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RESEARCH ARTICLE

ODONTOGENIC KERATOCYST: CASE REPORT AND LITERATURE REVIEW

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Abstract

Odontogenic keratocyst is a peculiar benign intraosseous lesion, recognized for its local and progressive invasive potential and high recurrence rate, which have led to its classification and reclassification. It constitutes 10-20% of odontogenic cysts and is the third most common cyst of the jaws, with peak incidence in the second and fourth decades. The aim of this report is to present a case study of an odontogenic keratocyst in a 27-year-old female patient, and to provide an overview of this lesion, highlighting their pertinent clinical, radiographic features as well as its surgical management.

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Introduction:-

Odontogenic keratocyst (OKC) was first described by Philipsen in 1956 (Borghesi et al., 2018; Mehdi et al., 2022; Ravi et al., 2022). It has been one of the most controversial benign pathological entities of the maxillofacial region since the mid-20th century, due to its unique clinical features, especially in relation with its potentially aggressive behavior and relatively high recurrence rate (Borghesi et al., 2018; Khan et al., 2019; Kshirsagar et al., 2019; Mehdi et al., 2022).

According to the World Health Organization (WHO), OKC is defined as an odontogenic cyst characterized by a thin, regular coating of stratified parakeratinized squamous epithelium (Motaleb et al., 2022; Nath et al., 2020).

It accounts for 10 to 20 % of all odontogenic cysts (Boffano et al., 2022; Borghesi et al., 2018; Lafuente-Ibáñez de Mendoza et al., 2022; Motaleb et al., 2022; Titinchi, 2022) with a peak incidence occurring in the second and fourth decades of life (de França et al., 2021; Nath et al., 2020; Titinchi, 2022). It is the third most common odontogenic cyst (Ravi et al., 2022), and shows a mal predominance (Borghesi et al., 2018; Roman et al., 2022; Titinchi, 2022). The OKC mostly affects the mandible with an incidence rate reaching 70% (Boffano et al., 2022; Borghesi et al., 2018; Rajendra Santosh, 2020), and is associated with impacted teeth in particularly wisdom teeth in 30% of all cases (Borghesi et al., 2018; Rajendra Santosh, 2020; Roman et al., 2022).

Observation:-

A 27-year-old woman attended the oral surgery department of the dental consultation and treatment centre of Casablanca. She reported a slow-growing right mandibular swelling evolving for 9 months. The patient mentioned having undergone a non-professional extraction in the right mandibular region at the age of 18.

The patient was breastfeeding, in apparently good health, with no particular medical history apart from a caesarean delivery.

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The extra-oral examination showed a slight facial asymmetry, objectified by a non-delimited low genital swelling, depressible and painless upon palpation, with no adenopathies observed (Fig 1).

The intra-oral examination (Fig 2) revealed swelling of the buccal and lingual cortical plates, extending from 44 to 47, covered by normal appearing mucosa. It was painless and depressible upon palpation. The 45 was missing.

The vitality test was negative for 46 and 47.

The panoramic radiograph revealed a well-defined, bilobed, radiolucent image, extending from 43 to 48, displacing the root of 44, and enveloping the residual root of 45, with the disappearance of the inferior alveolar canal (IAN) (Fig 3).

The cone-beam computed tomography (CBCT) showed thinning and rupture of both the buccal and lingual cortical plates, and revealed that the IAN is encompassed within the image (Fig 4).

The location of the lesion led us to consider a keratocyst and an ameloblastoma, while the relationship with the necrotic teeth suggested a radicular cyst.

Clinical treatment consisted of cystic enucleation, with extraction of 47, 46 and 45, followed by mechanical curettage (Figure 5).

An amoxicillin, a steroidal anti-inflammatory, a level 2 analgesic, a chlorhexidine-based antiseptic and vitamin B12 were prescribed.

Macroscopically, the surgical specimen was firm and measured 3.5 x 2 cm (Fig 6).

Microscopically, histological sections showed a fibrous cystic wall, lined with a thin, more or less uniform squamous epithelium, consisting of 5 to 6 layers of regular cells. The basal layer was palisaded. The wall was moderately inflamed and ulcerated in some areas, with little keratin detached from the lining (Fig 7).

This finding supported the diagnosis of a parakeratinized odontogenic keratocyst.

The 15-day follow up showed good mucosal healing (Fig 8). However the patient reported paresthesia in the right mandibular area.

A 6-month radiological follow-up showed the early signs of mineralization in the cystic lesion. Clinically, the patient reported occasional itching and paresthesia (Figure 9).

One-year and 2-year radiological follow up revealed signs of bone regeneration (Fig 10, 11), with no clinical symptoms reported.

Discussion:-

OKC is a uni or multi-locular, benign, progressive lesion with aggressive and invasive potential, emerging from the cellular remnants of the dental lamina (Serres debris) (Boffano et al., 2022; de França et al., 2021; Motaleb et al., 2022; Nath et al., 2020).

It was initially classified as a developmental odontogenic cyst of the jaw in 1971-1992, and reclassified as a benign odontogenic neoplasm or keratocystic odontogenic tumor in 2005, due to its genetic mutation related to the PTCH tumor suppressor gene, its aggressive behavior and its high recurrence rate. Since then, evidence has shown that PTCH genetic mutations do not occur only in these lesions, and that keratocysts can regress following marsupialization, which contradicts the neoplasm's characteristic of autonomy that continues to increase. Therefore, this led the WHO in 2017 to reclassify this lesion as an odontogenic cyst and to consider OKC and odontogenic orthokeratocyst (OOC) as two distinct entities, due to the non-aggressive behavior, low recurrence and favorable prognosis (2022; de França et al., 2021; Sabarinath et al., 2022; Titinchi, 2022; Wright & Vered, 2017).

According to the latest classification in 2022, the OKC is still classified as a jaw cyst as a distinct entity (Vered & Wright, 2022).

Clinically, OKC presents as swelling with or without pain, occasionally causing displacement of the teeth, as observed in our case, and paresthesia of the lower lip (Motaleb et al., 2022).

The OKC mostly shows minimal bone expansion (Borghesi et al., 2018; Kitisubkanchana et al., 2021), and can be either sporadic or solitary, as in our case, or syndromic (de França et al., 2021). It has been reported that 5% of all OKCs are associated with the nevoid basal cell carcinoma syndrome (NBCCS), in particularly multiple OKCs (Consolo et al., 2020; Motaleb et al., 2022).

Radiologically, OKC presents a well-limited, unilocular or multilocular radiolucency with smooth bony margins. It may be associated or not with an impacted tooth, and doesn't show tooth displacement and tooth resorption. (Alves et al., 2018; Boffano et al., 2022; Borghesi et al., 2018; de França et al., 2021; Rajendra Santosh, 2020).

The differential diagnoses include ameloblastoma, radicular cyst, myxoma, dentigerous cyst and central giant cell granuloma (Kshirsagar et al., 2019; Lafuente-Ibáñez de Mendoza et al., 2022; Rajendra Santosh, 2020).

The positive diagnosis is based entirely on anatomopathological examination (Borghesi et al., 2018; Kshirsagar et al., 2019).

The OKC is treated surgically. Several treatment approaches have been proposed, varying from a conservative approach with primary enucleation or initial marsupialization followed by secondary enucleation, to aggressive or invasive treatments, such as cystic enucleation associated with adjunctive therapies (Carnoy's solution (CS), electrocautery, 5-Fluorouracil (5-FU), cyclopamine, cryotherapy and peripheral osteotomy) or marginal or segmental surgical resection (Boffano et al., 2022; Mehdi et al., 2022; Motaleb et al., 2022).

Currently, there is no consensus on the best therapeutic approach, but the objective is to adopt the modality with the lowest risk of recurrence and the least morbidity (Mohanty et al., 2021).

Al Moraissi et al. 2023 show in their meta-analysis, that CS as an adjuvant to enucleation and peripheral osteotomy is the most effective agent unless proven otherwise (Al-Moraissi et al., 2023).

The choice of therapeutic approach depends on several factors, such as the patient's age, the lesion's size, its location, the radiological appearance, the cortical perforation, the soft tissue involvement and whether the lesion is primary or recurrent (Kshirsagar et al., 2019; Motaleb et al., 2022; Roman et al., 2022).

OKCs have a recurrence rate that varies from 0 to 60% after treatment (Kinard et al., 2019), frequently occurring within the first 5 years (Kshirsagar et al., 2019). This high risk is attributed to the difficulty of completely enucleating the thin, friable cystic wall of OKCs, and the tendency of OKCs to have small satellite cysts (Motaleb et al., 2022).

Various factors may influence the recurrence rate of OKCs: the size > 4cm, the syndromic form, the therapeutic approach, the location, the multilocular appearance, the cortical perforation, the wall fragility, the presence of satellite cysts and inflammatory infiltrate (de França et al., 2021; Kshirsagar et al., 2019; Mehdi et al., 2022; Motaleb et al., 2022; Rajendra Santosh, 2020; Titinchi, 2022).

Due to the recurrent nature of OKCs, a long term clinical and radiological follow-up are essential (Motaleb et al., 2022).

Our patient showed no recurrence at 2 years follow-up, and is still under follow-up.

Figures



Figure 1:- Extra-oral examination.



Figure 2:- Intra-oral examination showing the swelling of the buccal cortical plate.



Figure 3:- Panoramic radiograph showing a well-defined radiolucent image.



Figure 4:- CBCT, 4a : axial slices,4b : orthoradial slices.

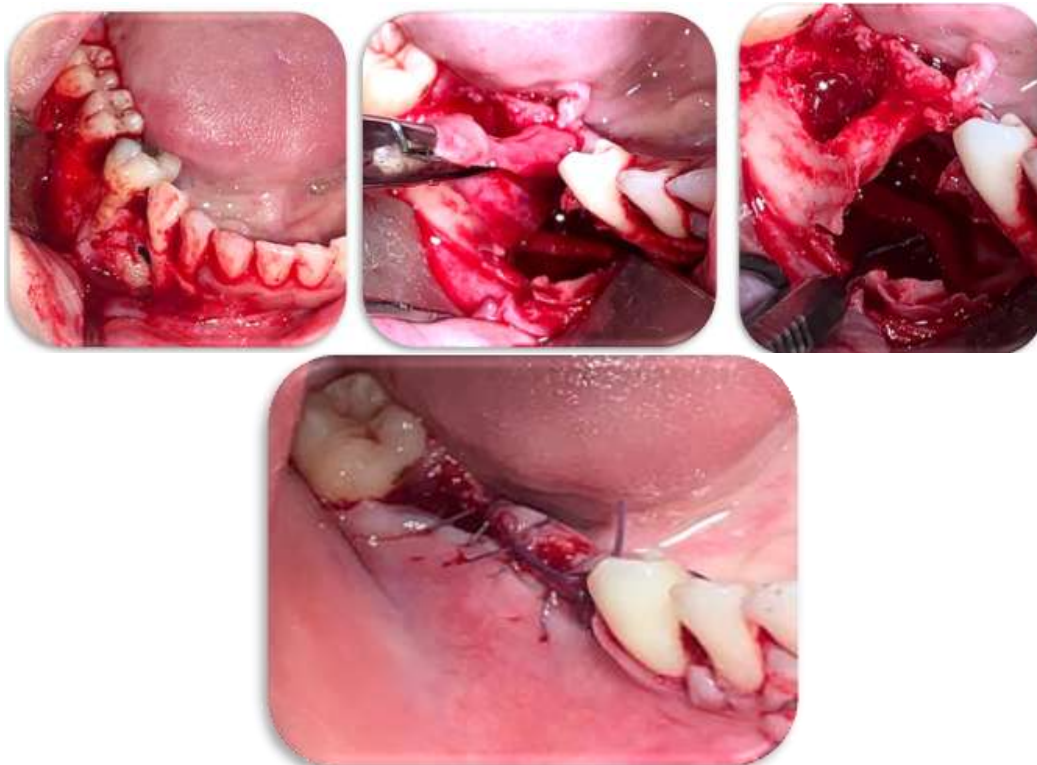


Figure 5:- Enucleation and extraction of 46 and 47.



Figure 6:- The surgical specimen.

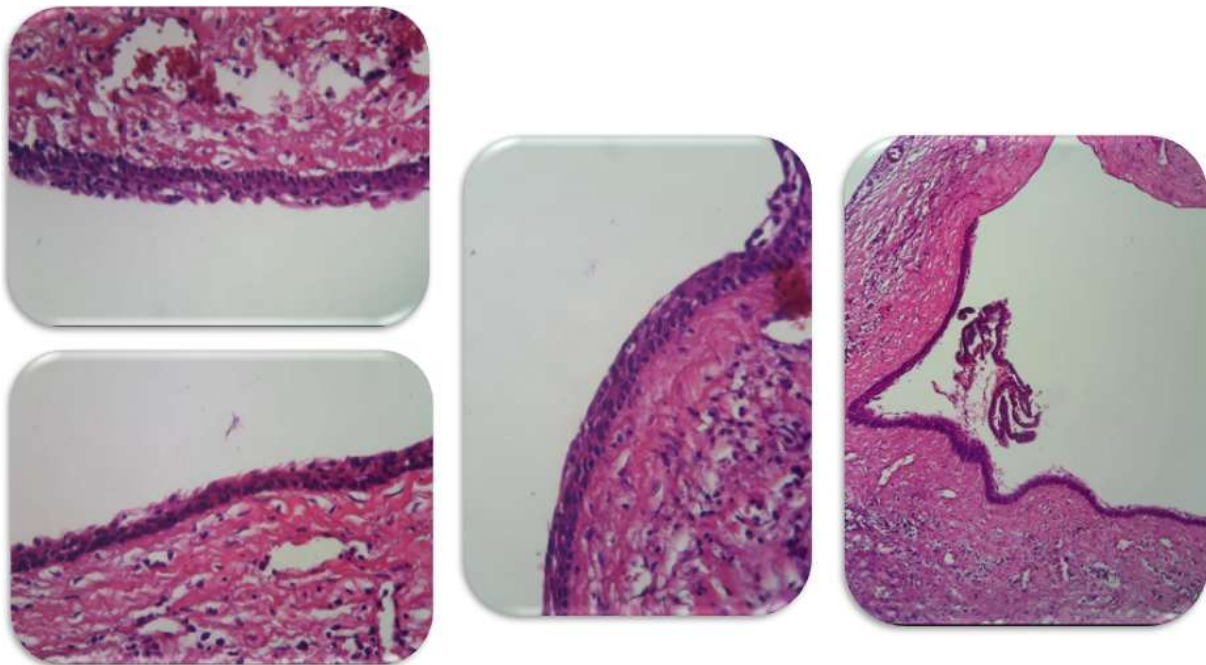


Figure 7:- Histological sections stained with hematoxylin and eosin.



Figure 8:- 15-day follow up.



Figure 9:- 6-month radiological follow-up.



Figure 10:- 1-year radiological follow-up.

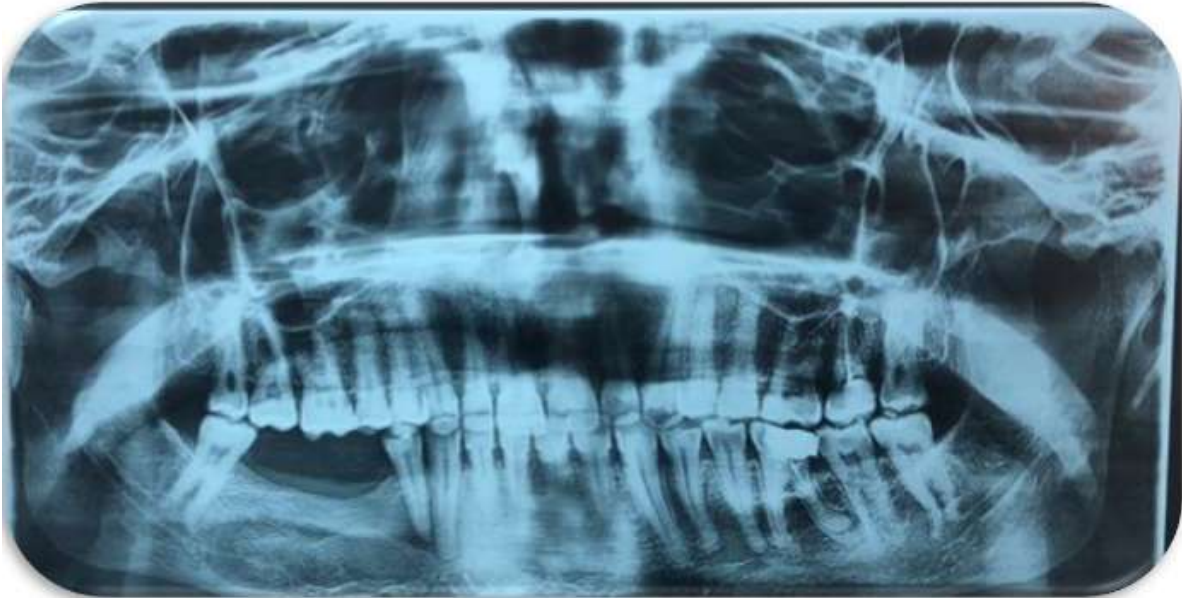


Figure 11:- 2-years radiological follow-up.

Conclusion:-

OKC is a unique cyst of its kind. It has generated considerable attention from researchers. The lack of standardization concerning the most effective therapeutic approach, and the dilemma concerning its classification, demonstrate the lack of understanding of this lesion despite scientific advances.

Conflict of interest

The authors confirm that this article content has no conflict of interest.

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