

# Bilateral keratocystic odontogenic tumor: A report of two cases

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## ABSTRACT

The designation “keratocyst” was used to describe any jaw cyst in which keratin was formed to a large extent. A rare incidence of bilateral mandibular cysts (odontogenic keratocysts) was related to third molar teeth. Herein, we report two cases of bilateral keratocystic odontogenic tumor in a 22-year-old male and 15-year-old female, which was diagnosed by a series of investigations and treated appropriately.

**Key words:** Aggressive, decompression, enucleation, keratocystic odontogenic tumor, marsupialization, odontogenic keratocyst, recurrence

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## INTRODUCTION

The term “odontogenic keratocyst” was introduced by Philipsen (1956). In this and in a subsequent paper (Pindborg *et al.*, 1962), and in a paper by Pindborg and Hansen<sup>[1]</sup> (1963), the designation “keratocyst” was used to describe any jaw cyst in which keratin was formed to a large extent.<sup>[2]</sup> Finally, in the latest World Health Organization classification, the knowledge regarding the treatment of the odontogenic keratocyst, now renamed as the Keratocystic Odontogenic Tumor (KCOT) was there.<sup>[3]</sup>

The OKC is a locally aggressive, commonly recurring (10% to 50%) odontogenic cyst, most common in the second and third decades, with an anatomic predilection for the mandible in an approximate 2:1 ratio. Radiographically, OKCs usually present as unilocular, well-circumscribed radiolucent lesions with smooth, thin, opaque margins. Multilocular lesions are not as common and are usually associated with higher recurrence rates. In contrast to the current case, OKCs tend to grow in an anteroposterior

direction within the medullary cavity of the bone with minimal jaw expansion.<sup>[4]</sup> Microscopically, two types of OKCs have been described: Orthokeratinizing and a more aggressive parakeratinizing type.<sup>[5]</sup> A rare incidence of bilateral mandibular cysts (Odontogenic keratocysts) is related to third molar teeth (ramus region).<sup>[2]</sup>

During the last 3 years, two cases of bilateral odontogenic keratocyst involving the mandible were treated with a marsupialization technique that resulted in complete resolution of the lesion without the need for any other definitive cystectomy treatment, successfully without recurrence.

## CASE REPORTS

### Case 1

A 22-year-old man presented with a chief complaint of “pain in lower right back tooth region since 3 months.” History of present illness revealed that the swelling was initially small and gradually increased in size. The swelling was also associated with dull, continuous pain that would subside on taking medications.

Extra oral examination revealed no evidence of facial swelling, but there was point tenderness in the region over the mandibular ramus region. On palpation, the swelling was firm and tender. Intraorally, there was tenderness in the buccal vestibule over the first molar,

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which was sensitive to percussion and had a vestibular obliteration. There was no obvious expansion of the buccal or lingual cortices in the involved area.

A panoramic radiograph showed a 3-4 cm anteroposteriorly, well-circumscribed, multiloculated radiolucency in the right body to ramus of the mandible. On the left side, 1-2 cm unilocular radiolucency distal to 38 which was round in shape was noted [Figure 1a].

**Case 2**

A 15-year-old female presented with complaint of a swelling extending gradually from the left mandibular canine to the mandibular ramus region. The swelling was slightly painful; it was first noticed about 5 months previously.

External examination revealed no evidence of facial swelling, but there was point tenderness in the region over the mandibular ramus region. On palpation, the swelling was firm and tender. Intraorally, revealed a buccal spherical and fluctuant swelling involving the left lateral incisor, premolars and first molar region with scalloped margins, a vertically impacted left second molar displaced inferiorly. There was no obvious expansion of the buccal or lingual cortices in the involved area.

A panoramic radiograph showed a well-circumscribed, multiloculated radiolucency in the left canine to ramus of the mandible. On the right side, 1-2 cm unilocular radiolucency distal to 47 which was round in shape was noted [Figure 2a].

**Treatment**

Under strict aseptic conditions, local anesthesia was obtained with bilateral inferior alveolar, lingual, and long buccal nerve blocks using 2% lidocaine containing 1:100,000 epinephrine. Treatment consisted of marsupialisation by excision of the overlying mucosa and opening of an appropriate-sized window (never less than 1 cm in diameter) into the cystic cavity and, where there was possible, suturing of the cyst lining to the oral mucosa. Once the cyst had largely filled in, histologic material was taken from the mucosa at the base of the residual shallow depression. Histology were performed to evaluate the cystic lining. The cavity was thoroughly irrigated with povidone-iodine solution mixed with saline. Then packed with iodoform gauze. The packing was replaced during the recall visits biweekly for 9 months following the initial surgery.

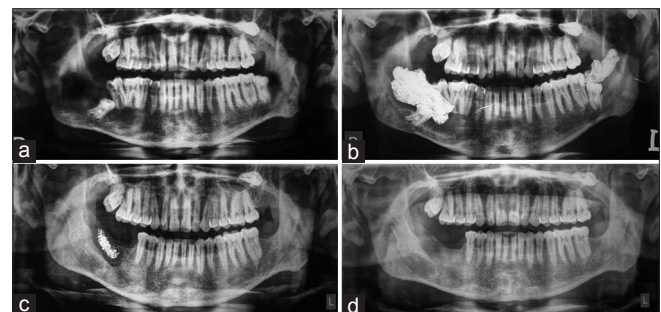
In case 1, after 9 months when the bone healing was appropriate than the impacted 38, 48 was removed along with the 46 and 47; and in case 2, after 12 months when the bone healing was appropriate than the impacted 37 was removed under local anesthesia.

All the cases radiographically show a complete resolution of the lesion [Figures 1b-d and 2b-d]. At the end of 2 years of follow-up period, no evidence of recurrence was noticed and radiograph showed complete bone healing.

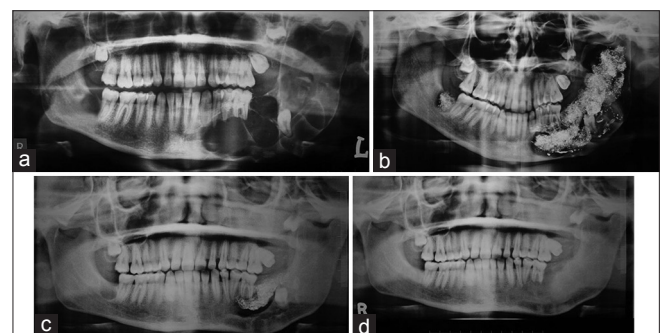
The histopathology of the cystic lining showed keratin floating in lumen immediately beneath a thin layer of parakeratin, which lined a squamous epithelial layer. Based on the clinical examination and investigations a final diagnosis of bilateral OKC was made [Figure 3].

**DISCUSSION**

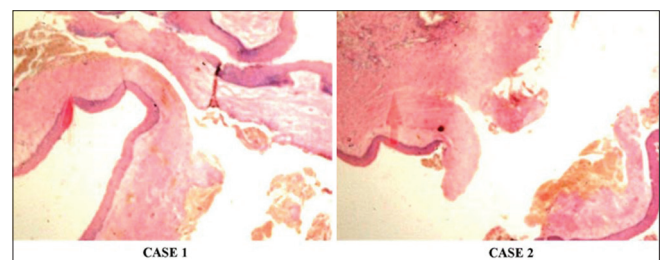
Over a period of 100 years, we have arrived at the conclusion that keratinization can occur in the lining of many different cysts but there is specific type in which the keratin is predominantly of the parakeratinizing variety.<sup>[5]</sup> In comparison to other cysts of the jaws,



**Figure 1:** (a) Preoperative radiograph, (b) postoperative radiograph (3 months), (c) postoperative radiograph (12 months), (d) postoperative radiograph (24 months)



**Figure 2:** (a) Preoperative radiograph, (b) postoperative radiograph (3 months), (c) postoperative radiograph (12 months), (d) postoperative radiograph (24 months)



**Figure 3:** Histopathological view

OKC is unusual because it shows characteristic clinical features, including potentially aggressive behavior, high recurrence rate, and an association with nevoid basal cell carcinoma syndrome. Various treatment options are available.<sup>[6]</sup> Enucleation alone is associated with the highest recurrence rates (range, 17% to 56%), especially when the cyst is removed in fragmented fashion.<sup>[7]</sup> Various adjunctive therapies have been advocated to decrease recurrence potential; these include peripheral ostectomy or treatment of the surgical site with Carnoy's solution, electrocautery, or cryotherapy.<sup>[8-11]</sup> Marsupialization has also been reported but is associated with recurrences in the range of 25% to 100%.<sup>[12,13]</sup> Large or recurrent lesions in difficult anatomic locations have been treated by en bloc or marginal resection.<sup>[14]</sup>

Decompression or marsupialisation has been recommended in a number of studies as a technique to allow partial resolution and decrease in size in the keratocyst so that teeth or the inferior alveolar nerve may be spared.<sup>[15-18]</sup> All of these reports, however, used decompression or marsupialization to decrease the size of the cyst, after which it was definitively enucleated. There is relatively little in the literature except for the occasional isolated case report in which marsupialization has been carried out for complete resolution of a keratocyst.<sup>[19-21]</sup> The technique has been described for complete resolution of other types of benign dental cysts<sup>[22]</sup> and was originally described by Partsch in the late 1800s,<sup>[23,24]</sup> as a definitive treatment for cysts at a time when antibiotics were not available and enucleation and primary closure most often led to wound breakdown and infection. They are still described as Partsch I procedure. This treatment was put forward at that time as definitive treatment for the cysts, and it consisted of the removal of the overlying epithelium and the deroofing of the cyst, keeping it open with a drain of some kind. This management is often advocated to stop subsequent growth and cause some decrease in size of the cyst to take it away from vital structures such as inferior alveolar nerve or adjacent teeth in the mandible or maxillary sinus or nasal cavity in the maxilla. It also allows subsequent enucleation to be performed without complication and complete bone fill can be expected. Marsupialization, on the other hand involves converting the cyst into a pouch. This implies a more definitive treatment for the oral cavity and the exposure of the cyst lining to the oral environment.

This type of treatment requires commitment and compliance on the part of patients over an extended period of time. Regular recall visits are required to ensure cyst involution and opportunity for appropriate treatment should there be any evidence of recurrence.<sup>[6]</sup>

This treatment may require at least 9 months. However, chances of recurrence and surgical morbidity are less

with this procedure. OKCs respond to one of the most noninvasive treatments of all which reinforces the paradox that accompanies new evidence that they may represent true cystic tumors and they even respond to simple treatment.<sup>[25]</sup>

This report describes the marsupialization technique used that resulted in complete resolution of the lesion without the need for any other definitive cystectomy treatment.

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