



MANDIBULAR ORTHOKERATINIZED ODONTOGENIC CYST (OOC): A CASE REPORT OF AN UNCOMMON ODONTOGENIC CYST

Dr Krishna kishor

Senior Lecturer, Dept Of Oral & Maxillofacial Surgery, Buddha institute of dental sciences & hospital, Patna

Dr Hiralal Ash

Professor & Head, Dept Of Oral & Maxillofacial Surgery, Buddha institute of dental sciences & hospital, Patna

Dr Kumar Arunesh

Professor, Dept Of Oral & Maxillofacial Surgery, Buddha institute of dental sciences & hospital, Patna

Dr Heera*

BDS, MDS, Oral pathologist, Patna*Corresponding Author

ABSTRACT

Orthokeratinized odontogenic cyst (OOC) is an odontogenic cyst was initially termed as the uncommon orthokeratinized type of odontogenic keratocyst by the World Health Organization (WHO). Mandible is the commonest site of OOC. Numerous studies shown that OOC has some similar characteristic clinico-pathological features when compared to other developmental odontogenic lesions such as dentigerous cyst and the keratocystic odontogenic tumour (KCOT). It was noted that after the surgical removal of orthokeratinized odontogenic cyst (OOC) having low rate of recurrence. The purpose of this case report of OOC that arised in the posterior mandible and emphasize on differentiating it from the KCOT and dentigerous cyst.

KEYWORDS : ODONTOGENIC CYST, ORTHOKERATINIZED ODONTOGENIC CYST, KERATOCYSTIC ODONTOGENIC TUMOUR

INTRODUCTION

Wright (1981) first identified the orthokeratinized variant of odontogenic cyst which is now known as orthokeratinized odontogenic cyst [1]. The term orthokeratinized odontogenic cyst coined by the Li et al which also reflect its most plausible histologic origin [2]. Wright explained that OOC having different histopathology and reduce chance to recur than odontogenic keratocyst (OKC) [3]. According to reclassification of odontogenic cyst by WHO in 2005, Odontogenic keratocyst (OKC) to keratocystic odontogenic tumour (KCOT) as parakeratinized type and stated "cystic jaw lesion that are lined by orthokeratinized epithelium do not form part of the spectrum of keratocystic odontogenic tumour (KCOT) [4]. The frequency of keratocystic odontogenic tumour (KCOT) in oral cavity is eight times more than that of orthokeratinized odontogenic cyst (OOC).

CASE REPORT

A 27 years old female reported to our institution with a chief complaint of swelling in the lower right back region of jaw for the past 9 months.

On extra oral examination no gross facial asymmetry was present. Diffuse swelling was present in the right parasymphysis region of 3 cm × 2 cm approx in size, hard in consistency and non tender on palpation. Buccal cortical plate expansion was evident with crepitus at some area.

On Intra-oral examination mild swelling was present extending from lower right lateral incisor to left first molar region. Orthopantomogram showed a well-defined radiolucency with corticated border in the body of the mandible extending from 43 to 46 regions and extending up to 2mm above the inferior border of the mandible with impacted premolar. Over-retained deciduous molar was present in lower right mandible.

Cone beam computer tomography showed an expansile osteolytic lesion with buccal cortical plate.

Provisional diagnosis of dentigerous cyst with impacted

premolar was suggested with differential diagnosis of KCOT, unicystic ameloblastoma and orthokeratinized OKC.

After all routine laboratory investigation incisional biopsy was planned. All laboratory findings were within normal limit.

Incisional biopsy was done under local anesthesia 2% lignocaine with adrenaline. Histopathological finding was orthokeratinized odontogenic cyst (OOC).

Due to low recurrence rate than KCOT surgical enucleation of the lesion was planned. Surgical enucleation with impacted premolar removal was done through intraoral buccal vestibular approach under general anesthesia. After the enucleation gross examination of the excised specimen revealed a thin cystic sac with luminal surface. The lumen also contained white cheesy material.

Microscopic histopathological examination of the specimen revealed an orthokeratinized odontogenic epithelium and underlying connective tissue. Overlying hyperplastic epithelium was 6-8 cell layered with flat interface between epithelium and connective tissue.

DISCUSSION

Orthokeratinized odontogenic cyst (OOC) is a developmental cyst of odontogenic origin and was initially defined as the uncommon orthokeratinized variant of odontogenic keratocyst (OKC). Orthokeratinized odontogenic cyst having some similarity with keratocystic odontogenic tumour on microscopic basis i.e equal epithelium lining with keratinization, these two cysts are different from various aspects as orthokeratinization and parakeratinization.

According to previous literature the incidence of orthokeratinized odontogenic cyst (OOC) ranging from 5.2-16.8% among cases which had been previously coded as KCOT [5]. OOC has been reported to occur in the third and fourth decades of life [3]. The mandible involvement is more common than that of maxilla [6]. According to reported literature, OOC was located in the body of mandible, starting from lower canine region involving lower premolars and lower first molar.

Swelling is the main clinical feature with or without pain in cases of OOC. In present case the lesion extending from lower right lateral incisor to lower right molar area [7].

The size of the orthokeratinized odontogenic cyst varies from greater than 1 cm and less than 7 cm [8].

The character of Orthokeratinized odontogenic cyst resembles clinically and radiographically representing dentigerous cyst as they most often involved an unerupted mandibular third molar [9].

Swelling is the most frequent symptom and is accompanied with pain although in most of the cases, the lesion is asymptomatic. Large lesions can cause cortical expansion [10].

On radiological examination orthokeratinized odontogenic cyst appears well circumscribed unilocular or multilocular radiolucency that some time associated with impacted tooth or with the tooth causing root resorption, with or without expansion of cortical plate/displacement of inferior alveolar canal [10].

On histological examination orthokeratinized odontogenic cyst is lined by 4-9 layers of stratified squamous epithelium which is uniformly distributed and basal layer cell that exhibits palisade cuboidal or flat cells, with nuclear hyperchromatism [11]. There are so many other lesions of jaw like dentigerous cyst, unicystic ameloblastoma and KCOT resembles with orthokeratinized odontogenic cyst but OOC usually shows no root resorption of involved teeth, which is a characteristic feature of ameloblastoma [7].

Etiopathogenesis

The orthokeratinized odontogenic cyst (OOC) is a developmental odontogenic cyst relatively rare; arising from the cell rests of the dental lamina.

Kotwany S et al., had shown the role of reduced enamel epithelium in formation of dentigerous cyst that had completed its tooth-forming function and which had the capability to keratinize under appropriate stimuli. This is how the true dentigerous cyst with keratinization may be formed [12]. According to Zhu study, they stated that like KCOT, which arise from the dental lamina with the presence of the dental papilla, the OOC may arise from oral epithelium under the influence of the dental papilla or only the oral epithelium. Due to this different histogenesis of KCOTs and OOCs further investigation is required. Other studies suggested that KCOT originate from dental lamina. This would explain their common occurrence in the posterior mandible, because the dental lamina is more active in this area at the age when many patients develop their cysts [9,11].

Immunohistochemistry

The KCOT is clinically more aggressive than other forms of odontogenic cyst and tends to recur after surgery. The recurrence in KCOT has varied from 12% to 60%. These may be due to significantly less expression of p63 in OOCs than KCOTs. p63 which is a member of the p53 tumor suppressor gene family, plays a major role in the terminal differentiation of epithelial stem cells as well as their maintenance. Due to less significant expression of p63 in OOC, epithelial cells have less proliferative and self-renewal potential. p63 expression usually seen be more intensive and diffuse in malignant odontogenic tumors and benign odontogenic tumors, which showed local aggressiveness in comparison of other odontogenic tumors. Even recent immunocytochemical results demonstrated fewer Ki-67-positive proliferating cells, which are mostly confined to the basal cell layer in the epithelial linings of OOC than KCOT. Thus, KCOT lining demonstrate more prominent suprabasal proliferative activity than that of ooc [13,14].

Treatment

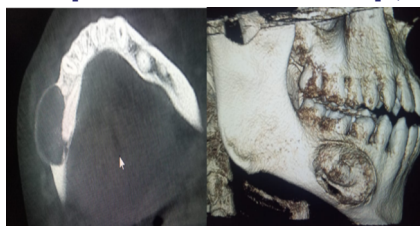
According to reported literature Surgical enucleation of lesion with removal of involved impacted teeth is the line of treatment. Follow-up of the enucleated lesion showed no recurrence. In this case we also enucleated the lesion with removal of associated impacted teeth [15].

Very limited information is present about orthokeratinized odontogenic cyst due to very low prevalence rate and due to lack of specific clinical and radiographic features. More study is required for orthokeratinized odontogenic cyst to find out etiopathogenesis and clinical-radiographic feature of this lesion. OOC should be considered always in the differential diagnosis of all the radiolucent lesions involving impacted teeth.



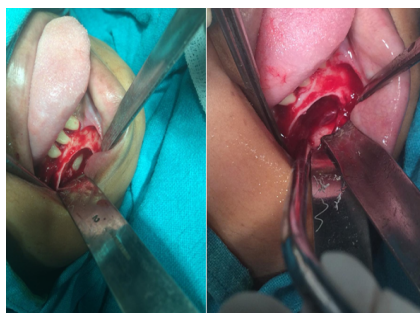
Pre op facial profile

Pre op (intra oral)



Pre op CBCT

Pre op CBCT



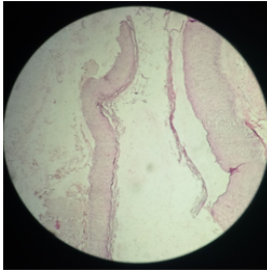
Intra operative (a)

Intra operative (b)



Tissue after enucleation

Impacted tooth



Histopathologic image showing cystic epithelium lined by orthokeratin (H & E) Low Magnification

BIBLIOGRAPHY

1. Wright JM. The odontogenic keratocyst: orthokeratinized variant. *Oral Surg Oral Med Oral Pathol* 1981;51:609-618.
2. Li TJ, Kitano M, Chen XM, et al. Orthokeratinized odontogenic cyst: a clinicopathological and immunocytochemical study of 15 cases. *Histopathology*. 1998;32(3):242-251
3. Sandhu SV, Rao SK, Brar RS, Kakkar T. Orthokeratinized odontogenic cyst of the mandible: A case report. *Int J Oral Maxillofac Pathol*. 2012;3:69-73.
4. Philipsen HP. Keratocystic odontogenic tumour.
5. Barnes L, Eveson J, Reichart P, Sidransky D (eds) WHO Classification of Tumours. Pathology and genetics of tumours of the Head and Neck. International Agency for Research on Cancer (IARC), Lyon, 2005, pp 306-307.
6. Dong Q, Pan S, Sun LS, Li TJ. Orthokeratinized odontogenic cyst: A clinicopathologic study of 61 cases. *Arch Pathol Lab Med* 2010;134:271-5.
7. Swain N, Shilpa P, Poonja LS, Pathak J, Dekate K. Orthokeratinized odontogenic cyst. *J Contemp Dent* 2012;2:31-3.
8. MacDonald-Jankowski DS, Li TK. Orthokeratinized odontogenic cyst in a Hong Kong community: The clinical and radiological features. *Dentomaxillofac Radiol*. 2010;39:240-5. [J]
9. Neville BW, Damm DD, Allen CM, Bouquot JE. Odontogenic cyst and tumors. In: Neville BW, editor. *Oral and Maxillofacial Pathology*. 3rd ed. New Delhi: Reed Elsevier India Private Limited; 2013. p. 683-8.
10. Sivapathasundharam B, Rajendran R. Cysts and tumors of odontogenic origin. In: Rajendran R, editor. *Shafer's Textbook of Oral Pathology*. 7th ed. New Delhi: Reed Elsevier India Private Limited; 2012. p. 263-7.
11. González Galván Mdel C, García-García A, Anitua-Aldecoa E, Martínez-Conde Llamas R, Aguirre-Urizar JM. Orthokeratinized odontogenic cyst: A report of three clinical cases. *Case Rep Dent* 2013;2013:672383.
12. Philipsen HP. Keratocystic odontogenic tumour. In: Barnes EL, Eveson JW, Reichart P, Sidransky D, editors. *Pathology and Genetics of Head and Neck Tumors*. World Health Organization Classification of Tumors. Lyon, France: IARC Press; 2005. pp. 306-7.
13. Pillai AK, Gupta MK, Mhaske SJ, Satpathy M, Singh SK. An aggressive orthokeratinized odontogenic cyst of the mandible: A case report. *IOSR J Dent Med Sci*. 2013;12:45-8.
14. Li TJ, Browne RM, Matthews JB. Epithelial cell proliferation in odontogenic keratocysts: A comparative immunocytochemical study of Ki67 in simple, recurrent and basal cell naevus syndrome-associated lesions. *J Oral Pathol Med*. 1995;24:221-6
15. Senoo M, Pinto F, Crum CP, McKeon F. p63 is essential for the proliferative potential of stem cells in stratified epithelia. *Cell*. 2007;129:523-36.
16. Li TJ, Browne RM, Matthews JB. Quantification of PCNA + cells within odontogenic jaw cyst epithelium. *J Oral Pathol Med*. 1994;23:184-9