

Surgical Treatment of Massive Keratocystic Odontogenic Tumour and its Prognosis: Review of Treatment Modalities

Venkata Krishna I^{1*} Giridhar Kumar V² Karuna Sree P³ K Sravan Kumar Reddy⁴ Phanitej G⁵ J Laxmi Sravya⁶

¹Reader, Department of Oral and Maxillofacial Surgery, Panineeya Institute of Dental sciences, Hyderabad, Telangana, India.

²Professor, Department of Oral and Maxillofacial Surgery, Panineeya Institute of Dental sciences, Hyderabad, Telangana, India.

³Post Graduate Student, Department of Oral and Maxillofacial Surgery, Panineeya Institute of Dental sciences, Hyderabad, Telangana, India.

⁴Post Graduate Student Department of Oral and Maxillofacial Surgery, Panineeya Institute of Dental sciences, Hyderabad, Telangana, India.

⁵Post Graduate Student, Department of Oral and Maxillofacial Surgery, Panineeya Institute of Dental sciences, Hyderabad, Telangana, India.

⁶Post Graduate Student Department of Oral and Maxillofacial Surgery, Panineeya Institute of Dental sciences, Hyderabad, Telangana, India.

ABSTRACT

Background: Although keratocystic odontogenic tumour (KCOT) or odontogenic keratocysts (OKCs) are benign, they are often most aggressive odontogenic cysts with a high recurrence rate. This was explained by histopathological features like a thin, friable wall, which is often difficult to enucleate from the bone as a whole, presence of small satellite cysts within the fibrous wall. Multiple surgical approaches were followed including decompression, marsupialization, enucleation with or without adjunct (Carnoy's solution) and resection. As recorded in literature, KCOT can be conservatively treated with enucleation and application of Carnoy's solution. This can be used specially in the large lesions where resection results in loss of continuity of the jaw. Here we present a case of massive KCOT which is treated by enucleation and chemical cauterization with Carnoy's solution. At one year follow-up, the patient was comfortable and complete resolution of the radiolucent pathology was evident.

Keywords: Keratocystic odontogenic tumour, odontogenic keratocyst, benign, conservative treatment, Carnoy's solution.

INTRODUCTION

The odontogenic keratocyst (OKC) is a developmental cyst of odontogenic origin, which is derived from the dental lamina. Odontogenic keratocyst OKC was first described by Philipsen (1956). OKC is now referred to by the World Health Organization (WHO) as a keratocystic odontogenic tumour KCOT, defined it as "a benign uni-cystic or multi-cystic, intraosseous tumour of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium and potential for aggressive, infiltrative behaviour¹.

The KCOT is one of the most aggressive odontogenic lesions of the oral cavity. It has a high

recurrence rate and is associated with the basal cell nevus syndrome^{2,4}. Different studies have shown the incidence of KCOT to be 3–11% of the odontogenic cysts³. The distribution between sexes varies from equality to a male to female ratio of 1.6:1. The age distribution appears to be bimodal. There appeared to be two incidence peaks between 25 and 34 years, and 55 and 64 years of age. KCOTs may occur in any part of jaws, with the majority occurring in the mandible, most commonly in the angle of the mandible and ramus. If it occurs in the maxilla, it mostly occurs in the anterior region. This lesion can be associated with an impacted third molar. Radiographically, it appears as a well-defined unilocular or multilocular lesion with a smooth or scalloped contour, often with corticated margins⁵.

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*Correspondence Dr. Venkata Krishna I.

Department of Oral and Maxillofacial Surgery, Panineeya Institute of Dental sciences, Hyderabad, Telangana, India.

Email: drkrishna@gmail.com

Obvious bone expansion is not seen in many cases because KCOTs tend to grow in an anteroposterior direction within the medullary cavity of the bone. Displacement of teeth adjacent to the cyst occurs more frequently than resorption⁶. When they are very large, they tend to expand bone.

KCOTs are treated conservatively or aggressively. Conservative treatment includes simple enucleation, with or without curettage, marsupialization and aggressive treatment includes peripheral ostectomy, chemical curettage with Carnoy's solution, cryotherapy, or electrocautery and resection⁷. The choice of treatment should be based on multiple factors like patient age, size and location of the cyst, proximity to the vital structures, soft tissue involvement, history of previous treatment and a histological variant of the lesion. The treatment modality should be such that, It carries the lowest risk of recurrence and the least morbidity⁸.

CASE REPORT

A 28 year old female patient visited to the Oral and Maxillofacial Surgery unit of Panineeya Dental College, Hyderabad with a chief complaint of pain and swelling on the lower left side of the face since 1 month. On palpation, it was tender and soft in consistency. Skin surface over the swelling was normal, mouth opening was adequate. There no alteration in sensation over the mental nerve distribution, Left submandibular lymph nodes were tender and palpable.

Intra orally, the buccal sulcus was obliterated [Figure 1] by a swelling extending 34 to 37. It is soft in the center and hard at the periphery and tender on palpation. Surface is rough with papillary lesion. Orthopantomogram revealed well-defined unilocular lesion with a scalloped contour and corticated margins in left mandibular region extending from lateral incisor to the sigmoid notch involving body and ramus associated with impacted 38 and pathological resorption of 35, 36, 37 [Figure 2]. Under local anesthesia, incisional biopsy was carried out in the left body region of the mandible. The specimen was sent for histopathological examination. Histopathological report revealed "Keratocystic odontogenic tumour". Considering patient's age and to reduce the morbidity, it was planned for enucleation of the cyst followed by

chemical cauterization with carnoy's solution under general anaesthesia.

Surgical procedure

Case was performed under general anesthesia. A crevicular incision was placed from 42 to 37 and extended onto the anterior border of the left ramus region and a releasing incision was placed mesial to 43. Mucoperiosteal flap was raised [Fig.3]. Small perforation in the buccal cortex was present [Fig.4]. Enucleation of the cyst was performed, followed by chemical cauterization with carnoy's solution [Fig 5,6,7].The specimen was sent for histopathological examination.

The histopathologic report revealed that the cystic lining of the lesion was parakeratinized stratified squamous epithelium of uniform 6-8-cell thickness (Fig 8). The basal cell layer is hyperchromatic with basal cell hamartias at few areas. The lumen is filled with keratin, cholesterol clefts and hyaline bodies. The connective tissue wall shows 3-4 micro cysts, cholesterol clefts and inflammatory cells at few areas. Follow up was done on 1st, 2nd, 3rd, 6th and one year postoperatively. The post-operative period was uneventful without any complications. Patient is asymptomatic since then without any complaints [Fig.9].



Fig 1: Intraoral view showing obliteration buccal sulcus.

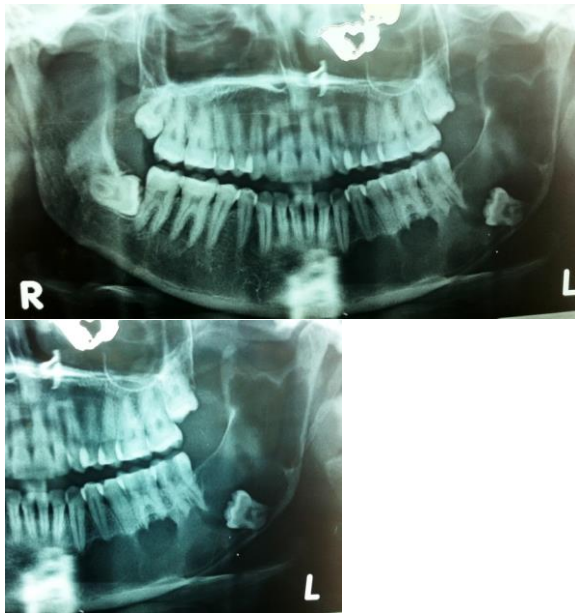


Fig 2: Orthopantomogram reveals an unilocular and scalloped borders in left mandibular region extending in relation from lateral incisor to the sigmoid notch involving body and ramus associated with impacted 38 and pathological resorption of 35, 36, 37.



Fig 3: Exposure of the lesion.

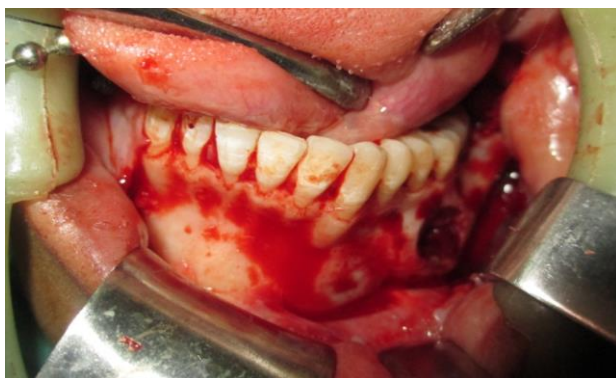


Fig 4: Perforation of the buccal cortex seen after mucoperiosteal flap elevation.

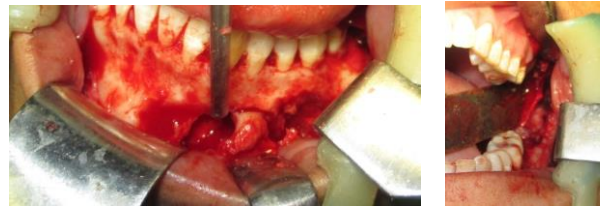


Fig 5: Intrabony osseous defects before cyst enucleation.



Fig 6: Cystic lining was removed intact along with the impacted third molar.

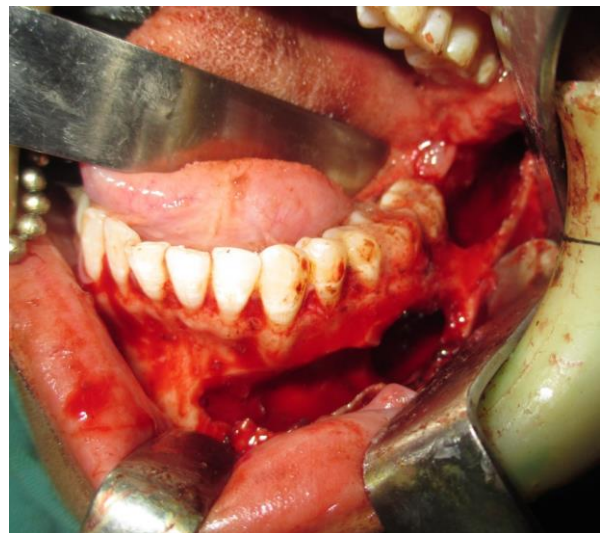


Fig 7: Intrabony osseous defect after cyst enucleation.

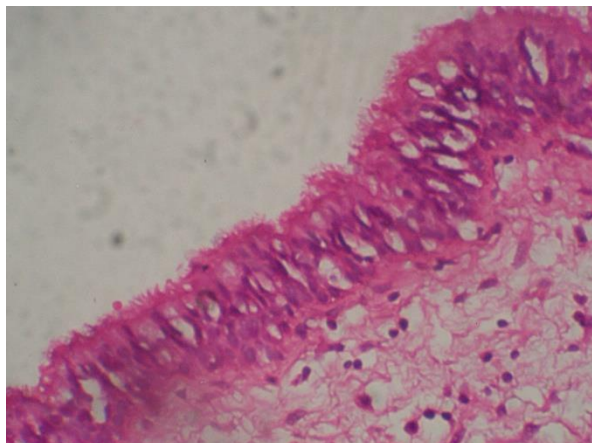


Fig 8: Section stained with hematoxylin and eosin (400×) showing cystic lining with parakeratinized stratified squamous epithelium of uniform 6–8-cell thickness.



Fig 9: One year orthopantomogram reveals new bone formation and no signs of recurrence.

DISCUSSION

Treatment of KCOT has always been challenging, in view of the marked recurrence rate. KCOT is usually treated by enucleation and curettage. Complete enucleation is however difficult, because of the thin, friable nature of the cyst wall, limited surgical access, cortical perforation, and the desire to preserve adjacent vital structures.

The frequency of recurrence ranges from 5% to 62%. This wide variation in recurrence rate is thought to be due to the different treatment approaches, ranging from simple curettage to resection of the affected jaw. Conservative treatment includes marsupialization, simple enucleation, with or without curettage. Aggressive treatment includes peripheral ostectomy, chemical curettage with carnoy's solution and resection. The treatment goals should involve eliminating the

potential for recurrence while also minimising the surgical morbidity.

Recurrence occurs due to the following reasons. The first reason involves incomplete removal of the original cyst's lining. Secondly, it involves growth of a new KCOT from small satellite cysts of odontogenic epithelial rests left behind by the surgical treatment. The third reason involves the development of an unrelated KCOT in an adjacent region of the jaws, which is interpreted as a recurrence⁹

Decompression and marsupialization

Any technique that relieves the pressure within the cyst is decompression, as this pressure is the way by which the cyst grows by expansion. Decompression can be performed by making a small opening in the cyst and keeping it open with a drain.

Marsupialization was first described by Partsch in 1882 for the treatment of cystic lesions which is based on the externalization of the cyst. In this technique, a surgical window is created to expose the cystic wall. The borders of cyst wall are sutured to create an open cavity that communicates with the oral cavity. Relieving pressure from the cystic fluid, allowing reduction of the cystic space and facilitating bone apposition to the cystic walls¹. Mandibular cysts are normally marsupialized into the oral cavity, while maxillary cysts can be marsupialized into oral cavity or maxillary sinus or nasal cavity. Marsupialization is described as a Partsch I procedure (the Partsch II procedure is enucleation and primary closure)¹⁰.

In a number of studies Decompression or marsupialization has been recommended as a technique that allows partial decrease in size in the KCOT so that vital structures like teeth or the inferior alveolar nerve can be preserved, then the KCOT was certainly enucleated. Brondum and Jensen(1991) reported a recurrence rate of 25% in 32 (KCOT) patients treated with decompression of the lesion¹¹. On the other hand, other studies have shown that marsupialization of KCOT can be followed by total resolution of the lesion without any further surgery.

Studies have shown that when the KCOT is open to the oral cavity by marsupialization, a

number of changes occur in the cyst lining. The lining appears to become thicker and easier to enucleate, and histologically it does appear to change and resemble normal oral mucosa¹².

Enucleation with and without adjuncts

To enucleate is “to remove whole or clean, as a tumour from its envelope.” Curettage is defined as “the removal of growths or other material from the wall of a cavity¹³. Curettage can be done by mechanical techniques (hand, rotary) alone or in combination with a chemical solution (Carnoy’s)¹⁴ or cryosurgical agents (liquid nitrogen)¹⁵.

Enucleation and treatment of the bony defect with Carnoy’s Solution

Enucleation of the thin, friable wall of the KCOT as a whole is difficult (especially in multilocular cyst). Because of the small satellite cysts, treatment should aim to eliminate all vital cells left behind in the defect. Cauterizing agent like Carnoy’s solution {consists 3 ml of chloroform, 6 ml of absolute ethanol, 1 ml of glacial acetic acid and 1 g of ferric chloride}⁵ is used. In case of lingual or buccal cortical perforation, some studies recommended electro-cauterization or complete excision of the overlying mucosa to decrease the recurrence, as microcysts and epithelial islands may be associated with overlying attached mucosa¹⁴.

Frerich et al. (1994) first reported the effects of Carnoy’s solution on the inferior alveolar nerve and he did not observe axonal damage during the first three minutes of direct application¹⁶.

Enucleation and liquid nitrogen cryotherapy

Liquid nitrogen has the ability to devitalize bone in situ while leaving the inorganic framework untouched. Cryosurgery results in cell death by direct damage from intracellular and extracellular ice crystal formation plus osmotic and electrolyte disturbances¹⁷. The standardized technique is enucleation of the cyst following which surrounding tissues are protected with sterile wooden tongue blades and gauze, and the cavity is sprayed with liquid nitrogen twice for 1 min, with a 5-min thaw between freezes. Mucosa is closed with watertight sutures. Bone graft can be placed in the defect simultaneously.

The advantages of liquid nitrogen are (1) the bone matrix is left in place to act as a clean scaffold for new bone formation, (2) a bone graft can be placed immediately to accelerate healing and minimize the risk of a pathologic fracture, and (3) decrease of bleeding and scarring. Disadvantages are (1) difficulty in controlling the amount of liquid nitrogen applied (2) Nerve paresthesia or anesthesia. However, the axon sheaths are left intact and nerve regrowth is normal such that most patients obtain partial or complete return of sensation in three months. The recurrence rate of 3–9% has been reported following enucleation and liquid nitrogen cryotherapy.

Block resection, with or without preservation of the continuity of the jaw

Resection, either segmental resection (surgical removal of a segment of the mandible or maxilla without maintaining the continuity of the bone) or marginal resection (surgical removal of a lesion intact, with a rim of uninvolved bone, maintaining the continuity of the bone)¹⁸ is an extreme technique, that results in considerable morbidity, particularly because reconstructive measures are necessary to restore jaw function and aesthetics (Stoelinga, 2005).

In a systematic review done by Blanas et al. (2000), the authors reported that resection was found to have the lowest recurrence rate (0%) but the highest morbidity rate, while enucleation followed with application of Carnoy’s solution for three minutes appears to be the least invasive procedure with the lowest recurrence rate comparable to that of resection¹⁸.

In this case, we have followed minimal invasive technique of enucleation and chemical cauterization with Carnoy’s solution, considering the patient’s age and to reduce the morbidity. At one year follow-up, the patient was comfortable and complete resolution of the radiolucent pathology was evident. Periodic examination is required because of the high rate of recurrence of KCOT.

CONCLUSION

As literature suggests aggressive nature and high recurrence rates of KCOT, there is a need to select a treatment modality which has got

potential to reduce the recurrence rate with least morbidity. The present treatment modality serves as a reliable option, as it serves to eradicate pathologic lesion, reduction of potential for recurrence and Preservation of the continuity of the mandible.

CONFLICT OF INTEREST

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

REFERENCES

1. Barnes, L., Eveson, J.W., Reichart, P., Sidransky, D. (Eds.). Pathology and Genetics of Head and Neck Tumours. 2005; IARC Press, Lyon, WHO classification of tumours series.
2. Oda D, Rivera V, Ghanee N, Kenny EA, Dawson KH. Odontogenic keratocyst: the northwestern USA experience. J Contemp Dent Parct. 2000;1:60-74.
3. Chuong. R., Donoff, R.B., Guralnick, W. The odontogenic keratocyst. J. Oral Maxillofac. Surg.1982;40:797-802.
4. Payne. T.F. An analysis of the clinical and histologic parameters of odontogenic keratocyst. Oral Surg. Oral Med. Oral Pathol. Oral Radiol. Endod.1972;33: 536-546.
5. Faustino SE, Pereira MC. Recurrent peripheral odontogenic keratocyst: A case report. Dentomaxillofac Radiol. 2008;37:412-4.
6. Unusual CT. Appearance in an odontogenic keratocyst of the mandible: Case report. AJNR Am J Neuroradiol.2001;22:1887-9.
7. Morgan, T.A., Burton, C.C., Qian, F. A retrospective review of treatment of the odontogenic keratocyst. J. Oral Maxillofac. Surg.2005; 63: 635-639.
8. Rogerson, K.C.Gorlin's syndrome: an update on diagnosis and management. Oral Maxillofac. Clin. North Am.1991; 3: 155.
9. Tae GI, Hoe-Kyung H. Diagnostic ability of differential diagnosis in ameloblastoma and odontogenic keratocyst by imaging modalities and observers. Korean J Oral Maxillofac Radiol.2006;36:177-82.
10. Pogrel, M.A. Treatment of keratocysts: the case for decompression and marsupialization. J. Oral Maxillofac. Surg.2005; 63:1667-1673.
11. Brondum, N., Jensen, V.J.Recurrence of keratocysts and decompression treatment. A long-term follow-up of forty-four cases. Oral Surg. Oral Med. Oral Pathol.1991;72: 265.
12. August, M., Faquin, W.C., Troulis, M et al. Differentiation of odontogenic keratocysts from nonkeratinizing cysts by use of fineneedle aspiration biopsy and cytokeratin-10 staining. J. Oral Maxillofac. Surg. 2000; 58:935-940.
13. Giuliani, M., Grossi, G.B., Lajolo, C., Bisceglia, M., Herb, K.E. Conservative management of a large odontogenic keratocyst: report of a case and review of the literature. J. Oral Maxillofac. Surg. 2006;64:308-316.
14. Stoelinga, P.J.W.Excision of the overlying, attached mucosa, in conjunction with cyst enucleation and treatment of the bony defect with Carnoy solution.The odontogenic keratocyst. Oral Maxillofac. Surg. Clin. North Am. 2003;15: 407.
15. Jensen, J., Sindet-Pedersen, S., Simonsen, E.K. A comparative study of treatment of keratocysts by enucleation or enucleation combined with cryotherapy. J. Craniomaxillofac. Surg.1988;16: 362.
16. Frerich, B., Cornelius, C.P., Wietholter, H.Critical time of exposure of the rabbit inferior alveolar nerve to Carnoy's solution. J. Oral Maxillofac. Surg. 1994; 52 (6): 599-606.
17. Schmidt, B.L., Pogrel, M.A., The use of enucleation and liquid nitrogen cryotherapy in the management of odontogenic keratocysts. J. Oral Maxillofac. Surg.2001; 59:720.
18. Blanas, N., Freund, B., Schwartz, M., Furst, I.M. Systematic review of the treatment and prognosis of the odontogenic keratocyst. Oral Surg. Oral Med. Oral Pathol. Oral Radiol. Endod. 2000; 90: 553.

