

Odontogenic Keratocyst (OKC) Of Maxilla- A Diagnostic Dilemma

¹Dr. Kiruba Shankar K, ²Dr. Mahabaleshwara Chalathadka,
³Dr. Sumanth Unakalkar, ⁴Dr.Sahya S Lal,

¹Post graduate student, Department Of Oral and Maxillofacial Surgery, KVG Dental College and Hospital, Sullia, Karnataka, India.

²Professor, Department of Oral and Maxillofacial Surgery, KVG Dental College and Hospital, Sullia, Karnataka, India

³Senior Lecturer, Department of Oral And Maxillofacial Surgery, KVG Dental College and Hospital, Sullia, Karnataka, India.

⁴Post graduate student, Department Of Oral and Maxillofacial Surgery, KVG Dental College and Hospital, Sullia, Karnataka, India.

*Corresponding Author: ¹Dr. Kiruba Shankar K,

ABSTRACT:- Odontogenic keratocyst (OKC), dentigerouscyst & radicular cyst are most common jaw cyst encountered by maxillofacial surgeons. Among this, OKC are known for their peculiar behaviour & unique tendency to reoccur. Clinical and radiographic presentations of OKC possess a major diagnostic dilemma as they can be located at various sites mimicking other pathologies. Maxillary OKCs are more prone for misdiagnosis because of its rare occurrence and asymptomatic nature. Moreover OKC requires aggressive surgical management as compared to other odontogenic cysts especially in maxilla. Hence it is important to accurately diagnose this lesion. Here we present a case of OKC which was provisionally diagnosed as residual cyst, treated by complete enucleation and the diagnostic dilemma while managing.

KEY WORDS: Odontogenickeratocyst, OKC, Residual cyst, Enucleation.

I. INTRODUCTION:

Cysts constitute about 17% of the total tissue specimens that are submitted to oral pathology for biopsy¹. OKC were recently known as keratocysticodontogenic tumour because of its aggressive behaviour pattern and now is reconsidered as OKC. Most of the oral cystic lesions have similar clinical and radiological features. Hence, correlations between clinical, radiographic and histopathological findings are necessary to conclude a proper diagnosis. Distinguishing OKC from other cysts is most important as they have the tendency for high recurrence rate, aggressive growth pattern and chance for malignant potential. OKC is a developmental cyst originating from remnants of dental lamina, occurring between second and third decades of life, which affects the mandible at a proportion of 2:1 in relation to the maxilla². Maxillary OKC's are most commonly located in canine region than in posterior maxilla and can be easily mistaken for inflammatory cysts. There are conflictory findings in predominant location of OKC in maxilla. Some authors suggest that OKC occurs equally among anterior and posterior region³. Some suggests that there are more anterior lesions than posterior lesions⁴ and others conclude that posterior region is the predominant site⁵. OKCs involving maxilla should be treated with great care because reports have shown the ability of this cyst to extend into adjacent areas and high recurrence rate because of more cancellous bone. The purpose of this article is to report a case of OKC of posterior maxilla misdiagnosed as residual cyst, various investigations done for the same and the dilemma in diagnosis & treating the lesion.

II. CASE REPORT:

A 33 year old male patient reported to department of OMFS, K.V.G Dental College and Hospital, Sullia with a chief complaint of pus discharge and bad breath from upper left back tooth region since 6months. On eliciting the past history, patient gives history of extraction 6 months back but the pus discharge had not reduced even after it. Past medical history was unremarkable. On extra oral examination, no gross asymmetry of face and no obvious extra oral swelling present. Intra oral examination revealed missing 27 due to previous extraction. There was no evidence of oroantral communication or obliteration of vestibule noted (Fig1). Orthopantomogram(OPG) showed radiolucency of approximately 3x3 cm size over the periapical region of 26,27,28. Sclerotic borders of the radiolucency could not be appreciated on the OPG, obliteration of left maxillary sinus is also noted (Fig 2). FNAC was carried out which showed a cheesy white aspirate material

mixed with blood which turned out as suggestive of an inflammatory cystic lesion(Fig3).Protein estimation was carried out which showed cystic fluid protein content as 7g/dl. Pulp vitality test was done for 26 which suggest that the tooth was vital.To assess the extent of the cystic lesion,bony and maxillary sinus involvement, CBCT was carried out which showed a well-defined radiolucency of about 2.7x2.4cm in size with obliteration of left maxillary sinus along with resorption of buccal cortical plate in relation to 27 & 28 region (Fig4).Provisional diagnosis of the case was infected residual cyst when FNAC,protein estimation and CBCT was taken into consideration. After all routine preoperative investigations & with all aseptic conditions, surgical enucleation of the lesion with secondary closure using betadine soaked gauze roll done under general anesthesia (Fig 5).The pack was removed after 24 hours and sutures placed. Histopathological specimen revealed the lesion as infected OKC.One Year follow up of the patient had no recurrence.Patient recalled for bimonthly follow up for next 5-10 years considering histopathological diagnosis, location and complete enucleation is the only treatment carried out in this case.

III. DISCUSSION:

OKC is an epithelial developmental cyst and was first described by Phillips in 1956⁶. The incidence of male to female ratio is about 1.6:1⁷. OKC may occur in any part of the jaw, majority occurring in the mandible, most commonly in the angle of the mandible and ramus⁸. Incidences of occurrence in maxilla are about 31.3% and are more common in canine region. It occurs most commonly in second and third decades of life. Multiple OKCs are frequently associated with nevoid basal cell carcinoma syndrome⁹. OKC are generally thought to be derived from either the epithelial remnants of the tooth germ or the basal cell layer of the surface epithelium¹⁰. It is a benign developmental odontogenic tumour with many distinguishing clinical and histopathological features¹¹. Clinically, OKC can easily be misdiagnosed as inflammatory cysts because majority of the cases presents with common inflammatory signs such as pain, swelling and pus discharge. Diagnosis based on clinical information alone will be problematic. Radiographically, patients with unilocular radiolucency in maxilla can also be misjudged. Even with the clinical and radiological findings, OKC's becomes difficult to diagnose especially when the lesion is located adjacent to non-vital teeth/missing tooth. In our case, patient gave a history of tooth extraction six months back due to complaint of pus discharge from sinus. But patients complain of pus discharge & foul smelling remained even after extraction indicating underlying pathology. This gives importance of clear history taking for extraction of sound tooth/carious tooth or non-vital tooth. In our case, history was inconclusive. Use of advanced diagnostic aid such as CBCT is crucial in cases like our patient as it gives the exact size, extension of the lesion & bony involvement. CBCT provides more detailed information for evaluation of the borders of the lesion and the relation of the lesions with adjacent anatomic structures¹². Both clinical and radiological appearance of OKC mimics other pathologies and poses a major diagnostic dilemma. FNAC had been carried out which gave the result as "suggestive of inflammatory lesion". Protein content of the aspirated material is done to distinguish the lesion between OKC and inflammatory lesion as the cystic protein content in OKC will be less than 4g/dl. But, the result turned out to be 7 g/dl which again gave a clue to consider inflammatory cyst. Our experience in this case states that over reliance on FNAC/protein estimation may give rise to misdiagnosis. Protein content in our case was high may be due to more pus content in the aspirated material. Differential diagnosis of this case includes periapical cyst, residual cyst, dentigerous cyst and ameloblastoma. Using all these investigations in our case, patient had been provisionally diagnosed with residual cyst and was treated by enucleation with secondary closure. But, histopathology of the specimen confirmed the lesion as infected OKC. Treatment options for OKC are enucleation with primary closure, enucleation with secondary closure, enucleation with cryotherapy, enucleation with chemical cauterisation using Carnoy's solution and en bloc resection. Brannon stated that the recurrence rate of OKC, which was treated with enucleation alone, was 12%. Recurrence rate reported for resection is 0-2%. However, resection can have high morbidity¹³. Prognosis of OKC becomes poor when treated with enucleation alone due to its thin friable nature of the cyst lining and presence of daughter cysts. Special attention should be given to the dentate area if the enucleation is chosen as treatment, due to higher rates of recurrence found in OKC associated with teeth¹⁴. Prognosis of our case is fair and chance of recurrence becomes high as chemical cauterisation was not performed since the lesion was provisionally treated as residual cyst. Regular follow up with radiographic examination is necessary with OKCs because of the potential for recurrence. OKCs usually recur within five years after surgery but they can recur more than 15 years also¹⁵. Since our case of OKC was in maxilla, chance of recurrence is even high.

IV. CONCLUSION:

To conclude, any cystic lesion of jaw should be evaluated carefully because of possibility of OKC. Clinical and radiological picture alone is not sufficient enough to diagnose OKC's. Since recurrence rate of OKC is high, has high tendency for malignant transformation and its aggressive growth pattern, surgeons should give best surgical treatment in the first surgery itself. OKC's are commonly misdiagnosed as radicular

cyst/dentigerous cyst and attempt to be made to accurately diagnose the lesion prior to primary surgical treatment. Hence careful history & in depth evaluation & investigations such as CBCT is extremely useful in accurate diagnosis of OKC. Prevention of recurrence by taking additional measures along with complete enucleation is needed to manage OKC.

REFERENCES:

- [1]. Ali M, Baughman RA. Maxillary odontogenic keratocyst: a common and serious clinical misdiagnosis. *J Am Dent Assoc* 2003;134:877-83.
- [2]. Cawson RA, Odell EW. *Essentials of oral pathology and oral medicine*. 6th ed. New York: Churchill Livingstone; 1998:97.
- [3]. Payne TF. An analysis of the clinical and histopathologic parameters of the odontogenic keratocyst. *Oral Surg Oral Med Oral Pathol* 1972;33:538-46.
- [4]. Panders AK, Hadders HN. Solitary keratocysts of the jaws. *J Oral Surg* 1969;27:931-8.
- [5]. Brannon RB. The odontogenic keratocyst: a clinicopathologic study of 312 cases, part I—clinical features. *Oral Surg Oral Med Oral Pathol* 1976;42:54-72.
- [6]. Shear M, Speight P. *Cysts of the Oral and Maxillofacial Region*. 4th ed. Singapore: Blackwell Munksgaard; 2007. p. 223.
- [7]. Haring JI, Van Dis ML (1988) Odontogenic keratocysts: a clinical, radiographic and histopathologic study. *Oral Surg Oral Med Oral Pathol* 66:145-153
- [8]. Brondum N, Jensen VJ (1991) Recurrence of keratocysts and decompression treatment. A long-term follow-up of forty-four cases. *Oral Surg Oral Med Oral Pathol* 72:265-269
- [9]. Myoung H, Hong SP, Hong SD, Lee JI, Lim CY, Choung PH, Lee JH, Choi JY, Seo BM, Kim MJ (2001) Odontogenic keratocyst: Review of 256 cases for recurrence and clinicopathologic parameters. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 91(3): 328-333
- [10]. Chandrashekar R, Bande M, C. Prashant, Bharat Sumbh, P. K. Pandilwar. Prevalence, Treatment and Recurrence of Odontogenic Keratocyst in Central India. *J Maxillofac Oral Surg*. 2010 Jun; 9(2): 146-149
- [11]. Shubhankar Dasgupta, Shalu Rai, Deepankar Misra, Sapna Panjwani, Nivedita Singh. Diagnostic dilemma: Radicular cyst or keratocystic odontogenic tumor? *SRM Journal of Research in Dental Sciences*. 2015 July-sept; 6(3)
- [12]. Koçak-Berberoğlu H, Çakarer S, Brkić A, Gürkan-Koseoglu B, Altuğ-Aydil B, Keskin C. *Med Oral Patol Oral Cir Bucal*. 2012 Nov 1; 17(6): e1000-5.
- [13]. Blans N, Freund B, Schwartz M, Furst IM. Systemic review of the treatment and prognosis of the odontogenic keratocyst. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2000;90:553-8
- [14]. 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;101(1):5-10.
- [15]. Vedtofte P, Praetorius F. Recurrence of the odontogenic keratocysts in relation to clinical and histological features: 20 year follow-up study of 72 patients. *Int J Oral Surg* 1979;8:412-20.

***Corresponding Author: ¹Dr. Kiruba Shankar K,**
¹Post graduate student, Department Of Oral and Maxillofacial Surgery, KVG Dental College and Hospital, Sullia, Karnataka, India.