


CASE REPORT

Odontogenic Keratocyst Mimicking a Radicular Cyst

Soukaina Essaket , Laila Benjelloun, Saliha Chbicheb
Faculty of Dentistry, Mohammed Vth University in Rabat, Rabat, Morocco

ABSTRACT

The odontogenic keratocyst (OKC) is a benign intraosseous cyst of the odontogenic origin, characterized by an aggressive behavior with a relatively high recurrence rate. OKC is mostly presented at the 2nd and 3rd decades and predominantly affect males. It affects mandible more than maxilla and is frequently found at the angle of mandible. OKC can clinically and radiographically mimic other benign and less aggressive lesions. It is therefore important to differentiate them from other cysts and tumors, for a proper treatment and reduction of recurrences. We report the case of a 19-year-old woman who presented with a swelling in left mandible. The clinical and radiographic features were in favor of a radicular cyst. Histological examination, however, contradicted this diagnosis, indicating a case of OKC.

KEYWORDS: Odontogenic Keratocyst; Radicular Cyst; Odontogenic Cysts.

Correspondence: Dr Essaket Soukaina, Consultation Center and of Dental Treatment, Faculty of Dental Medicine, Avenue Allal el Fassi, Mohammed Jazouli Strert- Cité Al Irfane - BP 6212 Rabat, Morocco. Email: Soukainaessaket93@gmail.com

Copyright © 2020 Essaket S et al. This is an open access article distributed under the [Creative Commons Attribution 4.0 International](https://creativecommons.org/licenses/by/4.0/), which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited.

INTRODUCTION

The odontogenic keratocyst (OKC) is a benign intraosseous cyst of the odontogenic origin, with aggressive and invading behavior (1). Previously classified under developmental odontogenic cyst of the jaw by the WHO in 1971 and 1992, OKC was reclassified and renamed as keratocystic odontogenic tumor by the WHO classification of jaw tumors in 2005 because of its aggressive nature and growth pattern and mutational changes. However, since 2017, the keratocystic tumor have again been classified as odontogenic keratocyst due to their pathogenic cystic process (2, 3, 4, 5)

After radicular and dentigerous cysts, odontogenic keratocysts are the third most common cyst and represent approximately 12-14% of all odontogenic cysts of the jaws. (1, 6)

OKC is mostly presented at the 2nd and 3rd decades and predominantly affect males. It affects mandible more than maxilla and is frequently found at the angle of mandible. (2, 7)

OKCs are characterized by their local aggressive potential and their high recurrence rate after treatment but can clinically and radiographically mimic other benign and less aggressive lesions. It is therefore important to differentiate them from other cysts and tumors, for a proper treatment and reduction of recurrences. (2,3,4)

The aim of this article is to report the case of a 19 years old patient who presented with an OKC mimicking clinically and radiologically a radicular cyst.

CASE REPORT

A 19-year-old female patient presented with the chief complaint of a swelling in left mandible. Her medical history was noncontributory.

She first noticed the swelling about 5 months ago with episodes of abscesses that resolved following a course of antibiotic but with the persistence of a mild swelling.

On extraoral examination, a swelling on the left cheek region was noticed, covered with a normal skin. (Figure 1)



Figure 1: Extraoral Image Showing Swelling on the Left Cheek Region

Intraoral examination showed a vestibular swelling which extends anteriorly from the mesial aspect of 35 to the distal aspect of 37 posteriorly. The color of the overlying mucosa was normal. On palpation, the swelling was fluctuating due to possible perforation of the cortical vestibular bone. (figure2).

A panoramic radiograph was prescribed and showed a well circumscribed unilocular lesion in the left premolar-molar region of the mandible, with sclerotic margins, located in the periapical region of tooth 36, which was root

state. The lesion extended from mesial of tooth 37 to mesial of tooth 33. The mental foramen and the mandibular canal could not be clearly seen.

On CBCT, multiplanar reconstructions showed a 35mmx20mm hypodense, well-circumscribed lesion in the left region of the mandible which was homogenous with perforation of the vestibular and lingual cortical bone and pushing back the mandibular canal and the mental foramen (figure 4).



Figure 2: Intraoral Image Showing a Swelling in the Left Premolar-Molar Region of the Left Mandible.



Figure 3: Panoramic Radiograph Showing a Unilocular Radiolucent Lesion in the Periapical Region of 36.

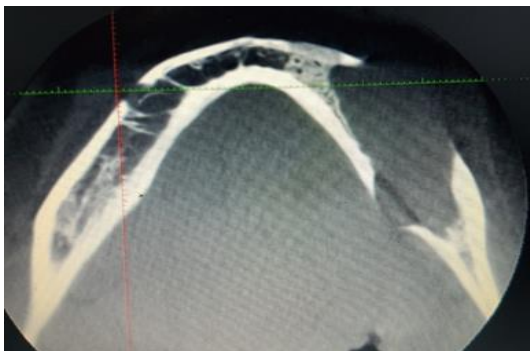


Figure 4: Axial View of the Cbct Showing A Hypodense Lesion in the Left Region of the Mandible with Perforation of the Vestibular and Lingual Cortical Bone.

An aspiration test of the lesion with a 2,5 syringe was also done that showed the presence of a citrus cystic liquid. Based on clinical and radiographic presentation of a well-defined radiolucency, located in the periapical position relating to the roots of non-vital tooth 36, and with liquid content, the first differential diagnosis was radicular cyst, but other lesions such as keratocyst or a unicystic ameloblastoma couldn't be eliminated.

Because of the extension of the lesion and its close contact to the mental nerve and inferior alveolar nerve, and the

young age of the patient, treatment by decompression was decided.

Histopathological examination revealed the diagnosis of an Odontogenic Keratocyst (figure 5).

A follow up after 5 months of decompression showed a great result, and the almost total disappearance of the lesion (figure 6). A complementary enucleation was necessary to avoid recurrences.

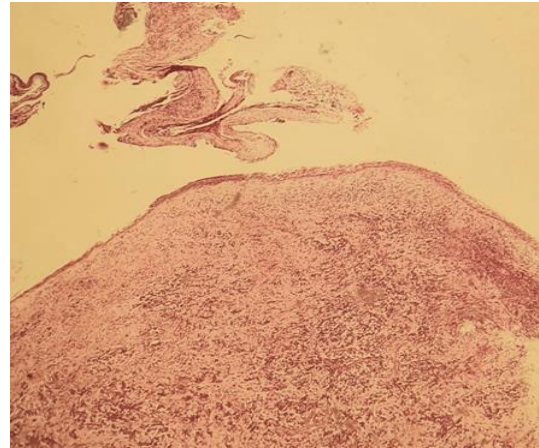


Figure 5: Histological Picture: Cyst Wall With a Keratinizing Squamous Border Resting on an Inflammatory Base (Hematoxylin-Eosin X40).



Figure 6: Panoramic Radiograph 5 Months after Decompression.

DISCUSSION

The term Odontogenic Keratocyst (OKC) was first coined by Philipsen in 1956 (Eryilmaz et al., 2009) and its characteristic features were first described by Pindborg and Hansen in 1963 (Pindborg and Hansen, 1963). Over the years, many researchers have been trying to understand the nature, identification, and management of the OKC leading to classify, and reclassify the disease. In 2017, the new WHO classification of Head and Neck pathology classifies it into the cystic category.

According to WHO, it is a benign uni or multicystic intraosseous tumor of the odontogenic origin with characteristic lining of stratified squamous epithelium and potential for aggressive and infiltrative behavior, and high recurrence rate (25% to 62.5%) (2.8)

OKC's arise from the proliferation of remnants of the dental lamina, or from proliferation of cells from the basal layer of oral epithelium into the mandible or maxilla, as an

intraosseous lesion associated or not with an unerupted tooth, mainly in the tooth bearing area, mainly in the posterior region of the jaws (3rd molar, angle and ramus in the mandible and 3rd molar/maxillary tuberosity in the maxilla). (9) Location is most commonly seen twice in mandible as compared to maxilla. (8)

OKC accounts for approximately 7.8 % of all cysts of the jaw. It occurs at all ages with peak incidence in 2nd and 4th decade of life. It predominantly occurs in white population with male: female ratio of 1.6:1. (8)

OKC is usually asymptomatic when the lesion is small and is often discovered only during radiographic examination (10). However, as the cyst enlarges, it may present with swelling, pain and purulent discharge if infected. Expansion of the buccal and lingual cortex is often not observed in OKC, in the initial stages, as it tends to grow in an anterior posterior direction within the medullary cavity of the bone (7,11). Patients may develop trismus in case of larger OKC's in the mandibular ramus. Larger OKC's in the maxilla may expand into the maxillary sinus and cause nasal obstruction on the same side. (9)

In our case, the OKC presented with buccal swelling and episodes of infections.

The presence of multiple OKCs is typically associated with the nevoid basal cell carcinoma syndrome (NBCCS), also known as Gorlin-Goltz syndrome, an autosomal dominant multisystemic disease (12).

Multiple keratocysts have also been reported in certain syndromes, such as oral-facial-digital syndrome, Noonan syndrome, Ehlers-Danlos syndrome, and Simpson-Golabi-Behmel syndrome. Non-syndromic cases are rare (4).

Radiographically, OKCs appear as a well-defined unilocular or multilocular radiolucency bounded by corticated margins, with minimal buccolingual expansion. Unilocular lesions are predominant, whereas the multilocular variant is observed in approximately 30% of cases (12).

OKCs may show tooth displacement and rarely root resorption. (12) It may have no relation with a tooth or the crown may be completely inside the cyst, or they may be positioned adjacent to the lateral aspect of the tooth.

Differential diagnosis represents an important and complex phase of the clinical process. Diagnosis can often be no more than hypothetical, despite thorough anamnesis, accurate clinical and radiological analysis. (13)

The radiographic features of OKCs are not pathognomonic, particularly in smaller unilocular lesions. When a small unilocular OKC occurs in the anterior sextant of the maxilla, it may simulate other odontogenic and non-odontogenic cysts, such as radicular cyst, lateral periodontal cyst or nasopalatine cyst (12,14).

Larger odontogenic keratocysts frequently mimics other pathologic entities, such as the dentigerous cyst, and the ameloblastoma. (15)

When an OKC is associated with an impacted tooth, it may simulate a dentigerous cyst. Similarly, when an OKC is multilocular and located in the posterior sextant or the ramus of the mandible, it may mimic an ameloblastoma. Finally, when an OKC has a periapical position or involves an edentulous area, it may be mistaken for a radicular cyst. The imaging features which are more effective for

suggesting a diagnosis of radicular cyst rather than of OKC are: round or pear-shaped unilocular osteolytic lesion and the epicenter of the lesion at the apex of a non-vital tooth. (12)

As a result, dentigerous cyst, ameloblastoma and radicular cyst are considered the most common odontogenic lesions in the differential diagnosis of an OKC.

In the case reported, the radiographic presentation of the lesion as a well-defined radiolucency in periapical position relating to the roots of a non-vital 36 was in favor of a radicular cyst. Histological examination of the biopsy, however, contradicted this diagnosis, indicating a case of OKC.

Histopathological assessment by means of an incisional biopsy is the best way to diagnose an OKC before the surgery. The cyst wall is fibrous and lined by a folded, thin, regular parakeratinized epithelium 5-8 cell layers thick, without rete ridges. The parakeratin surface is typically corrugated, and the basal layer is well defined and often palisaded, with hyperchromatic nuclei and focal areas showing reversed nuclear polarity (9).

The management of OKCs aims to reduce the risk of recurrence while minimizing, at the same time, the morbidity for the patient. At the moment, there is no consensus about the best treatment modality. Treatment generally varies from conservative methods such as marsupialization, enucleation, curettage, chemical cauterization, peripheral osteotomy to much aggressive treatments such as marginal or segmental resection. (1)

The reported recurrence rates for OKC vary from 5% to almost 70%, depending on the therapeutic procedure. There are several theories explaining OKC recurrence, including the incomplete removal of the epithelial cyst lining and the growth of new cysts from small satellite cysts or odontogenic residues left behind during the operation. (16)

While resection has the lowest rate of recurrence, it shall be kept as last option, and should be reserved for retreatment of patients suffering from multiple recurring lesions. This is because of the benign nature of the cyst and its resulting morbidities.

However, enucleation can be performed for small cysts provided that adjunctive measures such as Carnoy's solution is used. This treatment regime greatly reduced recurrence rate to about 8%. For larger lesion, it is recommended to perform decompression or marsupialization followed by enucleation with adjunctive measures to reduce surgical morbidity.

In the contrary of the OKC, conservative treatments of radicular cysts are more indicated and recurrences are rare. The relatively high recurrence rate of OKC, especially after conservative surgery, makes it necessary to perform a periodic radiographic monitoring of patients at least for the first 5 years. (7, 17)

CONCLUSION

Because of the aggressive behavior and the frequency of recurrences of the OKC, preoperative assessment of OKC is important to avoid misdiagnosis with other cysts and benign tumors, and for planning management, as OKCs require a more aggressive treatment than other low-attenuating lesions having similar radiological appearance.

ACKNOWLEDGMENTS

None.

AUTHORS' CONTRIBUTIONS

The participation of each author corresponds to the criteria of authorship and contributorship emphasized in the [Recommendations for the Conduct, Reporting, Editing, and Publication of Scholarly work in Medical Journals of the International Committee of Medical Journal Editors](#). Indeed, all the authors have actively participated in the redaction, the revision of the manuscript, and provided approval for this final revised version.

REFERENCES

- [1] Mohammad Asifur Rahman, Tarin Rahman, Ismat Ara Haider. A clinicopathological study and management of odontogenic keratocyst. Update Dental College Journal. 2019 April; 9(1): 8-15
- [2] John M Wright , Marilena Vered .Update from the 4th edition of the World Health Organization classification of head and neck tumours: odontogenic and maxillofacial bone tumors. Head Neck Pathol 2017;11(1):68-77)
- [3] Priyangana Nath, Col Suresh Menon, M. E. Sham, Veerendra Kumar, S. Archana.Conservative management of odontogenic keratocyst in a tertiary hospital. Ann Maxillofac Surg 2020;10:122-6.
- [4] P.M. Brechard ,G. Hervé, V. Descroix, G. Lescaille, A. Guyon. Short Case Report A non-syndromic case of maxillo-mandibular keratocysts. J Oral Med Oral Surg 2020;26:8)
- [5] Nalini Tomar, Kuldeep Singh Bhadauria. Odontogenic keratocyst- Mimicking residual cyst in maxilla. Journal of Oral Medicine, Oral Surgery, Oral Pathology and Oral Radiology 2020;6:98–105.
- [6] Auluck A, Pai KM. Treatment of recurrent odontogenic keratocyst: a known but forgotten point. Br J Oral Maxillofac Surg 2006; 44: 74-75.
- [7] Daniel Lim, Umar Kamali. Radicular Cyst Or Odontogenic Keratocyst : A Case Of Diagnostic Dilemma. Malaysian Dental Journal Vol 2/2019)
- [8] Deepak Passi, Deepika Singhal, Mahinder Singh. Journey of OKC from cyst to tumor to cyst again: Comprehensive review with recent updates on who classification (2017)Research, 9, (07), 54080-54086.
- [9] Slusarenko da Silva Y, Naclério-Homem.Conservative Treatment of Primary and Nonsyndromic Odontogenic Keratocyst: An Overview of the Practice. Int J Oral Dent Health (2018) 4:070.
- [10] Daniel Antunes FREITAS,Daniela Araújo VELOSO,Alisson Luís D'Afonseca SANTOS,Vinicius Antunes FREITAS.Maxillary odontogenic keratocyst: a clinical case report.RevGaúchOdontol, Porto Alegre, v.63, n.4, p. 484-488, 2015
- [11] Godhi, S.S. ,P. Kukreja, Keratocystic odontogenic tumor: a review. J Maxillofac Oral Surg 2009;8(2):127-131
- [12] AndreaBorghesi and al.Odontogenic keratocyst: imaging features of a benign lesion with an aggressive behaviour. Insights Imaging (2018) 9:883–897)
- [13] Borgonovo AE, Bernardini L, Francinetti P, Rizza F, Re D. Odontogenic Keratocyst Mimicking Parodontal Cyst. Case Rep Dent 2014;2014.
- [14] MacDonald D.Lesions of the jaws presenting as radiolucencies on cone-beam CT. Clin Radiol 71:972–985,2016
- [15] R. Kirthika, Ch. Uma Reddy, Br. Sathvikalakshmi, L. Chandrashekar,R. Sudarshan and A. Feroz Khan. Odontogenic Keratocyst: A Case Series .Asian Journal of Medicine and Health 11(4): 1-5, 2018;
- [16] Naida Hadziabdic and al.Case Series Nonsyndromic Examples of Odontogenic Keratocysts: Presentation of Interesting Cases with a Literature Review. Hindawi Case Reports in Dentistry Volume 2019, Article ID 9498202, 12 pages
- [17] Johnson NR, Batsone MD, Savage NW. Management and recurrence of keratocystic odontogenic tumor: a systematic review. Oral Surg Oral Med Oral Pathol Oral Radiol 2013; 116:e271-e276

COMPETING INTERESTS

The authors declare no competing interests with this case.

FUNDING SOURCES

None.

PATIENT CONSENT

Written informed consent was obtained from the patient for the publication of this case report.