

DIAGNOSIS AND CONSERVATIVE MANAGEMENT OF AN EXTENSIVE ODONTOGENIC KERATOCYST: ABOUT A CASE REPORT



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ABSTRACT

Odontogenic keratocysts (OKCs) are benign intraosseous lesions of odontogenic origin that are classified as developmental cysts derived from the dental lamina. They may occur at any age, but usually between the second and third decade of life. Men are more commonly affected than women are. The OKC accounts for approximately 11% of all cysts in the jaws and is most often located in the mandibular ramus and angle. They exhibit aggressive behavior with local invasive and evolutionary potential and a high level of recurrence. Radiographically, it appears as a unilocular or multilocular lesion with a scalloped contour. The aim of this article is to report the case of a large odontogenic keratocyst diagnosed in a 43-year-old patient who underwent surgical enucleation in the Oral Surgery Department of the Dental Consultation and Treatment Center of Casablanca, and to underline the importance of a conservative approach in managing OKCs.

Keywords: *odontogenic keratocyst; diagnosis; conservative management.*

1. INTRODUCTION

Odontogenic Keratocyst (OKC) is a benign lesion that is currently considered an odontogenic cyst in the latest classification of Head and Neck tumors by the World Health Organization (WHO) in 2022, based on its clinical and histopathological features. It is one of the most commonly researched cysts due to its aggressive clinical behavior and high recurrence rate. Most OKCs (up to 80%) are located in the mandible, particularly in the area between the mandibular angle and the third molar, with the cysts growing towards the ramus and body. The incidence of OKCs is broad, ranging from 8 to 82 years, with the highest prevalence occurring in the second and third decades of life, and men are more frequently affected than women [1]. Clinically, OKCs are typically asymptomatic and can grow considerably, with many being detected during routine radiological examinations.

Radiographically, OKCs appear as unilocular or multilocular radiolucent lesions that can vary in shape and size. Histological confirmation is necessary for a definitive diagnosis. This article aims to describe the diagnostic and treatment approach for a large mandibular OKC in a 43-year-old patient treated in the oral surgery department of the dental consultation and treatment center of Casablanca.

Case report

We report the case of a 43-years-old woman, received in the consultation of the Oral Surgery Department of the Dental Consultation and Treatment Center of Casablanca complaining of oral swelling on the right side of the mandible evolving for 6 months. The patient was a non-smoker and had no underlying disease.

The extra oral examination revealed a slight facial asymmetry with increased volume of the right posteroinferior hemiface [Figure 1].



Figure 1: Extraoral examination showing a slight facial asymmetry with increased volume of the right posteroinferior hemiface.

The intra-oral examination revealed a swelling in the right mandibular area, painless, non-tender, covered with a normal mucosa, extending anteriorly from the body of the mandible to the ramus posteriorly [Figure 2].



Figure 2: Intraoral examination showing a swelling in the right mandibular area, painless, non-tender, covered with a normal mucosa, extending from the body of the mandible to the ramus posteriorly.

A panoramic X-Ray revealed a multilocular radiolucent lesion extended across the right posterior region of the body of the mandible and ascending ramus without involvement of the condyle [Figure 3].



Figure 3: Preoperative panoramic X-Ray showing a multilocular radiolucent lesion extended across the right posterior region of the body of the mandible and ascending ramus without involvement of the condyle.

A cone beam computed tomography (CBCT) scan was then performed to better evaluate the lesion and to show its true extent and its relation with anatomic elements. The scan revealed an extensive hemi-mandibular lesion causing a great resorption of both vestibular and lingual cortical bone throughout the extent of the cyst, measuring a thickness of only 1.5 mm [Figure 4].

After a thorough clinical and radiological examination, a presumptive diagnosis of parakeratinized odontogenic keratocyst and ameloblastoma was made.

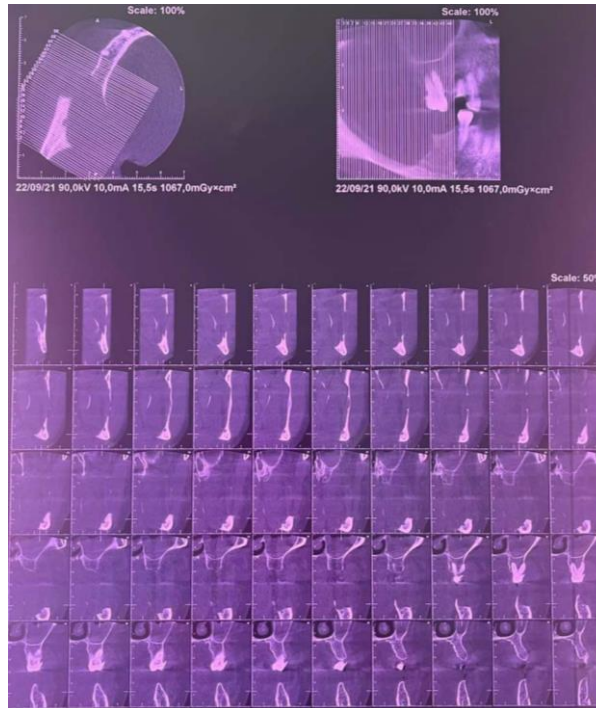


Figure 4: Preoperative CT scan showing an extensive hemi-mandibular lesion causing a great resorption of both vestibular and lingual cortical bone;

It was decided to perform an aspiration puncture of the lesion with a 2.5 cc syringe for subsequent pathological analysis. Thereby, a citrus cystic liquid was obtained [Figure 5].



Figure 5: An aspiration puncture of the lesion with a 2,5 cc syringe showed a citrus cystic liquid.

Surgical treatment was performed under local anesthesia. It consisted of enucleation of the lesion. It was removed entirely, followed by rigorous mechanical curettage and lavage of the bone cavity with abundant sterile saline solution and chlorhexidine digluconate 0.2% [Figure 6,7].



Figure 6: The figure presents the intraoperative photographs.



Figure 7: The figure presents the cystic lesion.

The final diagnosis established by histopathological examination was OKC [**Figure 8**].

The patient was informed about the diagnosis, and the follow-up program was planned for every 3 months in the first year and every 6 months after that. As a postoperative sequel, our patient reported a temporary paraesthesia of the inferior alveolar nerve (IAN) that recovered completely after 6 months.

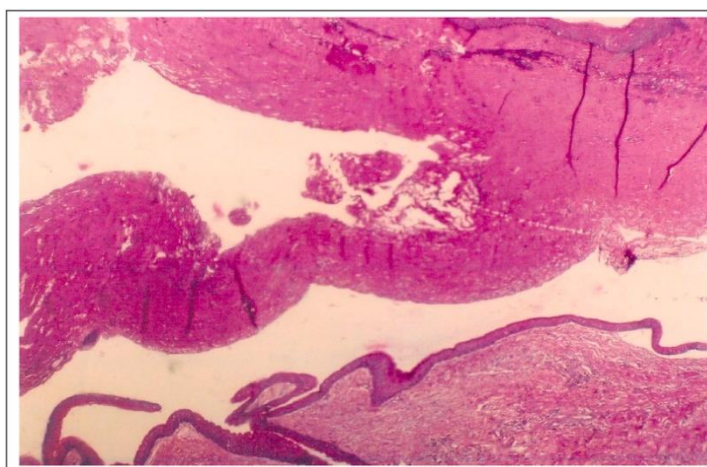


Figure 8: the figure presents the histopathological examination.

After approximately 18 months of regular clinical and radiographic follow-up, no recurrence has been observed and satisfactory bone healing was achieved [**Figure 9**].

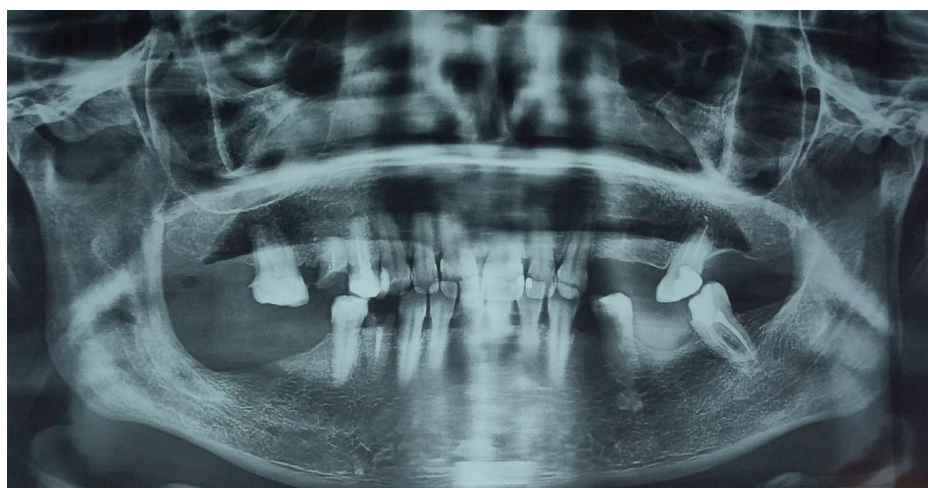


Figure 9: 18 months postoperative panoramic radiograph.

2. DISCUSSION

Odontogenic keratocysts (OKCs) are non-cancerous lesions that grow within the bone and have a tendency to recur after treatment due to their locally aggressive behavior [2]. These cysts are named after the keratin produced by their lining and are a rare type of developmental odontogenic cyst that arise from the dental lamina. They contain clear fluid and a cheesy material resembling keratin debris [3]. However, there is an ongoing debate regarding the biological behavior and classification of OKCs. The World Health Organization (WHO) classified OKC as an odontogenic cyst in

1971 and maintained it in 1992. In 2005, due to its aggressive behavior, high recurrence rate, and association with the mutation of the PTCH tumor suppressor gene, OKC was reclassified as a tumor and named keratocystic odontogenic tumor (KCOT) [4]. However, in 2017, the WHO reclassified KCOT as a cyst because research has shown that the PTCH gene mutation can also be found in non-neoplastic lesions like the dentigerous cyst.

In the latest edition of 2022, the WHO still consider OKC as an odontogenic cyst. It occurs at all ages with a peak incidence between the second and the third decade of life. They are commonly seen in the mandible with the majority occurring in the angle of the mandible and ramus [5, 6]. OKC is usually asymptomatic and it's discovered on a routine dental radiograph until the cyst reached a large size and involved the entire ascending ramus, including sometimes the condylar and coronoid processes or even the maxillary sinus. Then, it may become symptomatic and it manifests as inflammation, pain, discomfort, or spontaneous discharge of cystic fluid through fistulization. Our patient was complaining of oral swelling on the right side of the mandible [7, 8]. The radiological image of OKCs is usually described as a well-defined unilocular or multilocular radiolucent lesion with few intralésional bony septa, thin borders, with or without a peripheral sclerotic halo. The mandibular canal is usually marked by large lesions, and the roots of adjacent teeth are displaced but rarely resorbed. In some cases, they resemble dentigerous cysts on radiographs when associated with impacted teeth.

Most OKC are single lesions, but multiple lesions may occur. This is known as the Gorlin-Goltz syndrome or "nevroid basal cell carcinoma syndrome" [9]. Diagnosis is confirmed through the radiographic appearance of the lesion, its clinical behavior, and most definitively, with histopathology which reveals two variants of OKC: orthokeratinized odontogenic cyst (OOC) and parakeratinized odontogenic cyst (POC) [10].

Differential diagnosis includes ameloblastoma, dentigerous cyst when associated with impacted teeth, traumatic cyst, giant cell granuloma and odontogenic myxoma [11]. In the case reported in this article, the patient was a woman in the 4th decade. Clinically, she presented a swelling in the right mandibular area which gradually increased in size. The panoramic X-Ray revealed a large multilocular radiolucency extended from the right posterior region of the body of the mandible to the ascending ramus. Histopathological features revealed an odontogenic keratocyst.

Different treatment modalities have been described, ranging from conservative techniques including conservative enucleation alone or associated with additional measures (ostectomy, Carnoy's solution, and cryotherapy), marsupialization and decompression to radical surgeries (marginal or segmental resection) [12]. However, there is no consensus or evidence for determining which is the most appropriate to treat OKCs [13]. The choice of conservative or radical treatment is based on multiple factors: patient age, size, location of the lesion, unilocular or multilocular form, soft tissue management and presence of cortical bone perforation. Enucleation is the most used treatment modality for decades. However, several studies concluded that about 17–56% of the recurrence rate has been reported for this procedure [14]. For that reason, many authors suggested the combination of enucleation and adjunctive therapies to eliminate any residual cyst lining and islands within the cyst wall. The main adjuvant used is the Carnoy's solution, which destroys cyst remnants using chemical cautery.

It is composed of 3 ml of chloroform, 6 ml of absolute ethanol, 1 ml of glacial acetic acid, and 1 g of ferric chloride. Marsupialization is another conservative modality, as well as decompression followed by enucleation [15]. In extensive or recurrent cases, an aggressive surgical treatment based on a radical resection with reconstruction would be indicated. Although this procedure is the treatment modality with the lowest recurrence rate (between 0% and 8.4%), this approach constitutes a severe mutilation. It can produce important aesthetic and functional sequelae which can significantly alter the patient's quality of life [16]. In our case, we opted for a conservative approach which consisted of enucleation. Even though OKC is a benign lesion, its biological characteristics make this cyst to be considered as an aggressive entity because of its high tendency to recur after surgical treatment. The mechanism of recurrence is still insufficiently understood [17]. One of the main reasons given for recurrence has been the incomplete removal of the cyst due to the thinness and friability of its capsule. Other reasons that would explain the recurrences could be the persistence of satellite cysts that would remain during surgery, the presence of cystic debris in the adjacent bone or mucosa, or the existence of epithelial islands in the lining mucosa. Recurrences were also found in the area associated with teeth that were not removed during the surgical treatment [18].

Therefore, the implementation of a close clinical control as well as a careful radiographic evaluation is important for successful treatment. After approximately 18 months of regular clinical and radiographic follow-up of our patient, no sign of recurrence has been observed and satisfactory bone healing was achieved.

3. CONCLUSION

The OKC (Odontogenic Keratocyst) is a commonly occurring lesion in clinical and histologic contexts that displays unique and aggressive behavior. Surgeons should conduct a comprehensive examination of each case and offer the most effective and appropriate treatment options to their patients. In order to guarantee long-term clinical success and

prevent recurrence, it is advisable to conduct periodic radiographic monitoring after treatment, in addition to an appropriate and ongoing long-term follow-up care.

Conflict of Interest: The authors declare that they have no conflict of interest.

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