Case Report

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Keratocystic Odontogenic Tumor of Mandible with Reconstruction Using Free Fibula Vascular Graft

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ABSTRACT

Background: Odontogenic kertocyst is one of the very well know form of odontogenic cyst.What makes the odontogenic keratocyst uncommon from other forms of cysts (of jaw) is its histopathological character and clinical features i.e. aggressive behavior, very high recurrence rate and an association with nevoid basal cell carcinoma syndrome. The characteristic histologic feature i.e. the presence of parakeratin, is unique amongst all the different inflammatory and developmental cysts that occur in the jaws. Many treatment modalities have been advocated for its treatment, but some controversies still remain. The 2005 WHO classification now uses the term 'keratocystic odontogenic tumor'. We present a case reportof the KCOT followed by reconstruction using free fibula vascular graft.

Keywords: Dentistry, odontogenic kerotocyst, developmental cyst.

INTRODUCTION

Philipsen in 1956 first described a cyst of the jaws lined by keratinizing epithelium which was known as odontogenic keratocyst,[1] surgeons have been researching to find an ideal treatment for it. The knowledge regarding the treatment of the odontogenic keratocyst, now renamed as the Keratocystic Odontogenic Tumor (KCOT), has been ever increasing over the past few decades, and yet, the issue is still a debate in oral and maxillofacial surgery. Different treatment modalities have been tried for the successful treatment of the KCOT, ranging from simple enucleation to resection, but none has been regarded as the ideal treatment. The KCOT behaves like a tumor in many ways, e.g. involvement of large areas of the bone, high recurrence rate, distinctive histopathological features of the lesion, dysregulation of the PTCH (patched) gene in both Nevoid basal cell carcinoma syndrome associated and sporadic odontogenic keratocysts etc. On the other hand, successful treatment by marsupialization denies its tumor characteristics. Truly, many cases of carcinoma arising in KCOT have been reported. In fact, recurrence of the lesion has been reported even in a bone graft. In this article, we present a case report of KCOT with reconstruction using free fibula vascular graft.

CASE REPORT

A 17-year-old female was admitted in oral and maxillofacial surgery department of Rama dental college,Kanpur, with chief complaint of swelling over left side of lower jaw since three months which was gradually increasing in size and painless. The swelling was initially small and gradually increased. There was no associated pain, paresthesia or discomfort. Extra oral examination revealed mild swelling in the mandibular left angle-ramus region. The swelling was non tender and the

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overlying skin appeared normal with no local rise of temperature. The temporomandibular joint showed normal movements and absence of tenderness and clicking. Intraoral examination showed expansion of left buccal cortical plate and obliteration of vestibule in region of 34-37 with thinning and 'Egg Shell Crackling' over the cortical plates. The associated teeth were non-carious and did not show mobility. OPG showed a well-defined, corticated multilocular radiolucent lesion in the left posterior mandible, extending anterio-posteriorily from the apical region of 33 to posterior border of ramus of mandible and superior-inferiorily from coronoid and condylar processes to lower border of mandible. Based on the clinical presentation and the radiographic findings provisional diagnosis of keratocystic odontogenic tumor was made. Differential diagnosis included ameloblastomna, odontogenic myxoma, central odontogenic fibroma and central giant cell granuloma. Aspiration of the lesion as done under local anesthesia, which revealed thick white cheesy viscoid fluid with keratin flakes. Cytochemical evaluation showed soluble protein content of 3.4md/dl. Computed tomography (CT) was done to study exact extent of the lesion for pre-surgical planning. CT revealed intraosseous expansile lesion involving left mandibular body and ramus with buccal cortical perforation distal to the mental foramen and lingually at the mylohyoid ridge. Cortical erosions at the angle of the mandible and neck of the condyle were also seen.

Treatment plan included resection of involved segment of mandible with disarticulation, reconstruction of the hemimandibulectomy defect with vascular free fibula graft. Under general anesthesia submandibular incision was used to approach the tumor. Subplatysmal dissection was done to reach the submandibular space. In the submandibular space Facial artery and vein were identified, divided, and secured with vascular clamps. Mandible was freed from all the attachments and osteotomy cuts were marked anterior to the mental foramen. Osteotomy cut was completed buccally and lingually. Osteotomized mandible segment was rotated laterally and inferior alveolar neurovascular bundle was identified and ligated. The left hemimandible was disarticulated and the specimen was excised. After resection of the mandible the soft tissue areas adjoining buccal and lingual perforations were electrocauterized to remove any possible residual pathology in situ. A fibula graft was harvested from limb of the patient after measuring the length of the defect. The graft was shaped as per the shape of patient's mandible. It was then fixated with titanium mini plates and then to the patient's remaining mandible with screws. Wound was closed in layers. Postoperative recovery was uneventful.

Histopathologic study of the excised specimen confirmed the diagnosis of KCOT and the anterior margin of the resected specimen was free of tumor. Patient was kept on regular follow up. On one year follow up, patient's face was symmetrical with 30 mm mouth opening. Occlusion was stable with normal mandibular movements with slight deviation of to left side on opening.

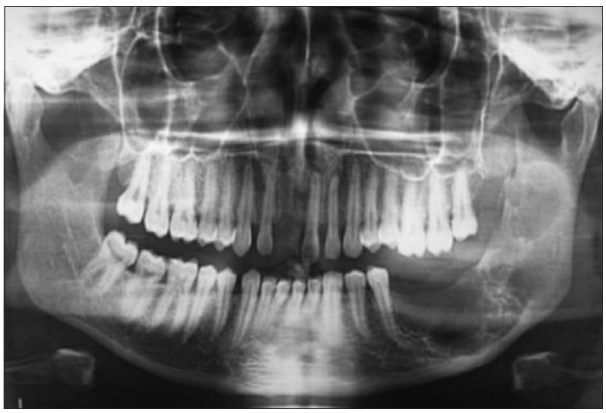

Fig 1: OPG

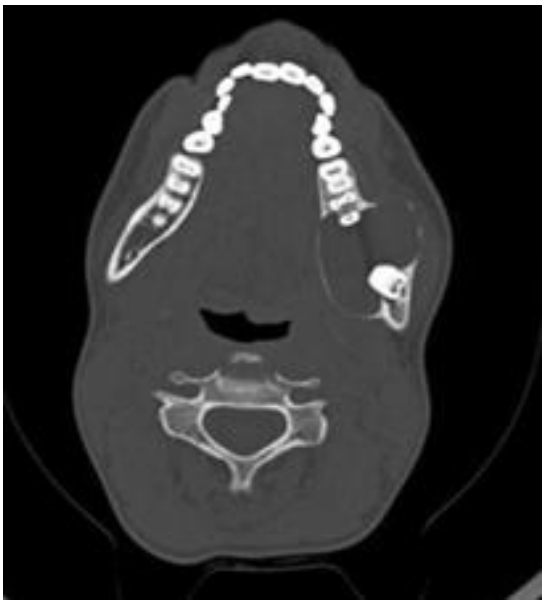

Fig 2: CT Scan

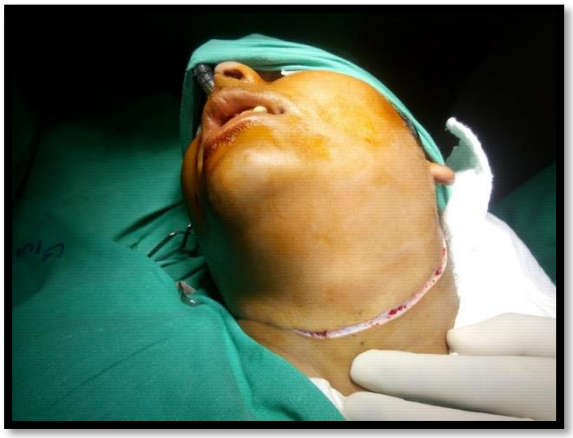

Fig 3: Incision line.

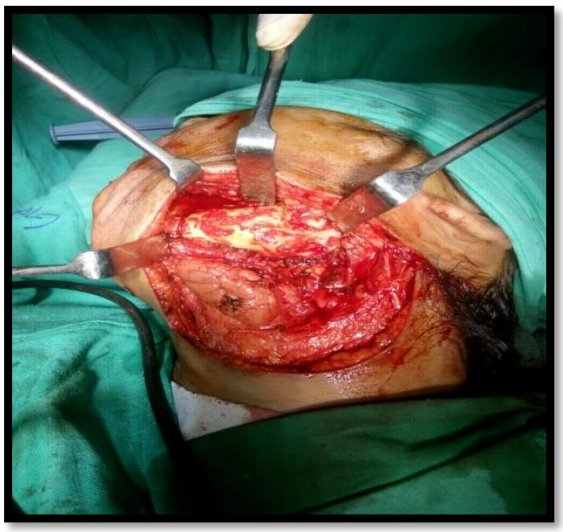

Fig 4: Exposre of Mandible

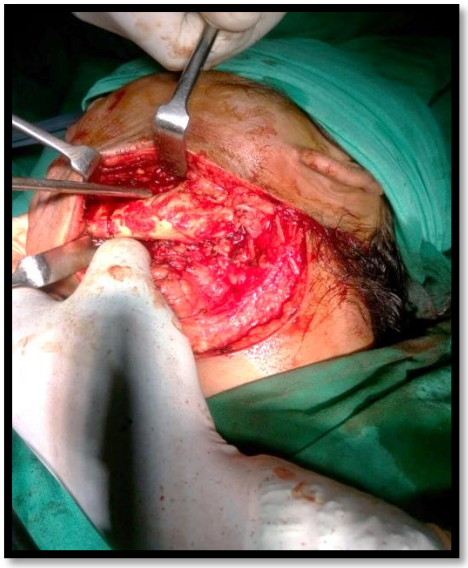

Fig 5: Osteotomy.

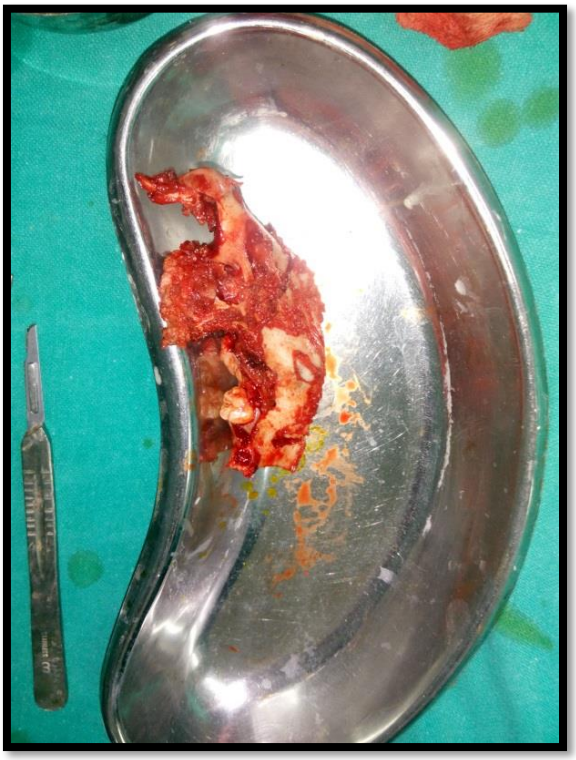

Fig 6: Primary lesion.

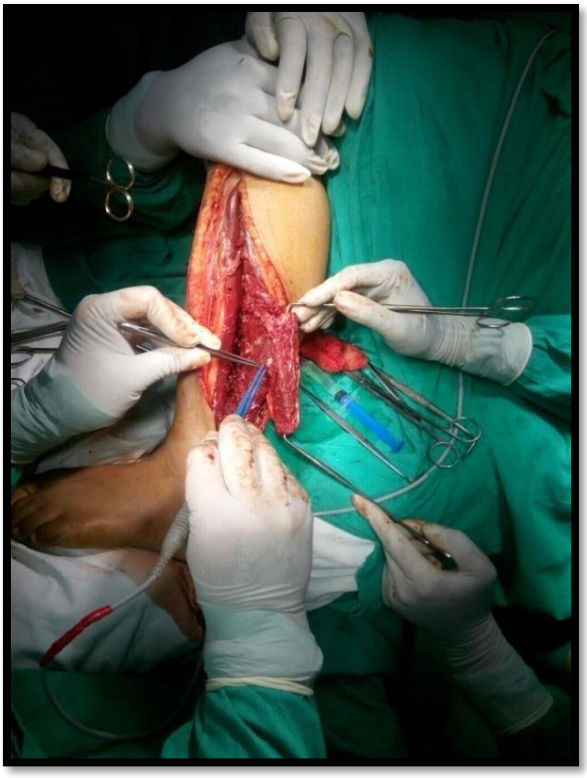

Fig 7: Reflection of fibula.

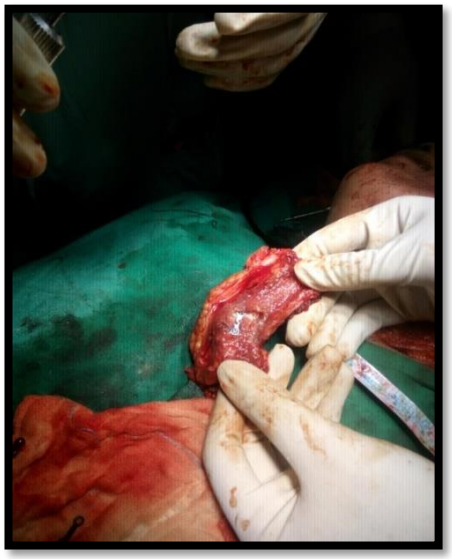

Fig 8: Free fibula graft.

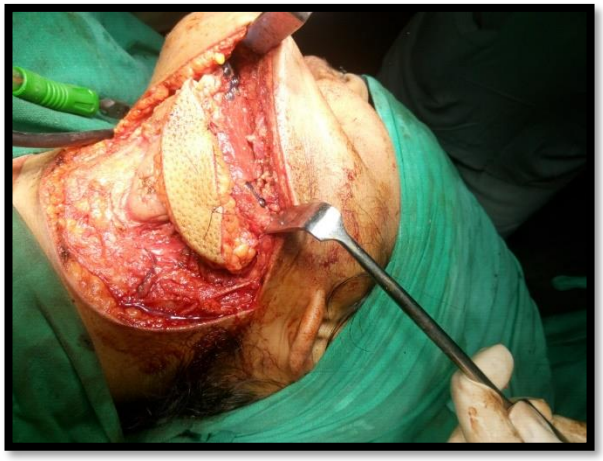

Fig 9: Recoturing and stablized with titanium plate and screw.

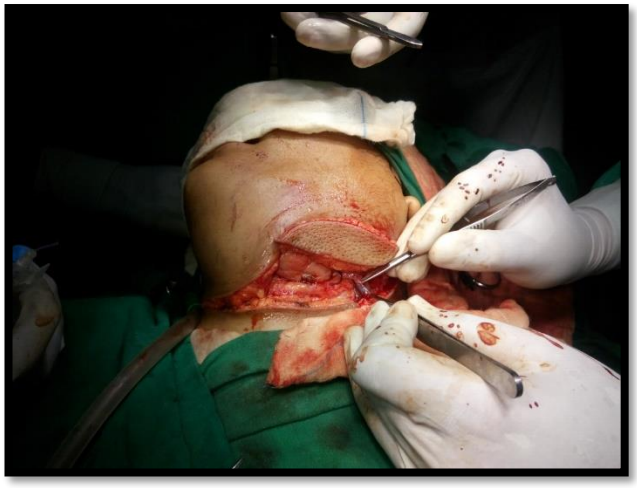

Fig 10: Closure in layers.

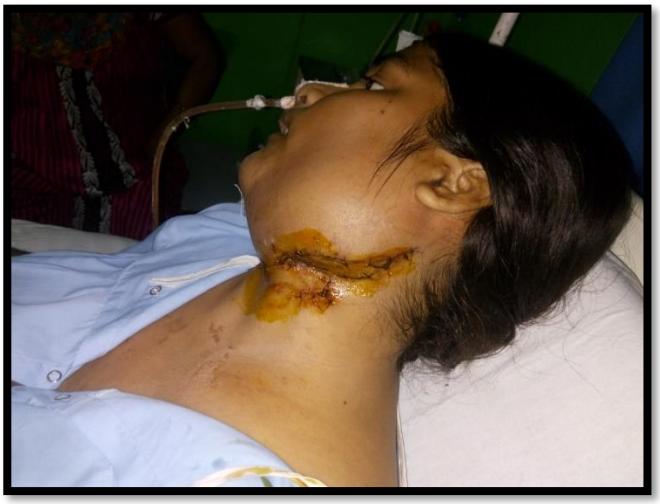

Fig 11: 1ST Post operative day.

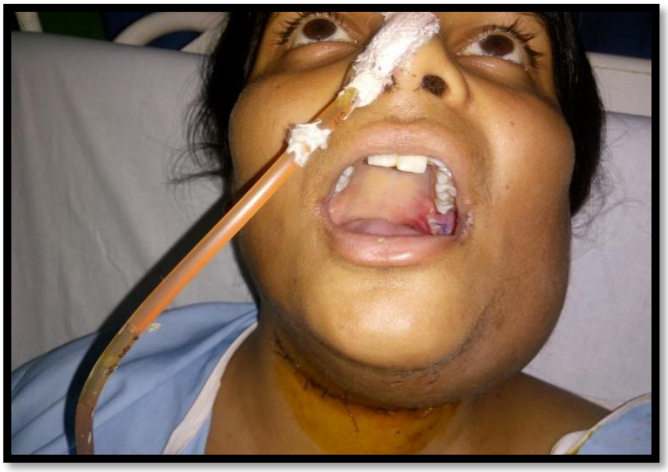

Fig 12: 5TH Post operative day

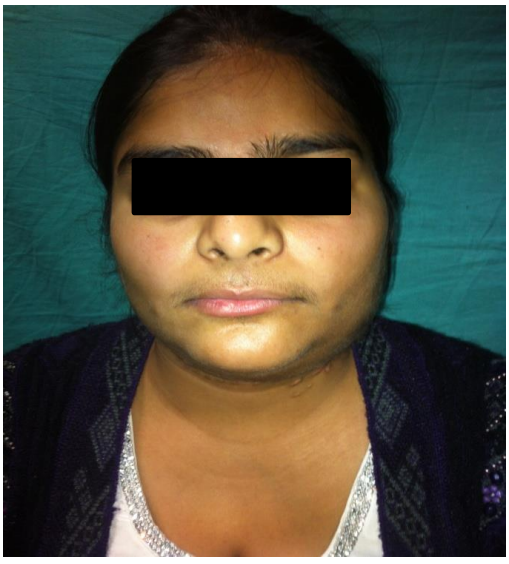

Fig 13: After 3 month note the symmetry of face.

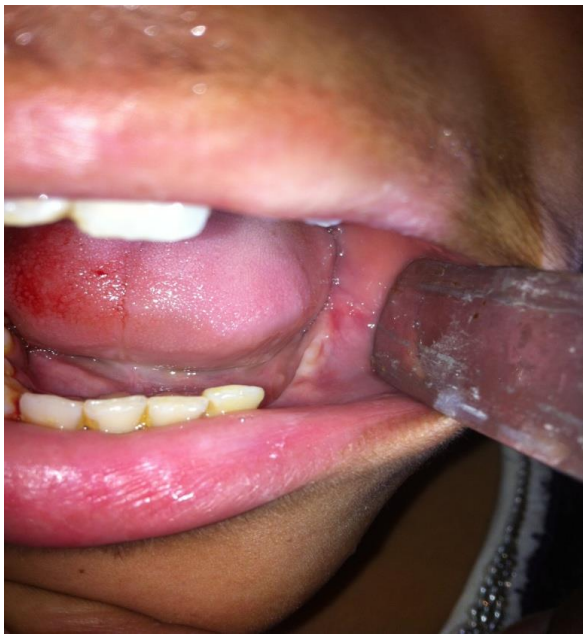

Fig 14: After 5 month intraoral view

DISCUSSION

WHO in 2005 reclassified Para keratinized variant of odontogenic keratocyst as an odontogenic tumor and the lesion was renamed as keratocystic odontogenic tumor (KCOT). Epithelial lining and connective tissue stroma of KCOT shows tumor like characteristics. The epithelial lining shows evidence of high mitotic activity with high turnover rate. There is also expression of various proliferation markers in the epithelium. The connective tissue of KCOT shows features of a tumoral stroma. There is high frequency of stromal myofibroblasts with presence of high enzymatic activity. Increased matrix metalloproteinases, mast cell tryptase and increased expression of receptor activator of nuclear factor and osteoprotegerin contribute to the aggressive tumor like nature of the lesion[3]. The over expression of p53 protein and mutations in p53 and PTCH genes (tumor suppressor genes) also justify tumor like behaviour of KCOT.[2-4] Owing to its tumor like behavior with high recurrence rate these lesions needs to be managed aggressively and followed up regularly. Reconstruction of the residual defect and dental rehabilitation helps in comprehensive management of the patient.

CONCLUSION

Treatment of KCOT should be meticulously planned. Choice of treatment should be based on multiple factors like patient age, size and location of the tumor, soft tissue involvement and history of previous treatment. The treatment modality that carries the lowest risk of recurrence and the least morbidity should be chosen depending on the case. Reconstruction of the surgical defect and prosthetic replacement of the missing teeth in postoperative period provides comprehensive functional and aesthetic rehabilitation of the patient. Long term follow-up is always advocated to rule out recurrence.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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