

Keratocystic odontogenic tumor: a case report

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ABSTRACT

The odontogenic keratocyst or keratocystic odontogenic tumor is a benign intraosseous neoplasm of the jaw. The main difference between odontogenic keratocyst and other jaw cysts is their potential aggressive behavior. Keratocystic odontogenic tumor shows a characteristic potential for infiltrative and aggressive behavior. This article reports a case of odontogenic keratocyst in left mandibular ramus and describes its clinical, radiographical and histopathological findings along with differential diagnosis.

Introduction

The odontogenic keratocyst (OKC) was first described in 1876 and named by Phillipsen in 1956. [1] OKC is now designated by the world health organization as a 'keratocystic odontogenic tumor' (KCOT) as it better reflects its aggressive and infiltrative nature. [2] KCOT 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 behavior. [2, 3]

OKCs of the jaw are developmental cysts arising from epithelial remnants of the tooth germ and another origin is from basal cells of the overlying oral epithelium. [4] Unlike other cysts, which are thought to grow solely by osmotic pressure, the epithelium in an OKC appears to have innate growth potential, much as in a benign tumor. [5]

Studies on KCOT have shown that the mandible is involved more often than the maxilla, while the posterior part of the mandible and ramus region is the most common location for OKCs. [6,7,8,9]

OKC is usually asymptomatic with mild swelling. Pain may occur with secondary infection. Aspiration by using a large-bore needle may reveal a thick, yellow, cheesy material (keratin).

Radiographic appearance of OKCs may range from a small to large radiolucencies with well-defined sclerotic borders, which can be either unilocular or multilocular. OKCs tend to grow in an antero-posterior direction within the medullary cavity of the bone without causing obvious bone expansion. [7,9,10]

Presence of multiple OKCs in 'nevroid basal cell carcinoma (Gorlin) syndrome' is well established. KCOT shows a high recurrence rate with 'nevroid basal cell carcinoma syndrome' and in multilocular lesion due to histological presence of one or more daughter cysts. [4].

The present report describes a case of KCOT in a 40-year old male patient.

Case Report

A 40 year old male patient presented with a complaint of pain and swelling in left cheek region since six months. The medical history was not contributory.

On examination a diffuse extra-oral swelling was noted on left side causing gross asymmetry of face [Fig.1]. The swelling was present in relation to left TMJ region that was firm and tender on palpation. Mouth opening was reduced to 5 mm.

Intra oral examination revealed a diffuse swelling with ill defined borders. The overlying mucosa was intact and was devoid of any abnormal changes. On palpation the inspeactory findings were confirmed, the swelling was hard in consistency, tender and no bruit or pulse was felt [Fig.2]. Aspiration revealed a thick, yellow, cheesy material [Fig.3].

On the basis of history and clinical features a provisional diagnosis of OKC and differential diagnosis of unicystic ameloblastoma was given.

The investigatory work up included complete hemogram, extra oral radiograph, computed tomography and excisional biopsy of the lesion.

Routine hematological investigation values were found to be within normal limits.

The Orthopantomogram revealed a well defined oval shaped radiolucency measuring approximately 2x3 cm in diameter, located in the left ascending ramus of the mandible, extending more towards the anterior border. The margins appeared to be sclerotic and the lesion had dense internal radiolucency [Fig.4].

The sagittal CT scan revealed hazy internal structures with interrupted and dispersed septae within the boundaries of the lesion [Fig.5].



Figure 1- Photograph showing extra oral view of the lesion.



Figure 2- Photograph showing the intra oral view of the lesion.



Figure 3- Photograph showing Cheesy white aspirate.

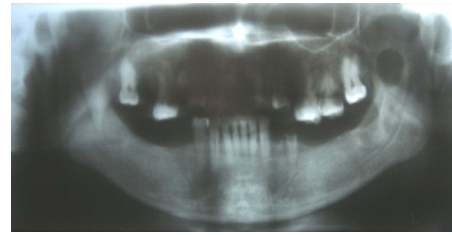


Figure 4- Photograph showing Orthopantomograph of the case.



Figure 5- CT reveals cystic cavity in mandibular ramus region.



Figure 6- Photomicrograph showing features of Keratocystic Odontogenic Tumor (H&E stain 40X).

The H&E stained sections revealed the presence of a parakeratinized stratified Squamous epithelium overlying fibro cellular connective tissue stroma. The parakeratinized epithelium appeared narrow and 5-6 layered with focal ameloblastomatoid proliferations of up to 10-15 layers at places. There was well defined palisaded basal layer containing columnar cells with intensely basophilic nuclei and increased mitotic figures both basally and supra basally. The superficial layer was corrugated and epithelial connective tissue interface appeared to be smooth with no rete ridges. The connective tissue stroma revealed satellite cyst suspended in edematous stroma with collagen fibers and plump fibroblasts. There was diffuse infiltration of chronic inflammatory cells along with endothelium lined blood vessels and RBCs [Fig.6].

Thus, a final diagnosis of KOCT was given.

Discussion

KCOT is seen in the second and third decade of life, with a peak

incidence between 11 and 40 years of age with a slight male predilection. Its common site is in mandible where the majority of the cysts occur in ramus and molar area.[11] While in maxilla, canine region is the most common location for KCOT [12] Unusual locations such as the anterior portion of maxilla, maxillary antrum, and maxillary third molar area, has also been reported.[12] In our case, the lesion was in left ramus region.

Conventional radiographic imaging, such as panoramic views and intraoral periapical films, in most cases are adequate to determine the location and estimate the size of an OKC. Advanced imaging techniques like computerized tomography and magnetic resonance imaging are important to assess full extent of lesion. [12, 13] In this case, both the panoramic and computed tomographic imaging was taken.

Radiographically KCOT appears as well defined radiolucency which may be unilocular or multilocular. [4] In this reported case, a unicystic, dense radiolucency with sclerotic margins was seen in panoramic view, while CT scan revealed hazy internal

structures with interrupted and dispersed septae within the boundaries of the lesion.

KCOT involves the cancellous bone in early and the compact bone in much later stages of its development therefore shows a very little expansion. [4] KCOTs are poor bone resorbers, because of this, scalloped margins related to the regional resorption of the surrounding bone is reported in long standing cystic lesion. [4, 14] In this reported case, no scalloping seen with the margins.

A definitive diagnosis of KCOT cannot be made without histopathological examination as other cystic and neoplastic diseases can present with same radiological features. [11] Unilocular OKCs can be located periapically, simulating periapical cysts [12, 15] Surrounding the crown of unerupted teeth, simulating dentigerous cysts, between the roots of teeth, simulating lateral periodontal cysts or lateral radicular cysts, or in the maxillary midline, simulating nasopalatine duct cysts. Large unilocular OKCs are difficult to differentiate from cystic ameloblastomas. [12, 16]

Histologically OKC shows two types of keratinization, a parakeratinization (with nuclei) and an orthokeratinization. [4] The orthokeratotic subtype produces normal skin keratin, with a keratohyaline granular while the parakeratotic subtype has more 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. [11] At histopathologic examination, an OKC has a fibrous wall lined by epithelium with a thin layer of stratified squamous epithelium. This epithelium has a basal layer six to eight cells thick and a lining of flattened keratotic epithelial cells. The formed keratin lines the luminal surface of the epithelial cells in a slightly wavy or corrugated pattern. [17] The luminal content can have different consistencies described as a "straw-colored fluid", "thick pus like" material; or a caseous, thick, cheesy, milk white mass. [2, 17] The varying consistencies reflect various densities of keratinaceous debris. Reported case is a parakeratotic KCOT.

Conclusion

Histopathological examination of keratocyst is important, because the clinical feature and radiographic appearance of OKC, although highly suggestive, are not diagnostic and this may lead to a risk for clinical misdiagnosis. CT is important for assessing full extent of lesion.

As multiple OKCs are associated with Nevroid basal cell carcinoma syndrome, early diagnosis and follow up of the patient with OKC is important. To avoid the recurrence, accurate clinical, radiographic, histopathological examination and intraoperative observation along with proper follow-up is needed.

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