

Conservative Management of Keratocystic Odontogenic Tumour with Enucleation, Excision of the Overlying Mucosa and Electrocauterization – A Case Report

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ABSTRACT

Odontogenic keratocyst remains an enigma for clinicians and researchers, although gains in knowledge in recent years have improved the understanding of this interesting lesion. The diagnostic problems are mainly related to the relative lack of specific clinical and radiographic features that unequivocally point to a proper diagnosis. Of particular interest to clinicians is the biologic behaviour of keratocysts which includes high rates of recurrence and potential existence as a benign odontogenic neoplasm, keratocystic odontogenic tumour. Various surgical modalities have evolved in an attempt to reduce the recurrence rate, including curettage, peripheral ostectomy, removal of overlying mucosa in cases of cortical perforation and osseous resection in the form of marginal or segmental osteotomies. The present case report describes the conservative surgical management of a large keratocystic odontogenic tumour in a young patient with no evidence of recurrence at two years follow-up.

Keywords: Keratocyst, ostectomy, perforation, recurrence

Tratamiento Conservador del Tumor Odontogénico Queratoquístico con Enucleación, Escisión de la Mucosa Superpuesta, y Electrocauterización Un Reporte de Caso

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RESUMEN

El queratoquiste odontogénico sigue siendo un enigma para los clínicos e investigadores, aunque las ganancias en conocimientos en los últimos años han mejorado la comprensión de esta interesante lesión. Los problemas de diagnóstico están relacionados principalmente con la falta relativa de características clínicas y radiográficas que apunten inequívocamente a un diagnóstico apropiado. De particular interés para los médicos es el comportamiento biológico de los queratoquistes, que incluye altas tasas de recurrencia y la potencial existencia como un neoplasma odontogénico benigno, o tumor odontogénico queratoquístico. Varias modalidades quirúrgicas se han desarrollado en un intento por reducir la tasa de recurrencia, incluyendo el curetaje, la ostectomía periférica, la eliminación de la mucosa superpuesta en casos de perforación cortical, y la resección ósea en forma de osteotomías marginales o segmentarias. El actual reporte de caso describe el tratamiento quirúrgico conservadora de un tumor odontogénico queratoquístico grande, en un paciente joven sin evidencia de recidiva a dos años de seguimiento.

Palabras claves: Queratoquiste, ostectomía, perforación, recurrencia

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INTRODUCTION

In the plethora of cysts occurring in the jaws, the odontogenic keratocyst exhibits a unique feature with respect to its biologic behaviour. The term “keratocyst” was coined by Phillipsen [1956] (1) and later defined in 1990 by the World Health Organization (WHO) as a cyst “characterized by a thin fibrous capsule and a lining of keratinized stratified squamous epithelium, usually about five to eight cells in thickness and generally without rete pegs” (2). The lesion was later reclassified and renamed “keratocystic odontogenic tumour” (KCOT) in the WHO classification of head and neck tumours in 2005 (3).

Keratocystic odontogenic tumour is a benign lesion which originates from the dental lamina and its remnants and generally occurs in the second and third decades of life. Although a considerable predilection for the mandibular 3rd molar area and ascending ramus exists, KCOTs also occur in the dentate area both in the maxilla and mandible, presenting themselves as seemingly ordinary odontogenic lesions (4). Typical radiographic features include well defined unilocular or multilocular radiolucency with scalloped margins (5). Aspiration biopsies, revealing keratin flakes, are certainly helpful in diagnosis.

The lesion has aroused much interest because of its silent, infiltrative mode of growth and a strong tendency to recur after removal. The recurrence rate is variable and dependent on site, type of surgical procedure used and length of follow-up. The therapeutic approach varies in different studies from marsupialization, decompression and secondary excision, enucleation to enucleation and freezing or use of cauterizing agents, and marginal or even radical continuity resections (6, 7). The present case report describes the conservative management of a large odontogenic keratocyst *via* enucleation, excision of the overlying mucosa and electrocauterization with successful preservation of the natural teeth.

CASE REPORT

An 18-year old male patient reported to the Department of Oral and Maxillofacial Surgery complaining of swelling over the right side of the mandible since one and a half years. The patient had undergone incisional biopsy of the lesion along with extraction of the right lower first molar and left lower first and second premolars six months previously at another institute. Biopsy report was suggestive of odontogenic keratocyst.

Clinical examination revealed a diffuse, non-tender swelling approximately 4 x 4 cm, extending from the right body to the left angle region of the mandible. Intraorally, the swelling was seen obliterating the buccal vestibule extending from the right mandibular second premolar to the left lower first molar. It was bony hard and non-tender with expansion of the buccal cortex. Non-healing sockets of extracted teeth were noticed without any pathologic mobility in the mandibular teeth (Fig. 1). Radiographic examination in the form

of an orthopantomogram [OPG] (Fig. 2) and computed tomography (CT) scan revealed a well-defined, multilocular radiolucent lesion with sclerotic borders extending from the right mandibular first molar to the left third molar region with root resorption of involved teeth and thinning of the inferior border.



Fig. 1: Preoperative intraoral view with unhealed sockets (arrows) which are connected to the cystic cavity.

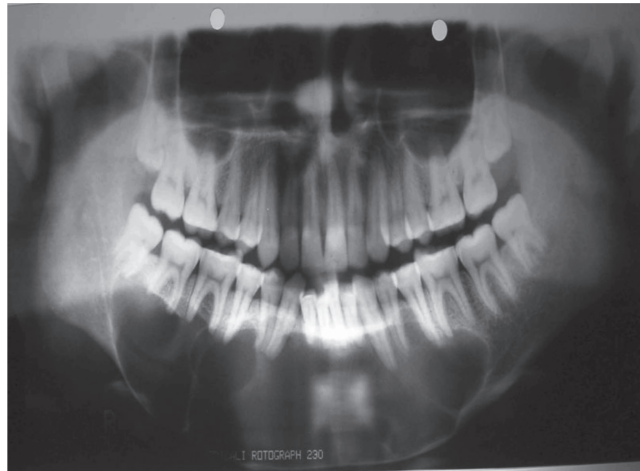


Fig. 2: Preoperative orthopantomogram (before extraction) showing multilocular radiolucency in mandible with root resorption.

A provisional diagnosis of odontogenic keratocyst was made based on these findings. Surgical enucleation of the lesion was planned under general anaesthesia after endodontic treatment of the involved teeth. After surgical exposure with a submarginal incision, the perforated buccal cortex was removed (Fig. 3) followed by enucleation of the underlying cystic cavity (Fig. 4). Apicoectomy of the involved teeth was done. Isolated perforations in the lingual cortex were cauterized with the help of unipolar diathermy.



Fig. 3: Removal of perforated labio-buccal cortex.



Fig. 4: Lesion enucleated in toto.

The attached mucosa overlying the lesion on the buccal aspect was excised before primary closure to minimize the possibility of recurrence.

The postoperative course of the patient was uneventful. Follow-up radiographs showed satisfactory healing (Fig. 5).

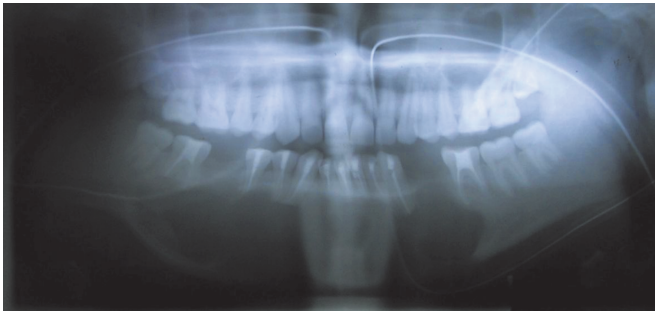


Fig. 5: Postoperative orthopantomogram.

The histopathological report of the specimen confirmed the diagnosis of odontogenic keratocyst (Fig. 6). The patient was followed-up every six months for two years with no evidence of recurrence.

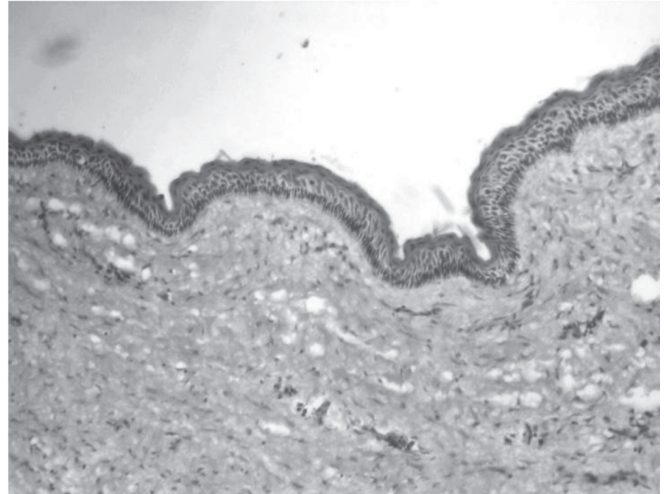


Fig. 6: Histology of the specimen showing parakeratinized epithelium with polarized, palisaded basal cell layer (haematoxylin-eosin stain; magnification x 20).

DISCUSSION

Keratocystic odontogenic tumour still poses a diagnostic and therapeutic challenge since its recognition as a separate entity. The objectives of treatment should include eradication of pathologic entity, while minimizing the risk of recurrence and patient morbidity. Both patient factors and lesion characteristics must be taken into consideration in the selection of the most appropriate treatment for KCOT. A discussion of conservative *versus* aggressive treatment must revolve around the risk to the patient of having a more extensive surgical procedure or the risk of developing untoward sequelae as a result of inadequate therapy (recurrence, extensive cysts involving the skull bone, etc).

Historically, various modalities have been employed in the management of KCOT including decompression, marsupialization, enucleation with excision of overlying oral mucosa, enucleation with adjuvant therapies (mechanical curettage, chemical [Carnoy's solution] curettage, cryosurgery), resection with or without continuity defect and disarticulation (8). Out of these, enucleation, curettage and marsupialization are commonly regarded as conservative aspects of surgical management (9), while resection with or without continuity defect and disarticulation represent the radical spectrum of treatment (10).

Many clinicians consider enucleation and curettage the minimal requirement, since earliest studies have shown that complete eradication of the lesion is necessary to decrease or eliminate recurrences. However, adherence of the keratocyst's thin lining to adjacent bone or soft tissues may result

in incomplete enucleation of the lesion (11). In such cases, several authors advise excision of soft tissue in continuity with the lesion (12, 13). It is thought that possible epithelial remnants in the lingual area may potentially give rise to untreatable recurrences in the soft tissues. Electrocauterization is probably more effective in eliminating these possible epithelial remnants in that its cauterizing effect reaches somewhat deeper than Carnoy's solution (5).

For the most part, KCOTs are accessible from an intraoral approach, lending them to a conservative surgical option. The obvious advantage of conservative approach is preservation of adjacent bone, soft tissues and dental structures which is of utmost importance in young patients (as in this case). Moreover, "recurrent" KCOTs are histologically similar to the "original" lesion and therefore are amenable to repeated conservative surgical interventions (9). Hence, we suggest that aggressive management of primary and secondary lesions should be reserved for the following cases:

- KCOTs involving the condylar process of the mandible which requires resection with disarticulation.
- When there is involvement of the lower border of the mandible with little likelihood of preservation of continuity.
- Odontogenic keratocyst of the maxilla with extraordinary size and involvement of the orbit, nasal cavity or pterygoid fossa.
- Lesions that have undergone malignant transformation.

It can be concluded that with the help of improved radiographic and other diagnostic aids, the conservative approach, if performed with diligence and an aim to remove the lesion completely, results in minimal recurrence. The radical approach should be reserved only for those cases that have undergone ameloblastic or carcinomatous change or for

recurrent lesions that are not surgically accessible by more conservative means. In any case, long-term clinical and radiographic follow-up is mandatory for keratocystic odontogenic tumours since recurrence has been reported even years later.

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