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Case Report

Odontogenic Keratocyst Mimicking Odontogenic Dentigerous Cyst. A Laborious Diagnostic and Therapeutic Challenge

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Abstract

Odontogenic keratocyst (OKC) is a developmental cyst of jaw which arises from odontogenic epithelium. OKCs has been considered to be cystic neoplasms and called keratocystic odontogenic tumors in WHO classification in 2005. Although reclassified as cysts by WHO at 2017, OKCs are characterized by clinically aggressive behavior and high risk of recurrence. Clinical and radiological features are not specific and can mimic other lesions of jaw as dentigerous cyst. This tumor is also a developmental cyst but is less aggressive and not recurring. The management differs depending on the nature of the cyst. This paper presents OKC case mimicking dentigerous cyst, also the histopathological characteristics specific to odontogenic keratocyst as well as the adapted management.

Keywords: Odontogenic Cyst; Odontogenic keratocyst; Dentigerous Cyst

Introduction

Odontogenic keratocyst (OKC) arises from odontogenic epithelium either in mandible or maxilla. Two sources of epithelium have been suggested: the dental lamina and its remnants and extensions of basal cells from the overlying epithelium. The majority of OKC (65 - 83%) occur in the mandible and half of them originate at the angle of the mandible [1]. Many authors do not share the WHO decision to reclassify OKC as cysts [2] and not as odontogenic keratocystic tumor as described in the 2005 WHO classification. Indeed, the aggressiveness and recurrence of odontogenic keratocysts make them require special care. Their presentation is not specific and can lead to a wrong diagnostic. This paper show OKC case with characteristics similar to dentigerous cysts.

Case Report

A 63-year-old female patient was referred from private clinic to oral surgery's department of "Centre de Consultations et de Traitements Dentaires" CCTD at Casablanca, to evaluate an incidental discovery of radiolucent lesion of jaw in the orthopantomogram (OPC). The patient had no particular medical history. Extraoral examination didn't show facial deformity or hypertelorism (Figure 1). Palpation of lymph nodes was unremarkable. During intraoral examination, there was no cortical expansion (Figure 2).

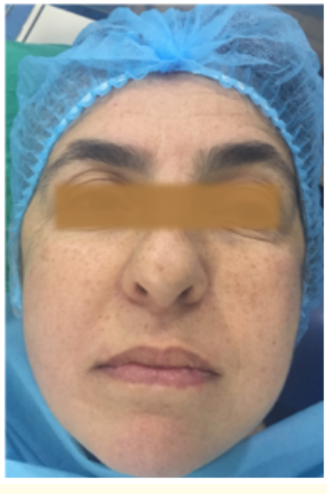

Figure 1: Extraoral view.

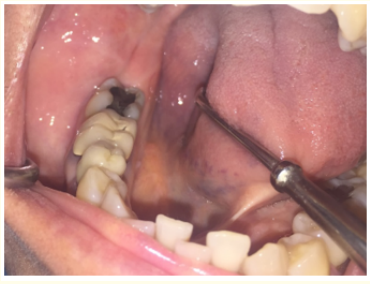

Figure 2: Pré-op intraoral view.

The orthopantomogram revealed a well-defined unilocular radiolucency, in the right angle of mandible, which surrounded entirely the impacted wisdom tooth (48) (Figure 3). Computerized tomography scan examination revealed an expansion of cortical bone with a rupture of both buccal and lingual corticals. These signs were in favor of dentigerous cyst. Under local anesthesia, enucleation of the lesion was performed supplemented with osseous curettage. The impacted tooth (48) was extracted as well as adjacent tooth (47) with poor prognosis (Figure 4). The removed lesion had two separate cystic cavities. The histopathological report revealed that the cystic lining was a thin keratinized stratified squamous epithelium. The underlying connective tissue capsule showed loose bundles of collagen fibers, inflammatory cells like lymphocytes. Thus, the final diagnosis was odontogenic keratocyst. The patient has been followed up and control orthopantomogram at 6 months and 1 year revealed signs of bone healing (Figure 5 and 6).

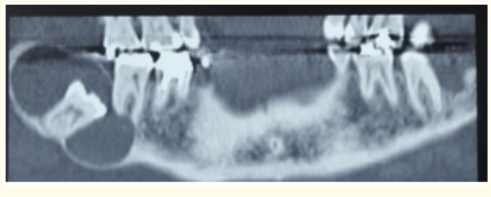

Figure 3: Orthopantomogram showing well-defined unilocular radiolucency, in the right angle of mandible.

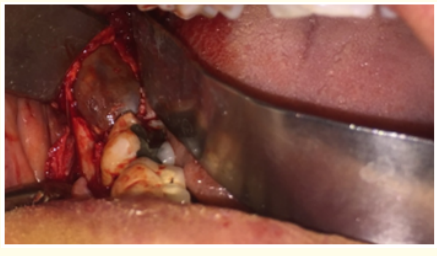

Figure 4: Intraoral view of the lesion.

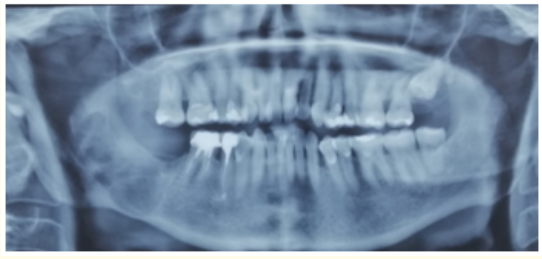

Figure 5: 6 months orthopantomogram showing bone healing beginning.

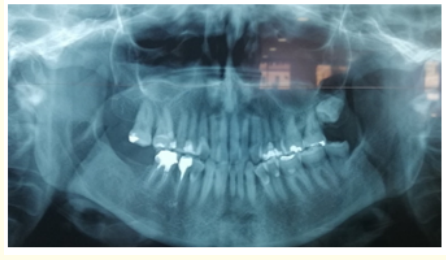

Figure 6: 1 year follow up revealed bone healing.

Discussion and Conclusion

Odontogenic keratocyst was first identified and described in 1876 and further characterized by Phillipsen in 1956 [3]. There is a wide reported age distribution (8 - 82 years), but the mean age of presentation is in the third decade of life [4].

A slightly higher prevalence of OKC is reported in males, with a ratio of 1.42 [5]. Odontogenic keratocyst occurs predominantly in the mandible and concerns the angle of mandible. Multiple OKCs can be found in syndromic patients as basal cell nevus syndrome. The case reported showed no signs in favor of Gorlin-Goltz Syndrome. In the recent World Health Organization classification of tumors of the head and neck [2], the name keratocystic odontogenic tumor (KCOT) has been changed again to odontogenic keratocyst (OKC). This decision has caused some confusions and not accepted by most of authors regarding this potentially aggressive lesion. OKCs are known as recurring tumors. Their potential aggressiveness has been well documented.

The case reported in this paper, was a 63-year-old female patient showing a keratocyst located at the angle of mandible.

Emerson., *et al*. [6], described the extension of two recurrent OKCs in the mediastinum and Abe., *et al*. [7] reported OKCs penetrating into the temporalis muscle. Makaria., *et al*. [8] and Yamamoto., *et al*. [9] found the same in the masseter muscle. Liu., *et al*.

[10] described a recurring OKC in an autogenous lyophilized bone graft, with extension into the masseter muscle. Odontogenic keratocyst, therefore, requires an accurate diagnosis and an appropriate management.

Clinical presentation of OKC depends on its stage of evolution. They are either locally destructive with facial deformation or they can grow to a large size undetected as reported in this case. Radiologically, OKCs are characterized by well-defined unilocular or multilocular radiolucent areas with a clear peripheral radiopaque rim. The borders are usually scalloped [11]. Odontogenic keratocysts can present 4 radiologic aspects: replacement, envelopmental, extraneous, and collateral. The OKCs formed in the location of a tooth are called the replacement type. OKCs that embrace an adjacent unerupted tooth are the envelopmental type. OKCs occurring in ascending ramus (away from the teeth) are extraneous type and OKCs occurring adjacent to the root surfaces are the collateral type [12,13].

OKC, in the present case, encountered unerupted wisdom tooth (envelopment type) and mimics strongly dentigerous cyst.

Dentigerous cyst is the second most common cyst of the jaw and has a developmental origin. Almost all of the dentigerous cysts enclose the crown of an unerupted tooth and the radiolucent area is attached to the tooth at the cementoenamel junction [14]. Dentigerous cysts are predominantly asymptomatic unless the condition is secondarily infected. In this case, the patient did not report pain or inflammation signs. Dentigerous cysts are classically characterized by a unilocular radiolucent areas associated with the crowns of unerupted teeth at the level of cement-email junction CEJ. The radiolucent cavity is well defined and well circumscribed with a sclerotic border (radiopaque) [11]. This presentation is similar to the case reported.

Differential diagnosis based on clinical and radiological features are challenging if not impossible. Histopathological examination therefore becomes necessary for the diagnosis of OKC. This examination requires special attention. The emphasis on keratinization was considered deceptive, masking other histological characteristics that were actually responsible for the biological behavior of the cysts [15]. In 1963, Hansen pointed out that the designation "keratocyst" was used invariably to describe any keratin-forming jaw cyst and highlighted the need for specific histological criteria [16]. This is more important nowadays, we know that certain lesions of jaw as dentigerous cyst can present keratin formation at histopathological examination [17]. Studies by Browne (1971) and Forssell and Sainio (1979) showed that the characteristic epithelial lining of OKC was unique and different from the metastatic keratinization in other jaw cysts, affirming its recognition as a discrete entity [15].

Microscopically, OKCs show a characteristic lining of parakeratinized stratified epithelium 6 to 11 cells thick. The surface of the lining epithelium is usually corrugated. The basal layer is lined by tall columnar cells with a nuclear palisading arrangement. The epithelium-fibrous wall interface is flat and detachment of portions of epithelium from the fibrous wall is also observed. Satellite (daughter) cysts can be found in the connective tissue wall. Chronic inflammatory cell infiltration can be seen in the fibrous wall [11].

Microscopically, dentigerous cyst is observed with thin (2 - 3 layers) nonkeratinizing cystic epithelium. Scattered mucous cells may be observed. The fibrous capsule is loosely arranged and may show small inactive-appearing odontogenic epithelial islands [11].

In this case, in view of anatomopathological examination, the diagnosis of odontogenic keratocyst is retained.

Treatment of odontogenic cysts includes enucleation (with or without peripheral ostectomy, treatment with Carnoy solution, or cryotherapy), marsupialization, or resection.

Some authors recommend radical treatment for OKCs, because of aggressive behavior and high recurrence rate, as the en-block resection, including the surrounding bone if the lesion is small enough to leave a rim of supporting bone. If the lesion is large, then bone resection and reconstruction with a composite free graft is required [1]. Authors are more conservative and advocate management of OKC via surgical care with careful complete excision of cyst and osseous curettage to prevent recurrences [11]. This last option was chosen in the present case reported.

A systematic review and metanalysis realized by Essam Ahmed Al-Moraissi., *et al*. [18] showed the lowest recurrence rate (RR) of 8,4% after treatment by resection and respectively 17% and 11% after treatment by enucleation with curettage and enucleation with Carnoy's solution. Considering the complications and undesirable effects of treatment by resection and except the extended

odontogenic keratocysts, the best compromise treatment would be enucleation with carnoy's solution and regular follow up.

Conflict of Interest

None.

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