



Unique Journal of Medical and Dental Sciences

Available online: www.ujconline.net

Case Report

ODONTOGENIC KERATOCYST INVOLVING THE ANGLE AND RAMUS OF THE MANDIBLE - A CASE REPORT

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Received: 29-07-2016; Revised: 27-08-2016; Accepted: 25-09-2016

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ABSTRACT

Odontogenic Keratocyst (OKC's) are known for their peculiar behaviour, varied origin, debated development, unique tendency to recur and disputed treatment modalities. Thus, it has been the subject of much research over the last 40 years. OKC received its new title as keratocystic odontogenic tumour (KCOT) by the WHO (2005) in order to suggest its aggressive and recurrent nature. It is a benign intraosseous neoplasm of the jaw. OKC most commonly occurs in mandible with predilection for the angle and ascending ramus and male predominance, which demonstrate a unilocular, round, oval, scalloped radiolucent area, while large lesions may appear multilocular. An important characteristic of OKC is its propensity to grow in an antero-posterior direction within medullary cavity of bone causing minimal expansion. Definitive diagnosis relies on histological examination. Treatment modalities included multiple surgical approaches that includes decompression, marsupialization, enucleation with or without adjunct (Carnoy's solution, enucleation) and resection. The objective of the present study is to report a clinical case of a surgical treatment of a parakeratinized odontogenic keratocyst by enucleation followed by tanning using carnoy's solution in a 18 year old male patient.

Keywords: Odontogenic Keratocyst, Keratocystic Odontogenic Tumor, Intraosseous Neoplasm, Enucleation

INTRODUCTION

Odontogenic keratocyst OKC is a developmental cyst that was first described by Philipsen in 1956¹. OKC since 2005 is 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"². WHO "recommends the term keratocystic odontogenic tumour as it better reflects its neoplastic nature"². KCOT arises from cell rests of the dental lamina³.

The most common location of OKC is posterior body of mandible and ramus⁴⁻⁶. Clinically, OKC reaches to a large size and usually remains asymptomatic, unless it penetrates the bone or becomes secondarily infected wherein it can manifest as a swelling and intraoral draining sinus⁷. OKC demonstrates a well-defined radiolucent area with smooth and often

corticated margins while large lesion may appear multilocular⁸. In 25-40% of the cases, it is found to be associated with an unerupted tooth⁹. Generally, the OKCs are solitary, multiple OKCs are frequently associated with nevus basal cell carcinoma syndrome, and the malignant transformation of OKCs has also been reported¹⁰.

The treatment of the OKC 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. 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 morbidity¹¹.

In this article we report a case of OKC in posterior region of mandible involving the body and the ramus of the mandible.

CASE REPORT

A 18 year old male patient had come to the department of oral and maxillofacial surgery at our institution with a chief complaint of pain in his lower left back tooth region since 1 month which was gradual in onset and continuous in nature, he also gives a history of mild swelling associated with pain a week after the pain was initiated but was reduced on taking medication. Now patient has again developed similar pain since a week. The medical history was non contributory with no history of trauma to the teeth or jaw. Personal history revealed that patient was vegetarian in diet and there was no history of any deleterious habit like smoking, tobacco or betel nut chewing, alcohol etc. On extra oral examination, the upper and middle 1/3rd of the face appears to be apparently normal. In lower left 1/3rd of the face mild swelling was seen in mandibular angle region (Fig 1). On palpation it was firm in consistency, tender, non-compressible, non-fluctuant and afebrile to touch. On Intraoral examination there were no inspectory findings, overlying surface was of same colour as that of surrounding mucosa. On palpation there was obliteration of buccal vestibule wrt distal to 37 teeth region. it was firm in consistency, tender and no discharge was present. Teeth were missing wrt 18,28,38,48 with normal healthy gingiva and periodontium. With the above clinical findings, provisional diagnosis of dentigerous cyst wrt 38 was given with differential diagnosis of odontogenic keratocyst.



Figure 1: Clinical photograph of the patient showing mild swelling on lower 1/3rd of the left side of face

Intraoral periapical radiograph (IOPA) was taken, which showed a radiolucency wrt impacted 38 tooth (Fig 2). OPG was taken which showed a well-defined radiolucency wrt left lower molar – ramus area which was extending from ramus region till mesial aspect of 36 on the body of the mandible with well-defined corticated borders on its anterior, superior, inferior and posterior aspect. Internal septae were present giving it a multilocular appearance (Fig 3). Radiographic diagnosis of odontogenic keratocyst was given.

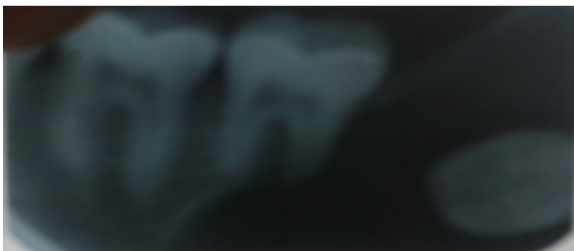


Figure 2: Intraoral periapical radiograph showing a radiolucency wrt impacted 38 tooth

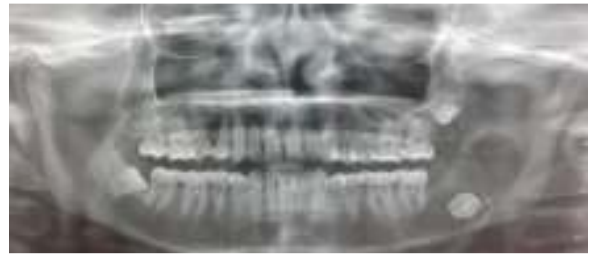


Figure 3: OPG showing the extent of the lesion

After taking the informed consent from the patient enucleation along with the extraction of the involved teeth 36, 37 and 38 was done under general anesthesia followed by application of Carnoy's solution (Fig 4). The excised specimen was sent for histopathological examination (Fig 5) which revealed that the overlying epithelium were odontogenic in nature with four to eight cell thick, with basal cell in palisading appearance. The superficial cell layer is parakeratotic in nature. The underlying connective tissue is loose to fibrous collagenous in nature, with chronic inflammatory cells spread throughout the connective tissue. In this case no daughter cyst or satellite cyst were seen (Fig 6), which were suggestive of odontogenic keratocyst.



Figure 4: Demonstrating surgical enucleation followed by application of Carnoy's solution



Figure 5: Excised specimen

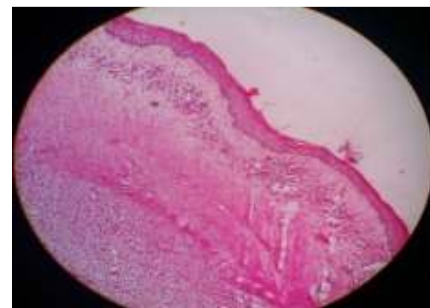


Figure 6: Histopathological view

The overall features were suggestive of odontogenic keratocyst based on clinical, radiological and histopathological findings. So final diagnosis of odontogenic keratocyst wrt left lower molar – ramus area was given.

The patient was followed up regularly for a period of 1 year and the healing was found to be satisfactory with no tendency for recurrence.

DISCUSSION

In the latest World Health Organization classification the former odontogenic keratocyst is added to the benign odontogenic tumours category. The new term is “keratocystic odontogenic tumor” (KCOT)¹².

The odontogenic keratocyst (OKC) is a known pathologic lesion of the jaws derived from rests of the dental lamina. It represents approximately 10 percent of all jaw cysts and may occur in a wide age range of patients. About 70 percent or more cases involve the mandible, especially in the molar, angle and ramus regions¹³. The present case also involved mandibular molar - ramus area.

Symptoms such as pain, swelling and drainage may be present, especially with larger lesions. However, at least half of all lesions are discovered as incidental radiographic findings. OKCs have a tendency for intraosseous growth, more often in a longitudinal than in a transverse direction (minimal expansion), as seen in this case, thereby replacing the bone marrow, rather than giving rise to periosteal bone formation, which would result in a bony swelling. Rapid growth does not allow enough time for the periosteum to lay down new bone¹⁴. In the present case there was pain with minimal swelling but no discharge was present.

An OKC usually occurs as a single lesion. Multiple lesions are associated with the nevoid basal cell syndrome (Gorlin–Goltz syndrome). Unlike other odontogenic cysts, OKCs have a high recurrence rate, reportedly ranging from 13 to 60%¹⁵. Recurrence occurs due to the following reasons. The first reason involves incomplete removal of the original cyst's lining. Secondly, it involves growth of a new OKC from small satellite cysts of odontogenic epithelial rests left behind by the surgical treatment. The third reason involves the development of an unrelated OKC in an adjacent region of the jaws, which is interpreted as a recurrence. Marx and stern believe that the two most common reasons for recurrence are incomplete cyst removal and new primary cyst formation¹⁶.

The radiographic appearance of OKC may resemble that of many other odontogenic lesions. If it is associated with a crown of an unerupted tooth, it may be indistinguishable radiographically from dentigerous cyst^{8,17}. The scalloped margin and multilocular appearance of OKC may resemble an ameloblastoma but the greater propensity to expand of ameloblastoma may help for differentiation of these lesions. OKC may show similarity to an odontogenic myxoma because of the mild expansion and multilocular appearance⁴. The above finding was in line with our case where radiographic features of the lesion represented a well-defined multilocular radiolucency involving mandibular impacted third molar tooth.

Histologically OKCs has been classified by some authors as parakeratotic or orthokeratotic subtypes. These types refer to

the histologic characteristics of the lining and the type of keratin produced. Compared to the parakeratotic subtype, the orthokeratotic subtype produces keratin more closely resembling the normal keratin produced by the skin with a keratohyaline granular layer, immediately adjacent to the layers of keratin which do not contain nuclei. The parakeratotic subtype has more disordered production of keratin; no keratohyaline granules are present and cells slough into the keratin layer. The keratin contains nuclei and is referred to as parakeratin. The parakeratotic subtype is the most frequent (80%) and has a more aggressive clinical presentation than the orthokeratotic variant. Some pathologists think that the orthokeratotic type should be classified as a separate entity and termed as orthokeratotic odontogenic cyst because of its distinct histological feature and substantially less-aggressive behaviour¹⁸. The lesion reported in this case is a parakeratotic OKC.

The treatment of the OKC remains controversial. Treatments are generally classified as conservative and aggressive. Conservative treatment generally includes simple enucleation, with or without curettage, using spoon curettes of marsupialization. Aggressive treatment generally includes peripheral ostectomy, chemical curettage with carnoy's solution and resection. Some surgeons believe that the cyst can be properly treated with enucleation if the lesion is removed intact. However, complete removal of the OKC can be difficult because of the thin, friable epithelial lining, limited surgical access, skill and experience of the surgeon, cortical perforation, and the desire to preserve adjacent vital structures. The goals of treatment should involve eliminating the potential for recurrence while also minimizing the surgical morbidity¹⁶.

Currently, treatment involving careful and aggressive enucleation with close follow-up has been advocated for the OKC. John and James described the use of enucleation in conjunction with a chemical cauterizing agent and excision of overlying mucosa as a means of reducing recurrence¹⁹. In the case discussed here similar treatment protocol of aggressive enucleation in conjunction with tanning with freshly prepared carnoy's solution (absolute alcohol, chloroform, glacial acetic acid, and ferric chloride) to prevent recurrence was used.

CONCLUSION

OKC is one of the most aggressive odontogenic cysts with a high recurrence rate. It has been the subject of much debate over the last 40 years with respect to its origin, its growth, and treatment modalities. Unfortunately, there is no consensus on a uniform treatment plan and the recommended surgical managements vary from marsupialization and enucleation to en bloc resection. The type of treatment chosen depends on several factors like patient age, lesion location and size and whether the OKC is primary or recurrent. In this case the obvious advantages of our treatment technique were:

- Complete removal of the pathologic lesion,
- Reduction of the potential for recurrence as the case was regularly followed for 1 year and no signs of recurrence were seen.

- Preservation of the continuity of the mandible, thus maintaining jaw function and shape.

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Source of support: Nil, Conflict of interest: None Declared