

# Conservative Management of Odontogenic Keratocyst in a Tertiary Hospital

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## Abstract

**Introduction:** Odontogenic keratocysts (OKCs) are benign intraosseous odontogenic lesions that have a locally aggressive behavior and exhibit a high recurrence rate after the treatment. The most appropriate surgical approaches for the successful treatment of OKCs remain controversial. **Aim:** The aim of this study was to evaluate the conservative management of OKCs by enucleation along with peripheral ostectomy and chemical cauterization in terms of recurrence rates after the surgical procedure. **Materials and Methods:** A retrospective study on 36 cases of OKCs treated at the Oral and Maxillofacial Surgery Department of a tertiary hospital from 2010 to 2017 was done. The demographic, clinical, radiographic, and histologic data were collected for each patient. All cases were surgically treated by enucleation followed by peripheral ostectomy and chemical cauterization using Carnoy's solution. The teeth that were involved in the lesion were extracted. The diagnosis was confirmed with excisional biopsy and histopathology reports. **Results:** Most of the OKCs were found in the mandible, except three which were present in the maxilla. A significantly higher incidence was seen in males in the age group of 21–30 years. Most of the cases (30 out of 36 cases) were accessed intraorally. Patients were followed up for up to 5 years. Recurrence of the operated OKCs was observed in five cases which were managed by enucleation with peripheral ostectomy and chemical cauterization again with good results. **Conclusion:** The results suggest that proper enucleation followed by peripheral ostectomy and chemical cauterization using Carnoy's solution may be the best and optimal approach for the management of OKC.

**Keywords:** Carnoy's solution, enucleation, odontogenic keratocyst

## INTRODUCTION

The odontogenic keratocyst (OKC) was first described by Philipsen,<sup>[1]</sup> and its characteristics were defined by Pindborg and Hansen.<sup>[2]</sup> Previously classified under developmental odontogenic cyst of the jaw by the WHO in 1971 and 1992, OKC was reclassified and renamed as 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 lining of stratified squamous epithelium and potential for aggressive and infiltrative behavior. The WHO proposed the terminology as KCOT, as it showed neoplastic nature.<sup>[3]</sup> However, in 2017, the new WHO classification of head-and-neck pathology reclassified OKC back into the cystic category.<sup>[4]</sup> OKC is so named because keratin is produced by the cystic lining. It is a parakeratin lined cyst-like lesion within

the bone. It is the one of the rare and distinctive developmental odontogenic cyst arising from the dental lamina, containing clear fluid, and a cheesy material resembling keratin debris. It occurs at all ages with a peak incidence in the 2<sup>nd</sup> and 4<sup>th</sup> decade of life. They are commonly seen in the mandible with the majority occurring in the angle of the mandible and ramus.<sup>[5]</sup> OKCs were clinically present as swelling with or without pain, discharge, displacement of teeth, and occasionally paraesthesia of the lower lip. The expansion of the cyst is very minimal in the initial stage, and it is due to the classical characteristic of

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the cyst to grow in anteroposterior direction in the medullary space of the bone. Expansion of the buccal cortex is seen in 30% of maxillary and 50% of mandibular regions.<sup>[6]</sup> 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.<sup>[7,8]</sup> Treatments modalities include enucleation with or without curettage, marsupialization, and peripheral ostectomy and chemical cauterization with Carnoy’s solution, cryotherapy, electrocautery, or resection.<sup>[9]</sup>

The aim of this study was to evaluate the conservative management of OKCs by enucleation along with peripheral ostectomy and chemical cauterization in terms of recurrence rates after the surgical procedure.

## MATERIALS AND METHODS

In this retrospective study, 36 cases treated in the Oral and Maxillofacial Surgery Department of a tertiary hospital from 2010 to 2017 were reviewed. The demographic, clinical, radiographic, and histologic data were collected for each patient. All patients reported with complaints of mobile teeth or had a radiological investigation done in the earlier dental center before reporting to this institution. Based on the clinical features, five of the patients had undergone extraction of the mobile teeth before reporting to this institution because of unhealed socket areas or discharge from the extraction site. Twenty-eight patients reported after a radiographic finding of a radiolucent lesion were seen in panoramic radiographs. Three patients underwent treatment for the pathology with persistent complaints of pain and discharge before reporting to us. In all cases, an incision biopsy was carried out, and the diagnosis of OKC was confirmed. The treatment protocol for these patients was enucleation followed by peripheral ostectomy and chemical cauterization using Carnoy’s solution through either an intraoral or extraoral approach depending on the extent and site of the lesion. Thirty out of 36 cases were accessed intraorally, including the three cases in the maxilla. Thirty-three cases involved the mandible, and three were found in the maxilla. Patients were followed up for up to 5 years. Recurrence of the operated OKCs was observed in five cases which were managed by enucleation with peripheral ostectomy and chemical cauterization again with good results.

When there was a probability of pathological fracture after the procedure, reinforcement with a reconstruction plate was given. All teeth involved within the pathology were extracted.

## RESULTS

Twenty-five of 36 patients were male and 11 were female. It was more commonly seen in the age group of 21–30 years which accounted for 27 cases. There were four cases in the age group of 11–20 years and five cases in the age group of 31–40 years.

Thirty-three out of the 36 cases were present in the mandible and three in the maxilla. In the maxilla, one involved the anterior maxilla, and two involved the posterior maxilla. Of the 33 cases in the mandible, 27 cases were seen in the posterior mandible [Figure 1], and six cases involved the anterior mandible, [Chart 1]. Fourteen out of the 36 cases were associated with an impacted tooth, 13 of them involving the mandible, ten related to the third molar, and three related to canine. Of the 33 cases in the mandible, 20 cases involved the right side and 13 involved the left side. Thirty of these cases were accessed intraorally and six extraorally. All cases accessed intraorally were through crevicular incisions followed by extraction of the involved teeth, accessing the pathology and enucleating the pathology, preserving the inferior alveolar neurovascular bundle in most cases. Chemical cauterization of the region with Carnoy’s solution was done for a maximum period of 3 min followed by vigorous irrigation with saline, and finally, peripheral ostectomy [Figure 2]. All maxillary OKCs were accessed intraorally. All cases were primarily closed without any attempt at obliterating the cavity. The six cases accessed extraorally were in the ramus region extending to the sigmoid region. The standard submandibular approach was used in these cases. In two cases, reinforcement with reconstruction plates was additionally given, both in the ramus/body region due to thinning of the inferior and posterior border after enucleation.

The most common postoperative sequela of the patients in the mandibular lesions was the temporary paraesthesia of the inferior alveolar nerve (IAN) that recovered completely in 6 months. The patients were followed up for 5 years with regular review after every 6 months. All the cases showed good reparative bone formation with filling up of the defect [Figure 3]. Five cases showed radiographic evidence of recurrence/persistence of the pathology in the form of well-defined small radiolucent areas of size 2–4 cm [Figure 4]. These were again subjected to enucleation with peripheral ostectomy and chemical cauterization. In one case, recurrence was seen within 6 months and in four cases after 2 years of surgery.

All cases of recurrence were followed up for at least a year and have not shown any signs of recurrence

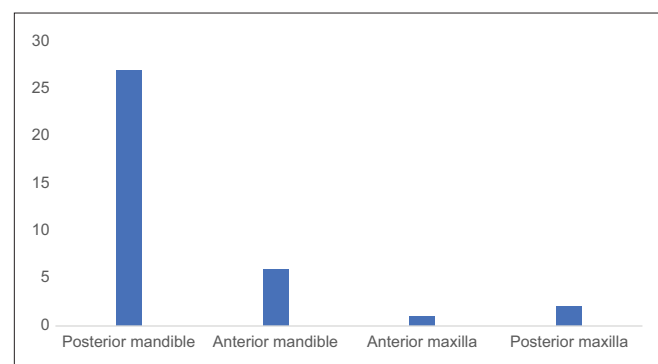
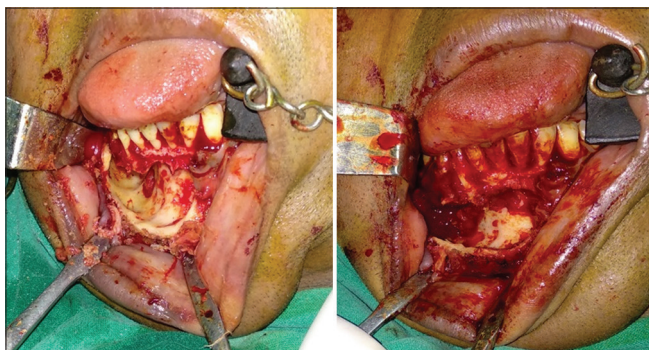


Chart 1: Sites of pathology



**Figure 1:** OKC with impacted tooth in the posterior mandible



**Figure 2:** Enucleation, peripheral osteotomy, and chemical cauterization using Carnoy's solution



**Figure 3:** Good bone formation after surgery

characterized by good bone formation in the site of the pathology [Figure 5].

## DISCUSSION

OKC is a unique lesion because of its locally aggressive behavior, high recurrence rate, and characteristic histologic appearance.<sup>[10,11]</sup>

The usual presentation of OKC is an asymptomatic lesion in the jaw associated with a tooth, discovered on a routine dental radiograph. The lesion can be multilocular or unilocular and can range from small to very large in size in the anteroposterior dimension, but it does not usually cause transverse expansion of the involved bone. The radiographic features of OKC can be the same as those of other common odontogenic cysts and tumors. Its diagnosis is most accurately made by histopathological examination, usually by incisional biopsy before definitive treatment of the lesion.

OKCs usually arise from the dental lamina and remnants of it after this organ has served its purpose.<sup>[12]</sup> This, however, does not explain the frequent appearance of the cyst in the ascending ramus of the mandible. There is plenty of evidence that most epithelial islands, as found in the wall of OKCs, are in fact located in the mucosa that is overlying the OKC and attached to it. This is the reason why it is thought that offshoots of the basal layer of the epithelium of the oral mucosa may also be involved in the etiology of OKCs. Whether they are from the original lining or derived from microcysts in the wall, they are bound to be located rather superficially in the defect. For this reason, a mild, not deeply penetrating, cauterizing agent such as Carnoy's solution is quite effective. In multilocular cysts, elimination of the bony septae will ensure complete removal of the pathology. Elimination of the epithelial islands and microcysts located in the overlying, attached mucosa should be assured by excising this part of the mucosa.

The success of this conservative treatment has been validated by the study by Stoeltinga<sup>[13]</sup> where enucleation along with application of Carnoy's solution and excision of the attached, overlying mucosa resulted in very few recurrences occurring within 5 years.

A comprehensive systematic review of published articles has established that enucleation and enucleation with adjunctive measures (other than Carnoy's solution) had recurrence rates of 25.6% and 30.3%, respectively. Marsupialization with adjunctive measures produced a recurrence rate of 15.8%, whereas enucleation with Carnoy's solution presented a recurrence rate of 7.9%.<sup>[14]</sup>

Resection generally has been reserved for patients who have undergone several surgical procedures to remove the same from recurring.

Another systematic review determined the overall and detailed recurrence rate of OKCs in relation to specific treatment methods.<sup>[15]</sup> One hundred and eight lesions found in the material were analyzed. Six treatment modalities were identified. The recurrence rates were 0% for resection, 0% for enucleation with peripheral osteotomy and Carnoy's solution, 18.18% for enucleation with peripheral osteotomy, 26.09% for enucleation alone, 40% for marsupialization, and 50% for enucleation with Carnoy's solution. The overall recurrence rate was 23.15%.<sup>[15]</sup>

One of the probable reasons given for recurrence has been the difficulty in completely eradicating the epithelial lining due to the friable nature of the thin wall. It has been shown that fragmentation of the cystic capsule during surgical excision does not affect the recurrence rate of OKCs.<sup>[16]</sup> Incomplete removal of the cystic lesion allows new cyst formation or epithelial islands in the wall of the original cyst remain in the surrounding bone or soft tissue. New OKCs can also develop from the basal layer of the oral epithelium. Recurrences were mainly found in the area associated with teeth that were not removed during the surgical treatment.<sup>[17]</sup>



**Figure 4:** Recurrence in the right body of the mandible

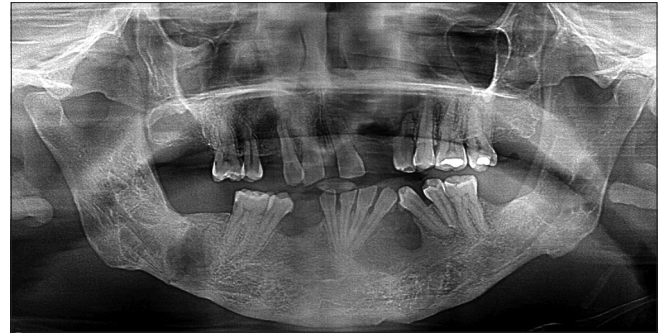
In evaluating recurrences, a study found that the median time for recurrence was 17.8 months. The 5-year disease-free estimate was 51.2%, and multiloculated lesions were 33.6 times more likely to recur than unilocular lesions.<sup>[18]</sup>

The use of chloroform containing Carnoy's solution was banned by the Food and Drug Administration in the USA. This has led surgeons in the country to adopt the use of "modified Carnoy's solution," which does not contain chloroform. However, a recent study compared the recurrence rate of OKCs treated with traditional Carnoy's solution to that of OKCs treated with the modified Carnoy's solution, and it was found that the modified formula had a significantly higher recurrence rate.<sup>[19]</sup> The additional effect of chloroform in causing necrosis of the cellular layer is probably responsible for the reduced recurrence potential as compared to the modified Carnoy's solution. This indicates that the traditional Carnoy's solution may still have a role in the treatment of OKCs as an adjunctive treatment to enucleation, reducing the recurrences.<sup>[20]</sup>

The various complications that may occur should also be considered while deciding on the treatment. Neurosensory deficit is a common complication of mandibular OKC treatment because of the proximity to the IAN. This nerve injury could be a consequence of nerve manipulation during cyst removal or of the cellular damage caused by the adjunctive treatment, such as the application of Carnoy's solution or cryotherapy.<sup>[21]</sup> Schmidt and Pogrel reported in detail the incidence of neurosensory deficits after mandibular lesion enucleation plus liquid nitrogen cryotherapy; this was found to be 100% in the early postoperative period, with 56% having a full or near full return of sensation at a mean time of 3 months.<sup>[22]</sup> In the present study, 93.3% of the patients with mandibular OKCs presented postoperative neurosensory deficits after enucleation and the application of Carnoy's solution. The majority of these patients with neurosensory deficits recovered within a short period of time.

## CONCLUSION

This retrospective study found a recurrence rate of 13.8% for OKCs treated by enucleation, peripheral ostectomy, and the application of Carnoy's solution. It is, therefore, recommended that patients treated with this method should be reviewed at



**Figure 5:** Healing of the recurrent lesion

6-month intervals for at least 5 years. Prospective studies to investigate the recurrence rate and effectiveness of different Carnoy's solution application protocols, especially in cases with a high risk of recurrence, may be worthwhile.

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## Conflicts of interest

There are no conflicts of interest.

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