



## UNCOMMON OCCURRENCE – ODONTOGENIC KERATOCYST: A CASE REPORT

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**(ABSTRACT)** Keratocystic odontogenic tumors (KCOT), often referred to as odontogenic keratocysts, are distinguished by their unusual behavior, diverse origin, contentious development, distinct propensity to recur, and contested treatment approaches. This article emphasizes the varied clinical presentation of Odontogenic keratocyst of a 52 year old female patient with history of trauma in relation to anterior mandible. In Orthopantomogram the lesion shows multilocularity mimicking botryoid cyst and CT reveals multilocular hypodense areas with buccal cortex perforations involving the inferior alveolar nerve canal. Histopathological findings of incisional biopsy confirms the diagnosis, after which patient was subjected to surgical enucleation.

**KEYWORDS :** Botryoid cyst, Odontogenic keratocyst, Multilocular radiolucency, Marginal resection.

### INTRODUCTION:

Odontogenic keratocysts (OKCs), first described by Philipsen in 1956<sup>[1]</sup>, are benign intraosseous lesions of odontogenic origin that account for about 10% of jaw cysts. OKC arises from the remnants of dental lamina either in mandible or maxilla. Posterior mandible is the most frequent site of involvement. OKCs are considered to be cystic neoplasms and termed as keratocystic odontogenic tumors in 2005<sup>[2]</sup>. They are characterised by an aggressive behaviour with a relatively high recurrence rate<sup>[3]</sup>. According to 4<sup>th</sup> edition of WHO classification of Head and Neck Tumours 2017, the term KCOT was replaced again with the term OKC<sup>[4]</sup>. This case report focuses on the 52 yrs old female patient presented with the mild swelling in the lower anterior region which is ultimately diagnosed as Odontogenic keratocyst.

### CASE REPORT:

A 52 yrs old female patient reported to the department of oral medicine & radiology at Meenakshi ammal dental college, Chennai, with the chief complaint of swelling in her right lower front teeth region for the past 10 days. Patient gave history of trauma in the lower front teeth region 6 months ago, which was associated with pain and pus discharge which subsided with medications. 10 days ago, patient developed swelling in same region which was associated with pain. Her past medical history was non-contributory. On clinical examination, evidence of altered vestibular morphology in 43,44 region measuring approximately 1.5x1cm in size extending anteroposteriorly. On palpation, it was soft in consistency, tenderness elicited and pus discharge from the gingival crevice in 44 region as shown in figure (1).



**Figure (1)** shows swelling in the attached gingiva of 43,44 region.

Correlating the history and clinical findings, provisional diagnosis of Infected dental cyst was given. Differential diagnosis of benign odontogenic cyst or tumor may be included.

### INVESTIGATIONS:

Electric pulp vitality testing was performed in which 42 showed delayed response (17 seconds) when compared with adjacent and contralateral teeth.

Fine needle aspiration revealed scanty blood tinged pus.

Orthopantomogram in 43,44 revealed evidence of multilocular radiolucencies with well defined cortical border and of size approximately 1.5cm in diameter in the interdental space and

periapical region of 42,43,44 as shown in figure (2) suggestive of odontogenic cyst or tumor.



**Figure 2:** (a) shows OPG, (b) Axial, (c) sagittal, (d) coronal sections of Computed tomography.

Patient was subjected to Computed Tomography of mandible reveals (Figure 2). In Axial section evidence of thinning and perforation of buccal cortical plates with intact lingual cortical plates. In sagittal section, evidence of multiple hypodense areas with internal septations in periapical region of 43,44 with intact inferior border of the mandible. In coronal section, evidence of multiple hypodense areas in the anterior body of the mandible with internal septations involving the inferior alveolar nerve canal with no displacement of the canal.

Routine blood investigations were performed, all the parameters were within normal limits.

Then patient was subjected to incisional biopsy and histopathological examination which revealed predominantly parakeratinized stratified squamous cystic lining epithelium exhibiting in foldings with surface corrugations, 3-4 cell layer thickness, consisting of cuboidal to columnar basal cells with hyperchromatic nuclei in palisaded pattern. Also focal areas exhibit ortho keratinized cystic lining epithelium. The cystic wall is dense fibro collagenous with 3-4 satellite cysts. Cystic lumen shows numerous flakes of keratin suggestive of odontogenic keratocyst.



**Figure 3** shows Postoperative OPG

Correlating the history given by the patient, clinical findings, radiographic features and histopathological diagnosis, final diagnosis of Odontogenic Keratocyst in relation to Right anterior mandible. Patient underwent marginal resection of the mandible in 41,42,43,44 region and splinting of the adjacent teeth were done. Patient was followed up and postoperative OPG (Figure 3) was taken after 1 month.

## DISCUSSION:

The Recent insights into the formation and biological behaviour of OKCs have prompted their reclassification from benign odontogenic tumors to developmental cysts of odontogenic origin<sup>[5]</sup>. They are more commonly observed in males, with a ratio of 2:1, and exhibit two notable peaks in occurrence typically around the ages of 30 and 60<sup>[6]</sup>. They predominantly manifest in the mandibular third molar region and ramus, the anterior mandible is a less frequent site for their occurrence<sup>[7]</sup>. OKCs originate from the epithelial lining of the dental follicle<sup>[8]</sup>. Furthermore, individuals with multiple lesions demonstrate a heightened recurrence rate (16.6% to 21.1%) for OKCs, indicating an elevated likelihood of malignancy<sup>[9]</sup>. OKC is frequently asymptomatic and is incidentally detected during routine radiographic assessment<sup>[10]</sup>. It appears as distinctly outlined radiolucent areas within the jaw bones, varying in appearance from unilocular to multilocular, and typically displaying a tendency for growth in the anteroposterior direction<sup>[11]</sup>. Different radiographic classifications of OKC include Replacement, Extraneous, Envelopmental, and Collateral types. Among these, the envelopmental subtype bears resemblance to dentigerous cysts, while the collateral subtype shares similarities with lateral periodontal cysts<sup>[12]</sup>. OKCs linked to retained teeth tend to exhibit higher recurrence rates, thus recommending the excision of both the tooth and the cyst to minimize recurrence risks<sup>[13,14]</sup>. Studies have indicated that Hounsfield unit (HU) values on CT scans are notably lower for OKCs compared to other types of odontogenic cysts. This distinction can enhance early-stage diagnosis, potentially averting recurrences in later stages<sup>[15]</sup>. In MRI, they often display thin and consistent rim-enhancement on T1-weighted images. MRI employing diffusion-weighted imaging (DWI) and the subsequent calculation of apparent diffusion coefficient (ADC) is sensitive to physiological factors such as tissue cellularity, nucleus-to-cytoplasm ratio, and the integrity of cell membranes. This technique offers insights into the microstructure of living tissues<sup>[16]</sup>. The apparent ADC value observed in OKCs typically presents lower compared to cystic or predominantly cystic ameloblastomas. Recurrences are often linked to the proliferative activity of satellite cysts, formed through basal cell budding within the epithelium. These cysts detach into the connective tissue, leading to subsequent cyst formation<sup>[18]</sup>. It has been suggested that inflammation leads to increased epithelial thickness contributes to recurrences in OKC<sup>[19]</sup>. In odontogenic conditions, hypoxia-inducible factor-1 $\alpha$  (HIF-1 $\alpha$ ) serves as an indicator of hypoxia and has been linked to invasiveness and cystic formation. Elevated levels of this transcription factor have been documented in OKCs<sup>[20]</sup>. Elevated Ki67 expression and an increased AgNOR count have shown a robust correlation with a heightened likelihood of OKC recurrence<sup>[21]</sup>. Stratifying OKCs based on their clinicopathological characteristics for recurrence risk assessment holds significant importance in patient management. Treatment options encompass a spectrum ranging from conservative approaches like enucleation (with or without curettage), decompression, and marsupialization, to more aggressive methods such as peripheral osteotomy, cryotherapy, application of Carnoy's solution, and mandibular resection<sup>[22,23]</sup>.

Lesions categorized as high risk should undergo more aggressive treatment approaches such as enucleation or resection, whereas low-risk lesions may be managed more conservatively through marsupialization/decompression or enucleation alone<sup>[24]</sup>. The objective of all these techniques is to eradicate the cyst and mitigate the risk of recurrence.

## CONCLUSION:

Understanding both common and unusual radiological characteristics of OKCs are vital for diagnosing and planning treatment. Therefore, a collaborative approach involving various disciplines is necessary to classify these features accurately, enabling the selection of the most suitable management approach and reducing the likelihood of recurrences.

**Patient Consent:** Obtained

**Conflicts Of Interest:** None

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## REFERENCES:

- Philipsen HP (1956) Om keratocyster (kolesteratomer) i kaeberne. Tandlaegebladet 60:963-971
- Sivapathasundharam B, Rajendran A. Shafer's textbook of oral pathology. Elsevier Health Sciences; 2012 Jun 30.
- Antonoglou GN, Sándor GK, Koidou VP, Papageorgiou SN (2014) Non-syndromic and syndromic keratocystic odontogenic tumors: systematic review and meta-analysis of recurrences. J Craniomaxillofac Surg 42:e364-e371
- El-Naggar AK, Chan JKC, Grandis JR, Takata T, Slootweg PJ (2017) WHO classification of head and neck tumours, 4th edn. IARC Press, Lyon
- Hornillos-de Villota M, Pampin-Martínez MM, Moran-Soto MJ, Cebrián-Carretero JL. Peripheral odontogenic keratocyst. A Case report. J Clin Exp Dent. 2023 Feb 1;15(2):e169-e172.
- Rajkumar, G C; Hemalatha, M1; Shashikala, R; Sonal, P. Massive keratocystic odontogenic tumor of mandible: A case report and review of literature. Indian Journal of Dental Research 22(1);p 181, Jan–Feb 2011. | DOI: 10.4103/0970-9290.80000
- Stoelinga PJ. Long term follow up on keratocysts treated according to defined protocol. Int J Oral Maxillofac Surg 2001;30:14-25
- Menon S. Keratocystic odontogenic tumours: etiology, pathogenesis and treatment revisited. J Maxillofac Oral Surg 2015;14(3):541-7.
- Chrcanovic BR, Gomez RS. Recurrence probability for keratocystic odontogenic tumors: an analysis of 6427 cases. J Craniomaxillofac Surg. 2017;45(2):244-51.
- Pazdera J, Kolar Z, Zboril V, Tvrdy P, Pink R. Odontogenic keratocysts/keratocystic odontogenic tumours: biological characteristics, clinical manifestation and treatment. Biomed Pap Med Fac Univ Palacky Olomouc Czech Repub. 2014;158(2):170-4
- Bilodeau EA, Collins BM. Odontogenic cysts and neoplasms. Surg Pathol Clin. 2017;10(1):177-222.
- Santosh AB. Odontogenic cysts. Dental Clinics. 2020 Jan 1;64(1):105-19.
- Cunha, J. F., Gomes, C. C., de Mesquita, R. A., Andrade Goulart, E. M., de Castro, W. H., & Gomez, R. S. (2016). Clinicopathologic features associated with recurrence of the odontogenic keratocyst: A cohort retrospective analysis. Oral Surgery Oral Medicine Oral Pathology Oral Radiology, 121(6), 629–635.
- Fidele, N. B., Yueyu, Z., Zhao, Y., Tianfu, W., Liu, J., Sun, Y., & Liu, B. (2019). Recurrence of odontogenic keratocysts and possible prognostic factors: Review of 455 patients. Medicina Oral Patologica Oral Cirugia Bucal, 24, e491–e501.
- Uehara, K., Hisatomi, M., Munhoz, L., Kawazu, T., Yanagi, Y., Okada, S., Takeshita, Y., Saito, E. A., & Asaumi, J. (2021). Assessment of Hounsfield unit in the differential diagnosis of odontogenic cysts. Dentomaxillofacial Radiology, 50(2), 20200188. <https://doi.org/10.1259/dmfr.20200188>
- Borghesi A, Nardi C, Giannitto C, Tironi A, Maroldi R, Di Bartolomeo F, Preda L. Odontogenic keratocyst: imaging features of a benign lesion with an aggressive behaviour. Insights into imaging. 2018 Oct;9:883-97.
- Sumi M, Ichikawa Y, Katayama I, Tashiro S, Nakamura T (2008) Diffusion-weighted MR imaging of ameloblastomas and keratocystic odontogenic tumors: differentiation by apparent diffusion coefficients of cystic lesions. AJNR Am J Neuroradiol 29: 1897–1901
- Bello IO (2016) Keratocystic odontogenic tumor: a biopsy service's experience with 104 solitary, multiple and recurrent lesions. Med Oral Patol Oral Cir Bucal 2(5):538–546
- de França, G. M., da Silva, L. B. A., Mafra, R. P., da Silva, W. R., de Lima, K. C., & Galvão, H. C. (2021). Recurrence-free survival and prognostic factors of odontogenic keratocyst: A single-center retrospective cohort. European Archives Otorhinolaryngology, 278(4), 1223–1231.
- Costa NMM, Abe CTS, Mitre GP et al (2019) HIF-1 $\alpha$  is overexpressed in odontogenic keratocyst suggesting activation of hif-1 $\alpha$  and notch1 signaling pathways. Cells 8(7):1–13
- Selvi, F., Tekkesin, M. S., Cakarar, S., Isler, S. C., & Keskin, C. (2012). Keratocystic odontogenic tumors: Predictive factors of recurrence by Ki-67 and AgNOR labelling. International Journal of Medical Sciences, 9(4), 262–268.
- Sigua-Rodriguez EA, Goulart DR, Sverzut A et al (2019) Is surgical treatment based on a 1-step or 2-step protocol effective in managing the odontogenic keratocyst? J Oral Maxillofac Surg 77(6):1210.e1–1210.e7
- Kinard BE, Chuang SK, August M et al (2013) How well do we manage the odontogenic keratocyst? J Oral Maxillofac Surg 71(8):1353–1358
- Titinchi F. Novel recurrence risk stratification of odontogenic keratocysts: A systematic review. Oral Diseases. 2022 Oct;28(7):1749-59.