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ABSTRACT:

Keratocysytic Odontogenic tumor is a lesion occurring in the oral cavity that has a high recurrence rate. This lesion has an ability to mimic other lesions affecting the jaw. The tumor has a varied clinical and radiographic presentation. This article presents a case report of a 45 year old male patient presenting with an asymptomatic swelling on the right lower jaw associated with an impacted tooth and depicts an unusual radiographic picture.

Key words: odontogenic tumor, keratocyst, recurrence,

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INTRODUCTION:

Keratocystic odontogenic tumor (KOT) defined by the World Health Organization (WHO), is a benign, intraosseous neoplasm of dental origin, with a characteristic lining of parakeratinized stratified squamous epithelium [1]. It was previously known as Odontogenic keratocyst and was first described by Phillipsen in 1956 [2]. The term Keratocystic odontogenic tumor was recommended by WHO which describes its neoplastic nature. It has a slight male predilection, usually between the second and third decades of life, but can occur at any age group. It can be located anywhere in the jaws,

most commonly occurring in the mandibular posterior region [3]. It can be considered as a benign odontogenic tumor with many distinguishing clinical and radiological features. The radiographic appearance of the lesion varies widely, which makes the diagnosis difficult. Herewith in this article, we report a case of Keratocystic odontogenic tumor in a 45 year old male patient who presented with an asymptomatic swelling in the right lower jaw that has varied distinguishing clinical and radiological features.

CASE REPORT:

A 45 year old male patient visited the department of Oral Medicine and Radiology, with a complaint of asymptomatic swelling in the left lower jaw since one month. He noticed the swelling one month back which gradually increased to the present size around 3×3 cm in diameter. Swelling began following a tooth ache and it was associated with throbbing type of pain which aggravates upon touch and during mastication. His medical and family history was noncontributory. Extra oral examination revealed the presence of diffuse swelling over left lower jaw measuring approximately 4×3 cm in size, extending inferiorly 0.5 cm below the inferior border of mandible. Swelling was non fluctuant, non-compressible and hard in consistency.

Intraoral examination revealed diffuse swelling involving left buccal vestibule extending posteriorly towards the retromolar region leading to obliteration of the left buccal vestibule Swelling was non tender and hard in consistency. Left mandibular canine was found to be missing and there was erosion of bone in relation to left mandibular lateral incisor and first premolar through which purulent discharge was visualized. Buccal cortical expansion was evident and there is thinning of bone lingually which made mylohyoid ridge more prominent. Severe cervical abrasion was noted in relation to left mandibular first premolar, second premolar and first molar. Aspiration revealed yellow coloured fluid. On vitality test, all mandibular teeth except mandibular right second premolar, first molar and second molar showed no response. Considering the history and clinical examination, provisional diagnosis of radicular cyst was made.

Panoramic radiograph revealed well defined hazy radiolucent lesion with a sclerotic margin involving the lower jaw (Fig. 1). Margins appear to be smooth, regular and non-corticated. Superior aspect of the lesion has a scalloped margin with respect to the teeth. The lesion extends from the distal aspect of left mandibular second molar, grows anteroposteriorly, and crosses the midline and reach till the distal aspect of right mandibular first premolar. Displacement of the teeth and resorption of roots of the teeth are evident. Presence of multiple septae with the radiolucent lesion gives a multilocular

appearance to the lesion. There was presence of tooth like radiopacity suggestive of an impacted tooth within the radiolucent lesion, located near the inferior border of the mandible. However the inferior border of the mandible was intact. Computed tomographic (CT) axial view showed a well-defined hyperdense area involving the mandible with its buccolingual and anteroposterior extension (Fig 2). The lesion crosses the midline to involve the other half of the jaw. Buccal cortical plates appear to be intact but there was perforation of lingual cortical plate on the left side of the jaw. There was also an evidence of hyperdense structure in the middle of the lesion suggestive of an impacted tooth.

The 3D reconstructed computed tomographic view showed the exact extension of the lesion with an evidence of through and through perforation of the mandible with respect to the lesion (Fig 3). On the basis of history, clinical and radiographic examination a final diagnosis of Keratocystic odontogenic tumor was made. In the present case seeing the extent of the lesion a surgical excision was planned. Surgical removal of the tumor was performed along with partial mandibulectomy and the specimen was sent for histopathological examination.

Histopathologic picture showed parakeratinized stratified squamous epithelium largely of a uniform thickness overlying a fibrous connective tissue showing focal areas of severe inflammation. Epithelial connective tissue interface was flat with basal cells of epithelium showing palisading appearance. Areas also showed proliferation and rete ridge formation with loss of surface keratinization. Histopathologic picture of the given section of the lesion was suggestive of infected odontogenic keratocyst (Fig 4).

Treatment carried out was enucleation of the lesion with partial resection of the mandible following which surgical plating was done. Patient was reviewed after 6 months, reported with no recurrence.

DISCUSSION:

Keratocystic odontogenic tumor (KOT) is a benign unicystic or multicystic, intraosseous tumour of odontogenic origin. KOT form approximately 11% of all jaw cysts and they have a very high recurrence rate.

The reason for the high recurrence rate may be due to the proliferation of islands of odontogenic epithelium that may be present in the wall giving rise to satellite microcysts. It has been reported that KOT most commonly occur between second and third decade of life but may be diagnosed at any age [3].

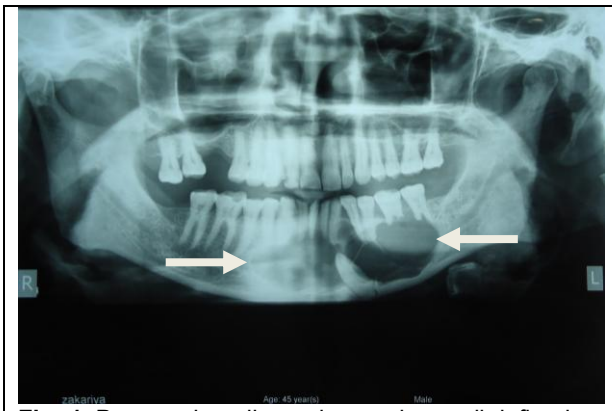

Fig: 1: Panoramic radiograph reveals a well defined hazy radioluscent lesion with superior scalloped margin with an evidence of resorption and displacement of teeth and an impacted tooth

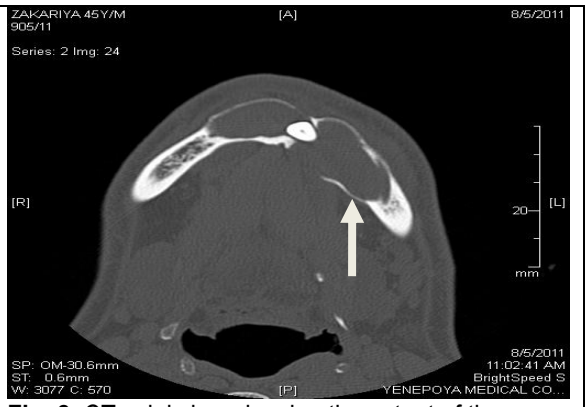

Fig. 2: CT axial view showing the extent of the lesion with the evidence of an impacted tooth and perforation of lingual cortical plate

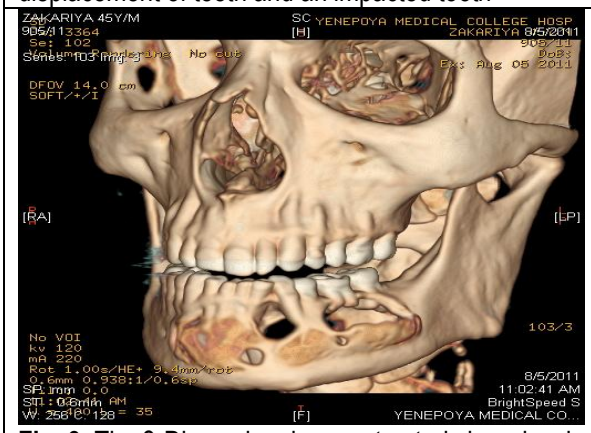

Fig. 3: The 3-Dimensional reconstructed view showing the extensive lesion with buccal & lingual cortical plate perforations. Shows the presence of bony septa within the lesion

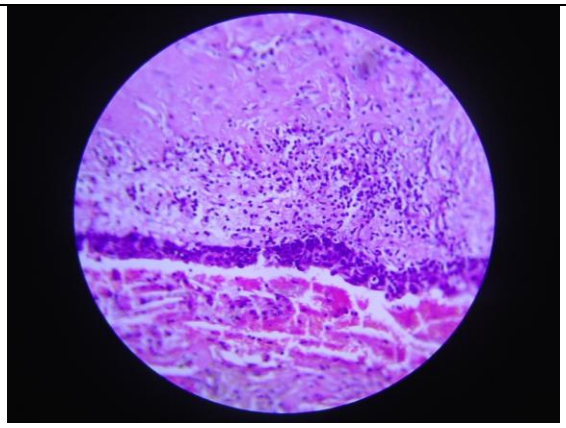

Fig 4: Histopathologic picture showing parakeratinized stratified squamous epithelium and basal cells of epithelium showing palisading appearance (10X zoom)

In another study, it was reported that the incidence was highest in the older age groups, and a decade younger in woman than in men [4]. The mandible is involved more frequently than the maxilla, the percentage of KOT occurring in the mandible ranges from 65-83% of cases [5]. The tumor can occur at any site in the mandible but most of the lesions arise in the posterior body, angle and ascending ramus of the mandible [4]. The lesion may be symptomless unless infected. It sometimes forms around an unerupted tooth; sometimes adjascent teeth may be displaced [1]. The present case was a 45 year old male patient with an asymptomatic swelling in the mandible with evidence of an unerupted tooth.

Unlike other lesion, the epithelium of KOT appears to have an innate growth potential. This difference in growth mechanism gives it a different radiographic condition. Conventional radiographs such as panoramic radiographs may be adequate in most of the cases to determine the location and estimate the size of KOT. Advanced imaging modalities such as

computerized tomography may be required to assess the full extension of the lesion [5]. In the present case, both the panoramic and computed tomographic imaging was taken.

KOTs occur as a well defined lesion which may be unilocular or as multilocular [6]. Most of the lesions of KOT are unilocular and often appears with smooth and regular borders. In the present case, panoramic view showed a well-defined multilocular hazy radioluscent lesion with superior scalloped border crossing the midline in the mandible with the presence of multiple septae making the lesion multilocular. The presence of septae within the bony cavity is the most striking feature of ameloblastoma, which serve to produce partial loculation of the cavity [7]. Scalloping of the sclerotic margin of the lesion that extends between the roots of the teeth is a characteristic feature of traumatic bone cyst. In the present case, scalloping of the margins was seen over the anterior part of the lesion which is a distinguishing feature. Since the present lesion was associated with an impacted tooth, dentigerous cyst should also be considered in the differential diagnoses [7]. Here the lesion emerges from the middle third root portion of the impacted tooth, and not from the cemento-enamel junction of the tooth, which is most frequently seen in case of a dentigerous cyst. The lesion in the present case extends from the posterior aspect of left mandibular second molar and was not localized surrounding the impacted tooth as in case of dentigerous cyst.

On clinical intraoral examination of the reported case, thinning of lingual cortical plate with increased fluctuation was felt, suggestive of perforation of the cortical plate. The perforation of the cortex was not well appreciated with two-dimensional image on the panoramic radiograph. However, the exact dimension of the lesion and the perforation of buccal and lingual cortical plates were visualized by CT scan sections and its three dimensional representation. Thus computed tomography plays an important role in imaging of an extensive lesion, thereby aids in the diagnostic process as well as treatment planning.

Histopathologic examination is usually required to arrive at a definitive diagnosis of KOT. Histologically KOTs can be classified into three categories: orthokeratinised, parakeratinised, or a combination of both [8]. The orthokeratinized subtype produces normal skin keratin with the presence of keratohyaline granules whereas the parakeratinized subtype involves disordered production of keratin without keratohyaline granules. The parakeratotic type has a more aggressive clinical presentation and is more frequent (80%) than the orthokeratotic variants [8]. The present case was a parakeratinized variety with areas of proliferation and rete ridge formation, which has a more chance for recurrence.

Many treatment options for KOT have been described in literature. The main goal of treatment is to reduce the risk of recurrence and morbidity of extensive resection [9].

Treatment modalities such as decompression, simple enucleation with or without curettage and resection have been employed in the treatment of KOT [10]. In the present case enucleation of the lesion along with partial resection of the mandible were carried out.

CONCLUSION:

The clinical and radiographic features of KOT's are not pathognomonic. It usually becomes difficult to distinguish with other lesions especially when the lesion is destructive with varied features. Advanced imaging is necessary for the visualization of its full extent following which definitive diagnosis is made histopathologically. The tumor requires a long term follow up since it has an aggressive and recurrent nature.

REFERENCES:

1. Philipsen HP. Keratocystic odontogenic tumour. In: Barnes L, Eveson JW, Reichart P, Sidransky D, eds. World Health Organization classifica-tion of tumours: pathology and genetic of head and neck tumours. Lyon: IARC Press. 2005: 306–7.
2. Philipsen HP. Om keratocystedr (Kolesteratomer) and kaeberne. Tandlaegebladet 1956; 60:963–71.
3. Myoung H, Song HP, Hong SD, Lee JI, Lim CY, Choung Ph, et al. Odontogenic keratocyst: review of 256 cases for recurrence and clinicopathologic parameters. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2001;91(3):328-333
4. Oda D, Rivera V, Ghanee N, Kenny MA, Dawson KH. Odontogenic keratocyst: the Northwestern USA experience. J Contemp Dental Pract 2000; 1(2):1-7.
5. Asokan G , Jeelani S, Parthiban J, Prabhushankar K., Prakash C, Shankar K.A. .Keratocystic Odontogenic Tumor – A Case Report And Review Of Literature. Int J Dent Case Reports 2012; 2(1): 87-91.
6. Gupta A, Rai B, Nair MA, Bhut MK. Keratocystic odontogenic tumor with impacted maxillary third molar involving the right maxillary antrum:An unusual case report. Indian J Dent Res 2011; 22:157-60.
7. H. M. Worth. Principles and Practice Oral Radiologic Interpretation. Year Book Medical Publishers Inc. Chicago.1988.
8. Cakur B, Miloglu O, Yolcu U, Goregen M, Gursan N. Keratocystic odontogenic tumor invading the right maxillary sinus: A case report. J Oral Sci 2008; 50:345- 9.
9. Leite TC, Meirelles V, Janini ME. Odontogenic Keratocystic Tumor: Clinical and Histopathologic Retrospective Study Based on the New WHO Classification. Int. J. Odontostomat 2011; 5(3):227-234.
10. Yildirim G, Ataoglu H, Kalayci A, Ozkan BT, Kuvuk K, Esen A. Conservative Treatment Protocol for Keratocystic Odontogenic Tumour: a Follow-up Study of 3 CasesJ Oral Maxillofac Res 2010;1(3);e7:1-11.