

## Case Report

## Keratocystic Odontogenic Tumour Mimicking as a Dentigerous Cyst – A Rare Case Report

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**Abstract:** Keratocystic odontogenic tumor (KCOT) formerly known as odontogenic keratocyst (OKC), is considered a benign unicystic or multicystic intraosseous neoplasm and one of the most aggressive odontogenic lesions presenting relatively high recurrence rate and a tendency to invade adjacent tissue. On the other hand Dentigerous cyst (DC) is one of the most common odontogenic cysts of the jaws and rarely recurs. They were very similar in clinical and radiographic characteristics. In our case a pathological report following incisional biopsy turned out to be dentigerous cyst and later as Keratocystic odontogenic tumour following total excision. The treatment was chosen in order to prevent any pathological fracture. A recurrence was noticed after 2 months following which the lesion was surgically enucleated. At 2-years of follow-up, patient showed no recurrence.

**Keywords:** Dentigerous cyst, Keratocystic odontogenic tumour (KCOT), Recurrence, Enucleation

### INTRODUCTION

Keratocystic odontogenic tumour (KCOT) is a rare developmental, epithelial and benign cyst of the jaws of odontogenic origin with high recurrence rates. The third molar region especially the angle of the mandible and the ascending ramus are involved far more frequently than the maxilla. The choice of treatment approach was based on the size of the cyst, recurrence status, and radiographic evidence of cortical perforation. It has been estimated that about 4%-9% of dentigerous cysts occur in the first decade of life [3, 12]. Keratocystic odontogenic tumor (KCOT) presents most frequently in the second, third and fourth decades of life (54.2%) with rare cases reported as early as the first, and as late as the ninth decade of life [2, 12]. Removal of the entire cyst associated with the extraction of the impacted tooth is the main treatment to prevent recurrence of odontogenic cystic lesions. However, when there is a destruction of the cortical bones, conservative treatment should be the choice. Treatment of KCOTs remains a controversial subject because of their great potential to recur. Sometimes, the first surgical approach in KCOT is decompression or marsupialization and when it gains reduced volume, enucleation has to be performed [5, 8].

### CASE REPORT

A 17-year-old patient reported to the OP with a chief complaint of swelling in relation to her lower left back tooth region for the past 1 week. Patient noticed the swelling 1 week ago, following which it reduced in size. The pain was present for the past 1 week, which was intermittent. Extraoral inspection revealed a diffuse swelling (Figure 1) present on the lower third of the face measuring approximately 2x3 cm in size.

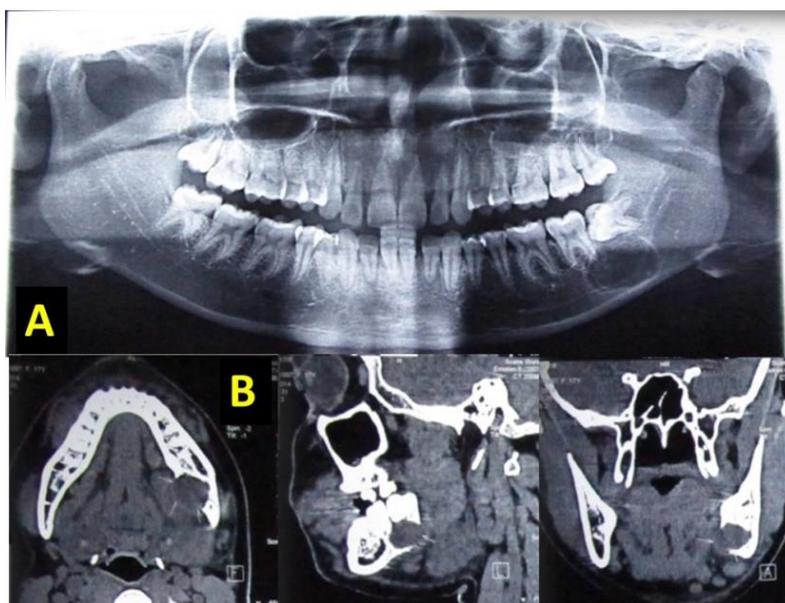


Fig-1: Extraoral inspection revealed a diffuse swelling

Surface of the swelling appeared normal with no evidence of any secondary changes. On palpation inspeactory findings regarding site, size, shape and extent were confirmed. The swelling was firm in consistency with tenderness present on palpation. Skin over the swelling was pinchable with no evidence of local rise in temperature. No evidence of paraesthesia. Intraoral inspection revealed a diffuse swelling (Figure 2) measuring approximately 2 x 2.5 cm on the buccal aspect of the clinically missing 38 region extending mesial to 36 region. Obliteration of the buccal vestibule on the lower left quadrant was evident extending from 36 to 38 regions.



**Fig-2: Pre-operative**

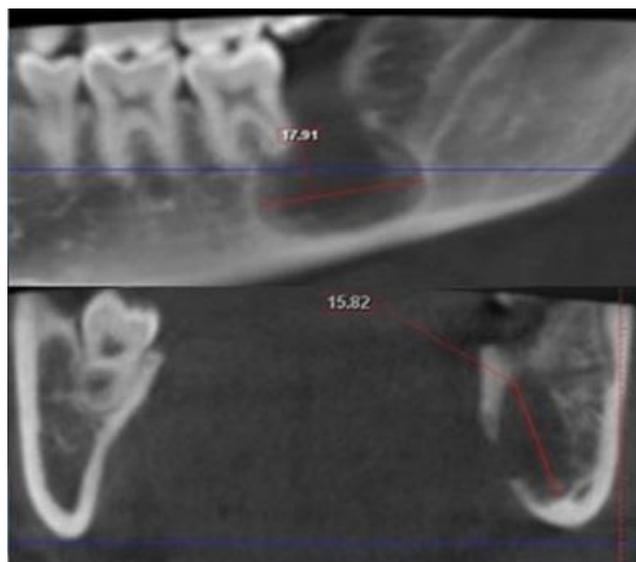


**Fig-3: A) OPG revealing the presence of a horizontally impacted tooth and a well-defined unilocular radiolucent lesion with a sclerotic margin and which is association with the lateral aspect of the impacted tooth near the CEJ, B) Axial, sagittal and coronal slices of CT showing a unilocular hypodense lesion in relation to 38 region with erosion of the lingual cortical plate.**

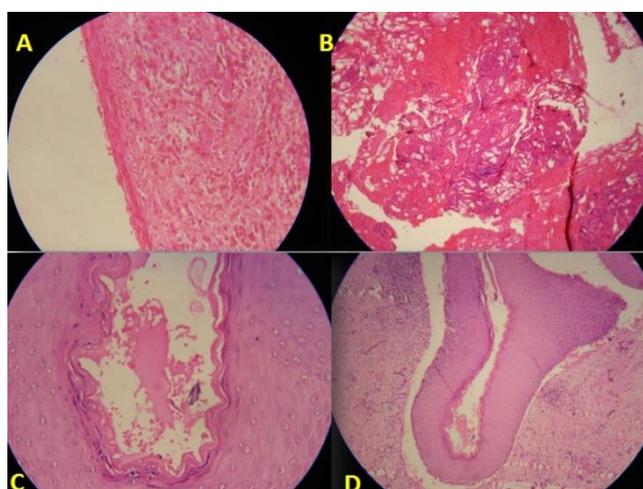
Mucosa appeared edematous and erythematous in the affected region. On palpation the swelling was tender. A provisional diagnosis of odontogenic cyst was made based on the history and clinical findings. OPG revealed horizontally impacted tooth of 38 with the presence of a well-defined unilocular radiolucent lesion with a sclerotic margin and which is association with the lateral aspect of the impacted tooth near the CEJ (Figure 3 A). Axial, sagittal and coronal slices of CT showed a unilocular hypodense lesion in relation to 38 region with erosion of the lingual cortical plate suggestive of an odontogenic cyst. Extraction of impacted teeth with enucleation of cyst was performed. The specimen was sent for histopathological investigation which returned with a diagnosis of

dentigerous cyst (Figure 5 a & B) and there was a recurrence of swelling in the same region 2 months later. CBCT was taken which showed a well-defined radiolucency in relation to the edentulous 38 region (Figure 4).

The lesion was completely enucleated and specimen was again sent for histopathological evaluation. It returned with a report of keratocystic odontogenic tumour (Figure 5 C & D). This turned out to be one of the rare instances in which a keratocystic odontogenic tumour mimicked as a dentigerous cyst. Patient is under follow-up for the past two and a half years without any recurrence.



**Fig-4:** CBCT images showing the breach of the cortical plate on the lingual aspect of the edentulous region



**Fig-5) A & B)** After 1<sup>st</sup> surgery 10 X magnification image of dentigerous cyst exhibiting 2 to 3 cell thick epithelial layer and connective tissue with inflammatory cell infiltrate & 40 X magnification image of dentigerous cyst exhibiting cholesterol clefts, **C & D)** After 2<sup>nd</sup> surgery 40 X image of Keratocystic odontogenic tumor exhibiting 6 – 8 cells thick parakeratinised corrugated epithelium & 100 X image of Keratocystic odontogenic tumor exhibiting presence of keratin.

## DISCUSSION

Keratocystic odontogenic tumour which was formerly known as odontogenic keratocyst is now classified as a neoplasm by the World Health Organization (WHO) in 2005, as a benign odontogenic tumor derived from the dental lamina that requires special surgical considerations because of its known aggressive behavior and high tendency to recur [2]. KCOTs have been suggested as a major diagnostic criterion of nevroid basal cell carcinoma syndrome [5]. The rate of recurrence ranges from 1% to 62% [11]. Recurrences occur within the first 5 years after treatment of the lesion [8]. Numerous surgeons advocate complete removal of lesion with extension margins and curettage of the surrounding tissues [2, 5, 6, 8-11, 13]. Recurrence is influenced by a variety of factors which are the length of the follow-up period, mode of treatment, size of the lesion and presence of

daughter cysts in histopathological analysis [6, 8]. Marsupialization is an advisable treatment in dentigerous cysts to preserve the cyst-associated teeth and promote their eruption [4, 7, 9]. In the case of KCOT, removal of the cyst including the associated tooth and surrounding bone is the treatment of choice [6, 13]. Many authors have reported that marsupialization can be an effective alternative [5, 8, 9, 13]. Decompression and secondary enucleation as first line treatment option for KCOT has been suggested [5, 6, 9]. Achieving a significant reduction of the lumen which can be confirmed through radiographic imaging, after which a complete enucleation is justified to prevent recurrence. The patient of this report was in the second decade of life and had cortical perforation associated with the KCOT. A decision of conservative management over aggressive surgery was made keeping in mind the perforation and preventing a pathological

fracture. Many molecular and clinical studies have shown that the parakeratinized and orthokeratinized KCOT were different in molecular area and recurrence rate; orthokeratinized KCOT had a lower recurrence rate [1, 6, 8]. Marsupialization is considered as an effective preliminary treatment for large KCOT and it does not affect the recurrence. In our case marsupialisation procedure here was done without the supplementary enucleation neither or application of Carnoy's solution. Reports suggest probable changes in growth characteristics when KCOT which are treated primarily with marsupialisation that makes it less aggressive [1, 6]. Many clinicians have observed epithelial dedifferentiation and loss of cytokeratin-10 production after marsupialisation [11]. As in our case a relationship between perforation of the lingual plate of the mandible and recurrence after treatment was observed by Borg *et al.* [14] (Shear ref). In many cases after marsupialization there is new bone formation in the area of cortical erosion. In our case, complete resolution of the lesion was not achieved by a conservative approach. Though preserving anatomy and function was a main concern, we suggest a surgical management for complete removal of the lesion to aid in a good prognosis.

## CONCLUSION

Proper biopsy specimen and care while transporting is mandatory to preserve the architecture of the specimen biopsied which will return back with a proper report. Each cyst has unique characteristics with respect to recurrence and malignant transformation. The aggressive nature of the KCOT warrants an aggressive treatment strategy & its recent reclassification by WHO as a neoplasm should further motivate clinicians in this direction. Thus, a correct diagnosis can pave way for proper care, treatment and follow-up of the patient.

## REFERENCES

1. August M, Faquin WC, Troulis MJ, Kaban LB. Dedifferentiation of odontogenic keratocyst epithelium after cyst decompression. *Journal of oral and maxillofacial surgery*. 2003 Jun 30; 61(6):678-83.
2. Chirapathomsakul D, Sastravaha P, Jansisyanont P. A review of odontogenic keratocysts and the behavior of recurrences. *Oral surgery, oral medicine, oral pathology, oral radiology, and endodontology*. 2006 Jan 31; 101(1):5-9.
3. Freitas DQ, Tempest LM, Sicoli E, Lopes-Neto FC. Bilateral dentigerous cysts: review of the literature and report of an unusual case. *Dentomaxillofacial Radiology*. 2014 Feb 13.
4. Hyomoto M, Kawakami M, Inoue M, Kirita T. Clinical conditions for eruption of maxillary canines and mandibular premolars associated with dentigerous cysts. *American Journal of Orthodontics and Dentofacial Orthopedics*. 2003 Nov 30; 124(5):515-20.
5. Hyun HK, Hong SD, Kim JW. Recurrent keratocystic odontogenic tumor in the mandible: a case report and literature review. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology*. 2009 Aug 31; 108(2):e7-10.
6. Kolokythas A, Fernandes RP, Pazoki A, Ord RA. Odontogenic keratocyst: to decompress or not to decompress? A comparative study of decompression and enucleation versus resection/peripheral ostectomy. *Journal of oral and maxillofacial surgery*. 2007 Apr 30; 65(4):640-4.
7. Koželj V, Sotošek B. case study: Inflammatory dentigerous cysts of children treated by tooth extraction and decompression—report of four cases. *British dental journal*. 1999 Dec 11; 187(11):587-90.
8. Maurette PE, Jorge J, de Moraes M. Conservative treatment protocol of odontogenic keratocyst: a preliminary study. *Journal of Oral and Maxillofacial Surgery*. 2006 Mar 31; 64(3):379-83.
9. Nakamura N, Mitsuyasu T, Mitsuyasu Y, Taketomi T, Higuchi Y, Ohishi M. Marsupialization for odontogenic keratocysts: long-term follow-up analysis of the effects and changes in growth characteristics. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology*. 2002 Nov 30;94(5):543-53.
10. Reichart PA, Philipsen HP. Oral erythroplakia—a review. *Oral oncology*. 2005 Jul 31; 41(6):551-61.
11. Shear M. The aggressive nature of the odontogenic keratocyst: is it a benign cystic neoplasm? Part 1. Clinical and early experimental evidence of aggressive behaviour. *Oral oncology*. 2002 Apr 30; 38(3):219-26.
12. Shear M, Seward GR. Cysts of the oral regions. John Wright; 1992.
13. Zhao YF, Wei JX, Wang SP. Treatment of odontogenic keratocysts: a follow-up of 255 Chinese patients. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology*. 2002 Aug 31; 94(2):151-6.
14. Borg GA, Noble BJ. Perceived exertion. *Exercise and sport sciences reviews*. 1974 Jan 1; 2(1):131-54.