

Keratocystic odontogenic tumor mimicking dentigerous cyst: A case reportNour-Sayda Ben Messaoud*¹, DMD,DDS, Hajer Hentati², PhD, Amine Derbel², Manel Njima³, MD, MPH, Jamil Selmi², PhD¹Oral Medicine Oral Surgery Unit, Department of Dentistry, University Hospital Farhat Hached, Sousse, Tunisia²Department of Oral Medicine Oral Surgery, University Dental Clinic, University of Monastir, Tunisia³Department of histopathology, University Hospital Fattouma Bourguiba, University of Monastir, Tunisia**Case Report*****Corresponding author**

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Abstract: Pericoronal radiolucencies are common radiographic findings in relation to impacted mandibular third molars .In most cases, they are associated to Dentigerous cysts since it's the most common developmental cyst of the jaw. we report a case of a 24-year-old female patient presenting with a mild pain in the mandibular posterior left side. The radiographic examination showed a well-defined pericoronal radiolucent lesion associated with the impacted 38. Enucleation and extraction of the 38 were performed under local anesthesia. The histopathological assessment has revealed a keratocystic odontogenic tumor. Pericoronal radiolucencies associated with the impacted mandibular third molars may have similar clinical and radiographic features, although they have different histopathological aspects, biological behavior, and treatment. This is especially true for dentigerous cyst, ameloblastoma and keratocystic odontogenic tumor. The differential diagnosis may be improved by careful analysis of the clinical and radiological features.

Keywords: Dentigerous Cyst, Molar, Third/radiography, Odontogenic Tumors

INTRODUCTION

Dentigerous cyst (DC) is the most common non-inflammatory odontogenic cyst commonly discovered as pericoronal radiolucency associated with impacted, embedded and unerupted permanent tooth on routine radiographic examination. The most affected teeth are respectively: the mandibular third molar, the maxillary canine and the mandibular second premolar. Dentigerous cysts are generally managed by enucleation or masupialization especially in pediatric patients and have excellent prognosis with low recurrence [1].

However many other benign and even malignant lesions may present radiographically as pericoronal radiolucency associated with an impacted tooth, resulting in different treatment and prognosis [2].The differential diagnosis of pericoronal radiolucencies associated with impacted mandibular third molars should especially include keratocystic odontogenic tumor (KOT), unicystic ameloblastoma, solitary bone cyst, odontogenic epithelial calcified tumor, and in rare cases intraosseous squamous cell carcinoma [3].

CASE REPORT

A 24-year-old female patient was presenting with mild pain in the mandibular posterior left side for the past 1 one year. The medical history was not otherwise significant. The Extra-oral examination showed mobile, tender submandibular adenopathies. The intra-oral examination has particularly revealed the absence of the tooth 38. The digital pressure in the retromolar area was painful and showed purulent selling in the distal area of the tooth 37(Fig-1).



Fig-1: Intra-oral examination showing purulent selling in the distal area of the tooth 37

A panoramic X-ray was performed and revealed a well-defined unilocular radiolucency that seemed surrounding the crown of the impacted 38. The

lesion was extending to the distal root of the tooth 37. The margins were well-defined without a clear sclerotic border (Fig-2).



Fig-2: Panoramic x-ray showing well-defined unilocular radiolucent lesion surrounding the crown of the impacted 38

A cold test was performed on the tooth 37 that responded positively. A cone beam computed tomography (CBCT) scan was assessed and has

particularly showed that the impacted 38 was encapsulated by the lesion (Fig-3).

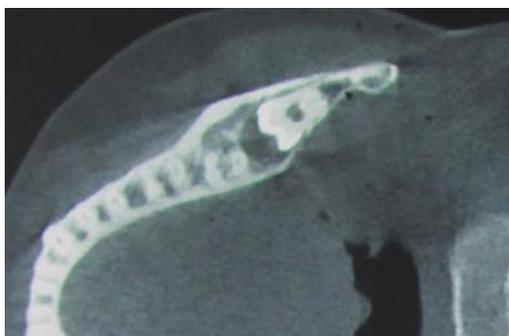


Fig-3: CBCT scan axial sectional view: the impacted 38 is involved by the radiolucent lesion

At first, a preoperative diagnosis of infected DC was established however KOT and unicystic ameloblastoma were considered as differential

diagnosis since the lesion was involving the tooth and not attached to the cement-enamel junction. The patient was prescribed antibiotic (amoxicillin 3 g/ day for 7

days), analgesic (paracetamol 2g/day) and chlorhexidine mouth rinse to manage pain and infection. The surgical enucleation of the lesion associated to the impacted 38 extraction was performed under loco-regional anesthesia (articaine in a 4% solution with epinephrine 1:100,000).

The histopathological analysis has showed that the cyst lumen was lined by parakeratotic squamous epithelium with palisaded, hyperchromatic basal layer. These features supported the diagnosis of KOT (Fig-3).

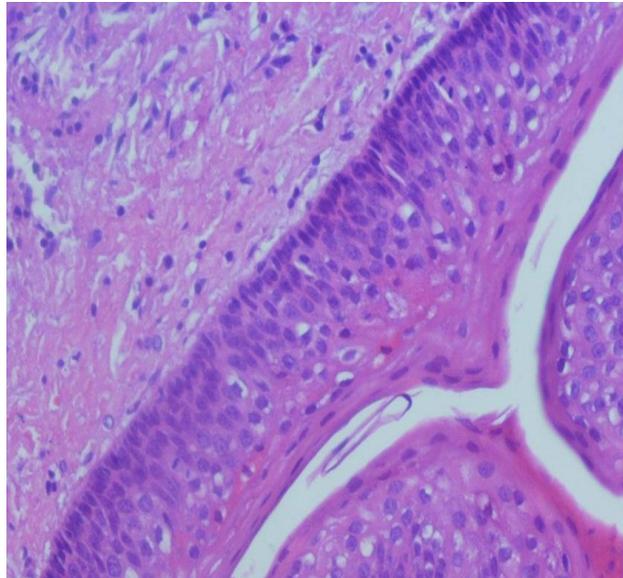


Fig-4: Histopathological section photomicrograph showing Fibrous tissue surrounding a cyst that is lined by parakeratotic squamous epithelium with palisaded, hyperchromatic basal layer (HE X 100)

The patient had uneventful postoperative recoveries. A panoramic X-ray was performed at 6

month post-operative and showed satisfactory bone healing (Fig-5).



Fig-5: post-operative panoramic X-ray at 6 month showed satisfactory bone healing

DISCUSSION

Dentigerous cysts are the second most common odontogenic cysts after radicular cysts, accounting for approximately 24% to 33% of all true cysts in the jaws [4, 5]. Most reports showed a peak incidence of DCs in the second and third decades of life [4, 5, 6, 7].

The pathogenesis of DCs involves the accumulation of fluid between the unerupted tooth crown and the surrounding follicle, giving rise to the characteristic radiographic finding of a cystic lesion surrounding the neck of the tooth at the cement-enamel junction [3]. In case of well-defined Radiolucent lesions that involve all the tooth, the diagnosis of DC has to be excluded suggesting benign odontogenic

tumors such as unicystic ameloblastoma and odontogenic keratocystic tumor in posterior mandibular location, odontogenic adenomatoid tumor, calcifying odontogenic cyst tumor in anterior location, and ameloblastic fibroma in pediatric patient. DCs may not always look radiographically typical as they may have multilocular aspect, enlarge and extend posteriorly or anteriorly to involve the root of adjacent teeth making the differential diagnosis more challenging [8]. Although even a typical DC can be something else, that's why a histopathological examination has to be performed to rule out a KOT, ameloblastic or in rare cases malignant transformation [2, 9, 10]

The total incidence of cyst and tumor development from impacted third molars seems to be relatively low, Stathopoulos P *et al* [3], have reported that the most frequent differential diagnosis of dentigerous cyst associated with Impacted and partially erupted third molars are KOT (17%), and ameloblastoma (5%).

Odontogenic keratocyst, was renamed KOT by the World Health Organization in 2005 since then it has been reclassified as benign epithelial odontogenic tumor to better account for its clonal nature and high recurrence rate [11]. Currently, there is ample evidence that the molecular and genetic alteration that affects some odontogenic keratocysts may affect their biological behavior [11, 12]. Actually Relapse rate of KOT could be related to the expression of specific biological markers in the epithelial cells, in the Epithelial-mesenchymal transition cells layer and in the fibrous capsule [12-14].

Recently, Cunha JF *et al* [13] in retrospective cohort analysis including 24 sporadic KOT have shown that recurrence was significantly associated with poor clinical response to decompression, remaining tooth with radiographic evidence of insinuation of the lesion between the dental roots, and the presence of budding basal cells layer together with epithelial islands in the fibrous capsule.

Currently, decompression or marsupializations, combined with stage-two curettage or enucleation, represent the commonly accepted treatment approach for KCOTs. The advantage of this therapeutic approach is not only minimal invasive approach and size decrease of the lesion, but also histological features of KCOTs markedly changed following decompression. This histopathological change may result in loss of the specific histological features that revert to a lining more like the oral mucosa [11, 15]. However, the mechanisms responsible for these changes have still to be elucidated.

CONCLUSION

Pericoronal radiolucencies are relatively common radiographic findings that are in most cases

related to DCs. Impacted Mandibular third molars are the most affected teeth [10]. Actually premolar-molars mandibular area is well known to be a predilection site of many benign odontogenic tumors. KOT and ameloblastoma should be considered in differential diagnosis especially in case of radiolucent lesion that does not enclose the crown of the tooth at the cement-enamel junction.

Conflict of interests

The authors declare that there are no conflicts of interest

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