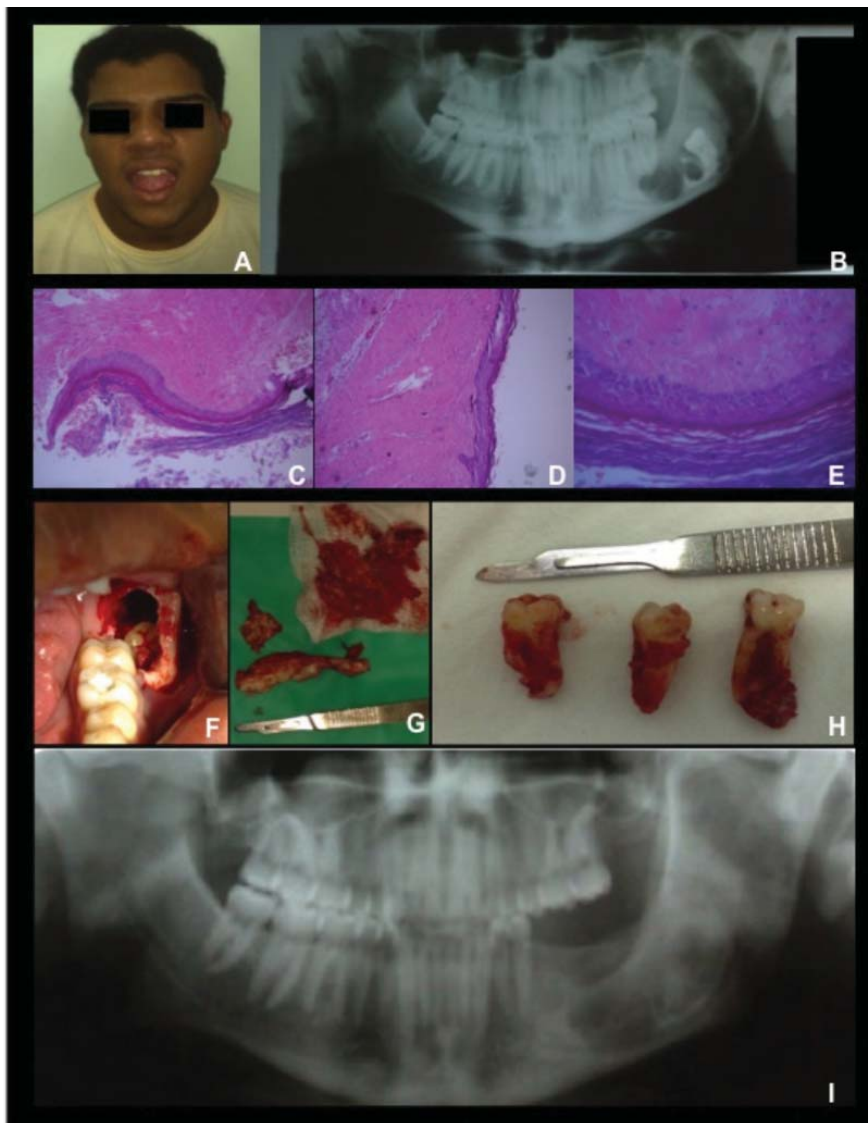


FIGURE 1 – A) Extraoral photograph showing unilateral swelling of the left cheek region; B) panoramic radiograph showing a unilocular, large radiolucent area associated with an impacted third molar; C-E) parakeratinized squamous epithelium filled with keratin lamellae; F-H) surgical procedure, excised specimen, and teeth no. 36, 37, and 38; I) panoramic radiograph evidencing healing and no recurrence at 12 months' follow-up.

## DISCUSSION

Odontogenic tumors comprise a rare group of lesions derived from the tooth-forming apparatus (6). Among odontogenic tumors, KCOTs are the second most frequently diagnosed lesion, with reported prevalence rates ranging from 19 to 38% (3). KCOTs are of great interest in clinical practice and require special attention due to their specific histopathological features, aggressive and infiltrative behavior, and tendency to recurrence (2,11).

KCOTs are usually observed in patients aged 10-29 years and show a slight preference for men (7,9,13,14). Lesions are mostly located in the mandible, especially in the posterior portion of the mandible and in the mandibular ramus, and are generally related with unerupted teeth (6,7,9,10,13,14). In our case, a KCOT was diagnosed in a 22-year old man of African descent, and it involved the root apex of teeth no. 36 and 37, in addition to unerupted tooth no. 38.

Patients with KCOTs may present various symptoms and features, e.g. swelling, pain, discharge, aggressive growth, invasion of adjacent structures, and recurrence (10,13). Myoung et al. (13) reviewed 256 cases of KCOT and observed that the most frequent clinical manifestations upon admission were swelling, pain, or a combination of both (82.4% of all cases). Conversely, Boffano et al. (14) reported that 62.8% of patients with KCOTs had no symptoms. It is important to note that KCOTs can be easily mistaken for inflammatory lesions, as patients usually show typically inflammatory symptoms, such as pain, swelling, and discharge (15). In our case, the patient did not spontaneously complain of pain, and was initially referred for surgical treatment of pericoronitis on tooth no. 37.

Conventional radiographic examinations such as panoramic and intraoral periapical radiographs are usually adequate to determine the location and estimate the size of a KCOT (7). Radiographically, most KCOTs present as a unilocular or multilocular well-circumscribed radiolucent lesion with smooth or scalloped margins and sclerotic borders (9,10,14). Because these radiological features are non-pathognomonic (2), differential diagnosis should include dentigerous cysts, ameloblastomas, radicular cysts, simple bone cysts, central giant cell granulomas, arteriovenous malformations, and fibro-osseous lesions (16,17). According to Güller et al. (7), one radiographic feature suggestive of KCOT is lesion growth in an anterior-posterior direction within the medullary cavity, with no obvious bone expansion.

Histologically, KCOTs present the following features: presence of a well defined, often palisaded, basal layer consisting of columnar or cuboidal cells; intensely basophilic nuclei of columnar basal cells oriented away from the basement membrane; parakeratotic layers, often with a corrugated surface; and mitotic figures frequently present in suprabasal layers (1). Some authors have classified KCOTs into parakeratotic and orthokeratotic subtypes (18), according to immunohistochemical and histologic features of the lining and the type of keratin produced (9,18). The parakeratotic type accounts for the majority of KCOTs (83 to 97%) (9) and has a greater potential for local destruction and extension

into adjacent tissues, rapid growth, recurrence, and multiplicity (7). The lesion identified in our case was a parakeratotic KCOT.

The treatment of KCOTs remains a controversial topic, and several treatment options are currently available, including decompression, curettage, marsupialization, enucleation, and resection (2,11,14,19). Johnson et al. (11) have conducted a review of the literature to provide an updated overview of the management and recurrence rates of KCOTs. Of the 2,736 articles retrieved by the authors, only eight met the inclusion criteria. In addition, because of a large number of variables assessed and the absence of randomized controlled trials available on the topic, a meta-analytic approach could not be used to analyze results. According to Maurette et al. (19), conservative treatment of KCOTs has important advantages: it is less traumatic for the patient, eliminates the need for medication, reduces hospitalization costs, and avoids the need for reconstruction using grafts or other bone extension methods. In our case, the KCOT was treated with enucleation associated with curettage, a conservative and effective treatment option. Healing was satisfactory, with no evidence of recurrence over the follow-up period.

In conclusion, KCOTs are benign uni- or multicystic intraosseous tumors of odontogenic origin and should be considered in the differential diagnosis of jaw lesions. Notwithstanding, even in the presence of clinical and radiological features indicative of KCOT, a definitive diagnosis cannot be made without microscopic analysis. Only a detailed investigation will allow to determine the most effective treatment and thus to avoid recurrences.

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