

Keratocystic Odontogenic Tumor of the Maxilla Revealed By a Diffused Cellulitis: A Case Report

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Case Report

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Abstract: Keratocystic odontogenic tumor (KCOT) is a unique cyst because of its locally aggressive behavior, high recurrence rate, and histological characteristic appearance. The maxillary presentation is rare. This paper reports a case of maxillary KCOT successfully treated with radical enucleation. A 23-year-old female patient was referred to our department to search for a possible dental etiology to a right upper facial cellulitis. Radiology showed a well-limited unilocular radiolucent area, between the teeth 15 and 16 which were displaced. Initially, a lateral periodontal cyst was suspected. An enucleation of the cyst was performed. Histology concluded on a maxillary KCOT. A follow up at 9 months showed complete healing with no signs of recurrence. No consensus exists concerning KCOT management. The most conservative treatment used is enucleation. However, it had a high recurrence rate of 56% up to 10 years after treatment compared with other methods which explain the need for long-term follow-up.

Keywords: Odontogenic tumors, odontogenic cysts, periodontal cyst, maxilla, therapeutics

INTRODUCTION

Keratocystic odontogenic tumor (KCOT) is a unique cyst because of its locally aggressive behavior, high recurrence rate, and histological characteristic appearance. In 62% of cases, KCOT is diagnosed incidentally during routine dental examination [1]. However, only 38% of the cases produce symptoms including pain, discharge, swelling and cellulitis [1, 2].

As for the site of predilection, the most common one is the mandible with 81%, most frequently affecting the body, angle and vertical ramus. The maxillary presentation is not pathognomonic and accounting for only 16% [1]. The aim of this report was to present a case of KCOT in the maxilla, discovered through a right facial cellulitis. The goal was also to provide the clinical and radiological diagnosis criteria of KCOT, its differential diagnosis and the treatments modalities.

CASE REPORT

A 23-year-old female patient, with non-contributory medical and surgical history, was referred to our unit by the Otorhinolaryngology (ENT) department at Farhat Hached Hospital Sousse to search for a possible dental etiology to a right upper facial cellulitis.

The medical history revealed that the cellulitis had been progressing since 5 days. Before being admitted to ENT department, she consulted a general practitioner who prescribed two injections of corticosteroids with no significant improvement.

On initial examination, an unlimited right cheek swelling covered with an erythematous and painful skin on palpation was noted. An extension to the eyelid region was evident.

Intra oral examination showed swelling of the mucosa in region 15-16 with a purulent discharge through a fistula (Figure 1). There was a first-degree mobility of these teeth with an increased periodontal probing depth (10 mm). No caries were noticed on these teeth which tested positive for vitality (ethyl chloride and cavity test).



Fig-1: Intra-oral clinical aspect: swelling of the mucosa in region 15-16 with a purulent discharge through a fistula.



Fig-2: Panoramic (a) and periapical (b) radiograph showing a well-defined unilocular radiolucent image without a cortical border, between the teeth 15 and 16 which are displaced

Panoramic and periapical radiography showed a well-defined unilocular radiolucent image without a cortical border between the teeth 15 and 16 which were displaced (Figure-2a and b).

A computed tomography (CT) scan of the maxilla with a contrast agent injection was performed. The CT scan with bone window shows a hypodense

unilocular lesion well-defined associated with the thinning of the maxillar cortex, the displacement of the maxillary sinus floor and thickening of the sinusal floor mucosa (Figure-3). The CT scan with soft tissue window and contrast agent injection shows the radiologic features of cellulitis: the maxillary abscess with soft tissue thickening and myositis.

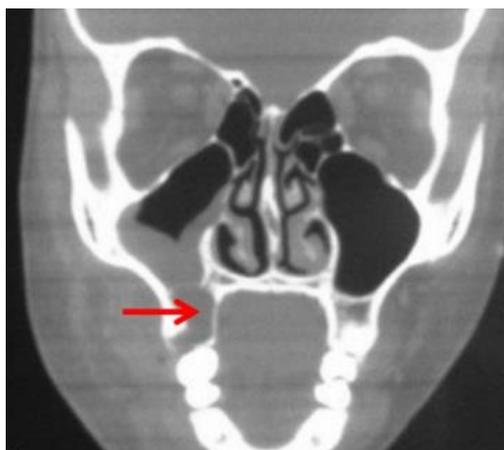


Fig-3: CT scan of the maxilla with bone window (coronal section) showing a hypodense unilocular and well-defined lesion associated with the thinning of the maxillar cortex, the displacement of the maxillary sinus floor and thickening of the sinusal floor mucosa (red arrow)

Based on clinical and radiographic features, the diagnosis of a lateral periodontal cyst and a keratocystic odontogenic tumor were suggested.

The patient was already under antibiotic treatment, prescribed by the department of Otorhinolaryngology, based on the association of

Amoxicillin, Clavulanic Acid and Gentamycin on intravenous.

Initially, an intraoral incision was performed for the drainage of the abscess and the antibiotic treatment was extended. At 3 weeks, a total regression of symptomatology was noticed. Then we performed an enucleation of the cystic lesion, under local anesthesia, with a preservation of the teeth 15 and 16. A thick whitish content mimicking keratin in the cystic cavity was found intraoperatively.

After total removal of the cyst, extensive cavity curettage with reduction of the surrounding bone, to remove residual cystic epithelium, was performed. Before suturing the flap, an internal dissection was done in order to remove the part of the mucosa attached to the cyst.

A post operative medication based on an association of amoxicillin-acid clavulanic, an antalgic and a mouth rinse chlorhexidine was prescribed. Anatomopathologic results concluded on a keratocystic odontogenic tumor (Figure-4).

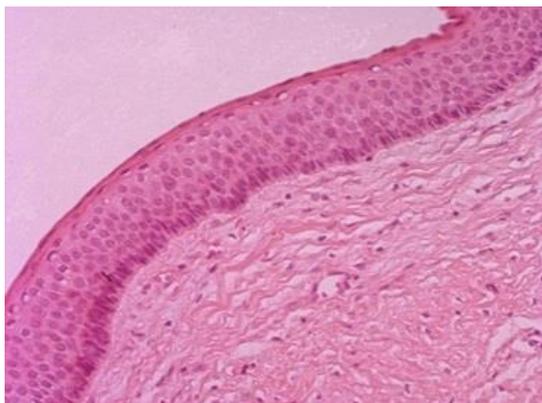


Fig-4: (HE *200) Odontogenic keratocyst showing thin parakeratinized lining. The basal cells have palisaded nuclei.

After 1 month, we noticed total healing of the site with no pain or signs of infection. A follow up at 9 months later shows a complete mucosal healing with no clinical signs of recurrence (Figure 5). Radiographic

and periapical radiographs (Figure-6a and b) follow up after 9 months show bone restoration in the area of the KCOT.



Fig-5: At 9 months: Complete mucosal healing with no clinical signs of recurrence

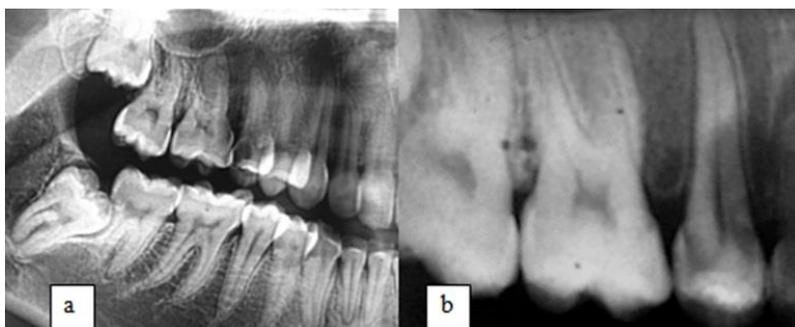


Fig-6: At 9 months: Panoramic (a) and periapical (b) radiographs follow up showing bone reconstitution in the area of the KCOT

DISCUSSION

In 1992, the World Health Organization (WHO) defined keratocysts as benign, uni- or multicystic intraosseous neoplasms of odontogenic origin. They have a potentially aggressive and infiltrating behavior. The well-known aggressive evolution of keratocysts, their histology, and new findings in genetics led the WHO in 2005 to reclassify these lesions as keratocystic odontogenic tumors (KCOTs) [3].

KCOT accounts for approximately 26, 15% of all odontogenic tumors of the jaws [4]. They are more frequent in males and have their peak incidence in the third decade of life. They may develop sporadically or accompany Gorlin syndrome (nevroid basal cell carcinoma syndrome, NBCCS) [5].

The most common site for KCOT is the mandible. Unusual locations have also been reported, such as the anterior portion of the maxilla, and the maxillary third molar area [6]. Racio *et al.* [1] in a retrospective study found that the overall mandibular to maxilla ratio of KCOT occurrence was 5:1, with 81% of the lesions located in the mandible, most frequently in the body (20%), angle (18%) and vertical ramus (10%). Only 16% of the lesions occurred in the maxilla, most in the posterior region (13%) and only 3% in the anterior one [1]. In our case, KCOT was located in the maxilla, which is a rare and an uncommon location. Its radiological aspect as a radiolucent lesion creeping, in a pear shaped, between the vital teeth 15-16 evoked a lateral periodontal cyst in the first place.

The KCOT tends to be found incidentally, during routine dental examination, because it shows no early symptoms and is painless. Racio *et al.* [1] reported that the frequency of such cases is up to 62% [5, 7]. While 38% of the cases produced symptoms and are discovered through an infectious process including pain, swelling and cellulitis [2]. That was the case of our patient where the presence of the KCOT was discovered through an infectious process with an upper right facial cellulitis.

The common KCOT radiographic features are unilocular or multilocular well-circumscribed radiolucent lesions surrounded by a thin sclerotic border [7]. KCOT characteristic feature is growth in the mesial-distal rather than vestibular-lingual direction which delays the symptoms of bone distension [5]. This last is less seen in maxilla since it is a pneumatic bone [7]. The only reliable radiographic parameter described is the lack of cortical expansion in most KCOTs compared with odontogenic cysts or ameloblastomas, tending to hollow the mandible and fenestrate the lingual cortex [1]. Similar features were noted in our case, with a broken buccal cortex and no cortical expansion.

Displacement of impacted or erupted teeth may be evident. Tooth resorption is rare but tooth dislocation is more common [7]. In our case radiologic features shows a displacement of the teeth 15 and 16 without root resorption.

Maxillary KCOT is difficult to differentiate from others lesions like: periapical cysts when it is located at the periapical region of teeth, dentigerous cysts when it envelopes the crowns of unerupted teeth and lateral radicular cysts or lateral periodontal cysts (LPC) when it's located between the roots of the teeth [7]. The LPC is a non inflammatory cyst on the lateral surface of the root of a vital tooth that occurs more often in the mandible [8]. It appears radiographically as a round or oval well-defined radiolucency. Keratocysts must be differentiated from the LPCs because of their aggressiveness and high potential for recurrence following surgical removal. It is important that the clinicians are aware of odontogenic keratocysts occupying a lateral periodontal position [9]. In our case the first diagnosis proposed was lateral periodontal cyst because of its radiological aspect and the vitality of the teeth 15-16.

The histopathology of the KCOT is pathognomonic: it shows a regular parakeratinized stratified squamous epithelium with 8-10 spinous cell layers thickness; palisading in basal layer with cubic cells not present in orthokeratinized odontogenic cyst. The spinous cell layer frequently exhibit intracellular edema and the lumen contains amounts of desquamated keratin [2, 7, 10]. Anatomopathologic results in our case were consistent with the literature and concluded on a KOCT.

No consensus exists concerning the management of KCOT. The goal of surgical treatment is to control potential recurrence with the least morbidity [1, 7]. The type of treatment depends on several factors including: patient age, lesion site and size, and whether the KCOT is recurrent or primary [2].

The conservative treatment includes marsupialization, decompression, enucleation associated or not with mucosal excision, and curettage. More aggressive approaches are based on peripheral osteotomy, marginal or radical resection associated or not with chemical curettage using solutions based on ethanol (Carnoy's solution) and liquid nitrogen for cryotherapy [2, 6].

The most conservative surgical treatment of KCOT used is enucleation. However, this treatment has a significantly higher recurrent rate of up to 56% than those treated with other methods. Based on the high rate of recurrence, most authors advocate radical enucleation for small unilocular keratocysts [6, 7]. Radical enucleation involves removal of the entire cyst lining together with any associated overlying mucosa, followed by extensive cavity curettage with reduction of the surrounding bone to remove residual cystic

epithelium. The recurrence rate with this surgical technique drop to 30,8% [11]. In our case, we performed a radical enucleation. We decided to conserve the teeth because the patient was young and the teeth were vital without mobility.

Some studies reports recurrence rates for intra-osseous odontogenic keratocysts ranging from 5% to 62% [2]. Racio *et al.* in 2014 [1] demonstrated the predictive KCOT recurrence factors in a 5 years follow-up: there is a higher rate of recurrence in men (31%) than woman (17%). KCOTs located in the mandible showed a higher recurrence rate (29%) than those in the maxilla (11%) with a predilection for mandibular body and ramus. Furthermore, 35% of the lesions involving more than one anatomical area developed a recurrence. In cases associated with one or more teeth, the recurrence rate was 40% whereas in the group with no tooth involvement it was 20%. The recurrence rate was higher in cases with third molar involvement (40%) than those without third molar involvement (17%) [1]. In our case the patient was a female, the site of the KCOT occurrence was the maxilla and the lesion was well-defined and unilocular, which are factors for a better prognosis. However, the close relation between the teeth 15 and 16 with the KCOT could increase the risk of recurrence.

Daughter cyst formation has also been associated with high recurrence rates. They are often found where the wall of the cyst is attached to the mucosa (13% of lesions) [1, 3]. Based on this theory, some authors have proposed the excision of an area of mucosa where the cyst is attached [1]. The use of chemical curettage, cryotherapy, or radical curettage has been advocated to ensure removing remaining epithelium. However, a recurrence was reported in the cases treated with enucleation in combination with Carnoy's solution. This may have been because of the ability of Carnoy's solution to fix a daughter cyst located in bone but not in gingival tissues [1]. In our case we had perform an internal excision of the mucosa attached to the cyst in order to prevent recurrence occurrence.

Recurrence has been described with approximately 20–30% up to 10 years after treatment, though it is more common during the first 5-7 years. The potential risk of recurrence and the long intervals reported explain the need for long-term follow-up [1, 2]. For our patient a clinical follow up at 9 months showed a total mucosal healing. The radiographic follow up shows bone reconstitution in the area of the KCOT.

CONCLUSION

KCOTs are rare tumors which most frequently develop in the mandible. They are often found incidentally. Despite their rare occurrence, they must be taken into consideration in radiological and clinical diagnostics. Due to the frequent recurrence of KCOT,

also after radical surgery, patients are recommended to be kept under long-term and close radiological supervision.

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