Case Series

Management of Odontogenic Keratocyst (OKC): A Case Series

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ABSTRACT

Odontogenic keratocyst (OKC) is significant in oral and maxillofacial surgery due to its diverse manifestations and high recurrence rate. The treatment protocol for OKC is controversial and treatment modalities range from enucleation to resection including adjunctive procedures like peripheral ostectomy, cryotherapy, and chemical cauterization. This paper discusses eight cases diagnosed with OKC which was confirmed by histopathological examination, and subsequently underwent enucleation along with chemical cauterization with modified Carnoy's solution. Kinds of literature have proved that enucleation with the application of modified Carnoy's solution has reduced the recurrence rate considerably. Our case series illuminates that the treatment approach adopted in our institute has decreased the recurrence rate while maintaining functional and esthetic aspects.

Key words: modified carnoy's solution, Enucleation, chemical cauterization, Odontogenic keratocyst (OKC)

The treatment of odontogenic keratocyst (OKC) is highly debated among oral and maxillofacial surgeons. Approaches range from simple enucleation for lesions smaller than 1 cm to extensive resection for cysts extending into the skeletal base. OKC occurs more frequently in men, the mean age group was the second and third decade of life and the most frequent site was the lower jaw and the recurrence rate was 20%. In 1869, Broca introduced a classification for odontogenic tumors that remained in use until 1914.[1] Thoma and Goldman later reclassified odontogenic tumors in 1946 into categories of ectodermal, mesodermal, or mixed origin.[2] The World Health Organization (WHO) first included cystic lesions in its 1971 classification, maintained through 1992, until Philipsen and Reichart reclassified them as tumors in 2005, naming them keratocystic odontogenic tumors due to their high recurrence rate, however, the World Health Organization (WHO) reclassified OKC back into the cystic category in 2017.[3,4]

The aggressive behavior of OKC has long puzzled surgeons. Its expansion is driven by osmotic tension on adjacent tissues, inherent epithelial forces, or enzymatic activity from the fibrous wall. The expression of Bcl-2 and cytokeratin 10 in the supra-basal and basal cell layers confers anti-apoptotic properties, leading to increased epithelial cell survival and significant mitotic activity.[5,6] The aggressive nature of OKC, with its para-keratinized epithelium, is managed using decompression followed by enucleation, including the removal of the overlying mucosa. This treatment is enhanced by the application of modified Carnoy's solution.[7,8] Modified Carnoy's solution works as a chemical cauterizing agent, essentially "fixing" and destroying any remaining epithelial tissue or small cysts around the edges of a surgical site. Primarily, it is used in the treatment of OKC, by causing cell necrosis through its combination of alcohol, acetic acid, and ferric chloride, effectively preventing recurrence by eliminating potential residual tumor cells. Bismuth Iodoform Paraffin Paste (BIPP) is a paste or gauze that is used to dress wounds and pack cavities. It works by making the gauze impermeable to blood and body fluids, which reduces the nutrition available to bacteria.[9]

This paper discusses cases diagnosed through biopsy, treated with decompression followed by enucleation, and the application of modified Carnoy's solution and electrochemical cauterization.

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CASE PRESENTATION

This report discusses eight cases of OKC, each presenting with extensive lesions due to anterior-posterior expansion, yet treated conservatively. A biopsy was performed to confirm the diagnosis and decompress the lesion, promoting thickening of the lining epithelium. Subsequently, the lesions were enucleated, with careful removal of the epithelial lining fragments from the cystic cavity and the overlying mucosa.

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The affected areas were then subjected to chemical or electrical cauterization. The summary of all cases are summarized in Table 1.

CASE 1

A 23-year-old woman presented to our department with continuous mild gnawing pain over the right angle of the mandible for the past two weeks, accompanied by progressive trismus limiting her mouth opening to 26 mm and signs of inferior alveolar nerve paresthesia. An orthopantomogram (OPG) revealed the extent of the lesion (Figure 1a). The aspirate consisted of a cheese-like creamy material. A biopsy with decompression was performed, and the histopathological diagnosis confirmed an OKC. The patient was advised to undergo cyst enucleation under general anesthesia. During the procedure, the cystic cavity was completely enucleated, and the inferior alveolar nerve was lateralized (Figure 1b). The cavity was then cauterized with modified Carnoy's solution for no more than two minutes. The bony margin forming the opening of the cystic cavity, along with the attached overlying mucosa, was removed, and the surgical site was closed primarily.

CASE 2

A 39-year-old male reported to our department with a chief complaint of mild pain and swelling at the right angle of the mandible that was gradually increasing in size. The patient exhibited inferior alveolar nerve paresthesia, showing its involvement. An OPG and computed tomography (CT) scan revealed a huge cystic lesion extending from the first premolar on the right to the distal aspect of the second molar involving the angle of the mandible and extending up to ramus region (Figure 2a). A CT scan was performed to assess the extension of the cyst and the relation of the cyst to the inferior alveolar nerve accurately. The Histopathological report (HPE) report from the incisional biopsy done at the apical region of left mandibular first premolar revealed it as ortho-keratinized OKC. The cystic lesion was enucleated along with chemical cauterization by modified Carnoy's solution under general anaesthesia (figure 2b) followed by packing of cystic cavity with Bismuth Iodoform Paraffin Paste (BIPP) soaked gauze. The excised specimen was sent for histopathological analysis and the report confirmed the initial diagnosis.

CASE 3

A 35-year-old female with a complaint of pain in the right third molar region for the past two weeks reported to our department. On examination, a swelling was observed on the right third molar region involving the angle of the mandible along with an impacted tooth which on palpation was found to be tender, non-fluctuant, and non-pulsatile with no buccolingual cortical expansion. OPG revealed it as a well-defined radiolucency extending from the distal aspect of 48 to the ramus of the mandible, antero-posteriorly. The HPE showed features suggestive of ortho-keratinized OKC. Treatment was administered in the form of enucleation along with the excision of the involved overlying mucosa (Figure 3a). Chemical cauterization with modified Carnoy's solution followed by primary closure was accomplished. Primary HPE revealed the lesion as OKC.

CASE 4

A 50-year-old male presented to our department with a concern about halitosis and fluid leaking intraorally from the right angle of the mandible. OPG revealed a well-scalloped radiolucent lesion extending from the mesial aspect of the third molar to the ascending ramus of the mandible. Incisional biopsy revealed it as suggestive of para-keratinized OKC that received surgical intervention in the form of enucleation with chemical cauterization under general anesthesia (figure 3b) with open dressing and debridement carried out meticulously as an outpatient. The patient was followed up at a regular interval. The final HPE was in accordance with the initial diagnosis.

CASE 5

A 25-year-old female presented with an ongoing concern of pain in the right mandibular second molar region for the last 3 months. On examination, a firm non-fluctuant swelling associated with pain was observed. OPG revealed an impacted tooth with a well-defined scalloped unicystic lesion extending from the distal aspect of the second molar to the angle of the mandible involving the inferior third of the ramus (figure 4a). The biopsy report revealed it as ortho-keratinized OKC. The cystic lesion was treated with enucleation followed by chemical cauterization and packing of the cystic cavity with the BIPP-soaked gauze. For one year, the gauze was replaced every two weeks, until the achievement of the desired healing.

CASE 6

A 35-year-old male presented with an ongoing concern of pain in the left mandibular third molar region for the last 8 months. On examination, a firm non-fluctuant swelling associated with pain was observed. OPG revealed an impacted tooth with a well-defined scalloped unicystic lesion extending from the distal aspect of the first molar to the angle of the mandible involving the inferior third of the ramus (figure 4b). The biopsy report revealed it as para-keratinized OKC. The treatment modality adapted was cystic enucleation followed by chemical cauterization.

CASE 7

A 49-year-old female reported to our department with a chief complaint of mild pain and swelling at the right angle of the mandible that was gradually increasing in size. The patient exhibited inferior alveolar nerve paresthesia, showing its involvement. An OPG and CT scan revealed a cystic lesion extending from the mesial aspect of the third molar involving the angle of the mandible and extending up to the ramus region. A CT scan was performed to assess the extension of

the cyst and the relation of the cyst to the inferior alveolar nerve accurately. A biopsy with decompression was performed, and the histopathological diagnosis confirmed an ortho-keratinized OKC (figure 5). The patient was advised to undergo cyst enucleation under general anesthesia. During the procedure, the cystic cavity was completely enucleated, and the inferior alveolar nerve was lateralized. The cavity was then cauterized with Carnoy's solution for no more than two minutes. The bony margin forming the opening of the cystic cavity, along with the attached overlying mucosa, was removed, and the surgical site was closed primarily.

CASE 8

A 45-year-old male with a complaint of pain in the right third molar region for the past two weeks. On examination, a swelling was observed on the right first molar region involving the angle of the mandible along with an impacted tooth which on palpation was found to be tender, non-fluctuant, and non-pulsatile with no buccolingual cortical expansion. OPG revealed it as a well-defined radiolucency extending from the distal aspect of the right mandibular first molar to the ramus of the mandible, antero-posteriorly. The HPE showed features suggestive of ortho-keratinized OKC. Treatment was administered in form of enucleation along with the excision of the involved overlying mucosa. Chemical cauterization with modified Carnoy's solution followed by primary closure was accomplished. The primary HPE report revealed the lesion as OKC.

Table 1: Comparison of all cases

Sl no.	Age	Gender	Site	Relation with inferior alveolar nerve	HPE report	Treatment	Recurrence
1.	23	Female	Right mandibular angle	Involve Paraesthesia present	Parakeratinized	Enucleation followed by chemical cauterization	No
2.	39	Male	Right mandibular ramus	Involved Paraesthesia present	Orthokeratinized	Enucleation followed by chemical cauterization	No
3.	35	Female	Right angle of mandible	No involvement	Orthokeratinized	Enucleation followed by chemical cauterization	No
4.	50	Male	Right angle of the mandible	No involvement	Parakeratinized	Enucleation followed by chemical cauterization	No
5.	25	Female	Right angle of the mandible	Involved Paraesthesia present	Orthokeratinized	enucleation followed by chemical cauterization	yes
6.	35	Male	right angle of the mandible	No involvement	para-keratinized	Cystic enucleation followed by with modified Carnoy's solution	No
7.	49	Female	right angle of the mandible	Involved Paraesthesia present	Ortho-keratinized	Cystic enucleation followed by with modified Carnoy's solution	No
8.	45	male	Right side of angle of mandible	No involvement	Orthokeratinized	Chemical cauterization with modified Carnoy's solution	No

Figure 1: (a) OPG revealed a large radiolucent lesion in right mandibular angle region; (b) Enucleation of the cyst in right mandibular angle region followed by lateralization of the inferior alveolar nerve and application of Carnoy's solution (Case 1)

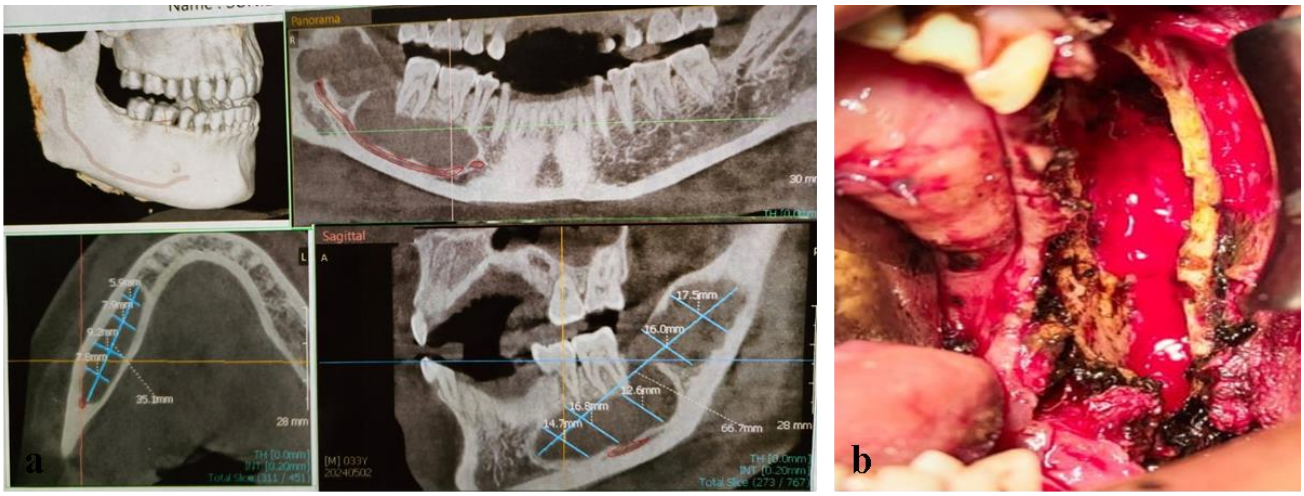

Figure 2: (a) OPG and CT scan revealed a radiolucent lesion extending anterior-posteriorly right side mandibular angle region and shows inferior alveolar nerve relationship with lesion; (b) cystic lesion of right mandibular angle region was enucleated along with chemical cauterization by modified Carnoy's solution (Case 2).

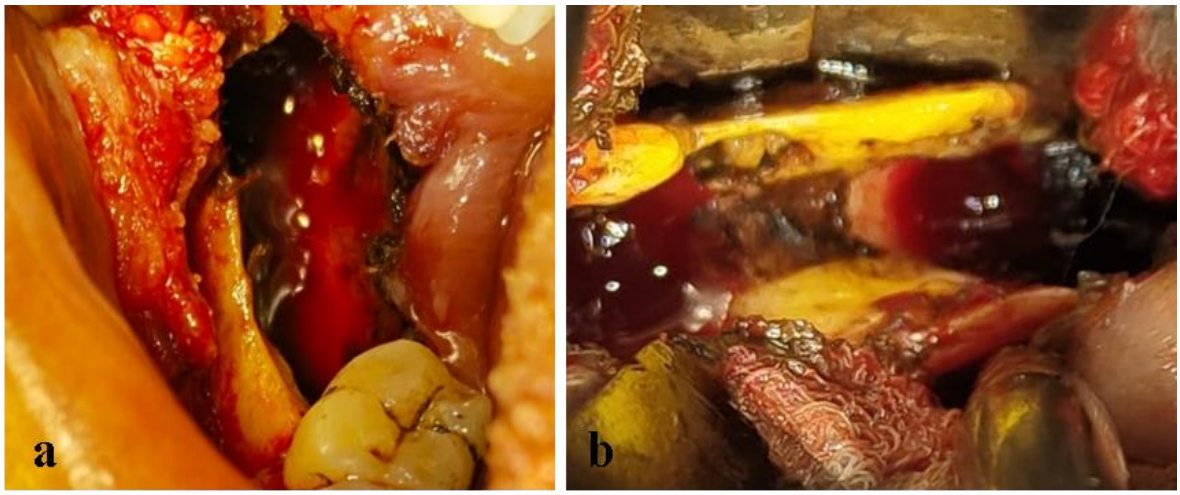

Figure 3: (a) Enucleation was carried out and chemical cauterization by modified Carnoy's solution in right side of mandibular angle and ramus region (Case 3); (b) Enucleation with chemical cauterization in the right side of mandibular angle and ascending ramus region (Case 4).

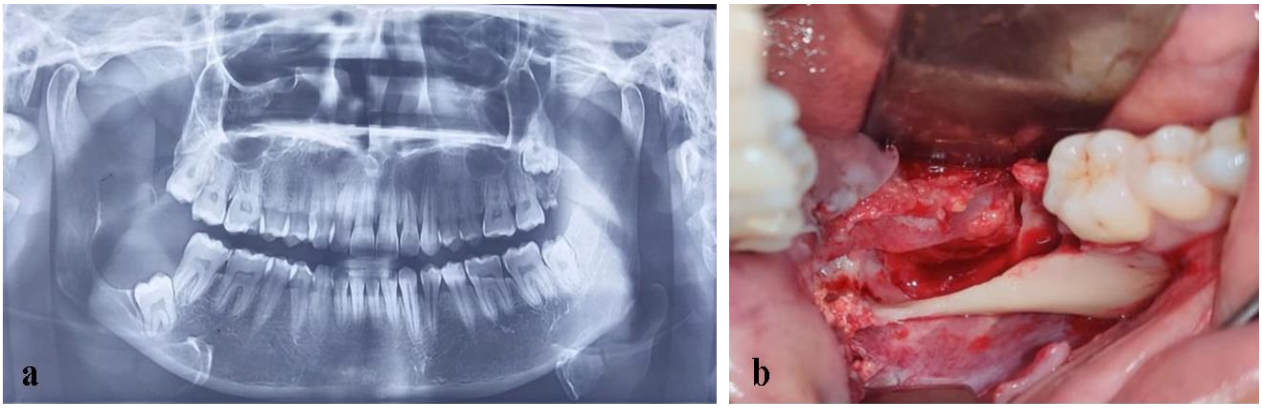

Figure 4: (a) OPG revealed that a scalloped bordered with radiolucent lesion in the right side of the mandibular angle region (Case 5); (b) bony margins forming the opening for the cystic cavity, along with its attached overlying mucosa, were removed in the right side of the mandibular angle region (Case 6)

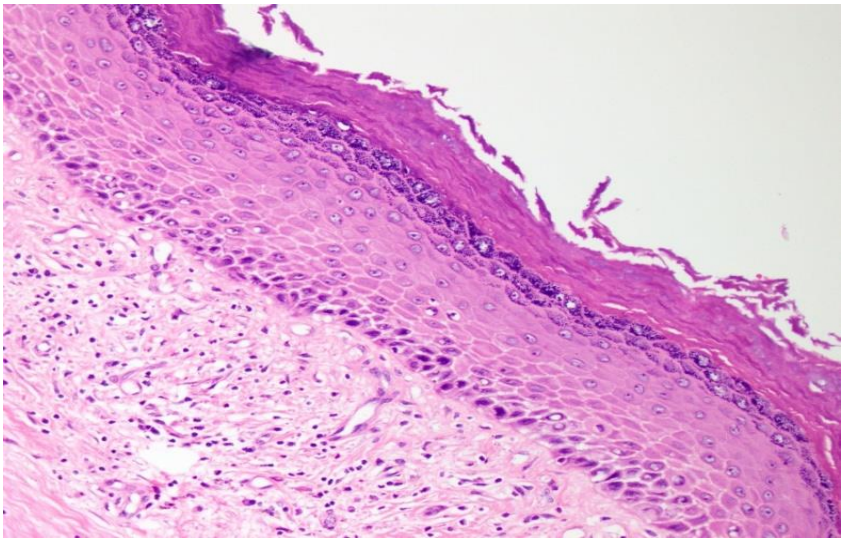

Figure 5: Non-inflamed fibrous cyst wall lined by thin uniform stratified squamous epithelium (H&e stain; 40X magnification)

DISCUSSION

The OKC is a jaw cyst known for its tendency to locally invade and recur. It is a unique type of developmental odontogenic cyst, and its effective management remains a topic of ongoing interest and debate in oral and maxillofacial surgery literature.[9] OKCs are characterized by rapid growth and a propensity to invade surrounding tissues, including bone. They have a high recurrence rate and are associated with basal cell nevus syndrome and Gorlin Goltz Syndrome.[10]

OKC occurs more frequently in men, the mean age group second and third decade of life, and can occur in any part of the upper or lower jaw, with the majority found in the mandible, particularly in the angle and ramus. In the maxilla, these cysts are generally smaller but can cause bone expansion when large. Advances in molecular biology and genetics have led researchers to suggest reclassifying OKCs as benign tumors, renaming them keratocystic odontogenic tumors (KCOT).[11, 12]

Various treatment options for OKC include enucleation and curettage, marsupialization, and adjunctive methods such as cryotherapy, modified Carnoy's solution, peripheral ostectomy, excision of overlying mucosa, 5-fluorouracil (5-FU), and resection.[13] Stoelinga and Van Hoelst proposed a conservative approach for large lesions, involving enucleation, excision of the overlying mucosa and/or muscle if attached, to eliminate epithelial rests and/or microcysts, followed by careful cauterization of the bony defect with Carnoy's solution.[14] Due to concerns about recurrence, modified Carnoy's solution has been suggested as an adjunctive treatment post-marsupialization. This solution comprises chloroform, absolute ethanol, glacial acetic acid, and ferric chloride. A systematic review of Carnoy's solution for treating OKCs categorized the evidence as grade C.[15] In 2017, Moraissi et al. conducted a meta-analysis to determine the best surgical treatment with the lowest recurrence rate, concluding that radical resection is the most reliable method for minimizing recurrence. Enucleation with the application of Carnoy's solution or cryotherapy is recommended as the first-line treatment to reduce recurrence.[16] Recurrence rates for OKCs range from 5% to about 50%. Postoperative follow-up should be conducted yearly for five years and then once every two years, provided the patient cooperates.[17,18]

CONCLUSION

Odontogenic keratocysts are generally believed to originate from either the epithelial remnants of the tooth germ or the basal cell layer of the surface epithelium. They can occur in any part of the upper or lower jaw, with mandibular predilection, particularly in the angle and the ramus region. This case series on OKC has shown that our treatment approach has helped in the elimination of the pathology along with decreasing the chances of recurrence while maintaining the functional and aesthetic aspects of the mandible.

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