 DOI: 10.18231/2455-6750.2017.0015

Large Keratocystic Odontogenic Tumor involving entire maxillary sinus: A common pathology in uncommon location

Sanika Kulkarni[1,*], Kumar Nilesh[2], Pankaj Patil[3], MI Parkar[4], Kamla KA[5]

[1]PG Student, [2]Reader, [3]Senior Lecturer, [4]Professor & HOD, [5]Associate Professor, Dept. of Oral & Maxillofacial Surgery, School of Dental Sciences, KIMSDU

***Corresponding Author:**
Email: ksanikag@gmail.com

Abstract

Keratocystic odontogenic tumor (KOT) is an odontogenic neoplasm of jaw which occurs more frequently in mandible. Its occurrence in maxilla is relatively infrequent. This article reports a case of large KOT of left maxilla involving the entire maxillary sinus extending up to the infra-orbital rim, in a middle aged person. The pathology was treated with surgical enucleation and chemical cauterization. Taking into the consideration the high recurrence rate of KOT, regular and periodic follow up is necessary in such cases. At 2 years follow up the present case showed normal healing with no recurrence.

Keywords: Odontogenic keratocyst, Jaw, Tumor, Maxillary Sinus

Introduction

Keratocystic Odontogenic Tumor (KOT) was previously known as Odontogenic Keratocyst.(1) It is a benign but locally aggressive pathology of odontogenic origin. In 2005, the World Health Organization renamed the lesion based on its clinical, histological and immunochemical parameters and classified it as tumor.(2)

KCOT presents as unicystic or multicystic intraosseous lesion. It mostly occurs in 2nd or 3rd decade of life and is commonly seen in mandibular ramus region.(2) KCOT differ from other odontogenic cysts in that they have biologically aggressive behaviour because of high proliferative activity of the lining epithelium, a tendency to expand in bony cancellous spaces and considerable high rate of recurrence. KCOT has created controversies with regards to its true nature. It has clinicopathological features of both simple cyst and benign neoplasm.(3)

Generally, KCOT of maxilla do not present any characteristic features. Though these lesions grow and reach large size, they are many times asymptomatic. The most common clinical presentation of the tumor is a localized asymptomatic swelling. There might be presence of pain and spontaneous drainage into the oral cavity in cases of infected cyst. Mobility and displacement of teeth can be noticed in aggressive lesions. Radiographically, they appears as unilocular or multilocular radiolucency with smooth and usually sclerotic margins.(5)

This article reports a case of KCOT involving entire hemi-maxilla and left maxillary sinus, extending up to the infraorbital region.

Case Report

A 42 years old male patient reported to Department of Oral and Maxillofacial Surgery with a complaint of painless swelling over the left side of face since past 2 months. There was mild facial asymmetry with diffuse swelling over left side of midface region. The swelling extended from below the lower eyelid region to ala-tragus line superoinferiorly and from malar prominence to the lateral part of nose mediolaterally (Fig. 1a). The swelling was non-tender and the overlying skin appeared normal. There was no local rise in temperature. Intraoral examination showed obliteration of buccal vestibule starting from 23 region to 28 region (Fig. 1b) All the associated teeth were non carious and vital with grade 1 mobility noticed with 25, 26, 27, 28. Based on these clinical features the provisional diagnosis made was cyst of maxillary sinus. Orthopantomogram showed diffuse radiolucency in left maxillary sinus (Fig. 2a). The paranasal sinus view showed haziness in the left maxillary sinus with loss of normal radiographic anatomy of maxilla (Fig. 2b). The differential diagnosis made was keratocystic odontogenic tumor, adenomatoid odontogenic tumor, ameloblastoma and central giant cell tumor. Under local anaesthesia incisional biopsy was carried out and the specimen was sent for histopathological examination. The haematoxylin and eosin stained section revealed, cystic lining showing corrugated, parakeratinized stratified squamous epithelium of 6 to 8 cell layer thick without rete ridges. The basal cell layer showed tall columnar cells with palisaded appearance, reversal polarity and nuclear hypochromatism. The underlying connective tissue revealed numerous collagen fibers, fibroblasts, sparse chronic inflammatory cell infiltrate, mainly lymphocytes. Odontogenic islands were also noticed at few places. Based upon these histological findings final diagnosis of keratocystic odontogenic tumor was made. Computed Tomography (CT) with three dimensional reconstruction was done to study the extent of the lesion. CT revealed intraosseous expansile lesion with loss of buccal cortical plate in the 23-25 region extending up to the left infraorbital rim.

Anteroposteriorly it extended from pyriform aperture on left side to the left zygomaticomaxillary suture. Root tips of involved teeth were present within the cystic cavity. Coronal section of CT scan revealed loss in the normal morphology of left side of maxilla with osteolytic lesion involving entire left maxilla (Fig. 3a, b, c). Taking into consideration the extent of lesion the treatment plan included enucleation and curettage followed by chemical treatment using carnoy's solution under general anaesthesia.

Fig. 1: Clinical photograph showing extraoral swelling (a), intraoral view showing vestibular swelling (b)

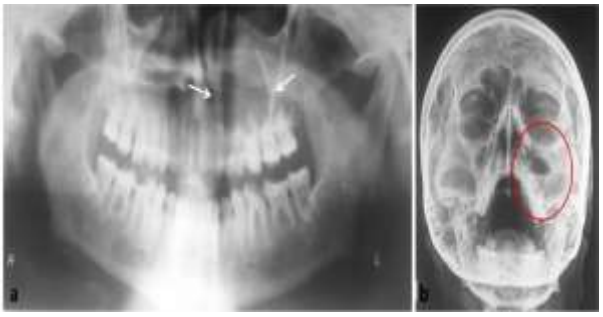

Fig. 2: Radiographic appearance of lesion on Orthopantomogram (a) and paranasal sinus view (b) showing radiolucency involving left maxillary sinus

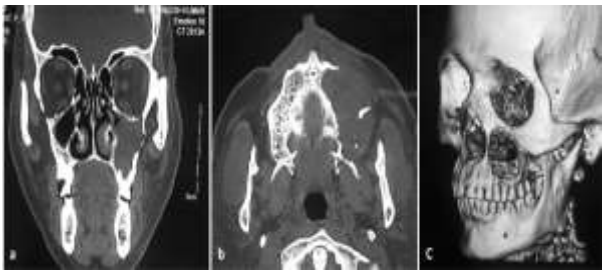

Fig. 3: Computed Tomography showing an osteolytic lesion involving entire maxillary sinus (a), with resorption of anterior wall of maxilla (b), formatted 3D image of lesion

The lesion was approached intraorally. Crevicular incision was given extending from 23 to 28 region with anterior and posterior releasing incisions given at 45º angle. Mucoperiosteal flap was reflected. The lining was found to be fragile and was removed in to with extraction of all involving teeth (Fig. 4a). Cystic lining was sent from histopathological examination. Chemical cauterization was done by keeping carnoy's solution soaked cotton inside the cystic cavity for three minutes (Fig. 4b). Primary closure was attained (Fig. 4c, d). Patient was kept on regular follow up. At two years' post op follow up healing was within normal limits with no sign of recurrence (Fig. 6a, b).

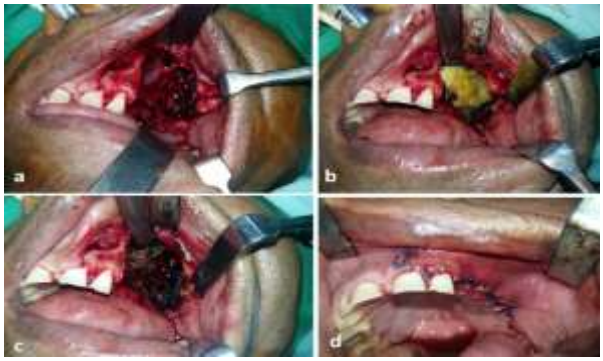

Fig. 4: Intraoperative images showing, enucleation of pathology (a), chemical cauterization of cyst defect (b), surgical site after cauterization (c) and primary closure

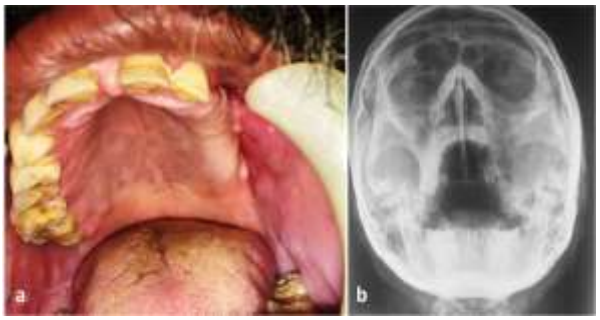

Fig. 5: Two year postoperative clinical (a) and radiological image (b) showing normal healing with no evidence of recurrence

Discussion

KOT is a benign but locally aggressive developmental odontogenic cyst. It has wide variety of clinical appearances. Among the jaw cysts KCOT is the third most common.[4] It occurs most commonly in the mandibular third molar and ramus region, followed by anterior maxilla, maxillary third molar area, mandibular anterior and mandibular premolar areas.[7] Overall maxillary involvement has been observed in ranges from 16.4% to 23.5% and only one percent of the lesions occur in the maxilla with sinus involvement.[8]

KOTs of maxilla are difficult to diagnose due to its lack of specific clinical and radiographic characteristics. In maxilla occurrence of KOT is relatively rare and the maxillary sinus invasion is unusual. Clinically they are asymptomatic but can present as toothache, swelling and bone destruction.

KCOT involving the maxillary sinus must be carefully assessed. The risk of orbital and nasal involvement and spread of associated infection from infected cyst can lead to local and systemic manifestations in the patient. Displacement of tooth and destruction of maxillary sinus, orbital floor, and lateral nasal wall can be seen in cases of maxillary KCOT. The present case showed diffuse extraoral, non-tender swelling over left side of midface. Intraorally the only finding was vestibular obliteration due to gingival swelling extending from 23 to 28 region.

As KCOT in the maxillary sinus is rare, its radiographic image in such situation may be misinterpreted. KCOT are poor bone resorbers and it involves only the cancellous bone in its early stages of development. A very little expansion is observed because compact bone is invaded late. Scalloped margins due to regional resorption, are seen in long standing cystic lesions.[9] On the CT scan, the lesion may appear as a large, well-defined, expansile, thin-walled cystic lesion extending cranially into the maxillary sinus. In cases of maxillary sinus involvement, an apparent radiopaque alteration may be found, differing from the classic description for this lesion. The radiopaque aspect is probably due to increased density of the cystic content in relation to the usual air filling of the sinus. Destruction of sinus floor and walls can also be seen in the sinus differently.[7] Present case showed haziness of left maxillary sinus. There was evidence of loss of buccal cortical plate with destruction of normal anatomy of left maxilla and presence of root apices of involved teeth inside the cyst cavity.

The treatment modalities for KCOT can be conservative or aggressive. Conventional management includes simple enucleation, with or without the curettage, or marsupialization. Aggressive management of KCOT commonly include treatment modalities such as peripheral ostectomy, chemical curettage with carnoy's solution, cryotherapy, or electrocautery and resection.[10] Various factors influence the choice of treatment in case of maxillary KCOT; affected individual's age, overall size and site of the cyst, involvement of soft tissue, record regarding prior management as well as a histological variation of the disease. The actual treatment objective should be to select a treatment method that has the minimum chances of recurrence and the slightest morbidity.[6] Hence in this case enucleation and curettage followed by chemical cauterization using carnoy's solution was done.

KOT frequently recur after enucleation, particularly within first 5 years.[5] Although, there is immense decrease in recurrence rate in case of complete enucleation of the keratocyst including the associated tooth.[5] The recurrence rate of only enucleation is 7.8% while that in marsupialisation is 33.3%.[11] Enucleation with adjunctive therapy such as cryosurgery and chemical cauterization has 1-8% recurrence rate.[9] Resection can have high morbidity. Postoperative follow-up with regular radiographic examination is important with KCOT because of the high potential for recurrence.

Conclusion

KOT is common odontogenic tumor of jaw bone. However large maxillary lesions involving the entire maxillary sinus are infrequent. Knowledge of clinical and radiological fracture of KCOT in its unusual location helps in early diagnosis and management, thereby reducing the post-operative morbidity and complications.

References

1. Pogrel MA. The keratocystic odontogenic tumor. Oral Maxillofac Surg Clin North Am. 2013 Feb 28;25(1):21-30.
2. Nagraja A, Anigol PS, Kamath W, Setlur KP. Keratocystic Odontogenic Tumor of Maxilla: Report of a Rare Case and review of Literature. World J Dent 2010;3(1):100-108.
3. Chirapathomsakul D, Sastravaha P, Jansisyanont P. A review of odontogenic keratocysts and the behavior of recurrences. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2006 Jan 31;101(1):5-9.
4. Gupta A, Rai B, Nair MA, Bhut MK. Keratocystic odontogenic tumor with impacted maxillary third molar involving the right maxillary antrum: An unusual case report. Indian J. Dent. Res. 2011 Jan 1;22(1):157.
5. Molon RS, Verzola MH, Pires LC et al. Five years follow-up of a keratocyst odontogenic tumor treated by marsupialization and enucleation: A case report and literature review. Contemp Clin Dent. 2015 Mar 1;6(5):106.
6. Mahadesh J, Kokila LB, Laxmidevi BL. Odontogenic keratocyst of maxilla involving the sinus-OKC to be a cyst or a tumour. J Dent Sci Res. 2010 Sep;1:83-90.
7. Myoung H, Hong SP, Hong SD et al. Odontogenic keratocyst: Review of 256 cases for recurrence and clinicopathologic parameters. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2001,91(3):328-33.
8. Brannon RB. The odontogenic keratocyst: a clinicopathologic study of 312 cases. Part I. Clinical features. . Oral Surg Oral Med Oral Pathol. 1976 Jul 1;42(1):54-72.
9. Ali M, Baughman RA. Maxillary odontogenic keratocyst: a common and serious clinical misdiagnosis. Journal Am Dent Assoc. 2003 Jul 31;134(7):877-83.
10. Singh M, Gupta KC. Surgical treatment of odontogenic keratocyst by enucleation. Cont Clin Dent. 2010 Oct 1;1(4):263.
11. Zhao Y, Liu B et al. Recurrent keratocystic odontogenic tumours: report of 19 cases. Dentomaxillofac Radiol. 2012;41(2):96-102.