KERATOCYSTIC ODONTOGENIC TUMOR A CASE REPORT

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ABSTRACT

Odontogenic tumors (OT) are lesions that derive from the tooth-producing tissues or their remnants that remain entrapped either within the jawbones or into the adjacent soft tissues. WHO's formal reclassification of Odontogenic keratocyst as a tumour underscores the fact that this lesion should not be managed as the simple cyst it was believed to be. The distinction and delineation from cyst to a tumor is an enigmatic part in the spectrum of progression of this disease. The purpose of this paper is to review the treatment of a patient with KCOT using marsupialisation combined with enucleation.

Key words:OKC, KCOT, TUMOR, ENUCLEATION.

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INTRODUCTION

Odontogenic tumors (OT) are lesions that derive from the tooth-producing tissues or their remnants that remain entrapped either within the jawbones or into the adjacent soft tissues. Odontogenic tumors comprise a heterogeneous group of lesions that ranges from hamartomas to benign and malignant neoplasms of variable aggressiveness[1].

First described by Philipsen in 1956[2] and its characteristics defined by Pindborg and Hansen (1963)[3], the odontogenic keratocyst (OKC) is now designated by the World Health Organization (WHO) as a kerato-cystic odonto-genic tumor (KCOT) and is defined as "a benign uni- or multicystic, intraosseous tumor 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 tumor as it better reflects its neoplastic nature (Barnes et al., 2005)[2]. The distinction and delineation from cyst to a tumor is an enigmatic part in the spectrum of progression of this disease.

KCOT incidence rates vary from 4% to 16.5% and it forms 7.8% of all cystic lesions of the jaws. KCOT generally occurs in the second and third decades[3,4] of life and affects slightly more men than women. Although the most common locations for this tumor are the angle / ramus of the mandible with a rate of 75%, unusual locations have also been reported such as pre-maxilla, maxillary 3rd molar region and maxillary sinus[3].

In this paper, the treatment of a patient with KCOT using marsupialisation combined with enucleation is reported.

CASE REPORT

A female patient aged 59 years, reported to the private practitioner with the chief complaint of numbness and vague pain on the right lower back jaw region since 6 months. On anamnesis, a history of an atraumatic extraction of a tooth was given on the right side related to the chief complaint, with no relief theresoever. She was well oriented to time, person and place upon presentation. Her medical history was also non- contributory.

On extra-oral examination, no evidence of any swelling on the right side of the lower jaw was seen. There was a moderate paresthesia of the right half of the lower lip and the skin above the parasymphyseal and symphyseal regions of the mandible with slight expansion of the right lower border.

On intraoral examination, the full compliment of maxillary teeth was present with missing left 2nd premolar and right 1st molar in mandible. A small lingual cortical expansion was noted on the right side of lower jaw without any buccal expansion or exudation. The gingiva was normal in texture, firm and resilient. There were proximal caries with tooth 45 & 47 and a restored 36.

The patient was advised for an IOPAR of right posterior sextant. The IOPAR revealed a well- defined, incomplete, corticated border immediately beneath right first molar. For confirmation and further examination, an Orthopantomogram (OPG) and an AP mandible view was advised to the patient. (Fig.1 & 2)

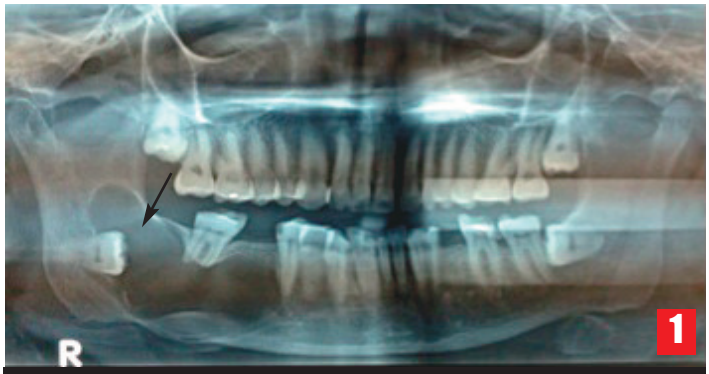


Fig. 1 - OPG revealing OKC extending into the right ascending ramus

Both OPG and PA mandible revealed a well-defined, large, unilocular radiolucent lesion on the right side of mandible with well-defined smooth and sclerotic borders and absence of scalloping. No internal septae were seen on the radiograph. All the four molars were

impacted with 48 pushed extremely to the posterior part of the ramus. The radiolucency measured 5cms × 4cms approx. and extended from distal aspect of 45 anteriorly to the posterior border of ramus involving the pterygomandibular raphe. The average surface area of the lesions measured radiographically was 20 sq.cm. It extended superiorly to the ascending ramus of mandible, completely involving it. There was no resorption of roots of the adjacent teeth. There was thinned out right lower of mandible with no evidence of pathologic fracture. The radiolucency engulfed 48 completely.

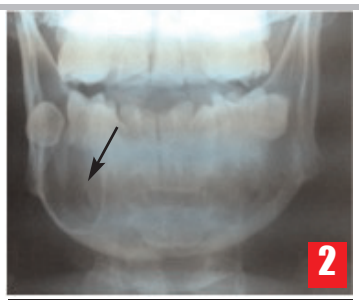
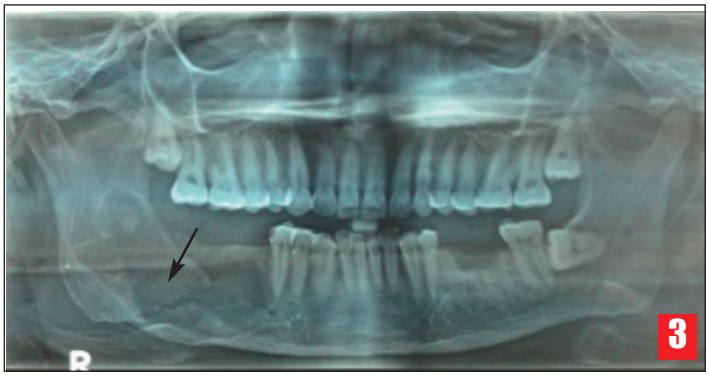
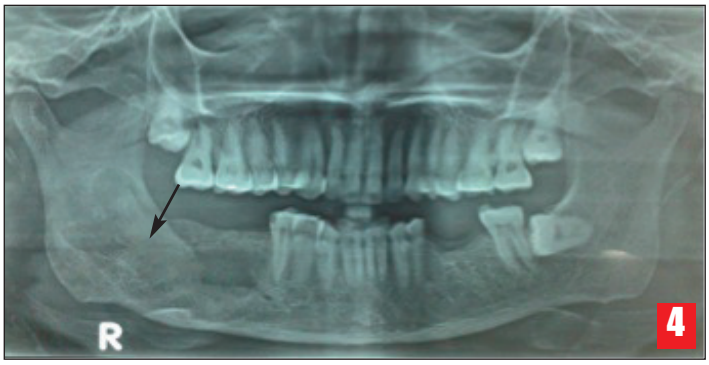


Fig. 2 - AP Mandible





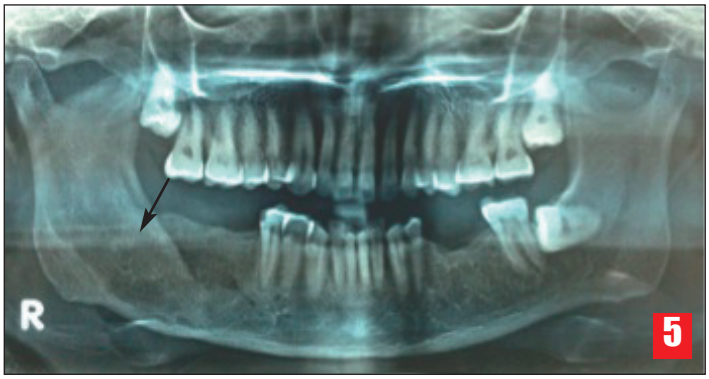


Fig. 3, 4 & 5 - OPGs after successive six monthly follow-ups showing healing of the defect.

The CT Scan was ruled out, as patient was not willing for the examination. Based on the OPG findings, radiographic diagnosis of central type of dentigerous cyst was made. Differential diagnosis included, odontogenic keratocyst, ameloblastoma and a central hemangioma of bone. The patient was advised an aspiration biopsy from the lesion and an incisional biopsy. Upon aspiration, about four ml of chocolate to reddish brown fluid with a semisolid consistency was obtained from the lesion.

Incisional biopsy was done from the affected side and histopathology report showed cytosmear with few inflammatory cells and cholesterol crystals. It was negative for any malignancy.

Based on radiology and histopathology reports of the lesion, patient was advised to undergo surgery for the same.

Surgery was performed under general anesthesia with extraction of 48 and decompression and masupialization followed by enucleation of the cyst with complete removal of the cystic lining and open packing. The cystic specimen was sent to pathology lab for final diagnosis. The final histopathology report confirmed it to be an odontogenic keratocyst.

The patient was put on regular follow up and successive OPG's were taken every six months for two years. The post-operative OPG's showed adequate healing with good bone formation and restoration of normal trabecular pattern. (Fig. 3, 4 & 5) Paresthesia was cured slowly after two years with good bone remodeling.

DISCUSSION

The KCOT (OKC) is an aggressive tumor, which is associated with impacted teeth and sometimes goes undetected if the patient does not inform about progressive symptoms, paresthesia and/or related discomfort. It may occur at any stage and the peak frequency occurs in second and third decades of life[3]. It is mostly detected on routine dental radiographic examination where there is enlargement at the expense of medullary space. The KCOT tends to grow in an antero-posterior direction within the medullary cavity without causing obvious bone expansion[4], and the tumor rarely expands in a bucco-lingual manner. Due to its propensity of longitudinal expansion, long standing cysts may be discovered as late as fifth decade as was found in our case. Odontogenic keratocysts appear to have an epithelial lining with an intrinsic growth potential and have a marked tendency to recur. This suggests an increased epithelial atypia[5].

At present, it is known that the potential sources to develop an odontogenic tumor are varied, and these include[1]:

1. The pre-functional dental lamina (odontogenic epi- thelium with ability to produce a tooth), which is more abundant for obvious reasons distally to the lower third molars.
2. The post functional dental lamina, a concept that covers those epithelial remnants such as Serre's epithelial rests, located within the fibrous gingival tissue; the epithelial cell rests of Malassez in the periodontal ligament and the reduced enamel organ epithelium, which covers the enamel surface until tooth eruption.
3. The basal cell layer of the gingival epithelium, which originally gave rise to the dental lamina.
4. The dental papilla, origin of the dental pulp, which has the potential to be induced to produce odontoblasts and synthesize dentin and/or dentinoid material.
5. The dental follicle.
6. The periodontal ligament, which has the potential to induce

the production of fibrous and cemento-osseous mineralized material.

In 1976, Brannon proposed three mechanisms for KCOT recurrence[2]:

i. Incomplete removal of the cyst lining,

ii. Growth of a new KCOT from satellite cysts (or odontogenic rests left behind after surgery) and

iii. Development of a new KCOT in an adjacent area that is interpreted as a recurrence.

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, cryo-therapy, or electro-cautery and resection[6].

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.

KCOTs have a high recurrence rate, reportedly between 25% and 60%[7].Recurrence has been reported as early as 10 months to as late as 25 years after surgery[8]. This could be contributed either due to retained fragments of the original cyst wall or the development of a new cyst from the 'satellite cyst'.The two years follow up in our case has shown good result and the non-recurrence of the cyst. The radiographic examinations revealed good bone and trabecular formation with an excellent healing of the cystic defect.

SUMMARY

KCOT is one of the most aggressive odontogenic cysts with a high recurrence rate. Multiple surgical approaches have been introduced including decompression, marsupilization, enucleation with or without adjunct and resection. As there is not enough experience to determine the proper therapeutic behavior in such cases; it is encouraged that the report of these kind of cases, as well as carrying out studies directed towards the identification of their pathogenesis and possible relationship with well established entities to know more about their biological behavior on the long term.

REFERENCES

1. Taylor AM. New findings and controversies in odontogenic tumors. Med Oral Patol Oral Cir Bucal 2008; 13(9):555-8.
2. Madras J. Lapointe H. Keratocystic odontogenic tumour: reclassification of the odontogenic keratocyst from cyst to tumour.JCDA 2008; 74(2): 165.
3. Ozkan A, Bayar GR, Altug HA, Sencimen M, Senel B; Management of keratocystic odontogenic tumour with marsupialisation, enucleation and carnoy's solution application- A Case Report. OHDM 2012; 11(2): 69-73.
4. Venkataraman BK. Diagnostic oral medicine. 1st ed. New Delhi (India):Wolters Kluwer Health; 2013 p. 536-37.
5. Özveren A, Tuskan C, Yaltirik M, Atalay B, Erseven G. Expression of the Tumour Suppressor Gene p53 in Odontogenic Cysts. Turk J Med Sci 2003; 33: 243-247.
6. Abdullah WA. Surgical treatment of keratocystic odontogenic tumour: A review article. The Saudi Dental Journal 2011; 23: 61-65.
7. Ongole R, Praveen BN. Textbook of Oral Medicine. 2nd ed. New Delhi (India): Elsevier; 2010 p. 407.
8. Langlais RP, Langland OE, Nortje CN. Diagnostic imaging of the jaws. Baltimore: Williams & Wilkins; 1995 p. 327-31.

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