Case Report

Indian Journal of Dental Education
Volume 10 Number 2, April - June 2017
DOI: https://dx.doi.org/10.21088/ijde.0974.6099.10217.14

Parakeratinized Odontogenic Keratocyst

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Abstract

The odontogenic keratocyst (OKC) is a commonly encountered developmental cyst of considerable importance because of its potential for aggressive clinical behavior and recurrence. Also, it may be a component of the nevoid–basal cell carcinoma (Gorlin) syndrome. Histologic features of OKC are pathognomonic. The common site of involvement is the mandibular ramus region but this case represents odontogenic keratocyst involving the mandibular anterior region. Most odontogenic keratocyst (60%) arise from dental lamina rests or from the basal cells of oral epithelium and are thus primordial-origin odontogenic keratocysts. The remaining 40% arise from the reduced enamel epithelium of the dental follicle and are thus dentigerous-origin odontogenic keratocysts.

Keywords: Corrugated parakeratinized epithelium; Mandibular anterior region; Odontogenic Keratocyst.

Introduction

The odontogenic keratocyst (OKC) was first described in 1876 and named by Phillipsen in 1956. It is one of the most aggressive odontogenic cysts of the oral cavity. OKC is known for its rapid growth 3-5 and its tendency to invade the adjacent tissues including bone.

Odontogenic keratocysts are generally thought to be derived from either the epithelial remnants of the tooth germ, or the basal cell layer of the surface epithelium [1,9]. 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.

It has a high recurrence rate and is associated with the basal cell nevus syndrome [2].

Odontogenic keratocysts may occur in any part of the upper and lower jaw with the majority occurring in the mandible, most commonly in the angle of the mandible and ramus region.

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Received on 10.05.2017, **Accepted on** 27.05.2017

Case Report

A 40 year old male patient reported with a chief complaint of discomfort and mild swelling in his lower front tooth region since 2 weeks, swelling was insidious in onset and gradually increased to the present size. Past dental history revealed , trauma to the lower front teeth region 25 years back and for which root canal treatment was done. No other associated symptoms were observed. Intra oral examination revealed , non tender swelling , which was roughly oval in shape approximately measuring around 3x4.5 cms crossing the midline extending from 35 to 44 causing buccal cortical expansion. There was no signs of bleeding, pus discharge or pain and had normal temperature. The patient was advised for orthopantomogram . The panoramic radiograph revealed a well defined unilocular radiolucency extending from the distal root of 43 to the mesial root of 35 with well defined cortical border.. Aspiration was done which was initially a straw coloured fluid but was not positive.

On the basis of clinical and radiographic findings , a provisional diagnosis of Radicular cyst was given. The patient was adviced for biopsy , for which routine blood investigation was carried out. All the hematological values were in normal limits. Enucleation of the cystic lesion was done under local anesthesia along with chemical curettage was done by applying Carnoy's solution to the enucleated cavity and the tissue was sent for histopathological

examination.

Hematoxylin and Eosin stained tissue section revealed corrugated parakeratinized stratified squamous epithelium of uniform thickness without any retepeg formation. Basal cells shows hyperchromatic nuclei arranged in a palisaded manner.

Connective tissue also shows numerous needle like spaces / Cholesterol clefts and chronic inflammatory cells predominantly of lymphocytes. On the basis of histopathological features, a final diagnosis of Parakeratinized Odontogenic Keratocyst was made.

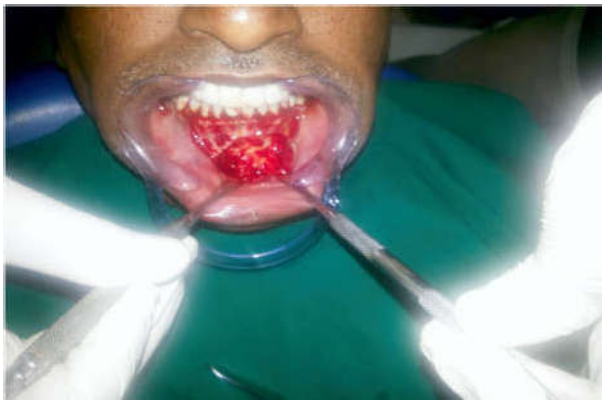

Fig. 1: Flap elevated in lesional area

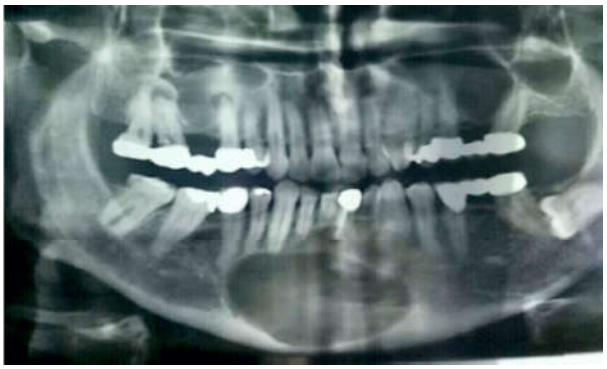

Fig. 2: Well defined Unilocular radiolucency in the mandibular anterior region

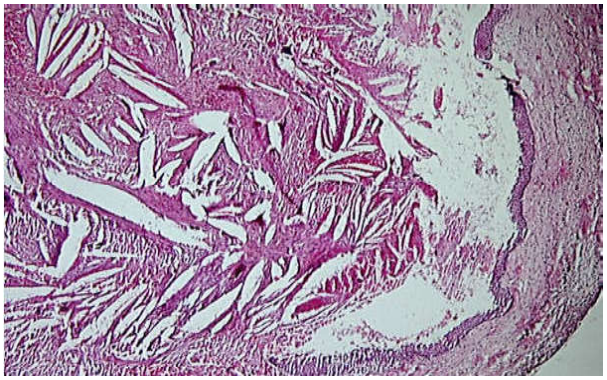

Fig. 3: Hematoxylin and eosin stained tissue section showing numerous cholesterol clefts

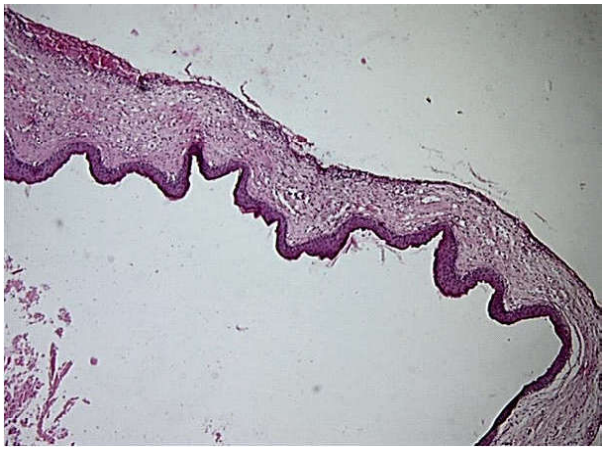

Fig. 4: Hematoxylin and eosin stained section showing corrugated parakeratinized epithelium without any reteridges

Discussion

The odontogenic keratocyst is derived from the remnants of the dental lamina with a biologic behaviour similar to a benign neoplasm. Because of this aggressive nature, recently World health organization used the term "keratocystic odontogenic tumor" to describe this cyst [3].

It is named keratocyst because the cystic lining produces keratin. The cyst occurs in any age group, but most commonly seen in the second and third decades of life with male predilection. The most common features are pain, soft tissue swelling, expansion of bone and parasthesia .

The odontogenic keratocyst may occur due to traumatic implantation or down growth of the basal cell layer of surface epithelium or reduced enamel epithelium of the dental follicle. Nohl and Gulabivala reported two cases of OKCs, and in their first case, tooth associated with OKC had history of trauma twenty years ago [4,5].

The most common site includes the posterior mandible but cases have been reported in anterior region also. Radiographically, most OKCs are unilocular with scalloped margin when presented at the periapex and can be mistaken for radicular or lateral periodontal cyst. When the cyst is multilocular and located at the molar ramus area it may be confused to ameloblastoma.

Multiple OKC's are usually associated with Nevoid basal cell syndrome, Gorlin-Goltz syndrome [6,10].

Recurrence rate ranges from 2.5 to 62% [7], in KCOTs and they occur due to incomplete removal

of the original cyst's lining, thin friable cystic lining, growth of the new OKC from small satellite cyst of odontogenic epithelial cell rests left behind by surgical treatment, or by development of an unrelated OKC in an adjacent region of jaw which is interpreted as a recurrence. Resection despite a recurrence rate of nil is not significantly better at elimination recurrences than enucleation plus Carnoy's solution or marsupialization plus cystectomy [8].

A case reported by Sulabha et al on Keratocystic OdontogenicTumour of mandible crossing the midline in a 11 year child has shown the various treatment modalities for the management of Odontogenic keratocyst [6].

In the present case, massive OKC was seen in mandibular anterior region crossing the midline and this case is different from others in relation to its site and etiology. To conclude, there is a need for further studies to better understand its characteristics for more accurate diagnosis and for the development and adoption of less aggressive therapeutic approaches that are perfectly adequate for each case in order to prevent its recurrence

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