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CASE REPORT

Keratocystic odontogenic tumor mimicking Radicular cyst: A Clinical dilemma

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ABSTRACT:

Jaw cysts are very common due to the presence of odontogenic epithelium remnants. The odontogenic keratocyst (OKC) is an epithelial developmental cyst of the jaws. This lesion is commonly found in the mandible, and can become quite large due to its rapid growth and its extension into the adjacent structures. The odontogenic keratocyst (OKC) is a distinct entity from other odontogenic cysts that deserves special attention due to its aggressive clinical behaviour and high rate of recurrence. WHO recommends the term keratocystic odontogenic tumor as it reflects its neoplastic nature. A case of odontogenic keratocyst involving the posterior mandible is presented in this article which was mimicking just like a radicular cyst. On closer view into the case proved the lesion to be an OKC.

Key words: ***Keratocystic odontogenic tumor, Odontogenic keratocyst, Periapical OKC, Jaw cysts, Odontogenic cysts***

Introduction

Odontogenic keratocyst was first described by Philipsen in 1956.[1] This lesion was recently renamed as Keratocystic Odontogenic Tumour (KCOT) and reclassified as an Odontogenic neoplasm in the World Health Organization's 2005 edition of its histological classification of odontogenic tumours. KCOT has been defined as ''A benign uni or multicystic intraosseous tumour of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium and potentially aggressive, infiltrative behaviour. It may be solitary or multiple. The latter is usually one of the stigmata of the inherited nevoid basal cell carcinoma syndrome (NBCCS).[2]

Odontogenic Keratocyst (OKCs) of the jaw are developmental cysts arising from cell rests of the dental lamina, the oral epithelial lining of the developing tooth follicle. The Odontogenic Keratocyst (OKC) is a distinct entity from other odontogenic cysts that deserves special attention due to its aggressive clinical behavior and high rate of recurrence. Due to unspecific clinical and radiographic features, it may be confused as ordinary cysts, leading to misdiagnosis and leading to inadequate treatment, resulting in unnecessary recurrences.[1] OKCs can be associated with a high recurrence rate, aggressive growth potential. OKCs have distinctive histologic feature that can distinguish them from other cysts.[3]

The odontogenic keratocyst (OKC) is now designated by the World Health Organization (WHO) as a keratocystic odontogenic tumour (KCOT) because of its neoplastic nature and is defined as "a benign uni or multicystic, intraosseous tumor of

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odontogenic origin, with a characteristic lining of parakeratinized stratified squamus epithelium and potential for aggressive, infiltrative behaviour."[4]

OKC comprise approximately 11% of all cysts of the jaws. They occur most commonly in the mandible especially in the posterior body and ramus regions. They almost always occur within bone, although a small number of cases of peripheral OKC have been reported. Patients may present with swelling, pain and discharge or may be asymptomatic. Distinctive clinical features include a potential for local destruction and a tendency for multiplicity.[1]

OKCs appear as well-defined radiolucencies, which can be either unilocular or multilocular. Unilocular OKCs can be located periapically, simulating periapical cysts. 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 can be indistinguishable from cystic ameloblastomas.[5] This paper tries to bring about confusion that a clinician can encounter when an aggressive lesion mimics an inflammatory periapical lesion.

Case Report

A male patient aged 31 years reported to the department of Oral Medicine and Radiology with a chief complaint of mild intermittent pain since 6 months in relation to 46 region of his lower jaw. Pain was non radiating in nature and was relieved by taking analgesics. Mild paresthesia was associated with of his lower lip on the right side since 3 months. Patients past dental and medical history were non contributory. On general examination, patient was apparently alright. Extra oral examination of right side did not show any gross facial asymmetry and lymphadenopathy.

On intra oral examination, normal complement of teeth were present including erupted third molars and all soft tissues appeared to be normal. Generalized attrition was noticed with anterior deep bite. Intraoral swelling was not seen and the periodontal status was apparently normal. 46, 47, were not responding for vitality test.

An IOPA radiograph was advised which revealed a unilocular scalloped radiolucency at the periapex of 46 and 47. Full extent of the lesion was not visible so an orthopantomogram was taken which showed an extensive, well defined uniloculor, oval shaped radiolucency which was surrounded by a thin sclerotic border with lesion extending from distal root of 43 up to distal root of 47. No resorption of the roots, displacement of the teeth or calcification was present with in the lesion. A Mandibular cross sectional occlusal radiograph was taken which did not reveal any buccal or lingual cortical expansion. Based on the clinical and radiographic examination a radiographic differential diagnosis of Radicular cyst, odontogenic keratocyst, unicystic ameloblastoma were considered.

Further on aspiration a yellow straw coloured fluid was obtained. Due to the confusing nature of the lesion a biopsy was under local anesthesia and under antibiotic coverage. Specimen was subjected for histopathological examination. Biopsy report of the specimen was suggestive of Odontogenic Keratocyst. Later surgical enucleation with curettage was done under local anesthesia and recalled for follow up.

Discussion

An insite into the history of odontogenic cysts goes back to the 19th century, when the odontogenic keratocyst (OKC) was first described in the year 1876 and was named by Phillipsen in 1956.[1] It is one of the most aggressive odontogenic cysts of the oral cavity. In 1963, Pindborg and Hansen described the essential features of this cyst. The most significant characteristic of this cyst is the exceedingly high recurrence rate.[6] This lesion was recently renamed as keratocystic odontogenic tumour (KCOT), because of its neoplastic nature[2] (and has been defined as "A begin uni or multicystic intraosseous tumour of

odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium and potentially aggressive, infiltrative behaviour). The periapical cyst is the most common odontogenic cyst (52.3-70.7 % of all odontogenic cysts) followed by the dentigerous cyst (16.6-21.3% of all odontogenic cysts) and odontogenic keratocyst, or OKC (5.4- 17.4 % of all odontogenic cysts).[5]

Odontogenic keratocyst are generally thought to be derived from either the epithelial remnants of the tooth germ, or the basal cell layer of the surface epithelium. The majority of patients are in the age ranges of 20-29 and 40-59, but cases ranging from 5 to 80 years have been reported,[7] This case of OKC was presented at the age of 31 years.

Patients with keratocysts may complain of pain, swelling, or discharge. Occasionally they experience paresthesia of the lower lip or teeth. Some are unaware of the lesions until they develop pathologic fractures. Other cysts have been discovered fortuitously during dental examination when radiographs were taken. In many instances, patients were remarkably free of symptoms until the cysts reached a large size and involved the maxillary sinus and the entire ascending ramus, including the condylar and coronoid processes. This occurred because the keratocyst tended to extend in the medullary cavity and clinically observable expansion of the bone occurred late.[8]

The mandible is involved far more frequently than the maxilla. The most common location for OKC is the posterior body of the mandible (90% occur posterior to the canines) and ramus (more than 50%) the epicenter will be located superior to the inferior alveolar nerve canal.[9] Similarly in our present case lesion is present posterior to the body of the mandible and epicenter is located superior to the canal. Multiple keratocysts are frequently associated with the bifid-rib basal cell nevus syndrome (Gorlin syndrome).

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Morgan and colleagues categorize surgical treatment methods for KCOT as conservative or aggressive. Conservative treatment is "cyst-oriented" and, thus, includes enucleation, with or without curettage, or marsupialization. Aggressive treatment addresses the "neoplastic nature" of the KCOT and includes peripheral ostectomy, chemical curettage with Carnoy's solution or en bloc resection. Aggressive modalities have generally been recommended for NBCCS cases, large KCOTs and recurrent lesions.[10]

However, irrespective of the modality of treatment, the recurrence rate of OKC is known to be high (from 5%—62.5%). This high recurrence rate has been attributed to the presence of epithelial remnants of satellite cysts in the osseous margin.[11] In 1976, Brannon proposed 3 mechanisms for KCOT recurrence: incomplete removal of the cyst lining, growth of a new KCOT from satellite cysts (or odontogenic rests left behind after surgery) and development of a new KCOT in an adjacent area that is interpreted as a recurrence.[7] For this present case enucleation was done and recalled for follow up.

Conclusion

The clinical and radiographic features of OKCs are not pathognomonic signs and may lead to a difficulty in diagnosis particularly when the lesion is in periradicular areas of nonvital pulps.

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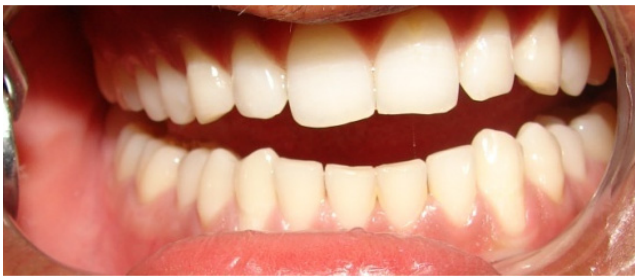

Figure 1: Clinical picture showing no swelling at the periapex of 45, 46, and 47.

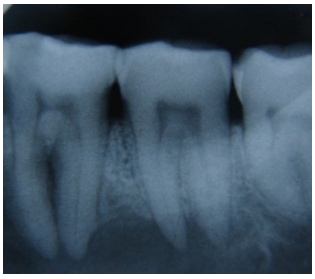

Figure 2: IOPA shows unilocular radiolucency at periapical area of 46, 47, with sclerotic border, Scalloping is seen at the roots of the teeth.

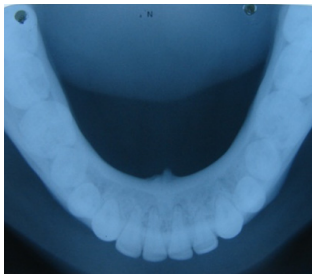

Figure 3: Mandibular cross sectional occlusal radiograph with no expansion in the right Posterior region

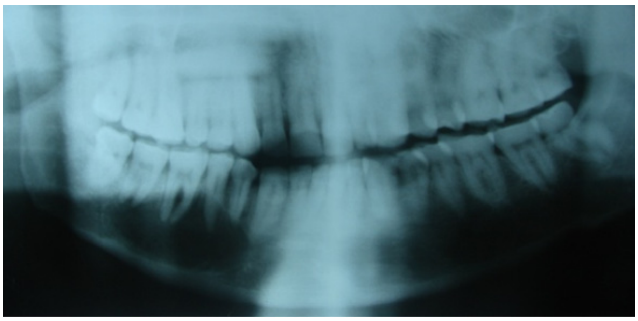

Figure 4: Orthopantomogram showing OKC involving periapical area of 44,45,46,47, tooth, Unilocular radiolucency with well defined sclerotic border, extending posterior 43 up to 48 representing the typical radiographic appearance of OKC.

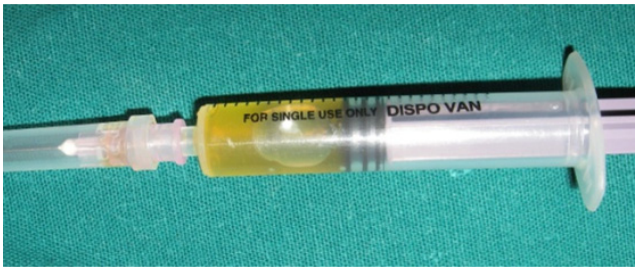

Figure 5: Syringe containing straw colored fluid on aspiration

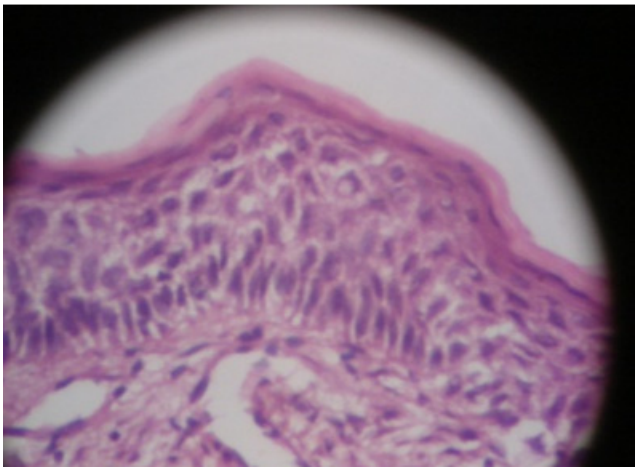

Figure 6: Thick parakeratotic stratified squamous epithelium and basal cells are tall columnar with polarized and palisaded nuclei. Suggestive of OKC.