



ORTHOKERATINIZED ODONTOGENIC CYST MIMICKING AS DENTIGEROUS CYST - CASE REPORT

Dental Science

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ABSTRACT

Orthokeratinized Odontogenic Cyst (OOC) is an uncommon developmental odontogenic cyst which exhibits peculiar clinical and pathological aspects in comparison with other developmental odontogenic cysts like Dentigerous cyst and Odontogenic keratocyst. The diagnosis of OOC is important as the pathologic behaviour, clinical outcome and the surgical management of OOC is disparate. The purpose of the article is to present a case of OOC radiographically mimicking Dentigerous cyst in a 20 year old male.

KEYWORDS

Orthokeratinized, odontogenic cyst, keratocyst, impacted, mandibular

INTRODUCTION

Orthokeratinized odontogenic cyst (OOC) is a uncommon developmental odontogenic cyst which arises from the cell rests of the dental lamina.^[1,2] In 1927, Schultz described OOC as an *orthokeratinized variant of the odontogenic keratocyst*. World Health Organization [WHO] in the year 1992 defined OOC as the uncommon orthokeratinized type of odontogenic keratocyst (OKC) and finally in the year 1998 Li et al. coined the term "orthokeratinized odontogenic cyst," which is accepted till date.^[3]

Since Odontogenic keratocyst is considered as a neoplasm and termed as Keratocystic odontogenic tumour [KCOT] by the WHO, it is necessary that both the clinicians and the pathologists have a thorough knowledge about the differences between the aggressive KCOT and OOC.

Here we report a case of OOC in the mandible radiographically mimicking lateral type of Dentigerous cyst.

CASE REPORT

A 20 year old male patient presented to our college with a chief complaint of dull intermittent type of pain in the left posterior region of the jaw for the past one month. His past medical and dental history was non-contributory and general physical status was good with all vital signs within normal limits. Routine blood investigations revealed no abnormality in any of the parameters.

Intra-oral examination revealed tenderness in the alveolar ridge in relation to partially erupted 38. Panoramic radiographic survey of the region revealed an oval shaped unilocular radiolucency partly surrounding the distal aspect of the crown of 38 and extending up to the middle third of the distal root of 38 [Figure 1].

Based on the radiographic evaluation, a preliminary diagnosis of lateral type of dentigerous cyst was made. Under local anesthesia, the lesion was surgically enucleated along with the extraction of 38 and submitted for histopathological analysis.

Gross examination of the specimen revealed an oval cystic sac attached to the distal aspect of crown of 38 and was measuring about 1 cm in diameter. The cut section of the sac showed white cheesy material within the central lumen [Figure 2].

Microscopic evaluation revealed a cystic cavity lined by thin orthokeratinized stratified squamous epithelium. The epithelial lining was about 5-6 layers thick with a distinct granular cell layer and the basal layer was cuboidal to flat with absence of palisading of nuclei. The surface of the lining epithelium showed sheaves of keratin arranged in many layers. The epithelial and connective tissue wall

interface was flat without any rete-ridges. The connective tissue wall was made of dense collagen fibers and was relatively free from inflammation [Figure 3,4].

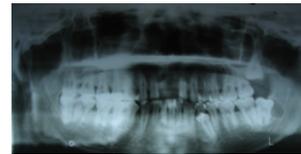


Figure 1: Orthopantomogram showing unilocular radiolucency in relation to an impacted 38



Figure 2: Enucleated cut gross specimen

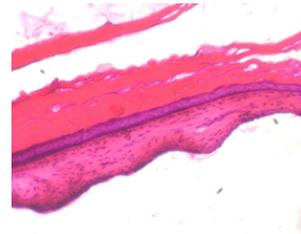


Figure 3: Cystic wall lined by Orthokeratinized Stratified squamous epithelium with many layers of sheaves of orthokeratin [H & E, 10X]

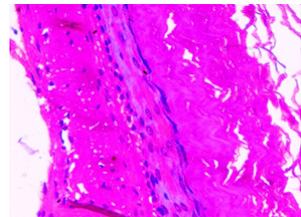


Figure 4: Cystic wall lined by Orthokeratinized Stratified squamous epithelium with prominent granulosum layer [H & E, 40X]

Based on the radiographic and histopathological features, diagnosis of orthokeratinized odontogenic cyst was made. The patient was followed up after 3 years and no sign of recurrence was noted.

DISCUSSION

Odontogenic cysts are the commonest cyst which are encountered in dental practise and comprise a major aspect in oral pathology. It is considered as developmental in origin if one of the rests of odontogenic cells such as the cell rests of Malassez, cell rests of Serres or the enamel organ gets activated and proliferates inside the bone or gingiva. Some odontogenic cysts are locally aggressive and are more prone to recur like keratocystic odontogenic tumor (KCOT), so therefore proper diagnosis of such lesions is essential for correct treatment and for ensuring adequate follow-up. [4]

Orthokeratinized odontogenic cyst (OOC) is rare developmental odontogenic cyst arising from the cell rests of Serres. OOC has been designated as a discrete clinicopathological entity by the World Health Organization based on its peculiar clinicoradiological and pathologic facets. [4]

OOC constitutes to about 11% of all jaw cysts and is about 5.2% to 16.8% of all cases which were previously diagnosed as OKC. [5] OOC is predominantly seen in males between the 3rd and 4th decades. It is mostly commonly seen in the molar – ramus region of mandible and usually associated with an unerupted tooth.

The lesion is usually asymptomatic and slow growing, and occasionally associated with pain. Radiographically it appears as a well defined unilocular or multilocular radiolucency that is occasionally concordant with an unerupted tooth or root. [6]

Histopathologically, the cystic cavity of OOC is lined by a thin orthokeratinized stratified squamous epithelium of 4-9 layers thick with a prominent granulosum layer. The basal cells appear either low cuboidal or flat and do not exhibit palisading arrangement like KCOT. [6,7]. The connective tissue wall do not exhibit satellite or daughter cysts like KCOT.

Recent Immunohistochemical studies between OOC and KCOT showed lower expression of P53, Ki-67, Bcl-2, and TGF- α in OOC than KCOT, reflecting the anti-apoptotic and reduced proliferative activity in OOC. [8]

Conservative surgical removal by enucleation and curettage is the treatment of choice for OOC with a recurrence rate of 2.2%. [6]

In our case, the lesion was conservatively enucleated and was periodically followed-up with no recurrence reported till date.

CONCLUSION

In our case, the lesion radiographically appeared as unilocular radiolucency surrounding partially impacted 38 and was contemplated to be Dentigerous cyst, but microscopically it fitted in favour of OOC.

In conclusion it is very challenging in diagnosing odontogenic cyst which gives a misleading radiographic appearance, and therefore a thorough histopathological and radiographic concurrence is necessary for obtaining the final diagnosis, and also for appropriate treatment planning.

Conflict of Interest:

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

REFERENCES

1. Byatnal A, Natarajan J, Narayanaswamy V, Radhakrishnan R. Orthokeratinized odontogenic cyst -critical appraisal of a distinct entity. *Braz J Oral Sci* 2013;12:71-5.
2. T.-J. Li, M. Kitano, X.-M. Chen et al., "Orthokeratinized odontogenic cyst: a clinicopathological and immunocytochemical study of 15 cases," *Histopathology*, vol. 32, no. 3, pp. 242-251, 1998.
3. Wright JM. The odontogenic keratocyst: Orthokeratinised variant. *Oral Surg Oral Med Oral Pathol* 1981;51:609-18.
4. Selvamani M, Devi AY, Basandi PS, Madhushankari GS. Prevalence and clinicopathological comparison of keratocystic odontogenic tumor and orthokeratinized odontogenic cyst in South Indian sample population: A retrospective study over 13 years. *J Pharm Bioallied Sci* 2014;6:S127-30.
5. 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.
6. González Galván Mdel C, García-García A, Anitua-Aldecoa E, Martínez-Conde Llamosas R, Aguirre-Urizar JM. Orthokeratinized odontogenic cyst: A report of three clinical cases. *Case Rep Dent*. 2013;2013:672383.

7. da Silva MJ, de Sousa SO, Corrêa L, Carvalhosa AA, De Araújo VC. Immunohistochemical study of the orthokeratinized odontogenic cyst: A comparison with the odontogenic keratocyst. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2002;94:732-7.
8. Marx RE, Stern D. Odontogenic and nonodontogenic cysts. In: Marx RE, editor. *Oral and Maxillofacial Pathology a Rationale for Diagnosis and Treatment*. Vol. II. 2nd ed. China: Quintessence Publications Co. Inc.; 2012. p. 616-31.