



## **Case Report**

# **A multi-disciplinary approach in management of undetected odontogenic keratocyst with three years follow up - a case report**

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**How to cite:** *Prachi M, Punit C, Ashwini B P, Deepak R, Amit, Ridhima G. A Multi-disciplinary Approach in Management of Undetected Odontogenic Keratocyst with Three Years Follow Up - A Case Report. Int J Endodontic Rehabil Volume 2022, Article ID 22041806, 9 pages.*

*Received:10.03.22*

*Accepted:25.03.22*

*Web Published:18.04.22*

### **ABSTRACT**

The keratocystic odontogenic tumor (KCOT) is regarded as the most aggressive and recurrent tumor of all. The lesion does not pose any clinical symptoms until significantly large in size. The different treatment modalities are always a matter of dilemma as to which one would yield good results with less recurrence. A Conservative treatment approach for moderately large lesions such as marsupialization and decompression were carried out in this case with regular follow-up sessions. The aim of this case report is to treat cases of KCOT with a conservative surgical approach.

**Keywords:** *Conservative surgical approach, Marsupialization, De-compression, Iodoform dressing*

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## INTRODUCTION

The dental lamina or the relics of the enamel organ are the sources of the uncommon developing cyst known as the odontogenic keratocyst (OKC).<sup>1</sup> The “odontogenic keratocyst” was defined by Philipsen in 1956, during his study of seven jaw cysts from cholesteatomas occurring in other cranial areas.<sup>2,3</sup> His research determined that these were non-inflammatory odontogenic cysts, and he created the term "odontogenic keratocyst" to describe them.<sup>4</sup> After that, it was discovered that many additional odontogenic cysts, including radicular cysts, follicular cysts, and lateral periodontal cysts, resemble OKCs morphologically.<sup>5</sup> Some observers claimed that the odontogenic keratocyst behaved more like a neoplasm than like a cyst.<sup>6</sup> The prior odontogenic keratocyst was categorised as a benign odontogenic tumour in the most recent World Health Organization classification. "Keratocystic odontogenic tumour" is the new term (KCOT).<sup>7</sup>

The recurrence rate of KCOTs is high, ranging from 13% to 80%.KCOTs may also undergo malignant transformation.<sup>8,9</sup> Recurrence of KCOTs occurs within the first 5 years after treatment.<sup>10,11</sup> Operative techniques are being improvised so as to reduce the high recurrence rate of KCOTs.<sup>12,13</sup> Many proponents of conservative medicine claim that marsupialization produces outcomes that are on par with those of more invasive procedures.<sup>14,15,16</sup>

## CASE REPORT

A 25-year-old young patient reported to the department of conservative dentistry and endodontics with the chief complaint of pain and swelling on the right side of the face [Fig.1]. The patient’s medical and dental history was non-contributory to the lesion.

**Figure 1:** Pre-op photograph showing swelling in right mandible area



Intraoral examination revealed no evidence of caries, exposure of pulp chamber to the oral cavity, or prior endodontic treatment. Cold tests and Electric pulp tests were performed for teeth involved in lesions. A negative response for percussion as well as pulp sensitivity tests was obtained. Following the above findings, a provisional diagnosis of aseptic pulp necrosis was made. The patient was advised to get a full mouth x-ray done(OPG). On the radiograph, a large irregular radiolucency was found extending from the lower right first molar to the lower left canine involving the lower border of the mandible with no pathologic fractures. The impacted canine was also present [Fig.2].

**Figure 2: Pre-op OPG showing large lesion in right side associated with impacted canine.**



The patient was referred to the department of oral and maxillofacial surgery for incisional biopsy as the multidisciplinary intervention was indicated for the case. Histopathologic report revealed presence of corrugated parakeratinized epithelium which is 6-10 cell thick with basal layer of cell depicting typical tombstone appearance pathognomonic of OKC. The report confirmed the presence of odontogenic keratocyst. A two-phase treatment protocol of Endodontic treatment followed by marsupialization with decompression was planned.

**First phase:** Endodontic treatment in the present case, OKC resulted in aseptic pulpal necrosis and therefore a single session endodontic treatment of all the teeth from lower right second molar (47) to left lower first premolar (34) prior to marsupialization [Fig.3] was performed. This was done so as to overall reduce the incidence of contamination in between treatment sessions.

**Figure 3: Endodontic treatment performed prior to surgery in involved mandibular teeth**



**Figure 4: Vestibular incision given**



A single visit of root canal treatment was carried out from 47 to 34 under local anesthesia and rubber dam isolation. Access opening was done with Endodontic access bur no. 2 (Dentsply Maillefer, Switzerland) and Working length was determined with 10K file (Dentsply Maillefer, Switzerland) using Root ZX II Apex locator followed by radiographic confirmation. Following which biomechanical preparation was done by 25-4% in 45,46 and by 20-06% in 44 to 34 with Hyflex CM rotary files (Coltène/Whaledent, Mumbai, India) using 15%

EDTA (Glyde, Dentsply Maillefer, Ballaigues, Switzerland) with thorough irrigation by 5.25% sodium hypochlorite (Medilise Chemicals, Kannur, India ) and copious saline. Canals were rinsed finally with Chlorhexidine (Dentachlor 2% Ammdent, Mohali, India) and were dried using sterilized paper points (Dentsply Maillefer, Ballaigues, Switzerland). Master cone fit was checked, and root canals were obturated with respective gutta-percha (Coltene/Whaledent, Mumbai, India) along with AH Plus resin-based root canal sealer (Dentsply Maillefer, Ballaigues, Switzerland). Composite (Filtek Z350 XT, 3M ESPE, Germany) was used as post root canal filling material so as to ensure coronal seal.

**Second phase:** Under General anesthesia vestibular incision [Fig.4] was given and a full thickness mucoperiosteal flap was raised, cystic content was removed partially [Fig.5] preserving the inferior alveolar neurovascular bundle. The impacted canine was also extracted. The remaining lining was sutured to oral mucosa and the defect was packed with iodoform dressing [Fig.6]. The initial dressing was changed for a month with an interval of 7 days for a month and later it was continued every fifteen days for a year.

**Figure 5: Full thickness flap and exposed lesion site**



**Figure 6: Iodoform dressing given**



Radiographic investigations were performed at one month [Fig.7], six months [Fig.8], twelve months [Fig.9], Eighteen months [Fig.10] and Three years [Fig.11] postoperatively. Postoperatively lesion was totally resolved with an intact lower border with good healing and bone formation in the defect.

**Figure 7: One month OPG showing iodoform dressing**



**Figure 8: Six months OPG showing healing of lower border of mandible and reduction in size of lesion**



**Figure 9: Twelve months OPG showing further reduction in size with good healing**



**Figure 10: Eighteen months OPG showing further reduction in size with good healing**



**Figure 11: Three years follow up shows good healing and new bone formation**



The patient is under further follow-up.

## DISCUSSION

Keratocystic Odontogenic Tumor can occur in any age group, but the prevalence is more in the second and third decades of life with a predilection for males. Frequently it occurs in the mandible than maxilla and that too in Ramus –third molar area.<sup>17</sup> There are no characteristic clinical manifestations of the keratocyst but commonly pain, soft tissue swelling, expansion of bone, and in the later period of time paresthesia of lip and teeth are seen. The radiographic appearance includes most commonly unilocular, well-defined peripheral rim. Multilocular radiolucency with satellite cysts is also observed but is infrequent and limited to large lesions. Additionally visible are the displacement of adjacent teeth and root resorption.

The position and extent of KCOT can be determined using conventional radiographic imaging techniques such as panoramic views (OPG) and intraoral periapical films. Large lesions involving the maxillary sinus and in the rare cases that extend to the skull base can benefit greatly from the use of advanced imaging techniques like computed tomography and magnetic resonance imaging.<sup>18</sup> Studies indicate that most of the KCOTs show clonal loss of heterozygosity of common tumor suppressor genes.<sup>19</sup> These studies support its origin as a neoplasm rather than developmental.<sup>20</sup> A tumour suppressor gene in the Sonic hedgehog pathway, PTCH1 is the human equivalent of the *Drosophila* segment polarity gene. Patients with nevoid basal cell carcinoma syndrome (NBCCS) as well as spontaneous neoplasms such as KCOT, basal cell carcinoma, and medulloblastoma

have displayed PTCH1 gene mutations.<sup>21</sup>

In accordance with studies by Grachtchouk et al. on transgenic mice, it is the epithelial expression of the Hh transcriptional effector Gli2 responsible for highly penetrant keratocyst.<sup>22</sup> But still, it is a controversial matter whether they are developmental or neoplastic in origin with recent studies supporting the latter more. The definitive and confirmatory diagnosis is made only after the BIOPSY of the lesion showing typical 6-10 cells thick lining epithelium giving "picket fence" appearance which is 83% of times parakeratinized. The lumen of the keratocyst is filled with a thin straw-colored fluid or thick creamy material.<sup>23</sup> KCOT is known for its high recurrence rate and aggressive nature invading adjacent tissues and bone.

So the treatment planning is aimed for complete and thorough removal of the lining epithelium. The treatment should be planned according to the size of the lesion and its vicinity to the neurovascular bundles, sinus, and lower border of the mandible. However, it was observed that lesions enucleated in one piece recurred significantly less as compared to several piece enucleation. The multilocular variant has a higher recurrence rate than the unilocular one due to the multiple satellite cysts in the former. The possible treatment which can be done in cases of KCOT is:

1. Marsupialization
2. Enucleation with open packing
3. Enucleation with closed packing
4. Resection

In the case presented in the department the more conservative treatment, i.e., marsupialization with decompression, was performed. The cyst cavity was entered, and the cystic fluid was drained. After the fluid drainage, the inner cyst lining was sutured to the outer wall or everted on the outer surface followed by thorough irrigation with a mixture of saline and povidone iodine solution. An iodoform dressing was given and patient was kept on regular follow ups for the evaluation of healing and change of dressings.

Experiments demonstrate that complete removal of large KCOTs of the mandibular ramus might be difficult.<sup>24,25</sup> For large-sized KCOTs and recurrent lesions, radical surgery that involves excision with or without continuity defects has been preferred. When excision is necessary for the treatment of KCOTs, the lesion is largely eliminated with little recurrence. Resection or enucleation combined with Carnoy's solution and peripheral ostectomy, as opposed to enucleation alone or marsupialization, leads in a lower recurrence rate than less invasive treatments. It is noteworthy that the recurrence rate following marsupialization and enucleation is not noticeably greater than that following the so-called more severe treatment techniques.<sup>26</sup>

Even though resection has a high cure rate, surgery comes with severe morbidity such loss of jaw continuity or facial deformity. Therefore, it should only be used for lesions that are aggressive or recurrent, or for individuals who cannot be closely monitored after receiving conservative therapy.<sup>27</sup> In cases of KCOTs, total enucleation, with or without a "peripheral ostectomy," is currently the most usual surgical technique.<sup>2,28</sup> Enucleation of the KCOTs followed by open packing is one example of a conservative surgical strategy that has been proposed.<sup>16,29</sup>

To reduce the risk of recurrence during each follow-up appointment, the cavity can be packed with iodoform gauze impregnated with bacitracin ointment and irrigated with a glass full of a solution of normal saline and chlorhexidine gluconate. Regular follow-up visits are necessary to verify cyst healing, and the chance to receive the proper therapy should be proof of recurrence. It is crucial to take annual radiographs for at least five years after surgery because the recurrence may be long delayed. Additionally, it is crucial to medically rule out the possibility of NEVOID BASAL CELL CARCINOMA SYNDROME in cases with numerous cysts.

Periapical lesions(Abscess, Granuloma, Cyst) are mainly sequelae to pulp necrosis triggered by infection. But there is certain periapical pathology like OKC that simulate the above-mentioned periapical lesions making overall diagnosis and treatment a challenge. Therefore, it is important for endodontists and other general practitioners to be aware and understand the importance of a multidisciplinary approach for such cases.

In the case presented above following intraoral examination and histopathology confirmation of OKC, endodontic treatment was prioritized before surgical approach and single visit root canal treatment was performed for non-vital teeth i.e., 46 to 34 for the following reasons:

1. To reduce inter-appointment contamination
2. To minimize overall treatment time
3. To decrease the chances of retrograde contamination following surgery thereby reducing the recurrence rate
4. To remove necrotic tissue and shape dentinal walls

The overall aim of endodontic treatment in our case was to remove necrotic tissue and eliminate any possibility of recurrence that might be possible with retrograde contamination.<sup>30,31</sup>

## CONCLUSION

With the limited evidence available in this review, it can be concluded that,

- Management of Odontogenic keratocyst require multidisciplinary approach to yield effective results.
- Endodontic treatment accompanied by marsupialization with decompression has significantly contributed towards positive outcome for the case as there was bone neof ormation with no sign of recurrence in 3 years follow up.

**Financial support and sponsorship** – Nil

**Conflicts of interest** - There are no conflicts of interest.

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