Comprehensive Management of Keratocystic Odontogenic Tumor of the Mandible: A Case Report

Ashwini Samant[1], Kumar Nilesh[2], M I Parkar[3], Shivsagar Tewary[4], Pronob Sanyal[5]

ABSTRACT

Introduction: Keratocystic Odontogenic tumor (KCOT) is a odontogenic neoplasm of jaw which has high recurrence potential.

Case Report: This paper reports a case of KCOT in left hemimandible with buccal and lingual cortical perforations. The case was treated by segmental resection with disarticulation and reconstructed with avascular fibula graft. Postoperative prosthetic rehabilitation was done with cast partial denture.

Conclusion: For complete management of KCOT involving hemimandible, a treatment plan of resection and reconstruction with long term follow up should be carried out. Also the patient's deformity should be prosthetically rehabilitated to give him a functionally and esthetically acceptable dentition.

Keywords: Odontogenic Tumor, Surgical treatment, Reconstruction, Fibula graft, Hemimandibulectomy.

INTRODUCTION

Keratocystic odontogenic tumor (KCOT), previously known as a cystic lesion was renamed as an odontogenic jaw tumor in 2005 by WHO.[1,2]

KCOT is clinically benign but locally aggressive lesion with a slight male predilection and commonly occurs in the second and third decades of life. KCOTs can occur in any part of the jaw, but majority of the lesions are seen in the mandible, most commonly in the posterior body and ascending ramus. It is considered to be an aggressive odontogenic tumor, owing to its high recurrence rate and tendency to invade adjacent tissue. The treatment modalities of KCOT's, vary from conservative procedure like marsupilization and enucleation, with or without adjuvant physicochemical therapy (such as cryotherapy, application of cornoy's solution) to more aggressive marginal or segmental resection of jaw. The recurrence rates are significantly high and differ in various published articles, varying from 0% to 62%, depending on the site of involvement, type of surgical procedure used, and length of follow-up. Most recurrences are seen in the first 5 years after surgery.[1,3]

In the present report, we describe a case of large KCOT originating in the left mandibular body and ramus region with buccal and lingual cortical perforations. The tumor was managed by segmental resection of mandible. Reconstruction was done using autogenous avascular fibula graft and prosthetic rehabilitation was done using cast partial denture.

CASE REPORT

A 34-year-old male patient reported to the Department of Oral and Maxillofacial Surgery, School of dental sciences, Krishna Hospital, Karad, in September 2014 with a chief complaint of swelling over lower left jaw for the past 3 months. The swelling was initially small and gradually increased to the present size. There was no associated pain, paresthesia or discomfort. Extraoral examination revealed mild swelling in the mandibular left angle-ramus region (figure 1a). The swelling was non tender and the 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 (figure 1b). The associated teeth were non-carious and did not show mobility. Electric pulp vitality test revealed positive vitality for all the left mandibular posterior teeth. Orthopantamogram showed a well defined, corticated multilocular radiolucent lesion in the left posterior mandible, extending anterio-posteriorily from the apical region of 35 to posterior border of ramus of mandible and superior-inferiorily from coronoid and condylar processes to lower border of mandible (figure 1c). 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 of the aspirate showed soluble protein content of 3.4md/dl. Incision biopsy was performed and a portion of the lesion was submitted for histopathological examination. Hematoxylin and eosin (H and E) stained section showed 6 to 8 cells thick parakeratinized stratified squamous epithelium with surface cell keratinization, hyperchomatic tall columnar basal cells with palisading and reversal of nuclear polarity. Separation of basement membrane from connective tissue at some places was appreciated. Based on the histological findings diagnosis of keratocystic odontogenic tumor was made (figure 2a, b). 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 men-

[1]Post-Graduate Student, [2]Reader, [3]Professor and H.O.D., Department of Oral and Maxillofacial Surgery, [4]Reader, [5]Professor and H.O.D., Department of Prosthodontics, School of Dental Sciences, KIMSDU, Karad, India

Corresponding author: Dr. Ashwini Samant, Department of Oral and Maxillofacial Surgery, School of Dental Sciences, Krishna Hospital, Karad, Satara 415110, Maharashtra, India

How to cite this article: Ashwini Samant, Kumar Nilesh, M I Parkar, Shivsagar Tewary, Pronob Sanyal. Comprehensive management of keratocystic odontogenic tumor of the mandible: a case report. International Journal of Contemporary Medical Research 2016;3(5):1258-1261.

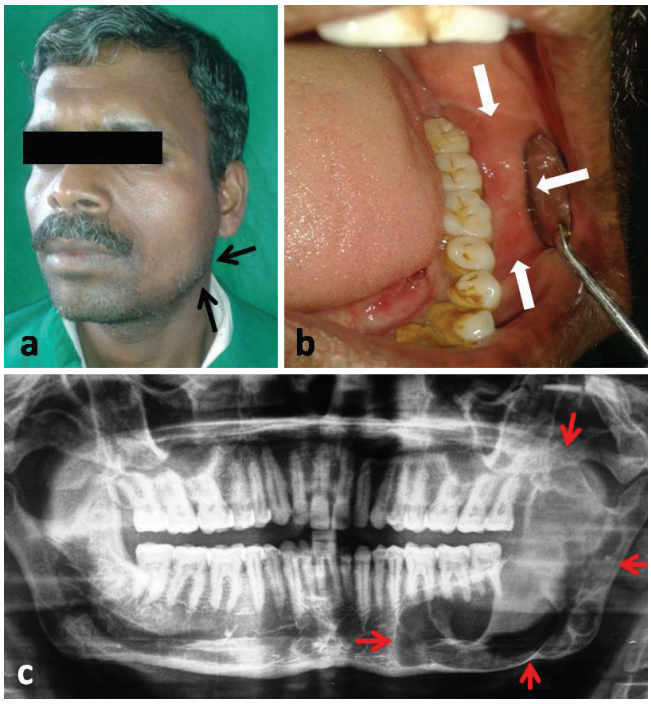

Figure-1: Clinical presentation of lesion; (a) diffuse swelling over left mandibular angle and ramus region, (b) intraoral view showing expansion of buccal cortical plate and obliteration of buccal vestibule**,** (c) Orthopantamogram showing well defined, corticated multilocular radioleucency in left mandibular body, angle and ramus region**.**

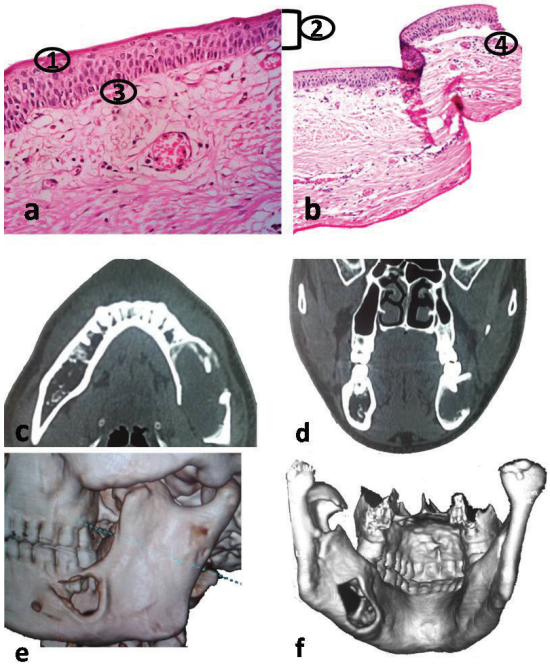

Figure-2: H and E stained section (10X magnification) showing; (a) cyst lining with surface cell keratinization(1), 6-8 layer thick parakeratinised stratified squamous epithelium(2), hyperchromatic tall columnar basal cells with palisading nuclei and reversal of polarity [Picket fence/Tombstone appearance](3); (b) underlying connective tissue showing separation from epithelium at certain places (4), Computed tomogram (c) Axial view, (d) Coronal view and (e, f) 3-D reconstruction showing expansile intraosseous lesion involving left posterior mandible with perforation of left buccal and lingual cortex, altered morphology of coronoid and condyle and cotical erotions over neck of the condyle and angle region.

tal foramen and lingually at the mylohyoid ridge. Cortical erosions at the angle of the mandible and neck of the condyle were also appreciated. There was altered morphology of the left coronoid and condylar process (figure 2c, d, e, f).

Treatment plan included resection of involved segment of mandible with disarticulation, reconstruction of the hemimandibulectomy defect with fibula graft fixated over titanium reconstruction plate with condylar head. 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 vessels were identified and ligated. 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

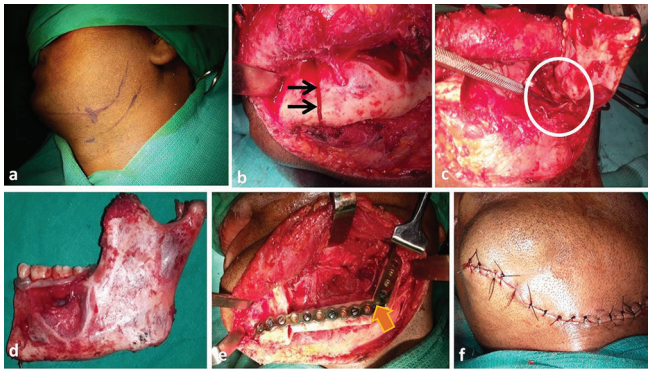

Figure-3: Surgical steps in management of lesion (a) submandibular incision marked, (b) osteotomy cuts placed anterior to mental foramen, (c) medial muscle attachments stripped and inferior alveolar bundle identified and ligated, (d) excised specimen, (e) fibula graft with reconstruction plate fixated to remaining mandible, (f) closure.

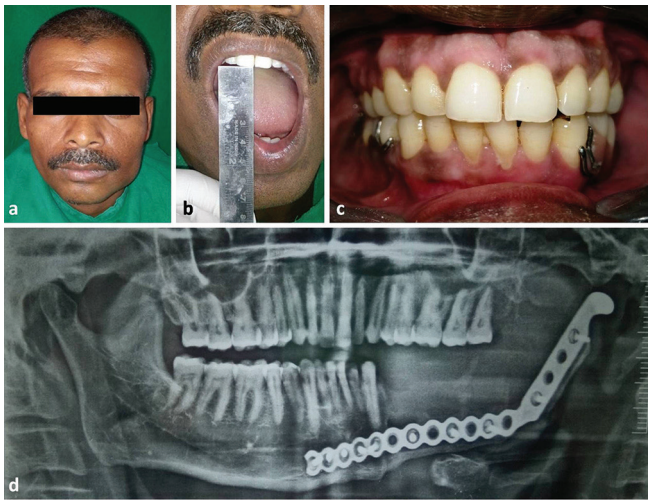

Figure-4: One year ear postoperative photographs showing (a) satisfactory facial contour, (b) adequate oral opening, (c) Intraoal view showing replacement of missing teeth with cast partial denture, (d) OPG showing osseointegration of fibula graft with mandible.

non dominant 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 reconstruction plate and then to the patient's remaining mandible with screws. Wound was closed in layers (figure 3). Postoperative recovery was uneventful and the patient was kept on intermaxillary fixation for 2 weeks for achieving initial stability of the graft, maintenance of occlusion and prevention of mandibular deviation. 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. OPG showed osseointegreration of the fibula with patient's mandible. The missing teeth were replaced with cast partial denture for prosthetic rehabilitation of the patient (figure 4).

DISCUSSION

WHO in 2005 reclassified parakeratinized 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. 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. Various treatment modalities have been reported for management of KCOT (table 1) including decompression, marsupialization, enucleation and resection.

Decompression / Marsupialization of lesion

KCOT enlarges by expansion due to pressure exerted by the fluid within the lesion. Decompression is a technique that relieves this pressure by making perforation in the lesion and keeping the opening patent to prevent build up of fluid inside. Marsupialisation exposes the lesion to the oral environment thus reducing the pressure and preventing its further growth. The exposed cavity is packed with antibacterial gauze like iodoform impregnated with bacitracin. This process not only prevents further growth of KCOT but also changes the friable parakeratinised epithelium into a thicker epithelium which can be easily removed later with enucleation. This treatment plan is preferred when the lesion is large and in vicinity of vital structures, in young patients to reduce post-operative morbidity, and in systemically compromised patients where more aggressive resection or administration of general anesthesia is contraindicated.[2,3]

1. Decompression or Marsupialization
2. Enucleation a. Enucleation without physicochemical treatment b. Enucleation with physicochemical treatment i. Peripheral ostectomy ii. Cornoy's solution iii. Electrocautery iv. Cryotherapy (liquid nitrogen) v. Methylene blue
3. Resection a. Marginal resection b. Segmental resection i. Segmental resection without disarticulation ii. Segmental resection with disarticulation

Table-1: Management options for KCOT

Enucleation of KCOT lining, with or without physicochemical treatment

Enucleation is removal a lesion by shelling it out of the bone. When used alone for management of KCOT, It has the highest recurrence rate (26.9% to 54.5%) and thus should be supplemented with physicochemical treatments like peripheral ostectomy, cornoy's solution, electrocauterization and cryotherapy (liquid nitrogen).[5-7]

Peripheral ostectomy is removal of few millimeters of bone beyond the visible margins of the lesion after enucleation. It can be carried alone or may be assisted by vital staining of the surgical bone defect.[2] Staining is done with help of stains like methylene blue or crystal violet. The dye penetrates into cells that have an abnormal increase in nucleic acids. This helps to identify areas that have been incompletely excised during peripheral ostectomy.[5] Cornoy's solution is a chemical used to cauterize retained pathological tissue. It is applied to the bone cavity after enucleation of the lesion. The chemical penetrates around 2 mm of bone (after application for 3 minutes), which is then removed by peripheral ostectomy. Cornoy's solution is neurotoxic and contact with nerves for more than 2 min may lead to paresthesia. When KCOT perforates the buccal or lingual cortex into the soft tissue, to avoid the recurrence electrocauterisation of the surrounding soft tissues must be done.[3] Liquid nitrogen cryosurgery has also been used to devitalize the peripheral bone after removal of lesion. Cryosurgery causes cell death by direct damage from intracellular and extracellular ice crystal formation along with osmotic and electrolyte disturbances. This technique involves rapidly freezing and slow thawing of the cavity walls after enucleation of lesion. The process affects around 1.5 mm of the bone and can cause paraesthesia if it comes in contact with nerve.[2,3]

Resection of jaw for management of KCOT

Resection of the lesion is preferred treatment modality for aggressive jaw tumors like KCOT. It provides more definitive treatment with less recurrence rates. Resection is of two types- marginal or segmental. In marginal resection part of mandible is removed maintaining the continuity at lower border. In segmental resection a part of mandible is removed without maintaining the continuity. When condyle is removed along with the segment it is termed as disartic-

ulation.[2,3]

KCOT shows high recurrence rate. The main reason for recurrence of KCOT is its thin lining, which is friable, and portions are easily left behind during its removal. KCOT also have daughter cysts beyond the lining of the cyst, between the cyst and the alveolar mucosa and in the area of alveolar mucosa wherever there is perforation. These further increase risk of recurrence in large cysts showing areas of perforation.[2] Segmental resection and electrocauterisation decreased the chances of recurrence in such cases. In the present case segmental resection of mandible with disarticulation was carried out. The soft tissue abutting the areas of lingual and buccal plate perforations were electrocauterised to remove any left out pathological tissue. Reconstruction was done with autogenous fibula graft. Fibula graft gives adequate length of bone which can be shaped easily as per the shape of the mandible. It offers a good quality of compact bone which can give stability to future prosthetic rehabilitation.

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 provide comprehensive functional and esthetic rehabilitation of the patient. Long term follow-up is always advocated to rule out recurrence.

REFERENCES

1. Liu B., Cai Y. Recurrent keratocystic odontogenic tumor in the masseter muscle overlying the boney perforations: a case report, Oral Surg Oral Med Oral Pathol Oral Radiol. 2012;113:e1-e5.
2. M.A. Pogrel. The Keratocystic Odontogenic Tumor, Oral Maxillofacial Surg Clin N Am. 2013;25:21–30.
3. Abdullah W. Surgical treatment of keratocystic odontogenic tumour: A review article, The Saudi Dental Journal. 2011;23:61–65.
4. Mendes R. Characterization and management of the keratocystic odontogenic tumor in relation to its histopathological and biological features, Oral Oncology. 2010;46:219–225.
5. Iwai T. Use of methylene blue for precise peripheral ostectomy of keratocystic odontogenic tumour, British Journal of Oral and Maxillofacial Surgery. 2011; 49: e84–e8.
6. Johnson N. Management and recurrence of keratocystic odontogenic tumor: a systematic review, Oral Surg Oral Med Oral Pathol Oral Radiol. 2013;116:e271-e276
7. Kaczmarzyk. A systematic review of the recurrence rate for keratocystic odontogenic tumour in relation to treatment modalities, Int. J. Oral Maxillofac. Surg. 2012; 41: 756–767.

Source of Support: Nil; **Conflict of Interest:** None

Submitted: 04-03-2016; **Published online**: 09-04-2016