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CASE REPORT

Radiographic features indicative of KOT (OKC) recurrence: A Case report

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Abstract

Background: Odontogenic keratocyst (OKC) is the third most common type of cyst in the jaws after radicular and dentigerous cysts.

Case Report: A 26-year-old female presented to the oral and maxillofacial radiology center for follow-up of OKC on her left mandible in April 2023, without any clinical symptoms. The radiographic view reveals expansion inferiorly to the border of the mandible, and septa were detectable in the lesion, which made recurrence a possibility.

Conclusion: Odontogenic keratocyst was confirmed by biopsy due to para-keratinized, stratified, and corrugated squamous epithelium and cheesy-like white keratin debris.

Key words: Odontogenic Keratocyst, Keratocystic Odontogenic Tumor, Radiographic Features, Mandible

Introduction

The odontogenic keratocyst (OKC) is particularly important due to its aggressive nature, high recurrence rate, and disputed treatment modalities. The term "odontogenic keratocyst" was first reported by Phillipsen (1). Odontogenic keratocysts (OKCs) account for 11.7% of all jaw cysts worldwide, making them the third most common type of cyst in the jaws after radicular and dentigerous cysts (2). In 2005, the WHO reclassified this pathology as a benign keratocystic odontogenic tumor (KCOT), however According to the WHO's last classification (2017), the keratocyst is an odontogenic cyst lined by "parakeratinized stratified squamous epithelium with palisading hyperchromatic cells" (3) Similar to other entities with an odontogenic origin, OKCs originate in tooth-bearing regions, and they occur twice as often in the mandible as in the maxilla (4). In the mandible, an OKC is usually present in the posterior area, which follows the angle and ramus of the mandible (5,6). Radiographically, OKCs usually appear as a well-defined unilocular or multilocular radiolucency bounded by corticated margins. Unilocular lesions are the predominant variant, whereas the multilocular variant is seen in approximately 30% of cases and mostly in the mandibular arch (7,8).

Okc can be confused with other jaw lesions such as ameloblastoma or dentigerous cysts (9). OKCs presenting with multilocular radiographic features such as visible septa on three-dimensional imaging are significantly more common in recurrent cases compared to primary lesions. The presence of septa/multilocular pattern on CBCT is associated with a higher risk of recurrence. These findings support that multilocular or septated OKCs on 3D imaging may require more careful follow-up due to their increased potential for recurrence (10). Histologically, OKC can be classified into two subtypes: parakeratotic and

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orthokeratotic. These subtypes are characterized by the type of keratin produced and the appearance of the epithelial lining. In the parakeratotic subtype, there is increased keratin production, and the cells lining the surface do not exhibit keratolytic granules.

Additionally, cells in the parakeratotic subtype tend to slough into the keratin layer. This classification is important because it helps further understand the characteristics and behavior of OKC lesions (11). Several options are available for the treatment of OKC depending on several factors. These options include: enucleation, curettage, resection, and marsupialization. The resection and enucleation are considered invasive surgical procedures, but they are the most common treatments (12). Marsupialization and decompression aim to reduce the size of the cystic lesion by lowering pressure and stimulating new bone formation. Peripheral ostectomy is a technique that involves enucleation of the lesion with peripheral bone removal using rotary instruments (13).

Case Report

A 26-year-old female presented to the oral and maxillofacial radiology center for follow-up of her left mandible in April 2023, without any clinical symptoms, such as pain, drainage, or neurosensory deficit, associated with the left inferior alveolar nerve. Patient has a history of surgery on the left mandible at the third molar region due to lesion extension from the distal of the second molar to the apical region of the wisdom tooth in 2016, which was evaluated by cone beam computed tomography and panoramic radiography (Figures 1, 2). The patient's pathology examination was performed on September27, 2016, and the results were as follows: Macroscopic Description: The submitted specimen consisted of a single piece of soft tissue, apparently cystic, cream-colored, measuring 30 × 10 × 5 mm.

Figure1. panoramic restoration view

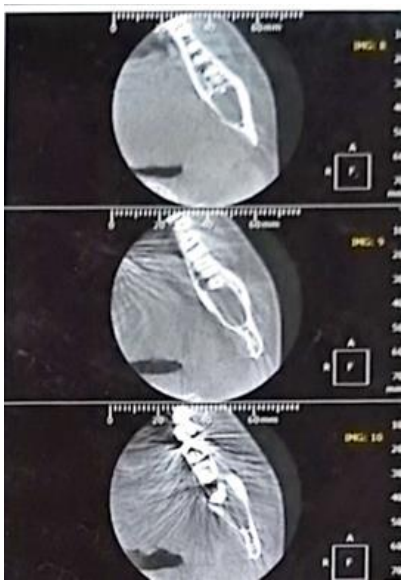

Figure 2. Axial view of the lesion

Microscopic Description:

Histological examination revealed a cyst lined by stratified squamous parakeratinized epithelium. The basal cell layer appeared columnar to cuboidal, exhibiting a palisading arrangement. The parakeratinized lining surface was corrugated. The cystic lumen contained clear serous fluid resembling transudate, along with keratin debris.

Diagnosis: Odontogenic keratocyst (Keratocystic odontogenic tumor) (Figure 3).

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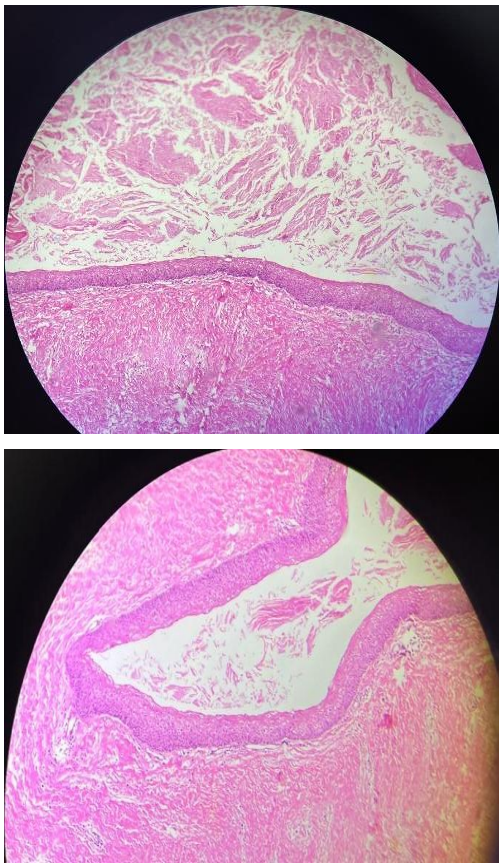

Figure 3. Histopathological view of odontogenic keratocyst tumor, demonstrating para-keratinized, stratified, and corrugated squamous epithelium, Red arrow. Yellow, palisading basal cells. Green, cystic lumen. Black, connective tissue. Light microscope 40x or 100x.

Recommendation: Given the high recurrence rate, clinical follow‑up every 6 months for 5 years is advised.

The patient was followed up in 2022 using three-dimensional imaging (CBCT) (Figure 4). The lesion extended from the root of the first left lower molar to the distal root of the lower left second molar, reaching the anterior border of the ramus and the alveolar crest to the inferior border of the mandible. The canal appeared to be displaced inferiorly. The borders of the lesion were well-defined and corticated. No obvious root resorption was noted in the involved region. Expansion of the buccal cortical plate was observed, and thinning of both buccal and lingual cortices was evident. The inferior border of the mandible appeared to be intact in the reconstructed sections.

Figure 4. Panoramic reconstruction view of odontogenic keratocyst tumor, on the left side of the body of the mandible, by lesion extension from the mesial of the first molar, towards the angle of the mandible.

The canal inside the lesion, from the mesial of the first molar to the distal of the second molar, was not visible, and canal discontinuity was noted in this region. The maximum dimensions of the lesion were approximately 41 mm anteroposteriorly and 11 mm buccolingually. Since the patient did not present with any specific clinical symptoms, follow-up was recommended for 6 to 12 months. If the lesion size remained unchanged, the possibility of recurrence should be considered, and appropriate treatment measures would then be undertaken. There was no clinical significance in 2023, either. Cone beam computed tomography was performed with a slice thickness and interval of 2mm. Examination of coronal, axial, and cross-sectional views illustrates an average multilocular radiolucency from the mesial of the first molar to the angle of the mandible. The lesion expands inferiorly to the border of the mandible, alongside displacing the inferior alveolar canal nerve to the same position (Figure 5a). Although the lingual cortex became thin in some areas, no perforation was detected. No root resorption was observed, but a septum was detectable in the lesion, which raised the possibility of recurrence (Figure 5b).

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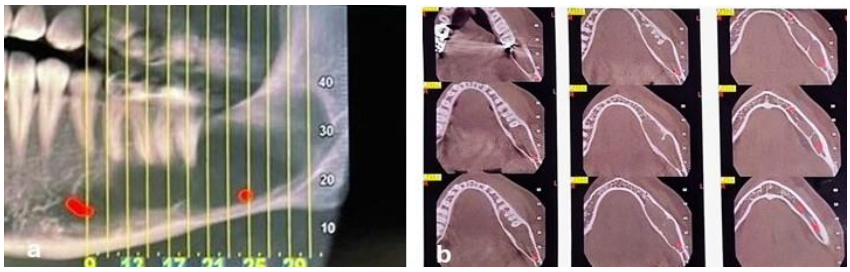

Figure 5. a) Panoramic reconstruction view. & **b)** Axial view of the odontogenic keratocyst tumor, on the left side of the body of the mandible, by lesion extension from the mesial of the first molar, towards the angle of the mandible.

The biopsy was performed, and the submitted specimen comprises a single fragment of soft tissue with a cystic appearance and a cream color, measuring approximately 2 cm in the anteroposterior dimension and 1.5 cm in the vertical dimension, fixed in 10% buffered formalin. The biopsy revealed para-keratinized, stratified, and corrugated squamous epithelium. The cylindrical or cubic basal cell layer was arranged in a palisade, parallel pattern the cyst lumen contained a lucid liquid resembling serum transudate, with cheesy, white keratin debris. Therefore, an odontogenic keratocyst was confirmed by biopsy. Enucleation was performed for the primary lesion. Eventually, owing to the patient's history and radiological findings, recurrent KOT was diagnosed, confirmed by incisional biopsy (Figure 6), and the patient was referred for surgical treatment. The first step of treatment was marsupialization due to the diminishing side effects of leaving the lesion at its actual size.

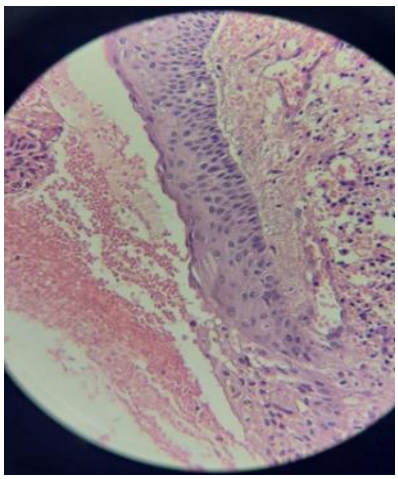

Figure 6. Histopathological view of odontogenic keratocyst tumor demonstrated para-keratinized, stratified, and corrugated squamous epithelium, Red arrow. Yellow arrow, palisading basal cells. Blue arrow, satellite cysts. Light microscope 40x or 100x

After 6 months, another CBCT and Panoramic radiograph were taken (Figure 7a, b). The lesion had shrunk; therefore, surgical removal was performed (Figure 8). Excision of the overlying mucosa with the cysts and curettage with Carnoy's solution has been performed in this case (Figure 9).

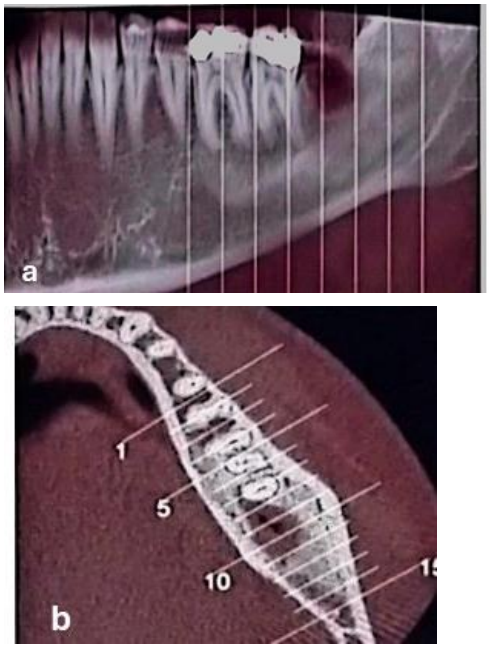

Figure 7. a)Panoramic reconstruction view. & **b)** Axial view of the odontogenic keratocyst tumor, on the left side of the body of the mandible, with the lesion limited to the distal region of the second molar.

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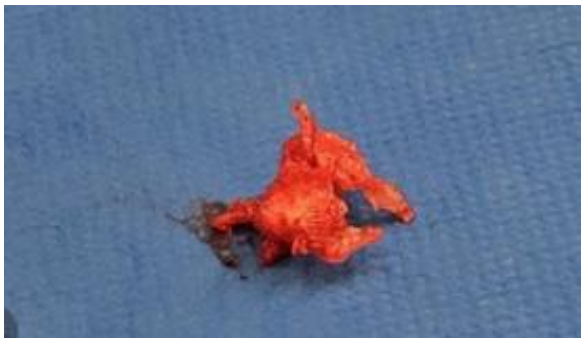

Figure 8. Enucleated Tissue of shrunk KOT, due to marsupialization

Figure 9. Intraoral photograph right lateral (after tissue enucleation), and placing a soaked gauze with carnoy solution for a short period of time, and then removed.

Discussion

OKC is a common developmental cyst, possibly originating from remnants of odontogenic epithelium, whether from the tooth germ or the dental lamina. Although in previous WHO classifications, OKC was considered to have dual features as a cyst or tumor, in the most recent classification, it is exclusively considered a cyst (14) due to insufficient evidence to categorize it as a neoplastic lesion (15). This cyst has aggressive behavior and a high recurrence rate (%5-%62.5) (14). Several theories have been proposed to explain the invasive and destructive nature of OKC. Growth in OKC is linked to unknown growth factors inherent in the epithelium itself or enzymatic activity in the fibrous wall (16). Its invasion and infiltration are attributed to the multicentric potential for cystic growth, driven by the proliferation of local epithelial cell groups (17). Moreover, few or incomplete septa may be seen in unilocular OKCs on mandibular panoramic radiographs, although this finding is more common in larger cystic lesions.

Approximately 30% of OKCs are usually associated with unerupted teeth, most commonly the third molars. According to the literature, OKCs may be located in the periapical, pericoronal, or lateral root areas. In approximately 30% of cases, there was no relationship to dental structures (18). In the present case, prior to recurrence, the lesion was associated with a semi-impacted wisdom tooth, which was removed along with the lesion. Treatment options include decompression, marginal resection, en bloc resection, and adjuvant therapy, like cryotherapy, peripheral ostectomy, and Carnoy's solution (19). A major shift occurred in our understanding and management of keratinous cysts in 2005 when they were classified as tumors. However, the reclassification of these lesions as odontogenic cysts in 2017 was based on new evidence regarding their behavior and histological composition, especially that they tend to regress after decompression. In recent years, many scientific papers have been published on the efficacy of marsupialization for the treatment of various types of cystic lesions in the oral region (20, 21), including cystic ameloblastoma (22). The marsupialization was described as a less aggressive option because it maintains the developing dentition, reduces cyst size (20, 22), minimizes damage to important anatomical structures (inferior alveolar nerve and sinus), and stimulates osteogenesis (22). Several studies have shown that the recurrence rate of keratinized cysts is highest when the lesion is managed by enucleation alone, yet it is the most common surgical option for the treatment of OKCs in the literature (23). The lowest recurrence rate of keratinized cysts in the literature was after resection (ranging from 1.85 to 2.2%) (24). Although it is

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associated with the lowest recurrence rate, it is the most aggressive procedure, which causes many postoperative complications and requires a specific plan for reconstruction.

Marsupialization is currently considered a non-definitive therapy in KCOT treatment by the majority of authors due to the presence of odontogenic epithelium g in the cyst cavity (25). Marsupialization requires complementary therapies, including simple enucleation, radical enucleation, enucleation with Carnoy's solution, enucleation with cryotherapy, and enucleation with peripheral ostectomy. The combination of enucleation with adjunctive techniques reduces the high recurrence rates observed with simple enucleation and optimizes treatment (20, 21). Simple enucleation differs from enucleation with peripheral ostectomy in that the enucleation with peripheral ostectomy is followed by the removal of 1.5–2 mm of bone with a handpiece in KCOT margins. Several studies reported that the combination of enucleation (26) with peripheral ostectomy yielded positive results, with low recurrence rates (26–29). A 2005 retrospective review found that enucleation followed by peripheral ostectomy was associated with lower recurrence rates. In 2007, Kolokythas et al. (27) reported a recurrence rate of 0% with enucleation followed by peripheral ostectomy.

Patient characteristics are major determinants, including gender, health status, age, and reliability of follow-up. It is advised to use resection for the more aggressive cysts with perforation of buccal and lingual plates (30).

The choice of treatment depends on the cyst's size, location, and characteristics. Based on these findings, it can be concluded that multilocular or septated OKCs on 3D imaging may warrant closer follow-up due to their increased risk of recurrence (31).

Conclusion

The choice of treatment depends on the cyst's size, location, and characteristics. Based on these findings, it can be concluded that multilocular or septated OKCs on 3D imaging may warrant closer follow-up due to their greater potential for recurrence. It can be concluded that the combination of treatment with enucleation and peripheral ostectomy, followed by the application of Carnoy's solution to the bony cavity for no more than 3 minutes, appears to be efficient for treating KCOT by diminishing the recurrence rate to 0% during the most critical period (the first 3 years), and its equal to that of the resection. The complications and morbidity originating from the application of Carnoy's solution occurred less frequently and were less serious than those associated with resection.

Conflict of Interests: The authors of this manuscript declare that they have no conflicts of interest, real or perceived, financial, or non-financial in this article

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