

CASE REPORT

Big Keratocystic Odontogenic Tumor of the Mandible: A Case Report

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ABSTRACT

BACKGROUND: *Keratocystic odontogenic tumor (KCOT) is a rare, benign, intraosseous tumor of odontogenic origin with a potential of aggressive and infiltrative behavior. It shows specific histopathological features, and has a high recurrence rate.*

CASE DETAILS: *The presented case was of a 30 years old man from South Ethiopia, with a giant keratocystic odontogenic tumor of the mandible.*

CONCLUSIONS: *Although the occurrence of KCOT is rare, attention should be given during its clinical diagnosis. In this report, we presented the aggressive surgical management of a KCOT in a 30 years old patient with no evidence of recurrence within six months of follow-up.*

KEYWORDS: *En bloc resection, Jimma, Keratocystic odontogenic tumor*

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INTRODUCTION

The term odontogenic keratocyst was first described by Philipsen in 1956. Keratocystic odontogenic tumor (KCOT) is a benign uni- or multi-cystic, intra-osseous tumor. It has odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium. KCOT originates as an extension of the basal epithelial cells or the dental organ due to degeneration of the stellate reticulum, or odontogenic epithelial remnants in the mandible or the maxilla. This tumor affects bearing areas of the teeth, and represents 2-11% of all mandibular cysts (1, 2). According to the 2005 World Health Organization classification, KCOT is grouped as an odontogenic tumor due to a number of features which reflect its neoplastic nature. In some cases, KCOT may undergo malignant transformation, i.e., transformation into a squamous cell carcinoma (3).

CASE PRESENTATION

A 30 years old male patient came from the southern part of Ethiopia with referral paper to the Department of Dentistry, St. Paul's Hospital, Addis Ababa, Ethiopia. This Department is one of the few centers which provide oral and maxillofacial surgery services to patients in Ethiopia. The patient had a complaint of progressively enlarging hard mass over the lower jaw for the past 15 years (Figure 1). The patient's case started as a painless swelling which grew slowly to attain the present size. During extra oral examination, a single ovoid swelling of about 16-17cm in size was seen in lower jaw extending from the right angle of mandible to the ramus of left side of the mandible. The skin over the swelling was normal, without visible pulsation or

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secondary changes. Palpation of the swelling was found to be non-tender, non-compressible, non-reducible, and firm in consistency. Intra-orally, there was buccal and lingual expansion extending from the angle of the mandible on the right side to the ramus of the mandible on the left side with obliteration of the alveolobuccal, alveololabial and alveololingual sulcus. Furthermore, the mass was covered by pink, intact and moist mucosa. Only 7 teeth, i.e. 47, 46, 43, 32, 34, 35 and 36 were present on the lower jaw. . Intriguingly, all of these seven teeth were mobile, vital and displaced towards the distal and lingual directions.

Nevertheless, no cervical lymphadenopathy was present.

On the other hand, neuro-sensory testing of the patient revealed normal mandibular nerve function and no other focal neurological deficit. Moreover, the general examination revealed that the patient was well developed, well nourished, but appeared distressed about his possible diagnosis. Based on the clinical findings of the patient, our differential diagnosis included KCOT, ameloblastoma and dentigerous cyst of the mandible.



Figure 1: Pre-operative picture of the patient

After taking history, and physical examination the patient was sent for radiological examination of PA and lateral oblique x-ray views of the mandible. The x-ray results showed multilocular radiolucent cystic-appearing lesion extending from

thr right angle of the mandible to the ramus of the mandible on the left side. In addition, the radiological findings showed a loss of continuity at the inferior border mandible (Figure 2).

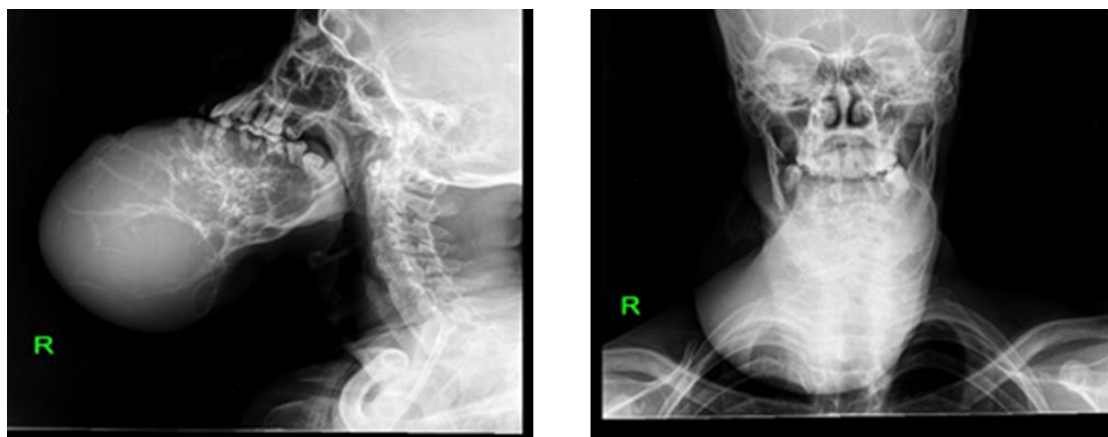


Figure 2: PA and lateral X-ray view of the patient's mandible before operation

In addition to the skull X-ray, other investigations such as complete blood count (CBC), chest X-ray, organ function and eco-tests were investigated, and all these were found to be normal. After careful interpretation of all the investigations, the surgery team decided to do mandiblectomy with decortication. Thereafter, the patient was taken up for surgery under general anesthesia. Consequently, total mandiblectomy was performed from the ramus of the mandible on the right side to total mandiblectomy with decortications on the left side. The specimen that was removed from the mandible contained a hard tissue and multi-cystic cavity. The lesion had a weight of 2.1 kgs and a size of 16x17 cm (Figure 3). The defect of the mandible was reconstructed with k-wire by making three holes on the ramus of the mandible

on the right side, and the k-wire was fixed with wire number 26. Similarly, the left side including the condyle was reconstructed by bending k-wire. Then, the k-wire was stabilized in the mandibular fossa by stitching temporalis and later pterygoid muscles together (Figure 4). Wound was secured with 3-0 vicryl for intraoral mucosa and subcutaneous tissue, and 4-0 black silk for skin. Finally, post-operative antibiotics and analgesics were prescribed. The patient was evaluated continuously for about 6 months both clinically and radiographically (Figure 5), and still the patient being under follow-up, there were no signs of recurrence and nerve deficits.



Figure 3: Resected mandible of the patient

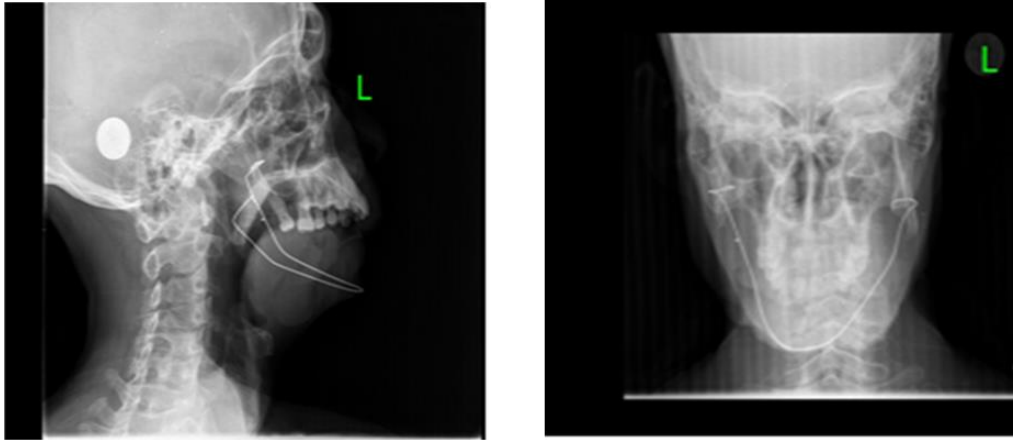


Figure 4: PA and lateral X-ray view of the patient's mandible after reconstruction with k-wire



Figure 5: Picture of the patient six months after operation

The mass that was removed from the patient was sent as excision biopsy for further histopathological investigation. The results showed features of a fibrous cyst wall lined by largely eroded stratified squamous epithelium of parakeratinized type. In addition, the luminal side had proteinaceous cyst content and macrophage. The surrounding cortical and woven bone cuffing had been seen.

Written informed consent was obtained from the patient for the publication of this case report, and for any accompanying images.

DISCUSSION

KCOT is a benign, intraosseous tumor of odontogenic origin. It occurs most commonly in

the mandible, especially in the posterior part of the mandible and ramus regions. KCOT is more common in males than in females, occurs over a wide age range, and is typically diagnosed during the 2nd, 3rd or 4th decades of life. KCOTs have a high recurrence rate, reportedly between 25% and 60%. KCOT is characterized by recurrence due to the presence of remnants of dental lamina, high mitotic activity in basal cell layer, friable capsule and inaccessible sites preventing complete removal, presence of daughter or satellite cysts (4,5,6).

KCOT tends to be found incidentally because it shows no early symptoms, and is painless. It is characterized by growth in the mesial-distal rather than vestibular-lingual direction, and upwards in the mandibular ramus areas, which delays the symptoms of bone distension. Furthermore, tooth resorption is rare while tooth dislocation is more common. Large tumors invade the soft tissue after damaging the compact external bone lamella (7). KCOTs are seldom large-sized; the dimension of our patient's lesion was 16 x 17 cm. Even though radiographic imaging did not provide the pathognomonic identification of KCOT, it is highly supportive for presumptive differential diagnosis, selection of the biopsy site and management decision.

Radiographically, KCOTs usually appear as asymptomatic uni-locular radiolucencies with corticated borders or multilocular radiolucencies, a finding similar to the radiologic result of our case, with a scalloped contour. The consequence on the surrounding structure causes bony expansion, displacement of un-erupted teeth accompanied by pain (4,6,8,9).

Based on the clinical presentations and radiological findings of KCOTs, its differential diagnosis includes dentigerous cyst, lateral periodontal cyst, ameloblastoma, adenomatoid odontogenic tumor, ameloblastic fibroma and central giant cell granuloma. However, histopathological investigation provides the most useful information for its proper diagnosis (3,9). Generally, the histological findings of KCOTs is classified as either orthokeratinized or parakeratinized epithelium. Our specific case showed parakeratinized type which has more recurrence (80%), and more aggressive than orthokeratinized variant (10).

The treatment of KCOT depends on many factors such as the size of the lesion, the age of the patient, the location of the lesion, involvement of soft tissues, history of previous treatment, and histological variant of the lesion. As such, the treatment can broadly be classified as conservative or aggressive. However, the treatment of choice among these two remains controversial (11).

Conservative treatment includes enucleation with or without curettage, or marsupialization. The main advantages of conservative treatment would be preservation of teeth, bone, soft tissue and avoiding damage to the adjacent anatomic structures (inferior alveolar nerve). These are significantly important facts concerning young patients. Moreover, there is a reduction in costs, as hospitalization and reconstruction procedures with the use of either bone grafts or fixation materials become unnecessary (12). In contrast, the disadvantage of conservative treatment is that it requires exhaustive cooperation of patients and parents. Nevertheless, conservative treatment can be associated with incomplete removal of the lesion, since it only decreases the size of the lesion by removing its content. Further complications of this management are malignant transformation and high rate of recurrences (8).

On the other hand, aggressive treatment includes enucleation with peripheral ostectomy, chemical curettage with Carnoy's solution or *en bloc* resection. As acknowledged by many authors, we did *en bloc* resection. However, the incomplete removal of epithelial lining of the tumor is known to play a major role in the recurrence of the case. Thus, *en bloc* resection with 1cm safe margin is

acknowledged to minimize the rate of recurrence and allow early reconstruction of the defect for improvement quality of life of the patient. On the contrary, *en bloc* resection produces a significant morbidity, the loss of the jaw continuity and facial disfigurement (5,13).

KCOT is a developmental, intra-osseous tumor of odontogenic origin. It is potentially aggressive, and is often found incidentally during routine dental examination. Treatment of KCOTs remains a controversial subject. However, aggressive treatment is often used by most surgeons to decrease the rate of recurrences. Late diagnosis and treatment of KCOT could lead to jaw dysfunctions and significant asymmetry of the face which in effect affects the quality of life.

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