Management of Keratocystic Odontogenic Tumour With Marsupialisation, Enucleation and Carnoy's Solution Application: A Case Report

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Abstract
The keratocystic odontogenic tumour (KCOT) is a relatively common oral and maxillofacial lesion that derives from remnants of the dental lamina. It is aggressive, grows rapidly and invades the surrounding tissues. Various treatment modalities and differing recurrence rates have been reported for KCOT. In order to treat a 24-year-old male patient with KCOT the first stage was marsupialisation. This was followed six months later by enucleation with the application of Carnoy's solution. The defect was filled completely with newly formed bone tissue after two years. This case shows that a large KCOT can be treated with a combination of conservative and aggressive methods.

Key Words: Keratocystic Odontogenic Tumor, Carnoy's Solution, Marsupialisation, Enucleation

Introduction

The odontogenic keratocyst was first described by Philipsen (1956) [1] and its characteristics were defined by Pindborg and Hansen (1963) [2]. With its aggressive behaviour, high recurrence rates and specific histo-pathological features, the odontogenic keratocyst was reclassified and renamed "keratocystic odontogenic tumour" (KCOT) in the World Health Organization (WHO) classification of head and neck tumours in 2005 [3]. KCOT is a benign, uni- or multicystic, intraosseous tumour, which originates from the dental lamina and its remnants with a characteristic lining of parakeratinised layered squamous epithelium. It has a potential for aggressive, infiltrative behaviour. Multiple KCOTs are indicative of the presence of the nevoid basal cell carcinoma syndrome [4]. KCOT incidence rates vary from 4% to 16.5% and it forms 7.8% of all cystic lesions of the jaws [5]. KCOT generally occurs in the second and third decades of life and affects slightly more men than women [6]. Although the most common locations for this tumour are the angle/ramus of the mandible with a rate of 75%, unusual locations have also been reported such as premaxilla, maxillary 3rd molar region and maxillary sinus [4,5].

Radiographically, KCOTS may appear as small, round or ovoid, radiolucent lesions, often with scalloped, multilobulated and distinct margins [4]. Lesions may be uni- or multilocular. Some 25% to 40% are multilocular [5]. KCOTS can involve an unerupted tooth which results in a clinico-radiographical diagnosis of dentigerous cyst [5]. Other differential diagnoses are ameloblastoma, radicular cyst, simple bone cyst, central giant cell granuloma, arterio-venous malformation and a number of fibro-osseous lesions [4].

In this paper, the treatment of a patient with KCOT using marsupialisation combined with enucleation and Carnoy's solution is reported.

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Case Report

A 24-year-old male patient was referred to the authors' clinic with the complaints of pain and swelling in the posterior region on the left aspect of his mandible and numbness of his lower lip. The patient stated that the problems first occurred one month previously. There was no history of systemic disease. During an extra-oral examination, a swelling was located at the left mandibular angle. It was also seen at the lower side of the second molar region during an intra-oral examination (*Figure 1*). There was no pain when the lesion was palpated. Panoramic radiography and computerised tomography scanning were performed. The radiological examination revealed a unilocular radiolucent lesion with well-defined sclerotic margins, including lower second molar tooth in it and it extended up to the mandibular notch. No resorption was seen on the roots of the tooth but destruction was observed on the lingual cortex (*Figure 2*). After fine-needle biopsy and histopathologic evaluation, the lesion was identified as a keratocystic odontogenic tumour.

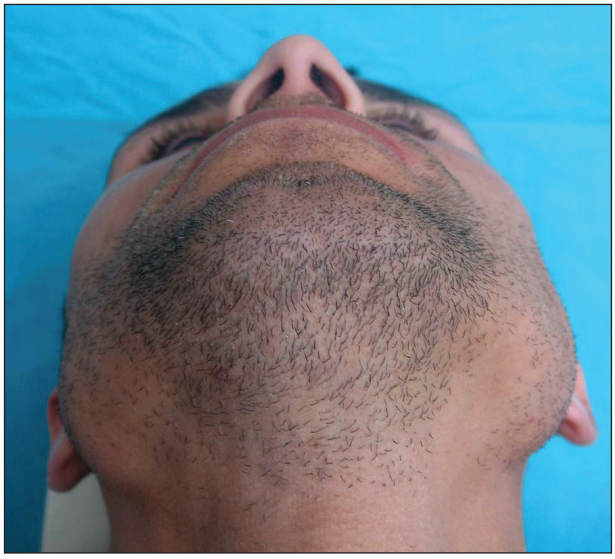


Figure 1. *Pre-operative extra-oral photograph showing swelling over the lower border of the mandible on the left side.*

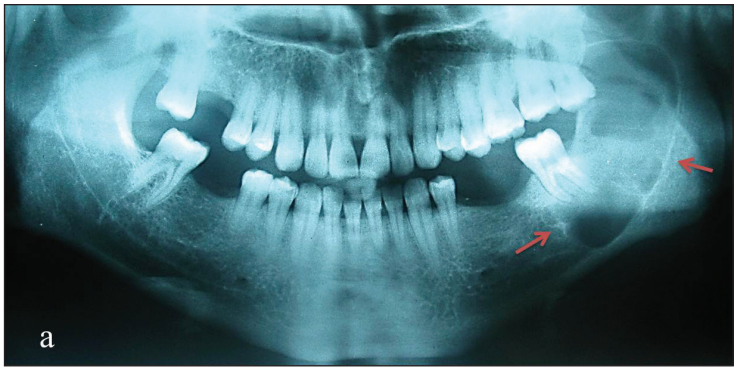
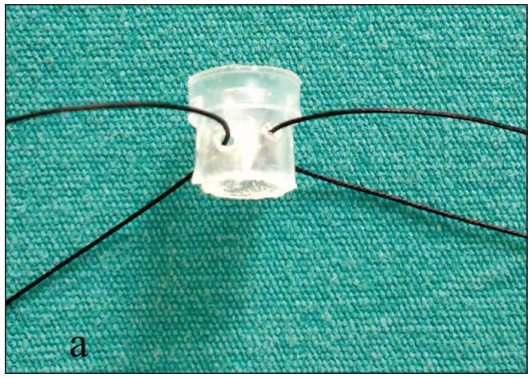


Figure 2. *(a) Pre-operative panoramic radiograph shows the unilocular radiolucency extending from the left second molar tooth to the mandibular notch (red arrows). (b) Pre-operative computed tomography scan coronal image showing destruction of the lingual cortex.*

Because of the dimensions of the lesion, relations with the anatomical features and the age of the patient, marsupialisation was chosen as the treatment option. Epithelium of the cyst was sutured to the oral mucosa after the extraction of the involved second molar under local anaesthesia. In order not to close the gap the inferior side of the injector, in which holes were drilled, it was sutured to the mucosa (*Figure 3*). One week after the operation, impressions were taken to fabricate an interim obturator prosthesis. The obturator prosthesis was inserted and checked for proper fit and final adjustments were performed. A histopathological examination was made of the tissue samples taken from the operation site. On the basis of the resulting histological appearance, a definitive diagnosis of the parakeratotic type of KCOT was made (*Figure 4*).

Post-operative follow up reviews were performed monthly, with clinical and radiological examinations. Six months after the surgery, it was seen on the computerised tomography scan that the lesion had become smaller (*Figure 5*). In addition,



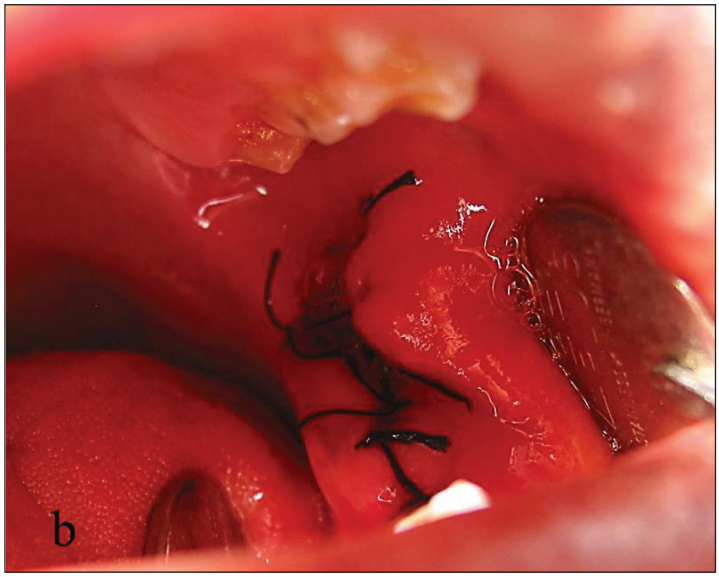


***Figure 3**. (a) Photograph of under-side of the injector.*
(b) A marsupialised keratocyst of the left posterior mandible with under-side of the injector sutured in place to maintain patency.

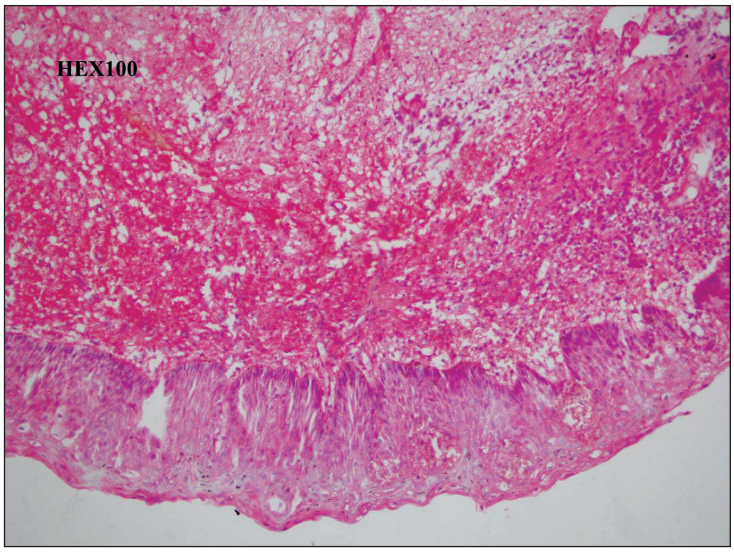


***Figure 4**. Typical histopathologic appearance of the KOCT showing 6 to 8 cells thick with a hyperchromatic and palisaded basal cell layer and a corrugated parakeratotic surface (H-E staining, x100).*

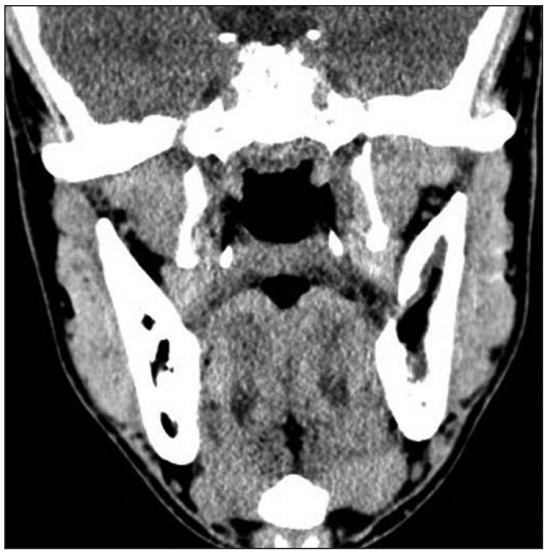


***Figure 5**. Computerised tomography scan showing that six months following the marsupialisation, the lesion was smaller.*

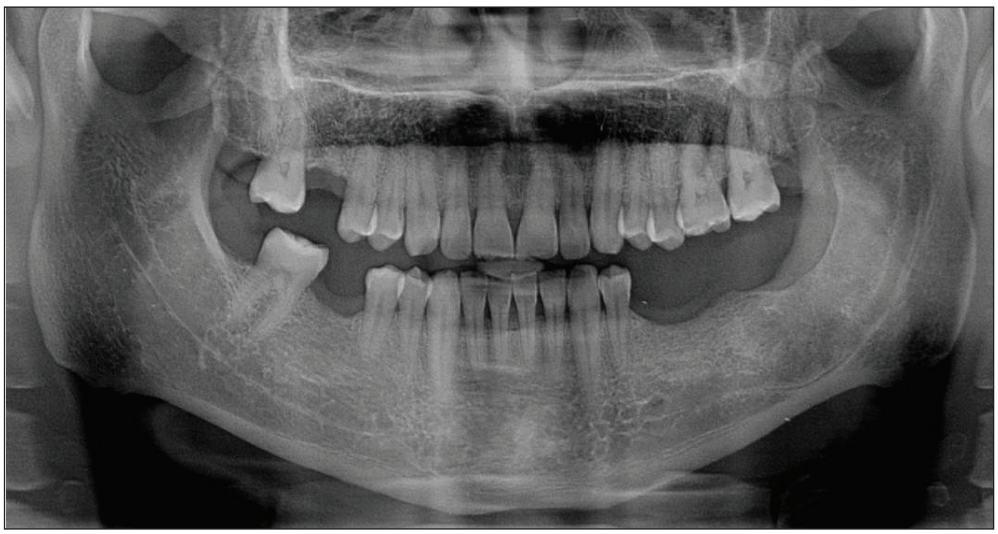


***Figure 6**. Panoramic radiograph at the end of two-year follow-up period, showing no signs of the recurrence.*

during the monthly reviews, it was noted that the paraesthesia of the lip had disappeared. At the second stage of the treatment, enucleation of the residual lesion was performed under local anaesthesia and the operation site was chemically cauterised for five minutes using Carnoy's solution, consisting of 300 ml of chloroform, 600 ml ethanol and 100 ml of acetic acid. No complication or recurrence has occurred after two years of follow up (*Figure 6*).

Discussion

Histologically, KCOTs have been classified as parakeratotic and orthokeratotic. These descriptions refer to the histologic characteristics of the lining and the type of keratin produced. The parakeratotic type is characterised by aggressive growth and a tendency to recur after surgical treatment. Because of these features, some authors have advised aggressive treatment methods [7]. The lesion reported in this case was a parakeratotic KCOT.

Several treatment options for the management of the keratocysts have been described in the literature. When choosing the treatment procedure, it is important to consider the age of the patient, the lesion's size, localisation and relationship with the surrounding soft tissue, and whether it is a primary or a recurrent lesion [8]. The treatment methods are categorised as conservative and aggressive.

Conservative treatment includes enucleation and marsupialisation. This method has the advantage of preserving anatomical structures and is applicable to patients from all age groups. Many authors have reported difficulties in the enucleation and curettage of KCOTs with or without cortical perforation due to adherence of the keratocyst's thin lining to adjacent bone or soft tissues. If enucleation is chosen as a surgical treatment, special attention must be given to the dentate area; tooth removal should be considered if there is any doubt of leaving pathologic tissue behind [4].

Aggressive treatment addresses the neoplastic nature of the KCOT. It includes chemical curettage with Carnoy's solution, peripheral ostectomy, and bone resection. These modalities are generally recommended for large KCOTs, as in the case reported in this paper, and recurrent lesions. However, in recent years, successful marsupialisation has been reported for the treatment of large lesions that were surrounded by anatomic structures [9]. For the case presented in this report, marsupialisation was performed primarily in order to minimise the lesion and to add some distance between the lesion and the anatomic features. Subsequently, enucleation was carried out using Carnoy's solution. Although the KCOT had grown large, the patient was treated under local anaesthesia.

The purpose of using Carnoy's solution is to provide a total elimination of epithelial remnants from the cyst walls, which may cause recurrences. Gosau *et al.* (2009) [10] stated that enucleation combined with application of Carnoy's solution reduced the recurrence rate of KCOTs compared with simple enucleation. Zhao *et al.* (2002) [11] reported that 29 of the 163 patients treated only with enucleation and two of the 29 patients treated with enucleation combined with Carnoy's solution fixation showed recurrence. No recurrence was seen in the 11 patients treated with marsupialisation and enucleation combined or in the 52 patients treated with resection. Madras *et al.* (2008) [15] reviewed the literature on the rate of recurrence KCOTs. They suggested that the recurrence rate is relatively low with aggressive treatment, whereas more conservative methods tend to result in more recurrences and after the combined therapy of enucleation and Carnoy's solution, the recurrence rate was found to be 9%. In this case, no sign of recurrence was detected after two years' post-operative follow-up. Some authors have suggested that if the inferior alveolar nerve is exposed in the cavity, Carnoy's solution can be used only once [12-14]. In our case, the inferior alveolar plexus was close to the cyst cavity and in order to prevent nerve damage the solution was applied once for five minutes.

Conclusions

Treatment of KCOTs remains a controversial subject. Many surgeons tend to treat large-sized KCOTs with an aggressive method. The outcome of this and other cases treated by the authors of this case report suggest that marsupialisation, enucleation and Carnoy's solution can be used as a reliable treatment option for large-sized KCOTs.

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Contributions of each author

- AO and GRB designed the study, collected data and wrote the paper.
- HAA wrote the introduction and references sections.

- MS gave advice and edited the paper.
- BS took and prepared the photographs.

Statement of conflict of interest

The authors are unaware of any conflicts of interest.

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