INTRODUCTION

The odontogenic keratocyst (OKC) has traveled a long journey from being known as “cyst” until in 2005 and it was designated by the World Health Organization as a keratocystic odontogenic tumor (KCOT). It is thus defined as a benign intraosseous tumor of odontogenic origin, which may be uni- or multicystic, and has a characteristic histopathological feature of parakeratinized stratified squamous epithelium lining with aggressive and infiltrative behavior. Slight male predilection is seen and usually occurs in the second and third decades of life. About three quarters of all KCOT occur in the body of the mandible, the most frequent in the molar region and vertical ramus. The atypical location has also been reported such as in the maxillary antrum, maxillary third molar area, and anterior portion of maxilla. Although among them canine region is the most frequent location reported for KCOT.

Case Presentation

A 30-year-old male patient came to the department of oral pathology and microbiology with chief complaint of pain and discharge in the right upper molar region of jaw. Pain was dull in nature and localized which was aggravating and relieved on medication. There was slight facial asymmetry with evidence of swelling over the cheek extra orally on the right side (Figure 1). The patient gave no significant medical history. Dental history revealed that he had undergone extraction of 16 (upper right 1st molar) couple of weeks back. On intraoral examination, there was a diffuse solitary swelling in the right buccal region extending from 15 to 16 region measuring 1.5 × 2.0 cm. The lesion was smooth and a purulent discharge through extraction socket was noted (Figure 2).

Investigations

An aspiration was done and it yielded a yellowish straw-colored fluid which was consistent with the diagnosis of a cystic lesion. Routine laboratory parameters were normal. Orthopantomogram was planned and it revealed unilocular radiolucency at the apical region of extracted 16 molar region. The border of the lesion was sclerotic and irregular in shape (Figure 3).

Provisional Diagnosis

The lesion was suspected primarily to be a radicular cyst based on the clinical and radiographic presentation of the lesion. Apart from that KCOT was also suspected. However, KCOT was diagnosed only after histopathological examination.

Treatment/Enucleation

In collaboration with oral and maxillofacial surgeon excisional biopsy was planned. The lesion was completely
enucleated with Caldwell Luc approach (Figure 4). The lining was found to be fragile in most places and a thorough curettage was carried out. Post-curettage the bony margins were trimmed with a large bone bur initiating bony bleeding. Areas of copious bleeding were controlled. No involvement of the maxillary sinus was noted and the operative site was closed. The patient is currently under post-operative periodic follow-up and no recurrence is observed until date.

**Histopathological Examination**
Microscopic features of enucleated cyst lining showed hyperplastic keratinized cystic lining with prominent palisading basal cell layer and nuclear hyperchromasia. Predominantly a remarkable uniformity of thickness of lining epithelium was noticed with palisaded, polarized basal layer of cells exhibiting tombstone appearance was appreciated. Connective tissue was found to be fibro-cellular with loose collagen fibers, spindle-shaped fibroblasts, infiltration of chronic inflammatory cells predominantly lymphocytes, and plasma cells. All these histopathological features were strongly suggestive of KCOT (Figure 5).

**DISCUSSION**
Philipsen first used the term “OKC” in 1956, while its important features were described by Pindborg and Hansen in 1963. The diagnostic metamorphosis of OKC into a recognized cystic neoplasm occurred after scrutinizing its biological behavior and the alliance of chromosomal and genetic abnormalities consistent with neoplastic sequence. KCOT is unique among odontogenic tumors because of its pathognomonic microscopic features, belligerent behavior,
and high recurrence rate. KCOT tends to grow in the anteroposterior direction within the medullary cavity of the bone without noticeable bone expansion causing its delayed observation by the patients. However, in maxilla, it has been reported to cause cortical plate expansion. Jackson et al. in 1993 reported a case of KCOT which extended from the maxilla and eventually involved the base of the skull, behaving like squamous cell carcinoma. Other authors have also reported about aggressive behavior of KCOT in maxilla, extending from the maxilla into the infratemporal fossa. Its aggressive character in maxilla may thus lead to destruction of the floor of orbit and proptosis of eyeball.

Some mechanisms have been suggested to elucidate its behavior and high tendency for recurrence such as the high proliferation of its cyst lining, difficulty to remove it in one piece due to its thin friable lining, small daughter cysts, and production of bone resorptive factors.

Several proliferative and apoptotic markers such as p53, Ki-67, and bcl-2 were used in many studies and KCOT epithelial lining showed an amplified expression of these markers as compared to other odontogenic cysts. However, the diagnosis of KCOT is primarily based on the histopathological features. The radiographic findings, although often highly indicative, are not diagnostic as KCOT may imitate number of radiolucent lesions such as radicular cyst, residual cyst, lateral periodontal cyst, and KCOT of the anterior midline maxillary region can also mimic nasopalatine duct cysts. Treatment protocol of KCOT has ranged from marsupialization and enucleation to en bloc resection with each giving success rates. Carnoy’s solution and cryosurgery have also been advocated as it kills epithelial remnants and dental lamina in the osseous margin. Carnoy’s solution being a tissue penetrates bone to a depth of 1.54 mm. There are no clear-cut guidelines regarding treatment plan of KCOT, however, the primary goal of the treatment is minimal recurrence of the tumor and it depends entirely on oral maxillofacial surgeon to which protocol he adopts, depending on his personal experience.

**CONCLUSION**

The re-classification of KCOT thus finally solved the controversies regarding its aggressive behavior and affinity toward recurrence. This is the main difference between KCOT and other jaw cysts. KCOT in maxilla is more aggressive than their mandibular counterparts and thus should be treated aggressively and the correlation between clinical and histopathological findings remains the gold standard in the diagnosis of KCOT.

**REFERENCES**


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