

## REVIEW ARTICLE

### Different treatment modalities for OKC of jaws

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#### ABSTRACT:

Odontogenic cysts are relatively common lesions and accounts to form a major part of total biopsies received by any pathology service. This diverse group of lesions exhibit varying presentations ranging from a small innocuous lesion, which may be detected accidentally or may present as a highly aggressive and destructive lesion that may even transform into a malignancy. Among the latter type most notorious are odontogenic keratocyst (OKC). OKC is the one of the rare odontogenic cysts, which attracts many researchers due to its unique characteristics. The treatment of OKC remains debatable. So many authors have described different modalities. Since OKC exhibits high recurrence rate 5-15% of all odontogenic cysts, the ultimate goal of treatment should be complete, adequate removal of the cyst. Treatment should be based on so many parameters such as extent, age, aggressiveness and size of the lesions.

**Key words:** Odontogenic keratocyst, Treatment

Received: 12 May, 2020

Revised: 19 May, 2020

Accepted: 26 May, 2020

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**This article may be cited as:** Gul S, Shah AA, Bashir S, Bashir S. Different treatment modalities for OKC of jaws. Int J Res Health Allied Sci 2020; 6(3):x-x.

#### INTRODUCTION

Odontogenic keratocyst (OKC) is relatively most common and most aggressive developmental odontogenic cyst of the jaw. Philipsen in 1956 first described this cyst and Pinborg and Hansen suggested the histopathological importance for the diagnosis of OKC in 1962. It was initially called as primordial cyst as tooth primordium was thought to be the origin of the lesion. It is commonly believed that the origin of OKC comes from dental lamina remnants in maxilla and mandible another origin of OKC is from basal cells of oral epithelium overlying it. Odontogenic keratocyst occurs as unilocular or multilobular radiolucency often with dentigerous cyst. It comprises of around 11% of all cysts of jaws. The histological characteristics are very thin orthokeratinized or parakeratinized stratified squamous epithelium, well defined basal cell layer without rete pegs, lumen containing desquamated keratin and fibrous capsule. In 97% of OKC parakeratinized squamous epithelial lining is seen.<sup>8</sup> Daughter or satellite cysts are seen in connective tissue walls. Cystic lining is thin, fragile, and difficult to enucleate in one piece. In 1960, Gorlin and Goltz described simultaneous occurrence of multiple basal cell carcinomas, multiple OKCs of mandible and maxilla bifid ribs, and other changes.<sup>1</sup>

Treatments are generally classified as conservative or aggressive. Conservative treatment generally includes simple enucleation, with or without curettage, or marsupialization. Aggressive treatment generally includes peripheral osteotomy, chemical curettage with Carnoy's solution, cryotherapy, or electrocautery and resection. The goal is to choose the treatment modality that carries the lowest risk of recurrence and the least morbidity.<sup>2</sup> Eyre and Zakrezewska in 1985, have stated the following treatment modalities for OKC/KOT-<sup>3</sup>

- Enucleation
  - With primary closure
  - With packing
  - With chemical fixation
  - With cryosurgery
- Marsupialization
  - Only
  - Followed by enucleation
- Resection

#### DECOMPRESSION AND MARSUPIALIZATION

Decompression of a cyst involves any technique that relieves the pressure within the cyst as this pressure is the

way by which the cyst grows by expansion. Decompression can be performed by making a small opening in the cyst and keeping it open with a drain. Marsupialization, on the other hand, involves converting the cyst into a pouch so the cyst is decompressed, but this is a more definitive treatment than decompression as it exposes the cyst lining to the oral environment. Mandibular cysts are normally marsupialized into the oral cavity, while maxillary cysts can also be marsupialized into the maxillary sinus or nasal cavity, as well as the oral cavity.

Although decompression or marsupialization was not recommended as treatment for the OKC by some authors, because it was thought that the pathologic tissue would be left in situ, decompression or marsupialization has been recommended in a number of studies as a technique that allows partial decrease in size in the OKC so that vital structures like teeth or the inferior alveolar nerve can be preserved, then the OKC was certainly enucleated.<sup>4-7</sup>

Yildirim et al. advocated a conservative treatment protocol for OKC. He suggested enucleation followed by open packing. The resulting cavity was irrigated with mixture of normal saline and chlorhexidine gluconate. The resulting cystic cavity was packed with iodoform gauze impregnated with bacitracin ointment to minimize the risk of recurrence in each recall visits. The benefit of this protocol lies in the minimal surgical morbidity, decreased incidence of damage to associated structures such as the inferior alveolar nerve and developing teeth.<sup>8</sup> In his study, Voorsmit has proven that marsupialization followed by enucleation has no significant recurrence rate. Furthermore, Voorsmit et al. in their 1981 study, established that enucleation with Carnoy's solution has a recurrence rate as low as 2.7% compared to 13.5% for an isolated enucleation. It was Stoeltinga who insisted that the removal of the overlying alveolar mucosa that forms the roof of the cystic cavity eliminates the presence of daughter cysts between the cyst lining and the alveolar mucosa.<sup>9,10</sup>

#### **ENUCLEATION WITH AND WITHOUT ADJUNCTS**

Enucleation with and without various adjuncts has been utilized for many years. Although enucleation/curettage has the advantage over marsupialization of providing a complete specimen for histopathologic analysis, it shows recurrence rates as high as 62.5%, which is no longer an acceptable treatment modality. This high incidence of recurrence is explained by the thin, friable wall of the OKCT, which is often difficult to enucleate from the bone in one piece, and the small satellite cysts within fibrous wall. Many clinicians consider enucleation and curettage as the minimal requirement in the treatment of OKC.<sup>11,12</sup>

#### **ENUCLEATION AND TREATMENT OF THE BONY DEFECT WITH CARNOY SOLUTION**

As a result of the difficulty of enucleating the thin, friable wall of the KCOT as one piece, and due to the small satellite cysts, therefore, treatment should aim to eliminate the possible vital cells left behind in the defect.

For this reason a mild, not deeply penetrating, cauterizing agent is used such as Carnoy's solution {consists 3 ml of chloroform, 6 ml of absolute ethanol, 1 ml of glacial acetic acid and 1 g of ferric chloride}. This should be enough to do cauterization of the remaining cells. In case the cyst has penetrated through the lingual or buccal cortex, authors described the use electrocauterization to avoid a recurrence in the soft tissues.<sup>13</sup>

Chemical cauterization with carnoys solution is used since 1933 and showing lowest rate of recurrence and low morbidity as compared to recurrence. Modified carnoys solution is used now a days because of carcinogenicity of chloroform in older preparation. According to animal study depth of penetration of carnoys solution in cancellous bone is 1.-0.54 mm. Carnoys solution must not be applied to the vital structure more than 3min. As literature accepts the enucleation of OKC as least accepted modality for treatment due to higher recurrence but in present case the treatment modality used was enucleation only and follow up was done for 5 yr with no recurrence in follow up period.<sup>14</sup>

According to Blanas N et al., simple enucleation was reported to have a recurrence rate of 17% to 56% while simple enucleation combined with adjunctive therapy, such as the application of Carnoy's solution or decompression before enucleation, was reported to have recurrence rates of 1% to 8.7%.<sup>12,13</sup>

The use of Carnoy's solution in combination with peripheral ostectomy may increase its safety margins and compensate for its deficiency when lesions are near soft tissues and between dental roots. Chow and Morgan et al demonstrated low recurrences (4.3% and 0%, respectively) when this combination of therapies was applied, but these authors did not specify the follow-up period that was used.<sup>14,15</sup>

#### **ENUCLEATION AND LIQUID NITROGEN CRYOTHERAPY**

Theoretically, the ideal treatment for the KCOT would be enucleation or curettage followed by treatment of the cavity with an agent that would kill the epithelial remnants or satellite cysts. In addition, the osseous framework should be left intact to allow for osteoconduction. Liquid nitrogen has the ability to devitalize bone in situ while leaving the inorganic framework untouched, as a result of this, cryotherapy has been used for a number of locally aggressive jaw lesions, including KCOT, ameloblastoma and ossifying fibroma. Cell death with cryosurgery occurs by direct damage from intracellular and extracellular ice crystal formation plus osmotic and electrolyte disturbances.<sup>16,17</sup>

#### **BLOCK RESECTION, WITH OR WITHOUT PRESERVATION OF THE CONTINUITY OF THE JAW**

Resection refers to either segmental resection (surgical removal of a segment of the mandible or maxilla without maintaining the continuity of the bone) or marginal resection (surgical removal of a lesion intact, with a rim of uninvolved bone, maintaining the continuity of the

bone) which is an extreme technique, that results in considerable morbidity, particularly because reconstructive measures are necessary to restore jaw function and aesthetics.<sup>18-23</sup>

### OTHER TREATMENT OPTIONS

Recently some researchers are working on the non-surgical management of odontogenic keratocyst. In 2000, Taipale., et al. shown that a plant derived teratogen cyclopamine shows the potent antitumor activity by blocking SHH pathway. In 2006, Zang., et al. was first to pointed out that intracystic injection of cyclopamine can be used as nonsurgical therapy for management of odontogenic keratocyst. Vemurafenib, dabrafenib, and trametinib are BRAF inhibitors can be used in management of OKC but above all drugs requires clinical trial.<sup>24, 25</sup>

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### CONCLUSION

Conservative surgical management of Odontogenic Keratocyst (OKC) with combined therapy using multimodal therapeutic approaches has been shown in the past literature as the first choice for treating this pathological condition. Therefore, an appropriate long-term follow-up must be done after the treatment performed in order to ensure clinical success described as an absence of signs of recurrent disease.

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