Case Report

Conservative Management of Mandibular Odontogenic Keratocystic with 5-Year Annual Follow-up: A Case Report and Literature Review

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Abstract

Odontogenic keratocyst (OKC) is a developmental jaw cyst that arises from odontogenic epithelium. OKCs were recently classified as an odontogenic cyst in the 2017 World Health Organization classification of Head and Neck tumors based on clinical and histopathological features. Because of their diverse nature and high recurrence rates, the management of OKC has been the subject of numerous debates, controversies, and speculations. The purpose of this report is to present a 21-year-old female patient with asymptomatic swelling on the left side of her lower jaw who was treated conservatively with peripheral osteotomy and chemical cauterization for mandibular OKC and to evaluate the radiological features postoperatively. Normal bone regeneration and no evidence of recurrence were noticed during a 5-year annual follow-up. The conservative treatment protocol was compared to the management strategies proposed in the literature.

Keywords: Odontogenic keratocyst, Classification, Recurrence, Conservative treatment.

Introduction

Odontogenic keratocyst (OKC) is a benign lesion that is distinguished by its clinical behavior. Philipsen reported and depicted the first case in the literature, naming it an OKC.[1] Pindborg and Hansen[2] defined its characteristics. OKC is a distinct lesion due to its aggressive behavior, high recurrence rate, and remarkable histologic appearance.[3]

OKC was previously classified as a developmental odontogenic cyst of the jaw by the WHO in 1971 and 1992, but it was reclassified and renamed keratocystic odontogenic tumor (KCOT) in the WHO classifications of head and neck tumors in 2005 due to its aggressive behavior, high recurrence rates, and specific histological characteristics. According to the WHO, it is a benign uni- or multisystem intraosseous tumor of odontogenic origin (dental lamina and its remnants) with a characteristic stratified squamous epithelial lining and the potential for aggressive and infiltrative behavior. The WHO gave the term KCOT because it demonstrated a neoplastic nature.[4] In 2017, the new WHO classification of headandneck pathology reclassified OKC back into the cystic category based on its clinical and histological features.[5] Payne[6] found no differences in the clinical diagnosis of the OKC based on gender predilection. Brannon[7] reported a predilection for male gender. These lesions could be diagnosed at any age; 60% of cases occurred in people aged 10–40 years.

The cystic expansion is very minimal in the initial stage, which is due to the cystic characteristic of growing in an anteroposterior direction in the medullary space of the bone. Buccal cortex expansion is seen in 30% of maxillary and 50% of mandibular regions.[8] Concerning the topographic sites for the occurrence of OKC, the majority of studies disclosed that they were diagnosed primarily in the body, ascending branch, and mandibular angle.[9] Conservative methods such as simple enucleation, decompression, or marsupialization, as well as aggressive or non-conservative approaches such as cryosurgery or chemical destruction, and radical surgery with bone resection are being employed in the management of OKCs.

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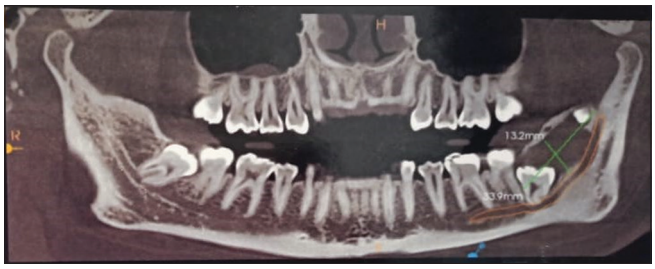

Figure 1: Pre-operative OPG showing unilocular radiolucent lesion distal to the left lower second molar. Impacted and supernumarary teeth bud are noted

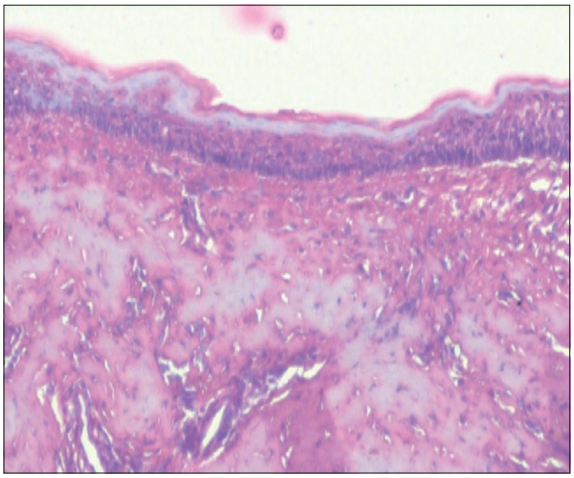

Figure 2: Histopathological image of the lesion showing parakeratotic cells

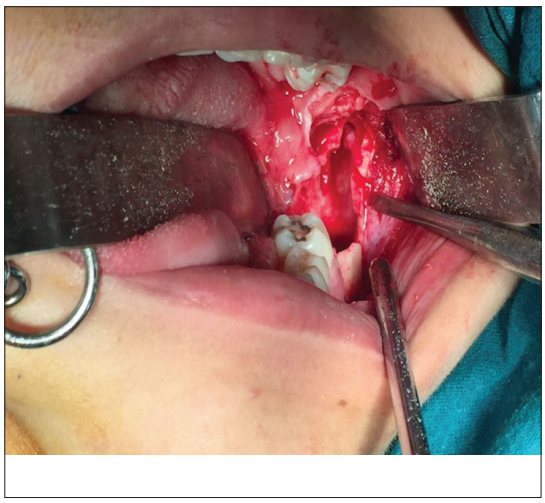

Figure 3: Transoperative picture of OKC enucleation

Figure 4: 6 months post-operative OPG

Figure 5: Twelve-month post-operative OPG

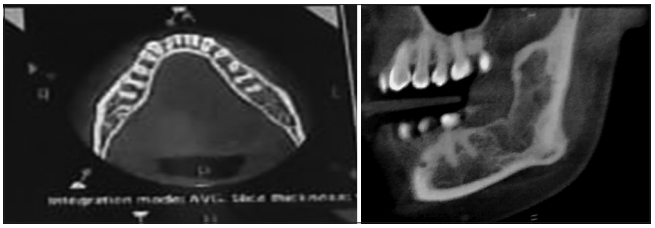

Figure 6: CBCT axial and sagittal slices of lesion 12-month postoperatively

Figure 7: OPG at 2nd post-operative year

The success of the conservative treatment was validated by a study by Stoelinga, in which enucleation, along with the application of Carnoy's solution and excision of the attached, overlying mucosa, resulted in very few recurrences occurring within 5 years.[10] During the first 5 years after treatment, the OKC has a high recurrence rate of around 7–28%. As a result, following the treatment of OKCs, a long-term follow-up is advised to determine the success of the procedure.[11] This case report aimed to use a 5-year annual study to evaluate the bone architecture and bone density in a patient diagnosed with parakeratinized OKC and to evaluate the success of conservative management.

Case Report

A 21-year-old female patient presented to the department of oral and maxillofacial surgery with an 8-month-old

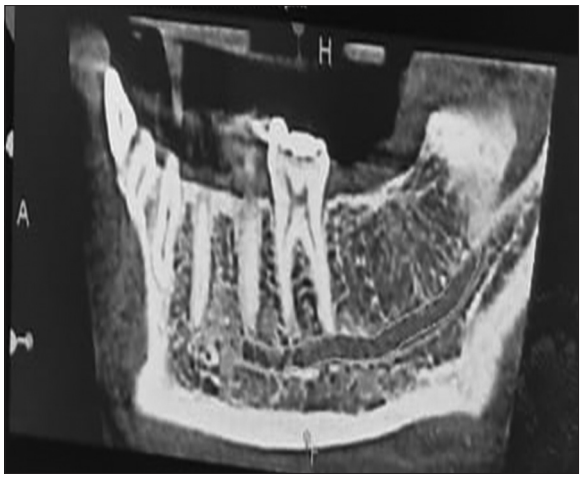

Figure 8: CBCT sagittal view at post-operative 2nd year

Figure 9: OPG at 3rd post-operative year

Figure 10: OPG at 4th post-operative year

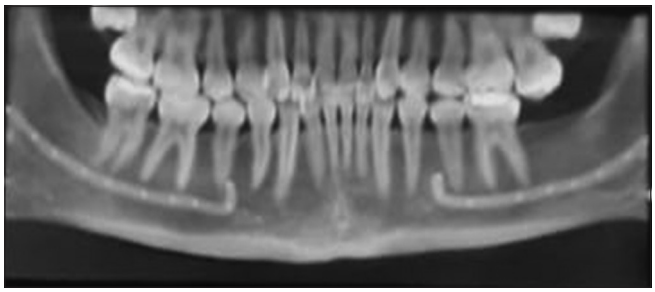

Figure 11: Panoromic view at 5th post-operative year

asymptomatic swelling on the lower left side of her face. There was no evidence of trauma. There was no systemic illness present, according to the medical history. When asked for a personal history, there was no history of deleterious habits.

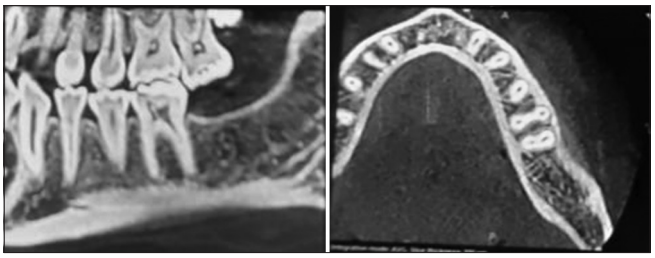

Figure 12: Sagittal and axial slices at 5th post-operative year showing good bone regeneration and no signs of recurrence

The patient's extra-oral examination disclosed a diffuse swelling on the lower left side of the face which extends anterio-posteriorly from the angle region to the ascending ramus. Distal to the left lower second molar, the overlying mucosa was inflamed and erythematous. Palpation revealed that it was hard in consistency, mildly tender, and non-fluctuating. Intraoral examination revealed buccal vestibular obliteration between 36 and 37 teeth.

The orthopantomogram and cone-beam computed tomography (CBCT) revealed a non-expansile well-defined hypodense bony lesion in the left retromolar anterior aspect of the ascending ramus. The lesion is encased in the pericoronal aspect of unerupted/bony impacted teeth, in relation to 38. There was a small tooth bud at the most superior aspect of (Figure 1) the sigmoid notch level. There was no evidence of cortical expansion or erosion. There was no root resorption. During the lesion, there were certain decortications at the superior border of the inferior alveolar canal.

A straw-colored cystic fluid was revealed during an aspiration test of the lesion. With the above clinical findings, a provisional diagnosis of OKC in this region was made, with dentigerous cyst, unicystic ameloblastoma, and calcifying odontogenic cyst as differential diagnoses. An incisional biopsy performed distal to 37 teeth revealed histological features resembling parakeratinized OKC with 7–8 layers of the squamous epithelial lining and a palisading basal layer. Parakeratotic (Figure 2) cells were found on the epithelial surface. In some areas, the cystic wall is fibrous and inflamed. Conservative treatment of OKC with enucleation and peripheral osteotomy, followed by chemical cauterization with Carnoy's solution (Figure 3), was meticulously planned and executed under general anesthesia. The impacted 38 teeth and additional supernumerary teeth were removed before chemical cauterization. Carnoy's solution was applied as a chelating agent which was embedded in a gauge for 3 min. The patient was subjected to periodic monitoring at regular intervals. For 5 years, radiological and clinical features were assessed for evidence of recurrence and bone regeneration using panoramic and CBCT imaging. A favorable healing process was noted with no signs of recurrences 5 years postoperatively.

Discussion

The literature has established a range of treatment modalities for OKCs, including conservative and non-conservative approaches (OKC) since 2005. Conservative methods such as simple enucleation, decompression, or marsupialization, non-conservative approaches such as cryosurgery or chemical destruction, and aggressive approaches such as radical surgery with bone resection. However, there is no agreement or sufficient evidence to determine, in which technique is the most suitable for treating OKCs. Lower recurrence rates were observed in patients treated with more aggressive treatments such as bone resection (0%), 7.8% for cases managed with enucleation and Carnoy's solution, and 11.5% for cases addressed with enucleation and liquid nitrogen.[11] Syndromes associated with multiple OKCs are nevoid basal cell carcinoma syndrome, Gorlin-Goltz syndrome, Marfan syndrome, Ehlers-Danlos syndrome, Noonan syndrome, orofacial digital syndrome, and Simpson-Golabi-Behmel syndrome.[12]

It was recently recommended and established in the previous literature that more hostile treatment has many disadvantages when compared to the clinical findings of the conservative approach[13] because the latter produced significant morbidity. Enucleation and peripheral osteotomy, followed by Carnoy's solution application, were used as the first line of treatment in this case report to reduce the possibility of OKC recurrence. Carnoy's solution, which was first used as a fixative agent in laboratory pathology,[14] is injected into the bone cavity to promote superficial chemical necrosis of up to 1.5 mm^2 to eliminate tumor tissue remnants.

Six months after surgery, the CBCT imaging reveals a healing post-operative defect with no signs of recurrence. One-year post-operative radiograph revealed an irregular (Figure 4) cystic defect with smooth bony margins (Figures 5-6) filled with osteopenic bone density and a few tiny and coarse irregular trabecular patterns with relatively type IV bone density. The thickness of the cortical plate was <1 mm. Second-year post-operative imaging shows cortication of the inferior alveolar canal near the (Figures 6-7) previous surgical site. A dense coarsely placed trabecular pattern resembled type III bone density. There was also bony continuity on the anterior border of the ascending ramus on the mandible's left side. The 3rd-year post-operative radiograph shows an enhancement in the trabecular pattern with cortical bone (Figure 8), that is., thickness of approximately 1 mm. In the 4th post-operative year, there was a regular arrangement of trabecular patterns, indicating type II bone density (Figure 9). When compared to the previous years, the 5th post-operative year shows a regular arrangement of trabecular pattern matching the surrounding adjacent bone with no evidence of recurrence and indicating good bone regeneration at the surgical site (Figures 10-11).

The majority of recurrence cases are seen within the first 5 years of treatment. The typical reasons for recurrence are parts of the lining being left behind, the presence of microcysts in the connective tissue of the cystic wall and the development of a new lesion from the epithelial island left behind the mucosa.[15] According to studies done by Stoelinga *et al*.,[15] a small KCOT where the margins can be accessed may be enucleated with adjunctive therapy such as Carnoy's solution which was similar to this present case report where enucleation, peripheral osteotomy, and Carnoy's application were carried out and showed a good post-operative bone regeneration with no evidence of recurrence.

Conclusion

Conservative management of OKCs with adjunct therapy may be the best alternative in the treatment of small OKCs. Even though bone regeneration is significantly reduced, the procedure is simple, effective, and prevents bone continuity loss that occurs in aggressive resections. Because of its peculiar aggressive behavior and high recurrence rate, a 5-year follow-up is recommended.

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