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Surgical Treatment of Odontogenic Keratocyst Tumour: A Review Article

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ABSTRACT

Odontogenic keratocyst is one of the most aggressive odontogenic cysts with a high recurrence rate, this was explained histopathologically as it typically shows a thin, friable wall, which is often difficult to enucleate from the bone in one piece, and have small satellite cysts within the fibrous wall. Multiple surgical approaches were introduced including decompression, marsupilization, enucleation with or without adjunct (Carnoy's solution, enucleation) and resection. Depending on other studies KCOT can be conservatively treated with enucleation and application of Carnoy's solution or cryotherapy. This can be used specially in the large lesions that when treated with resection, the continuity of the jaw will be interrupted. This technique shows comparable results to other more aggressive techniques.

Key words: Keratocystic odontogenic tumour, Marsupialization, Surgical treatment.

INTRODUCTION

Odontogenic keratocyst OKC is a developmental cyst that was first described by Philipsen (1956). OKC is now referred to by the World Health Organization (WHO) as a keratocystic odontogenic tumour KCOT, and WHO defined it as "a benign uni- or multi-cystic, intraosseous tumour of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium and potential for aggressive, infiltrative behaviour[1].

The KCOT is one of the most aggressive odontogenic cysts. It can become quite large because of its ability for significant expansion, extension into adjacent tissues and rapid growth[2]. Different studies showed the incidence of KCOT to be 3–11% of the odontogenic cysts. Generally, KCOT are solitary lesions unless they are associated with nevoid basal cell carcinoma syndrome[3] KCOT arises from cell rests of the dental lamina. Histopathologically, KCOT typically shows a thin, friable wall, which is often difficult to enucleate from the bone in one piece, and have small satellite cysts within the fibrous wall. Therefore odontogenic keratocysts often tend to recur after treatment[4]. Radiographically KCOT demonstrates a well-defined unilocular or multilocular radiolucency with smooth and often corticated margins. In 25–40% of cases, there is an unerupted tooth involved in the lesion. KCOT tend to grow in the anteroposterior direction within the medullary cavity of the bone without causing obvious bone expansion causing its delayed observation by the patients.

The treatment of the KCOT remains controversial. 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 ostectomy, chemical curettage with Carnoy's solution, cryotherapy, or electrocautery and resection[5].

The choice of treatment should be based on multiple factors; patient age, size and location of the cyst, soft tissue involvement, history of previous treatment and a histological variant of the lesion. The goal is to choose the treatment modality that carries the lowest risk of recurrence and the least morbidity[6].

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[7].

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[8].

Decompression and marsupialization of cysts is probably the earliest recommended treatment and was first suggested by Partsch in the late 19th century. In many parts of the world, marsupialization is still described as a Partsch I procedure (the Partsch II procedure is enucleation and primary closure)[9].

Although decompression or marsupialization was not recommended as treatment for the KCOT by some authors, because it was thought that the pathologic tissue would be left in situ[10], decompression or marsupialization has been recommended in a number of studies as a technique that allows partial decrease in size in the KCOT so that vital structures like teeth or the inferior alveolar nerve can be preserved, then the KCOT was certainly enucleated[10].

Those authors who are against the use of marsupialization or decompression for the treatment of KCOT depend on, that this technique does not remove completely the whole cystic covering, which would lead to a continuation of epithelial proliferation and facilitate the recurrence[(11)] reported a recurrence rate of 25% in 32 (OKCT) patients treated with decompression of the lesion. On the other hand, other studies have shown that marsupialization of KCOT can be followed by total resolution of the lesion without any further surgery[12].

The marsupialization technique was described by Pogrel (2005) as a window at least 1 cm in diameter is made into a cyst, and an attempt is made to suture the cyst lining to the oral mucosa. In the maxilla, the cyst is then often packed open with the packing protruding through the opening. The packing consists of iodoform gauze impregnated with bacitracin ointment. When it is removed in the maxilla, the cavity is usually self retaining and the patient needs to irrigate twice a day to prevent food accumulation or closure of the fistula. In the mandible, there is a greater tendency for spontaneous closure of the fistula and reformation of the cyst, particularly in the posterior mandible. In these cases, we have found that the use of a nasopharyngeal anaesthesia tube suitably cut down makes an excellent stent to keep the cyst open. Again, the cavity is irrigated twice daily (Pogrel, 2005).

Studies have shown that when the OKCT is open to the oral cavity by marsupialization, a number of changes occur in the cyst lining. Histologically, the lining of OKCT is only 5 or 6 cells thick and tears easily on attempted enucleation; which is one of the causes of the high recurrence rate. With decompression or marsupialization, the lining appears to become thicker and easier to enucleate, and histologically it does appear to change and resemble normal oral mucosa, both with routine histology and with immunohistochemistry[13].

Pogrel (2005) concluded that, decompression and/or marsupialization has at least as high a success rate as the other more aggressive treatments with lower morbidity and preservation of important vital structures.

Enucleation with and without adjuncts

To enucleate is “to remove whole or clean, as a tumour from its envelope.” Curettage is defined as “the removal of growths or other material from the wall of a cavity[(14)].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.[8,9] Many clinicians consider enucleation and curettage as the minimal requirement in the treatment of KCOT[15].

Regarding curettage, clinicians have advocated mechanical techniques (hand, rotary) alone or in combination with a chemical solution (Carnoy's) (Stoelinga, 2003) or cryosurgical agents (liquid nitrogen)[16].

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} (Morgan et al., 2005). 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[17].

Other studies showed that, although the defect was treated with Carnoy's solution. Microcysts and epithelial islands were always seen in the overlying attached mucosa. And so recurrence took place. So, the authors of these studies recommended the complete excision of the overlying mucosa to decrease the recurrence also reported in their study that the treatment with Carnoy's solution did not show a significant association with recurrence. Yet, Voorsmit et al. (1981) reported a decreased recurrence rate following treatment with enucleation and Carnoy's solution (2.5%) compared with enucleation alone (13.5%)[18].

According to (Blanas *et al*., 2000) enculation of KCOT followed with application of Carnoy's solution appears to be the least invasive procedure with the lowest recurrence rate. And they reported that adding Carnoy's solution to the cyst cavity for 3 min after enucleation results in a recurrence rate comparable to that of resection without unnecessarily aggressive surgery.

The effects of Carnoy's solution on the inferior alveolar nerve were first reported by Frerich *et al*. (1994)[19]. The authors did not observe axonal damage during the first three minutes of direct application. In contrast, another important study, Wolgen *et al*. (1999), noted that the alterations in neural conductivity developed after 2 min of direct application, with few signs of recovery after two weeks of follow-up. However, Júnior *et al*. (2007), reported that when a proper protocol is followed, the chemical treatment of the nerve can be accomplished without permanent functional damage.

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[(20)].Cell death with cryosurgery occurs by direct damage from intracellular and extracellular ice crystal formation plus osmotic and electrolyte disturbances.

According to Schmidt and Pogrel (2001) the standardized technique is as follows, the initial step in management of the lesion is enucleation of the cyst. The surrounding tissues are then protected with sterile wooden tongue blades and gauze, and the cavity is sprayed with liquid nitrogen twice for 1 min, with a 5-min thaw between freezes. Bone graft can inserted in the defect simultaneously, and then mucosa is closed with watertight sutures.

The advantages of liquid nitrogen over alternative methods of devitalizing the tissue beyond the visible lesion of the margin are that (1) the bone

matrix is left in place to act as a clean scaffold for new bone formation, (2) a bone graft can be placed immediately to accelerate healing and minimize the risk of a pathologic fracture, and (3) decrease of bleeding and scarring. However, because of the difficulty in controlling the amount of liquid nitrogen applied to the cavity, the resultant necrosis and swelling can be unpredictable. The recurrence rate following enucleation and liquid nitrogen cryotherapy has been reported at 3–9%.

When the liquid nitrogen cryotherapy is given around the inferior alveolar nerve, it is affected, and patients will suffer paraesthesia or anaesthesia. However, the axon sheaths are left intact and nerve regrowth is normal such that most patients obtain partial or complete return of sensation in 3 months.

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) [21]which is an extreme technique, that results in considerable morbidity, particularly because reconstructive measures are necessary to restore jaw function and aesthetics, wonders whether such aggressive therapy is warranted for a benign lesion that can be managed reasonably well with relatively simple means.

In a systematic review done by Blanas *et al.* (2000)[22], the authors reported that resection was found to have the lowest recurrence rate (0%) but the highest morbidity rate, while enculation with application of Carnoy's solution can result in a recurrence rate comparable to that of resection without unnecessarily aggressive surgery.

Multiple studies concluded that keratocysts might be treated with a conservative approach, the only disadvantages being the extended therapeutic time. Extensive resection of the mandible with its attendant morbidity may be too radical for large KCOT and even an overtreatment[23].

Summary

KCOT is one of the most aggressive odontogenic cysts with a high recurrence rate. Multiple surgical approaches were introduced including decompression, marsupilization, enucleation with or without adjunct (Carnoy's solution, cryotherapy), and resection. Depending on other studies KCOT can be conservatively treated with enculation and application of Carnoy's solution or cryotherapy. This can be used specially in the large lesions that when treated with resection, the continuity of the jaw will be interrupted. This technique shows comparable results to other more aggressive techniques.

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