**MASSIVE KERATOCYSTICODONTOGENIC TUMOR OF MANDIBLE:**

**A RARE CASE REPORT AND ITS SURGICAL MANAGEMENT**

**Abstract**

Keratocystic odontogenic tumor (KCOT), formerly known as odontogenic keratocyst, is considered a benign unicystic or multicystic intraosseous neoplasm arising from the dental lamina or its remnants and one of the most aggressive odontogenic lesions presenting relatively high recurrence rate and a tendency to invade adjacent tissue. KCOT possesses tumor-like characteristics because of its clinical behavior. In this report, we describe a case of large KCOT which was treated using conservative approach.

**Keywords:** Keratocystic odontogenic tumour, mandible, enucleation, marsupialization

**Introduction**

Keratocystic odontogenic tumor (KCOT) is a benign neoplasm of the maxilla and mandibula that originates from a dental lamina or from the primordial odontogenic epithelium1. KCOTs were first described by Phillipsen,2 who named them “odontogenic keratocysts”. The aggressive clinical behavior and high recurrence rate of KCOTs suggest a real neoplastic potential, and this potential guide the World Health Organization in 2005 to classify them as benign tumors containing an odontogenic epithelium with a mature and fibrous stroma and no odontogenic ectomesenchyme1.

KCOTs are seen in the mandible. (Philipsen 2005, ejasvi et al. 2010, Khanna et al. 2011, Mello et al. 2011) more than maxilla3,4. They have a predilection for males and occur mainly in the second and third decade of life5,6.

KCOTs radiographically appears as a unilocular or multilocular lesions with a scalloped-border. These characteristics are suggestive but not considered as a clearproof for the definitive diagnosis of KCOT because other lesions may reveal similar features7.

Histologic features of the KCOT include an epithelial lining of regular parakeratinized stratified squamous epithelium8.

The treatment methods of KCOTs, varying in different reports, include marsupialization and enucleation, which may be combined with adjuvant therapy, such as cryotherapy or application of Carnoy solution, and marginal or radical resection. The recurrence rates are significantly different in published articles detailing the treatment and prognosis of KCOTs, varying from 12% to 62% dependent on the site of involvement, type of surgical procedure used, and length of follow-up1-3,5-8; most recurrences occur in the first 5 years after surgery9-11.

In this report, clinical, radiographic and histopathological features and treatment prothocols -marsupialization and enucleation with Carnoy’s solution- of a 45-year-old patient with KCOT were described.

**Case Report**

A 45-year-old male patient referred for diagnostic evaluation of the radiolucent area at the left mandible. There was no relevant medical history. Patient gives past dental history of extraction of upper and lower left back tooth for the same pain.

On extra oral examination, normal facial color, contour and texture was present. There was no obvious swelling observed over the left cheek or pre-auricular region. TMJ examination was normal and there was no cervical lymphadenopathy. No obvious swelling or alteration of mucosa over the left side gingiva, buccal mucosa, vestibule and retromolar region was present (Figure 1).

Orthopantomogram revealed well defined radiolucency just below the coronoid process and sigmoid notch involving anterior left ramus and 1/3 of the corpus and upper right first molar was missing (Figure 2). Cone Beam Computed Tomography (CBCT) scan shows well defined, expansile lyticlesion causing cortical thinning and expansion with evidence of internal septation and internal solid content (Figure 3a-c,4a-e,5a,b). The lesion’s volume was 41,657 cm3 (Romexis-Planmeca).

On aspiration with 18 gauge needle achromatic fluid with cheesy consistency was aspirated. Type of aspirated fluid and clinical and radiological findings suggested odontogenic keratocyst of left ramus of mandible. Histopathological examination revealed that, it was confirmed to be a case of KCOT (Figure 6a,b). Treatment was planned as enucleation of the lesion with concomitant application of Carnoy’s solution following marsupialization.

Under local anesthesia patient’s lower left second and third molar teeth was extracted. After extraction the cavity was enlarged to posterior. For keep open the extraction area an appliance was made (Figure 7a). But because of some difficulties the patient couldn’t use. So he did another tiny silicon appliance (Figure 7b,c). With this appliance marsupialization was done approximetly 6 months .During the marsupialization three repeating OPGs (Figure 8a-c) were done which showed, marked reduction in the size of lesion and the risk of damage of inferior alveolar nerve was less. The lesion’s volume was 17,128 cm3 in the control CBCT. Hence, enucleation of the pathology was planned.

The patient was taken into operation under general anesthesia. After anesthesia is achieved incision was given over ramus and mucoperiosteal flap was reflected to expose the pathological site. By protecting the nerve the lesion was enucleated. Carnoy solution was applied. And the opening was closed primarily. After surgery 1gr amoksisilin + clavulanic acid combination, analgesic and oral rinse solution was prescribed to the patient. In the following controls there was no problem and healing was observed.

**Discussion**

KCOT, formerly known as OKC and classified as a neoplasm by the World Health Organization (WHO) in 2005, is a benign odontogenic tumor derived from the dental laminathat requires special surgical consideration becauseof its known aggressive behavior and high tendency to recurn12.

Clinical occurrence of KCOTs mostly seen in man­dible (65–83%) comparing with maxilla12.In our case the occurence of KCOT in the mandible was reported. Present casewas male, which maintains opinion the KCOTs are revealed males more than females5,6.

Radiologically, KCOTs demonstrate a well defined radiolucent area with smooth and often corticated margins and may be unilocular or multilocular. In 25 to 40% of cases, an unerupted tooth is seen in association with the lesion8. Radiographic findings in mandibular lesion showed a unilocular radiolucency in relation with an unerupted canine. In maxilla, a multilocular radiolucent lesion in relation with an unerupted second molar was seen; which had well corticated margins. In this present case the lesion appeared as multilocular radiolucent area.

The epithelium is thin, ranging from 6 to 10 cells thick, and lacks rete pegs, which produce the characteristic flat interface between the epithelium and connective tissue. Separation of the epithelium from the supporting connective tissue of the cyst is common and is caused by metalloproteinase-mediated degradation of collagen in the juxta-epithelial regions8.

Recently, it has been reported that marsupialization can be an effective alternative for begining5.Conventional options for surgery include enucleation and curettage, enucleation and peripheral osteoctomy, enucleation and liquid nitrogen therapy, enucleation and carnoy’s solution, Osseous resection without (ostectomy/ marginal resection) or with (segmental resection) continuity defect3. Some authors described theuse of decompression and secondary enucleation as first line treatment option for KCOT. Furthermore when achieving a significant reduction of the lumen which can be confirmed trough radiographic imaging, a secondary cystectomy is justified to prevent recurrence of the lesion14. In this report our patient treated with marsupialization, enucleation and curettage.

The rate of recurrence varies enormously, from 0% to 62%2,12, and the majority of recurrences occur within the first5 years after treatment13. Most surgeons support complete removal with extension margins or carefulcurettage of the surrounding tissues14,15. Recurrence rates may be influenced by avariety of factors, including the length of the follow-up period, treatment modality, lesion size, histopathological presence of daughter cysts13. In our case recurrence was not found.

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**Figure Legends**

Figure 1

Intraoral photograph reveals normal mucosal appearance.

Figure 2

Orthophantomogram shows well defined radiolucency below the coronoid process and sigmoid notch involving anterior left ramus and 1/3 of the corpus.

Figure 3(a-e)

Axial CBCT scan views show multilocular lesion that begins from left lower canine’s distal surface and reaches left ramus in the posterior. The lesion surrounded by cortical sclerotic bone tissue. It exhibits bucco-lingual expansion and in some region bucco-lingual perforation.

Figure 4(a-e)

Coronal CBCT scan views show multilocular lesion that spreads condylar and coronoid processes and causes bucco-lingual expansion and perforates in some region.

Figure 5(a,b)

3D CBCT scan views reveal large, multilocular and septating lesion makes bucco-lingual expansion and perforation.

Figure 6(a,b)

On histopathologic cross sections there is typical view of odontogenic kerathocysts that has a obvious palisaded basal layer surrounded by thin squamous epithelium.

Figure 7(a-c)

The appliance that made for keeping open the marsupialization fenestration area (Figure 7a). But because of some difficulties the patient couldn’t use and did another tiny silicon appliance (Figure 7b,c).

Figure 8(a-c)

The three repeating OPGs show marked reduction in the size of lesion and the risk of damage of inferior alveolar nerve was less.