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ORIGINAL ARTICLE

# Association of Treatment and a 3-D image follow-up on a Recurrent Keratocyst - Case Report

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## Abstract:

**Introduction:** Accordingly to the latest edition of the World Health Organization (WHO) the previously known Keratocyst Odontogenic Tumour (KCOT) has now returned to the simple odontogenic cyst (OKC) classification. We present a case successfully treated by a combination of minimal-invasive approaches. **Case Presentation:** A large OKC was identified extending from tooth 3.8 through the condylar process in the mandible and staged surgical conservative approaches were performed. Total healing was achieved and followed-up over 8 years. The case was well documented via panoramic radiographs, CBCTs, and a 3D image tool illustrates the cortical bone destruction (before treatment) and the cortical bone healing after treatment. **Discussion:** Agreement regarding terminology and treatment of OKC has been reached. In this case, a complete healing of a recurrent OKC was achieved by decompression, enucleation, and blurring of the bone walls. Rigorous follow-up enriched by a 3D reconstruction imaging allows an educational view of the healing. **Conclusion:** This case suggests that a staged surgery approaches concurrent to rigorous patient follow-up could be a feasible alternative to extensive OKT treatment. And, reinforces the importance of collaboration between orthodontist, pathologist, OMS, and the patient have crucial importance in the conservative management of the lesion.

**Keywords:** Odontogenic Cysts; Recurrence; Pathology, Oral; Treatment Outcome; Imaging, Three-Dimensional

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## INTRODUCTION

We have known Keratocystic odontogenic tumour (KCOT) as a neoplasm since the World Health Organization third edition -2015- classified it as a tumour. Although, recently the latest WHO edition-2017- renamed the KCOT as simple Odontogenic Keratocyst (OKC)[1].

Clinically, OKC grows silently and extensively through medullary bone fenestrating cortical bone. It is more prevalent in males than in females, and it is more common in the third decade of life. Radiographically, it may be seen as a multilocular or unilocular radiolucent lesion with a well-defined margin. Histologically, OKC has a unique feature showcasing a cystic cavity lined with a 6-10 cell corrugated ortho keratinized or para keratinized stratified squamous epithelium[2-4].

Cone-Beam Computer Tomography (CBCT) as a three-dimensional tool has been used in oral and maxillofacial surgery to aid diagnosis, treatment planning and follow-up controls Nevertheless, the follow-up imaging could be intercalated with digital panoramic imaging[5-7] to reduce ionising radiation exposure.

For a while since its first description by Philipsen in 1956 OKC ideal treatment was considered controversial[8], diverging between conservative and aggressive surgical treatments. Lately, the literature has suggested less invasive interventions[9].

In this case report, we present an extensive case of a recurrent OKC surgically approached through a conservative and minimally invasive approach that was successful. Rigorous patient long-term follow-up of over 8-years is presented with a technological 3D image of the healing bone.

## CASE PRESENTATION

A 27-year-old woman presented with a large tumour extending from the tooth 3.8 area to the ascending ramus, coronoid process and condylar process in the left mandibular side. The lesion had been incidentally found by an orthodontist and referred to an oral and maxillofacial surgeon who first examined the patient in May 2009.

Clinically, there was no cortical expansion, and no pain was referred by the patient. Neither family nor medical related history was identified. Three days after the finding, an excisional biopsy (50x20x10mm) was performed under local anaesthesia associated with nitrous oxygen sedation. The lesion was pretty friable which did not allow the surgical team to remove the whole lesion at once.

The material was sent to the pathologist under three differential potential diagnoses: ameloblastoma, keratocystic or dentigerous cyst due to its radiographic and clinical features. For the postoperative care, the patient was medicated and oriented to perform irrigation with saline solution for a few days following the biopsy. In addition, she was oriented to watch for her alimentation, rather choose soft food and avoid mastication on the left side.

A few days later, the pathologist confirmed the diagnosis of KOCT (Fig. 1). Contact with the patient was kept until May 2010 when contact was lost. On June 2012, we received a call from her orthodontist reporting that a recurrence of the lesion while performing an endodontic treatment had been noticed (Fig. 2 to 5).

At that time, there was no consensus on the literature regarding ideal treatment for recurrent OKC. This was discussed with a head and neck surgeon - who suggested a radical surgical approach in the area that would require a resection followed by reconstruction with micro-revascularization - and with the pathologist

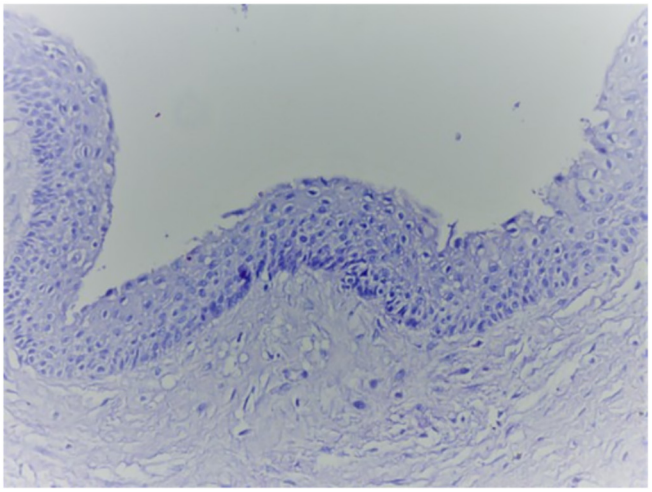

**Figure 1.** Histopathological view of parakeratinized 6-8 epithelium cells layer.

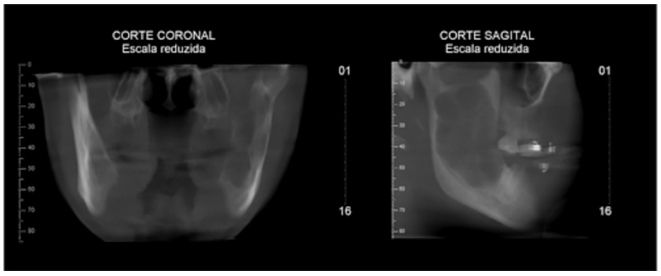


**Figure 2.** Coronal slice and axial slice (2012).

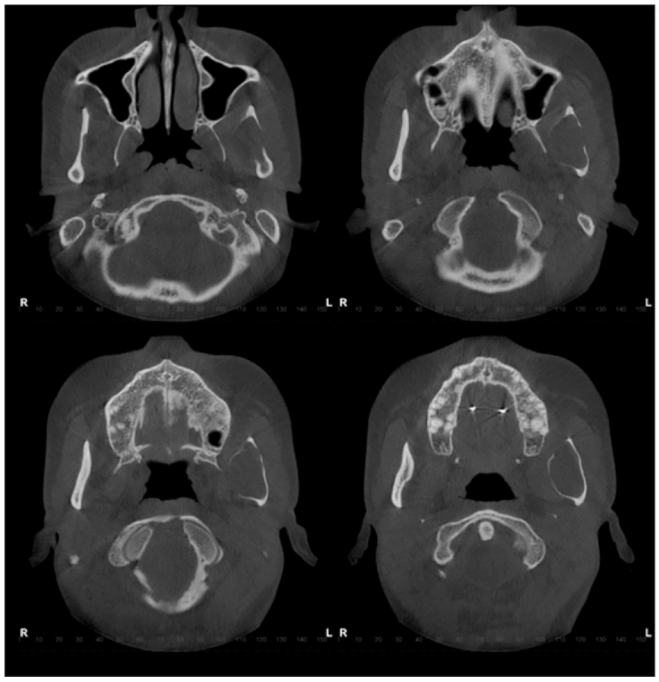

**Figure 3.** Axial image (2012).

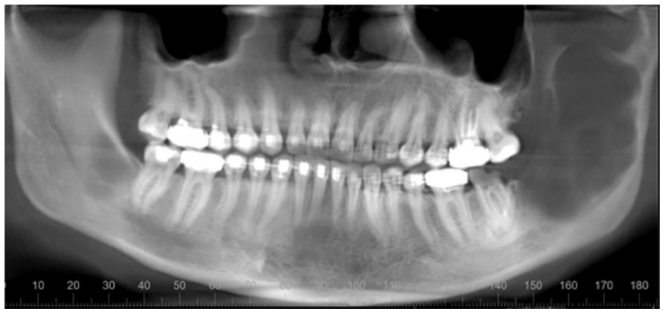

**Figure 4.** Secondary reconstruction of a DICOM - Panoramic view June 2012.

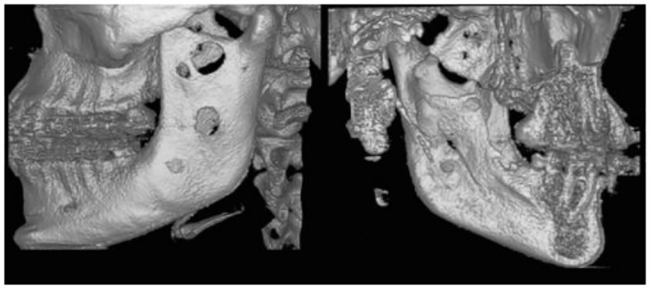

**Figure 5.** Hard tissue 3D DICOM reconstructions where cortical destruction can be seen (June 2012).

- who suggested a more conservative approach. Having disagreement among specialists and in the literature, the oral and maxillofacial surgeon agreed with the latest since we would count on the patient's follow-up collaboration.

We decided to defer a final decision to the patient. On a meeting with the patient and her husband surgical options were explained, and due to a variety of reasons, the patient's choice was to try the less radical approach, to adhere to the treatment and to follow-up all recommendations.

Thus, we planned a combination of conservative and aggressive approach, first decompression - in order to get some bone neoformation and minimise the sequel if a future resection and reconstruction was needed; secondly, by enucleation associated to a blurring of the peripheral bone.

At this second stage, we performed lesion aspiration plus incisional re-biopsy (20x20x6) and installed a tube for decompression which allowed the removal of a small cortical bone from the anterior face of the ramus. The re-biopsy histological analysis (June 2012) reconfirmed the diagnosis of OKC (Fig. 6). The irrigation with saline solution was kept for 5 more months, until December 2012, when there were no longer signs of secretion from inside the lesion. In October 2012, a radiographic image suggested that bone neoformation had occurred. On February 2013, under nasotracheal intubation, we performed the third intervention, a full enucleation of the lesion plus peripheral bone osteotomy.

The use of Carnoy's solution was rejected as the superior cortical of the Inferior Alveolar Nerve (IAN) was destroyed by the tumour, so it was considered safe not to use it. Hence an osteotomy was the harmless line of action. During the procedure, we could observe the increase in thickness of the lesion, which allowed a complete enucleation without rupture of the membrane.

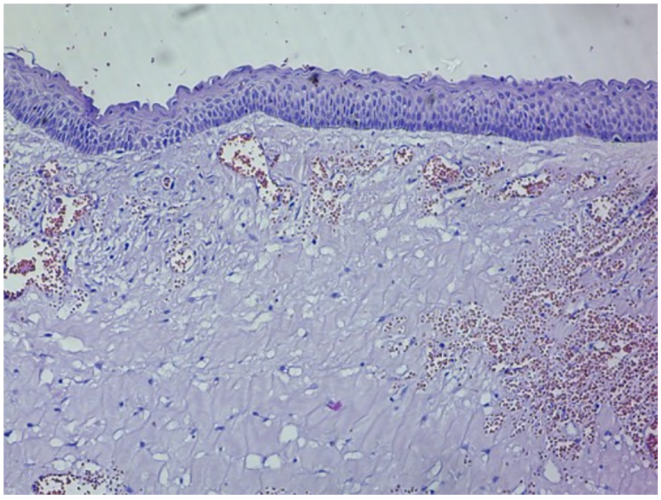

**Figure 6.** Histopathological view showing evident palisaded basal layer cells.

We've been following up the patient since then. Panoramic images obtained 8 months after the third surgery (Fig. 7) and two years after the intervention had shown no recurrence signs observed and a recorticatization of the IAN canal (Fig. 8). In 2016, her dentist performed an extraction of tooth 3.7.

A CBCT showed no recurrence signs. Unfortunately, this exam only caught the body region of the mandible (Fig. 9) because of its limited field of view (FOV). Because we would like to analyse the healing site tridimensionally (Fig. 10) a year later we requested a full volume CBCT in order to avoid misinterpretation of any hidden recurrence. This last exam was done in June 2017, where a complete healing can be seen in Fig. 11 and 12.

## DISCUSSION

The ongoing debates regarding bone lesions since the first international standard classification by WHO led to a cyst-tumour-cyst fluctuating classification of keratocyst[1]. Despite the having the term cyst on its name, Keratocyst was defined as an odontogenic tumour

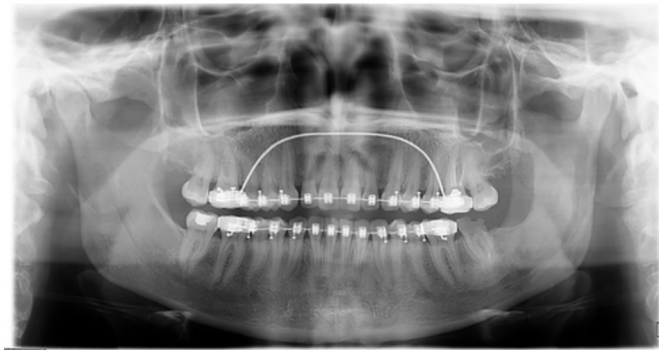

**Figure 7.** Conventional Panoramic view 8 months post enucleation (October 2013).

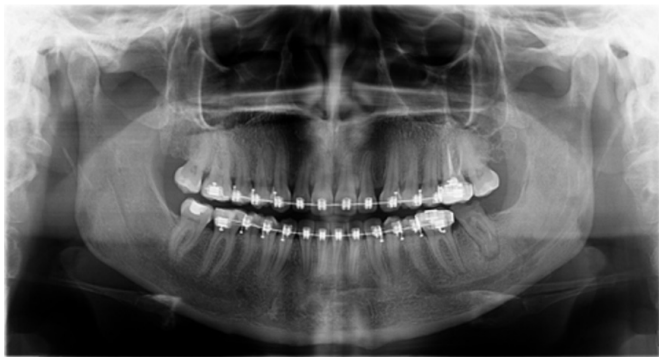

**Figure 8.** Panoramic view 2 ½ years post enucleation (September 2015). Visible re-corticalization of IAN canal.

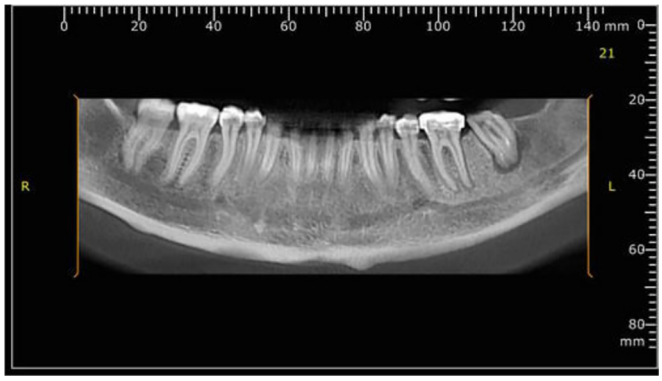


**Figure 9.** CBCT prescribed in 2016 where a apical lesion can be seen on tooth 3.7 but no ramus image can be seen.

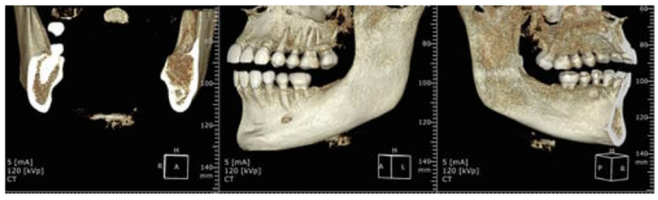

**Figure 10.** 3D reconstruction of the mandible total healing of the lesion (June 2017).

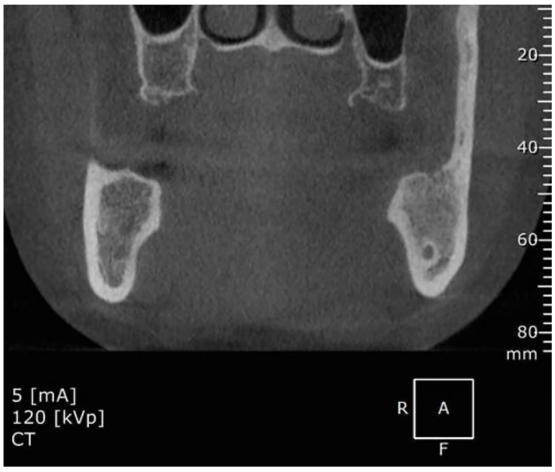


**Figure 11.** Total re-corticalization of the IAN canal's cortical (June 2017).

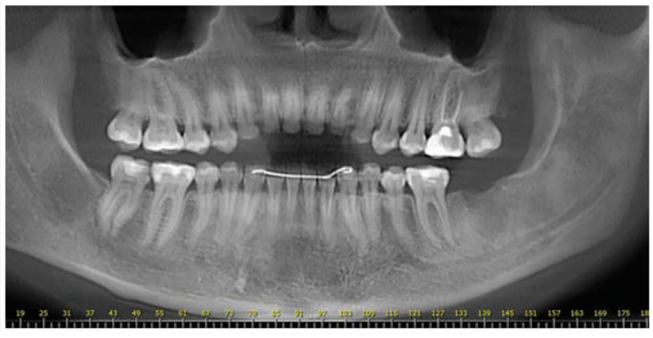

**Figure 12.** Panoramic view of a CBCT mandible showing healing and total re--corticalization including in the anterior ramus area where formerly we had made the fenestration.

with distinct histological and clinical characteristics[10]. Subsequently, the former Odontogenic Keratocyst (OKC), was renamed as the Keratocystic odontogenic tumour (KCOT) after 2005 WHO reclassified it as a tumour and defined as a benign intraosseous neoplasm of the jaws[9-11]. OKC has been considered by far the most potentially aggressive cystic lesion of the jaw[12] and the most frequent tumour[13].

Worldwide OKC is more prevalent in males than in females, although, there is an increased Brazilian female predisposition[2]. A common location is in the mandibular angle and ramus; asymmetry occurs occasionally and more likely in the third decade. Clinical symptoms are rarely observed, as it grows painless and silently through medullary bone fenestrating cortical bone and, it is more common in the third decade of life[10,14].

In the presented case no clinical symptoms were referred by the patient.

Three histological parameters have been reported in OKC findings: proliferating odontogenic epithelium, satellite cysts in the wall of suprabasal and mitoses in the lining epithelium. Although they are more frequently associated with multilocular lesions, a corrugated layer with a 6-10 layer of para-keratinized epithelium and a fibrous wall is suggested in the literature[2,15]. Fewer than six cell layer is also suggestive as a histological feature[16].

Authors had been shedding light into the importance of careful histological exam in extensive lesions, in order to prevent a poor diagnosis[8]. Thus, a biopsy is the first step in any extended mandibular lesion. In this specific case, we made use of this diagnostic tool three times during the treatment: one initially (2009), one after the recurrence (2012) and the last one after the enucleation (2013).

Imaging is very important in cases of OKC since they often are incidentally found during the routine radiographic examination[17]. OKC images could present lingual cortical bone perforations getting in contact with the soft tissue, but have neither the tendency to produce root resorptions nor teeth displacements[12].

In this case, the lesion was discovered through pre-orthodontic radiography records, and the bone fenestration could be easily seen through CBCT slices. The continuous use of imaging is mandatory to follow up any recurrence. Previous studies observed cystic lesion shrinkage in pathologies of the jaw and follow up controlled through panoramic and CBCTs[5,7,18-20]. We were able to use digital panoramic and CBCT imaging periodically during the whole case follow-up.

For a while since its first description by Philipsen in 1956 OKC ideal treatment had been controversial[8]. In the past, the literature recommended initial radical treatment since two cases of OKC had been reported with intracranial involvement leading to death in one of the cases[21]. Also, all cysts located in the ramus or into the ascending ramus (the same as in this case) should be treated as "potentially aggressive cysts", and resection had to be performed[12]. Nevertheless, this invasive surgical approach does not prevent a recurrence as long as a rate of 8.4% was shown in a recent meta-analysis[11].

Thus, due to the high morbidity of the resection procedure, clinical guidelines considered it as a last option to be performed only on those cases of multiple recurrences after previous more conservative treatments[22]. Hence lately, the literature has shown agreement into more conservative approaches[9,11].

Six modalities of treatment- among conservative and aggressive surgeries - were analyzed on a systematic review showing relapse rates of 0% for resection, 0% for enucleation + osteotomy + Carnoy's solution, 18.18% for enucleation + osteotomy, 40% for marsupialization and 50% for enucleation + carnoy's Solution and 26.09% for enucleation alone[14].

In addition, the use of decompression before enucleation has been associated with a decrease in the recurrence rate of the lesion since that procedure stimulates fibrotic changes to thicken the cystic membrane diminishing potential membrane tearing during the enucleation process[23,24]. Thus, our surgical conduct by enucleation + osteotomy has been shown as the same rate recurrence as a resection[15,25].

OKC must have a long-term and careful follow-up due to the high recurrence rate[12]. In our case, we have been controlling this case for over eight years.

One of the biggest challenges faced by OMS regarding OKC treatment is to meet the patient's expectations regarding minimising the adverse functional and esthetic side effects while at the same time to fully eliminate the pathology. So, diminishing recurrence with minimal morbidity should be the ideal treatment aiming for success[8]. It comes without saying that individual's self-esteem is affected once aesthetic is compromised.

Having a young and beautiful woman as a patient challenges the surgeon in how to solve the problem with minimal adverse sequels. We present this case because we suggest that literature guidance and patient adherence plays an important role in the decision-making process

by the surgeon, in this case for a more conservative treatment.

A previously decompression of the lesion shown in the literature as low recurrence rate[24] and a systematic review published in 2000[26] supported our approach in using enucleation plus osteotomy. Finally, patients collaboration allowed us been following up the case radiographically annually as suggested by the literature.

## CONCLUSION

Collaboration between orthodontist, pathologist, OMFS, and the patient was crucial for achieving success in this mixture of mini-invasive approaches. These allowed a good quality of life for the patient, recovering of her function with minimal damage to surrounding anatomical structures.

## ACKNOWLEDGEMENTS

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