



## **Techniques for Surgical Removal of an OKC or Keratocystic Odontogenic Tumor: A Dry Run**

Dr Rachana PB, <sup>1</sup>MDS; Dr Sumana Devadiga, <sup>2</sup>MDS; Dr Manoj Kumar Bhaskaran, <sup>3</sup>MDS; Dr Joseph Lijo, <sup>4</sup>MDS; Dr Arjun Shenoy, <sup>5</sup>MDS

**Keywords:** Odontogenic keratocyst; OKC; Keratocystic Odontogenic tumor; enucleation; marsupialisation; Carnoy's solution; cryotherapy; liquid nitrogen.

Assistant Professor, Department of Oral and Maxillofacial Surgery, KVG Sullia Dental College, Sullia, Karnataka, INDIA

Associate Professor, Department of Oral and Maxillofacial Surgery, KVG Sullia Dental College, Sullia, Karnataka, INDIA

Professor, Department of Oral and Maxillofacial Surgery, Sri Anjaneya Institute of Dental Sciences, Calicut, Kerala, INDIA

Associate Professor, Department of Oral and Maxillofacial Surgery, Malabar Dental College, Edappal, Kerala, INDIA

Consultant, Oral and Maxillofacial Surgery, Elite Mission Hospital, Thrissur, Kerala, INDIA

**How to cite this article:** Rachana PB, Sumana Devadiga, Manoj Kumar Bhaskaran, Joseph Lijo and Arjun Shenoy. Techniques for Surgical Removal of an OKC or Keratocystic Odontogenic Tumor: A Dry Run. Journal of Oral Medicine Surgery Pathology Biology. Dec- Jul 2016; 1(2): 1-8

### **Address for Correspondence:**

Dr Rachana PB, Assistant Professor, KVG Sullia Dental College, Sullia, Karnataka, INDIA

Email id: rachanadeepthi@gmail.com

**Running title:** Surgery for odontogenic cysts

**Clinical significance: Awareness about the mode of surgery commonly followed for Odontogenic cysts is helpful to Oral and Maxillofacial Surgeons**

## **ABSTRACT**

The OKC (odontogenic keratocyst)/ Keratocystic Odontogenic tumor (OKC/ KCOT) has been the subject of much debate over the last 50 years in relation to its origin, growth, and treatment methodologies. The treatment of OKC is controversial and no treatment till date has been considered as standard, as all the modalities of treatment practised, lead to recurrence. Recurrence of the cyst occurred either due to the incomplete removal of the original cyst's lining, or from small satellite cysts or odontogenic epithelial rests left behind after the surgical treatment, or they may have developed as a relatively primary OKC in another region of the jaw which was misinterpreted as a recurrence.

Treatment of Keratocystic Odontogenic tumor was either conservative or aggressive. Conservative treatment included simple enucleation, with or without curettage, and marsupialization. Aggressive treatment on the other hand included peripheral osteotomy, chemical curettage with Carnoy's solution and resection.

**Aims and Objective of the study:** The study aimed to understand the treatment protocols for the KCOT throughout the world. The objective was to trace the treatment modalities of OKC from the time it was called as a "primordial cyst" by Philipsen, through the time it went on to be called as an "odontogenic keratocyst" by Shear, to 2005, when it had been first introduced in the WHO Classification of Odontogenic tumors by a new terminology the 'Keratocystic Odontogenic Tumor'.

**Materials and Methods:** Online electronic databases were searched for articles published in English language literature pertaining to the treatment methods of OKC, accomplished by the surgeons throughout the world, and whether there have been any changes in treatment methodology that has been reported, since the "OKC" became "KCOT." Articles were appraised by two independent reviewers and the data was finally entered in the database which was further analysed.

**Results:** The treatment method for KCOT is still ambiguous and all methods have their own advantages and drawbacks. Therefore, the protocol should be based on the size, site, lesion accessibility and tissue destruction while planning the treatment.

**Conclusion:** The majority of cases of recurrence commonly occur within the first 5 years after treatment. Because of the problematic nature of these cysts, many attempts have been made to minimize the high recurrence rate by attempting to improve the surgical techniques. The goal of the treatment therefore should aim at eliminating the potential for recurrence and minimizing the morbidity. There has been no consensus on the appropriate treatment for this lesion.

## **INTRODUCTION**

Odontogenic keratocyst/ OKC, is a developmental, odontogenic cyst first described by Philipsen in 1956. It may arise as a solitary cyst or in association with the Nevoid Basal cell carcinoma syndrome. Histologically, the hematoxylin and eosin stained section of KCOT shows a thin, friable wall, which is

often not easy to enucleate from the bony cavity as one mass, as they comprise of small satellite or daughter cysts within its fibrous wall. <sup>1</sup>The 2005 WHO classification has included the OKC into odontogenic tumors and is now known as the keratocystic odontogenic tumour/ KCOT. The WHO has defined KCOT as “a benign uni- or multi-cystic, intraosseous tumour of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium with a potential for aggressive, infiltrative behaviour.”

The KCOT is one of the most aggressive odontogenic cysts. It enlarges in size and extends into adjacent tissues; it presents with a rapid growth and tendency for significant expansion in an anteroposterior direction.

The techniques used to treat OKC are multiple, as the recurrence of the cyst is still an issue, whatever be the treatment method practised.

Surgical treatment of OKC may be

- Conservative
- Aggressive



## Conservative

### Decompression and Marsupialisation

Decompression is any technique used, wherein the pressure is eased. The decompression of a cyst advocates the release of its contents by placing a drain through a small opening created in the cyst wall. This releases the pressure within the cyst and relaxes the pressure effect of the cyst on adjacent structures like the tooth root and nerves. Partsch in the late 19th century was the first to introduce the technique of decompression and hence is still called as the Partsch I procedure in many parts of the world. The enucleation and primary closure is the Partsch II procedure.<sup>2</sup>

Marsupialization is the exposure of the cystic lining to the oral environment by transforming it into a pouch in order to decompress the cyst. Pogrel first described the technique in 2005. The technique involves the suturing of the cyst lining to the oral mucosa. This treatment is more definitive than decompression. Maxillary cysts are exposed to the maxillary sinus or the nasal cavity and the mandibular cysts into the oral cavity. The cyst cavity is then filled with a packing material such as iodoform gauze impregnated with bacitracin ointment.

The authors in favor of these treatments argued that the enucleation resulted in the tear of the friable epithelial lining whereas the decompression and marsupialisation caused the lining to thicken and therefore resemble an oral epithelium; facilitating its complete removal.

This procedure suffers drawbacks such as:

- ✓ The contamination of the maxillary marsupialisation by food and other particles and

- ✓ In some cases, closure of fistula. Closure of fistula is more probable in the mandibular marsupialisation. This was overcome by use of a nasogastric tube acting as drainage. The cavity also required regular irrigation to flush out the food and other particles.

This procedure was said to retain the cyst wall, but the authors believed that, this was the main reason for the high rate of cyst recurrence.<sup>3</sup>

### **Enucleation without any adjunctive procedure**

Enucleation is “to remove whole or clean, a tumor from its envelope” and curettage is defined as “the removal of growth or other material from the wall of a cavity.” Removal of the cyst lining by curetting it out from the bony cavity in total, without the help of any other add-on is found to have a very poor success rate. Hence, this procedure is hardly recommended.

Curettage together with mechanical techniques foreg. Hand, rotary burs alone or together with a chemical solution such as Carnoy’s or cryosurgical agents (liquid nitrogen) have been suggested<sup>3</sup>

### **Enucleation and usage of the Carnoy’s fixative.**

John and James first suggested the technique of enucleation with Carnoy’s fixative as the treatment modality for OKCs, in order to reduce recurrences. Tanning, with Carnoy’s solution<sup>4</sup> (3 milliliter of chloroform + 6 milliliter of absolute ethanol + 1 milliliter of glacial acetic acid and 1 gram of ferric chloride) inside the bony cavity after enucleation or before enucleation<sup>4</sup> is said to cauterise the vital daughter cysts within the fibrous wall trapped within the jaw bone, which is said to be one of the primary reasons for the cyst recurrence.

The Carnoy’s solution taken on small gauze and picked up with a Kocher clamp, is applied generously on those areas where cyst is in contact with the hard tissues. Repetitive application is done to ensure that the entire bony wall is treated. Care is taken not to injure the lingual nerve. In cases where the inferior alveolar nerve is exposed in the bony defect, lifting it out of its canal before the Carnoy’s solution is applied is advisable. The bone, that becomes blackish after this treatment, needs to be washed off with saline before placing a pack to fill the defect. This pack, soaked in Whitehead’s varnish or impregnated with iodine-vaseline or any other ointment, is used for this purpose. The pack is replaced every week with a new pack. This procedure is repeated until the defect is completely epithelised, which usually takes about 3 weeks, based on the size of the defect.

In cases where the cyst has penetrated through the soft tissue covering of the buccal or the lingual plate, electro cauterisation is recommended.<sup>3</sup>

Abrading the cyst walls using coarse carbide or acrylic; surgical burs, is said to facilitate the removal of any daughter cysts present in the fibrous wall embedded within the bone, thereby minimizing the incidence of recurrence arising from a fragile cyst wall.<sup>4</sup>

Recurrence even after placement of Carnoy’s solution was corrected by completely removing the epithelial lining overlying the cyst.<sup>3</sup> The basal cells of the overlying epithelium are stated to be another cause for recurrence.<sup>1</sup> Epithelium is always present at the anterior portion of the ascending ramus and at the top of the tuberosity in case of a maxillary cyst. The presence of perforation can be identified by using a fine needle and the buccal and lingual walls exposed by stripping the mucoperiosteum.<sup>5</sup>

### **Enucleation and liquid nitrogen therapy**

Cryotherapy or liquid nitrogen therapy or cold therapy after enucleation, is said to devitalize bone in-situ<sup>3</sup> (organic tissue)<sup>2</sup> and enable the bony framework<sup>3</sup> (inorganic tissue)<sup>2</sup> to remain intact, facilitating osteoinduction and destruction of daughter cysts or cystic remnants. It has been reported that only the liquid nitrogen cryospray, that works at a temperature of about 20°C, through a radius of 1-2 millimetre around the lesion, would be able to remove the cystic remnants on a continuous basis. A thaw/ freeze technique is followed with a 1-minute freeze followed by a slow thaw, for each cycle. Protection, to both the soft tissues (9% tissue breakdown), protected by the wooden tongue blades and gauze, and the hard tissue [mandible (9% fracture rate) from pathological fractures] needs to be provided. This method is already in use to manage locally aggressive lesions like the ameloblastoma, and fibroosseous lesions.<sup>3</sup> Schmidt and Pogrel were the first to suggest this procedure in 2001. Bone graft is then inserted within the defect and the mucosa is closed with watertight sutures.

The following advantages are noted:

- ✓ the bone matrix that is left behind, acts as a clean scaffold for osteogenesis,
- ✓ a bone graft can be immediately inserted which would hasten healing and reduce the risk of pathologic fractures; and also simultaneously reduce bleeding and scarring.

The main disadvantage of this technique is that

- ✓ Liquid nitrogen weakens the bone with resultant fractures for which Pogrel recommended an immediate bone grafting to prevent this calamity.<sup>5</sup>
- ✓ Inconsistency in controlling the amount of liquid nitrogen applied, could result in necrosis and swelling,<sup>3</sup>
- ✓ Liquid nitrogen cryotherapy, when given around the inferior alveolar nerve may result in paraesthesia or anaesthesia, which would partially or completely return to normal within 3 months, following its regeneration from the intact axon sheath.<sup>3</sup>

The recurrence rate following enucleation and liquid nitrogen cryotherapy is said to be very low (9%).<sup>2</sup>

## **Aggressive treatment**

### **Block resection with or without continuity defect**

Resection may be either a

- The radical approach where the excision of a lesion includes a major area of investing bone. In the mandible this would be (segmental) or without (marginal) continuity defect or an en bloc osseous resection and
- Extend into a disarticulation where there is a temporomandibular joint involvement.
- In the case of the maxillary involvement it would be determined by the extension of the excision anatomically both in subtotal/ partial and total maxillectomy.<sup>6</sup>
- A more conservative surgical approach are the types of "peripheral" osseous management such as curettage or ostectomy using a curette or bur, where the lesion is removed together with a margin of surrounding bone. Peripheral ostectomy is primarily used as an adjunctive for

osseous removal when resections can be avoided. The drawback of this procedure is the inability to gauge the amount of bone to be resected. This can be overcome by topical application of Methylene blue, which penetrates bone and ensures proper clearance.<sup>7</sup>

Nevertheless, whichever aggressive technique is practised, there are chances of morbidity, especially as reconstructive measures are required to restore aesthetics as well as the jaw functions.<sup>3</sup>

Authors have argued in favour of this technique in terms of least number of recurrences, but with a risk of high morbidity. Other authors are not in favour of such an aggressive therapy for a benign lesion that can be treated reasonably well by using relatively simple means. The enucleation with application of Carnoy's solution results in a recurrence rate more or less like that of resection but without the unnecessary aggressive surgery.<sup>3</sup>

Techniques suggested by authors include a permutation and combination of the above given techniques such as

- Simple surgical tumorenuclation and marsupialization,
- Decompression by marsupialization followed by enucleation,
- Enucleation associated with mucosal excision,
- Peripheral osteotomy, and
- Chemical curettage, using solutions, containing ferric chloride ( $\text{FeCl}_3$ ) and ethanol (Carnoy's solution).
- ✓ Large KCOTs are often managed by marsupialization that allows new bone to fill in the lesional defect with minimal morbidity.<sup>8</sup>
- ✓ Small keratocysts in the dentate areas are mostly enucleated.
- ✓ Carnoy's solution for cysts in the maxilla have been advised to be used very selectively, since the delicate bony walls around the sinus may become necrotic. Application in the alveolar process is without such problems.<sup>5</sup>

Enucleation is a commonly used method for surgical treatment of KCOT in Europe and Asia. Blanas et al reviewed the literatures pertaining to the treatment and prognosis of KCOTs and found that simple enucleation had a recurrence rate of 17–56%, and enucleation combined with adjunctive therapy, such as the application of Carnoy's solution or decompression before enucleation, had a low recurrence rate (1–8.7%).

#### **Points to be noted:**

- KCOTs in the mandibular molar region, angle or ascending ramus had a higher recurrence rate. The logical explanation is that there is more chance for the epithelial rests of the cyst wall to be left behind after surgery, owing to the limited surgical access to the posterior part of the mandible.
- Patients with nevoid basal cell carcinoma syndrome, which had a higher occurrence of epithelial islands and/or microcysts in the cyst wall therefore, had a higher recurrence rate than the non-syndromic KCOT.



- In relation to the teeth associated with the cyst there is still no consensus with regard to its extraction. Chirpathomsakut et al noticed that the teeth being retained could be a probable reason for the recurrence. Hence, they were of the opinion to extract the teeth if enucleation is chosen as a surgical treatment, if they were unsure of assuring a complete pathological tissue removal. Tan et al reported that the majority of teeth involved, could be preserved when marsupialization together with secondary enucleation was the treatment of choice especially in case of the mandibular KCOTs, thus helping in retaining the masticatory function of the patient. This two-stage surgery was also found to minimize the damage to adjacent structures thereby reducing the recurrence rate especially when used for large or extensive KCOTs. Hence, in cases where multiple teeth are involved it seems appropriate to perform marsupialization or decompression at the first stage followed by enucleation. <sup>6</sup> Meiselman *et al.* was of the opinion to follow a conservative approach of 'enucleation, curettage, and marsupialization' and Williams *et al.* on the other hand suggested an aggressive treatment in addition to enucleation that included curettage (chemical, mechanical or physical) and/or resection either with or without loss of jaw continuity. <sup>5</sup>
- In cases when the tooth roots extend into the cyst cavity causing the incomplete removal of the epithelium around the tooth, it is recommended to either remove the affected tooth or treat it with apicectomy.
- Resection is advisable for the more aggressive cysts that have perforated the buccal and lingual plates.
- Endoscopy has been used by surgeons to accurately explore the operative field; and areas difficult to access. <sup>9</sup>
- Marsupialisation and decompression causes the cyst to become smaller; and Brondum and Jensen suggested a re-epithelialisation of the cyst lining by metaplasia, which resulted in excellent prognosis without any recurrence. <sup>5</sup>

### **Dredging Method**

This is a newly introduced conservative, surgical procedure wherein the lesion is deflated and enucleated or only enucleation is performed followed by repeated dredging: a procedure whereby new bone formation is accelerated by removing out the scar tissue from the bony cavity. This form of treatment has found reduced recurrences in case of the ameloblastoma and recurrent KCOTs; however, long-term follow-ups are necessary to confirm its success rate. <sup>10</sup>

Finally, a regular follow-up is recommended, on a yearly basis in the first 5 years for whichever technique that is implemented, followed by at least 15 years in which the patient is seen every 2 years. <sup>5</sup>

### **CONCLUSION**

Treatment of the keratocystic odontogenic tumor (KCOT) is one of the most controversial entities of the maxillofacial region. The complete removal of the OKC is difficult because of factors such as the thin, friable epithelial lining, limited surgical access, skill and expertise of the surgeon, cortical perforation, and the desire to preserve adjacent vital structures. The treatment modality should be based on the size

of the lesion, recurrence rate, site involved, and the morbidity. Despite the varied treatment options available, both clinical and radiographic follow-up is essential for years after surgery since there are chances of recurrence many years later.

## Footnotes

**Declaration of conflict:** The authors declare that they do not have any conflict of interest

**Source of funds:** The authors have received no funds

## REFERENCES

1. Merywn Shear and Paul Speight. OdontogenicKeratocysts. Pages 6-59. Cysts of the Oral and Maxillofacial Regions. 4<sup>th</sup> Ed. John Wiley & Sons. Blackwell Munskgard Publishing Ltd. 15 April 2008: 240 pgs.
2. M Anthony Pogrel. Treatment of Keratocysts: The Case for Decompression and Marsupialization. J Oral Maxillofac Surg. 2005; 63: 1667-1673
3. Walid Ahmed Abdullah. Surgical treatment of keratocysticodontogenic tumour: A review article. Saudi Dent J. 2011 Apr; 23(2): 61–65
4. Mamta Singh and K.C.Gupta. Surgical treatment of odontogenickeratocyst by enucleation. ContempClin Dent. 2010 Oct-Dec; 1(4): 263–267
5. Paul J. W. Stoelinga. The Management of aggressive cysts of the jaws. J Maxillofac Oral Surg. 2012 Mar; 11(1): 2–12.
6. Stephen E. Feinberg and Barry Steinberg. Surgical management of ameloblastoma. Current status of the literature. Oral Surgery, Oral Medicine, Oral Pathology. April 1996; 81(4): 383
7. Ann Arbor, Mich. Rafael Scaf de Molon, Mario H. Verzola, Luana C. Pires, Vinicius I. Mascarenhas, Rodrigo B. da Silva, Joni A. Cirelli, and Roberto H. Barbeiro. Five years follow-up of a keratocystodontogenictumor treated by marsupialization and enucleation: A case report and literature review. ContempClin Dent. 2015 Mar; 6 (Suppl 1): S106–S110.
8. NasimJafaripozve, ShahramJafaripozve, and MasoudAtaieKhorasgani. KerathocystOdontogenicTumor: Importance of Selection the Best Treatment Modality and a Periodical Follow-up to Prevent from Recurrence: A Case Report and Literature Review. Int J Prev Med. 2013 Aug; 4(8): 967–970.
9. Rawson K, Kallalli BN, Telkar S, Penumatcha MR. Keratocysticodontogenictumor of the right mandibular condyle: A rare case. J Indian Acad Oral Med Radiol 2014; 26:103-6
10. American Academy of Oral Surgeons- Conference. Paper 5658. 96th Annual Meeting Scientific sessions and Exhibitions. Nigel R. Johnson, Martin D. Batstone, Neil W. Savage. Management and recurrence of keratocysticodontogenictumor: A systematic review. Oral Surg Oral Med Oral Pathol Oral Radiol. 2013; 116: 271–276