Case Report

Odontogenic Keratocyst - An Interesting Case Presentation

Mahathi N*, Rajesh P**, DeepakV***, Manimaran S****

*Reader, **Professor & HOD, Department of Oral and Maxillofacial Surgery, Chettinad Dental College & Research Institute,

*** Senior lecturer, Department of Oral and Maxillofacial Surgery, Karpaga Vinayaka Institute of Dental Sciences

**** Student, Chettinad Dental College & Research Institute, Chennai, India.

Dr.Mahathi is an Associate Professor working in the department of oral and maxillofacial surgery, Chettinad Dental College. She did her MDS from Sri Ramachandra Dental College and Fellowship in Oral Oncology from Kidwai Memorial Institute of Oncology. Her area of interest is treating patients with oral cancer apart from treating maxillofacial fractures and pathologies.

Corresponding author - Dr. Mahathi N - (nerallamahathi@gmail.com)

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Abstract

An Odontogenic Keratocyst (OKC) first described by Philipsen in 1956, commonly affects body and ascending ramus of mandible with maxillary sites and anterior mandible being very rare. It is a locally aggressive tumor often but not always associated with an impacted tooth. Hereby, we are presenting a case of OKC with an uncommon clinical presentation.

Key Words: OKC, impacted tooth, ascending ramus of mandible.

Introduction

An Odontogenic keratocyst named by Philipsen in 1956 has had its fair share of controversies over the years leading to various disputes in defining it[1]. Some call it as a cystic lesion of odontogenic origin[2,3]. Even though many obscurities circle this enigmatic cyst, it is safe to say that OKC is an epithelial developmental cyst of jaw which takes it origin from Dental Lamina[4,5]. In 1967, various leading studies stated that owing to its aggressive behavior and high recurrence rate1 it is safe to say that OKC has an inherent neoplastic potential when compared with other cystic lesions[6]. WHO also backed up his statement by classifying OKC as a true benign tumor with odontogenic epithelium and mature fibrous stroma without ectomesenchyme[1]. Studies say that, it most commonly arises from the epithelial remnants of dental lamina - Cell Rests of Serrae [5,7]. Another peculiar feature about the OKC is its association with NBCC Syndrome in which multiple sporadic forms are evident most probably due to two-hit hypotheses or haploinsufficiency model [5,6]. When subjected to statistical analysis its incidence is 3-11% among jaw cysts [7,8]. As far as gender is concerned, there is a slight male predilection in the ratio of 1.42 : 11 . 65-83% cases are most commonly located in angle and ascending ramus of mandible [9]. Various studies such as immunohistochemistry, genetics, histopathology and others aid in understanding the aggressive nature and recurrence potential of OKC[4]. Radiographically it has unilocular and/or multilocular radiolucent appearance[4]. On aspiration biopsy it yields white keratin material, which is mostly diagnostic, sometimes purulent or bloody collection can also seen [4,7,8]. Histopathology of OKC reveals 5-8 layer thickness of epithelial cells with fibrous stroma and microcyst formation in connective tissue capsule [1,4]. Treatment options vary from simple enucleation to decompression, marsupialization, chemical / electrical cauterization, peripheral ostectomy and resection. Hereby we are presenting a case with unique clinical presentation[3,7,10].

Case Report

Patient is a 20 year old gentleman, who came to our hospital with the chief complaints of recurrent pain and swelling in the lower front region of the jaw of 6 months duration with a past history of trauma to the chin area with a ball while playing cricket a year back. There were no other associated complaints and past medical history was not significant.

On extra oral examination, a mild diffuse swelling with no definite borders , confined to the chin region was found. On intraoral examination, lower anterior labial sulcus was mildly obliterated with the swelling firm in consistency and interspersed with few soft areas. EPT of teeth from 36 - 46 was done and 31,33,35,36,45 found to be non-vital. Aspiration of the lesion from soft areas yielded bloody fluid.Preliminary investigation of Ortho Pan-Tomogram (OPG) revealed a well defined unilocular radiolucency of about 5×6 cm with sclerotic borders extending approximately from apical surfaces of 36-45 (Fig 1).

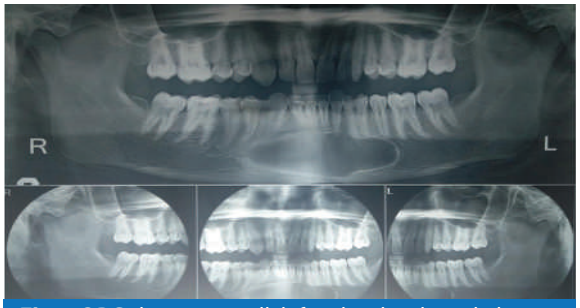

Fig 1: OPG showing a well defined unilocular radiolucency of about 5×6 cm with sclerotic borders extending approximately from apical surfaces of 36-45.

Based on the history and clinical findings a provisional differential diagnosis of Infected periapical cyst, Dentigerous cyst, Hemorrhagic bone cyst and OKC was made. Further CT scan evaluation revealed lingual perforation in the region of 32,33 (Fig 2,3). As blood was aspirated, we planned for excision under General Anesthesia instead of incisional biopsy under Local Anesthesia.

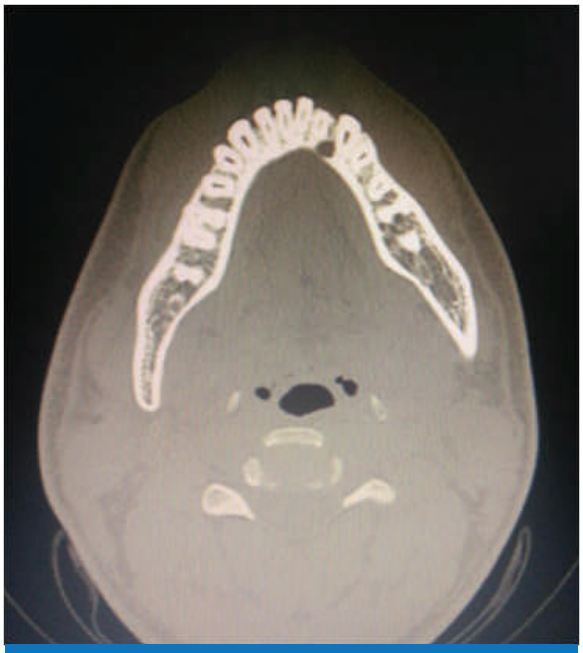

Fig 2: CT axial section showing lingual cortex involvement at the superior surface

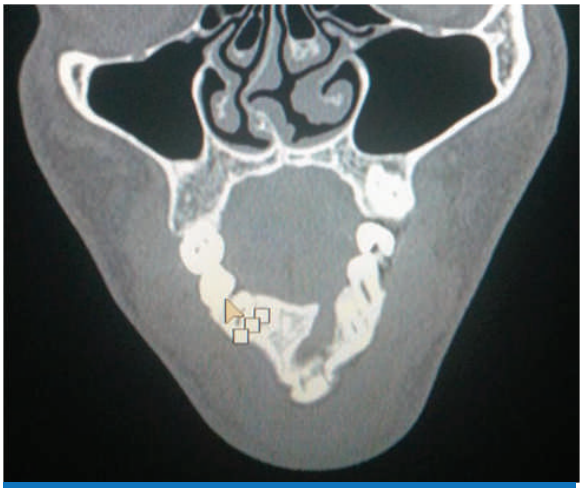

Fig 3: CT coronal section showing lingual cortex involvement

Intra operatively crevicular incision from 36 - 46 was given and envelope mucoperiosteal flap raised. The buccal plate was intact and hence we created a bony window for cyst enucleation wherein copious caeseous material was aspirated confirming the probability of OKC. Hence cyst enucleation in toto followed by peripheral ostectomy and chemical cauterization of the cystic cavity with freshly prepared Carnoy's solution was done (Fig 4). Cavity was closed with betadine soaked ribbon gauze .

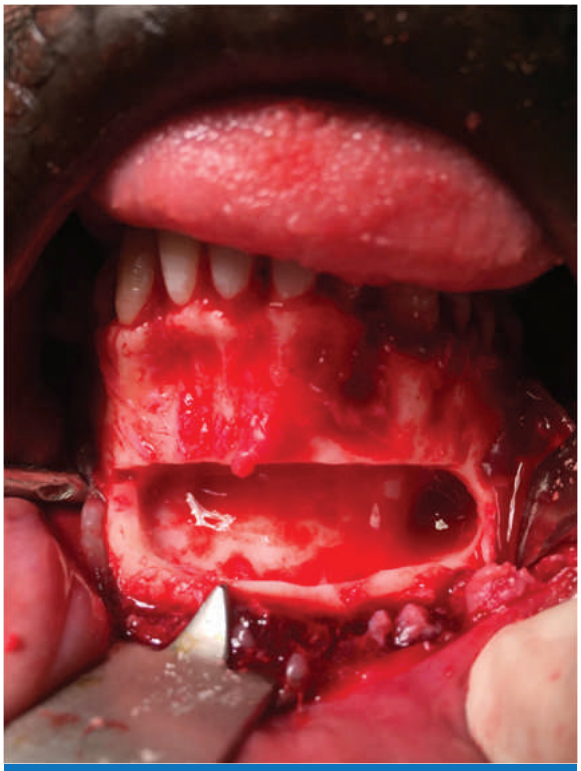

Fig 4: Intra operative photograph after complete cyst enucleation.

Post operatively histopathological evaluation confirmed OKC. On regular visits patient was given interim obturator and counseled for regular follow up.

Discussion

Odontogenic keratocyst usually shows predilection for the posterior mandible - the ramus and angle areas rarely in the anterior region. Confirmatory diagnosis can be framed only after histopathological evaluation, most cases are provisionally diagnosed with the aid of its clinical presentation.

Our case presented an array of peculiarities. A history of trauma, bloody aspirate and a radiographic evidence of unilocular pattern and presence in the anterior region of mandible and association with multiple non-vital tooth drove our diagnosis towards a infected periapical cyst. Peculiarities like these made diagnosis difficult.

On surgical exploration a white cheesy material was encountered suggesting the possibility of an OKC. Treatment shifted towards enucleation of the cyst in to with peripheral ostectomy and chemical cauterization with freshly prepared Carnoy's solution. The specimen collected from the surgical site was sent for histopathologic evaluation and a result showing positive signs of OKC with regard to WHO diagnostic criteria was received .

OKC has a tendency to recur[4]. This is commonly attributed to the presence of microcysts, incomplete removal of cyst lining which contains epithelial remnants[2-5]. Anatomical sites may pose a difficulty to us, in total removal of large cysts as a source of recurrence[4]. Many studies reveal a high chance of recurrence in mandible than in maxilla[7]. And also the

tooth in relation to cyst which are not extracted during surgery may cause recurrence[4,7] .

Another characteristic feature of OKC is its aggressive behavior which may be due to various etiologies like high collagenolytic nature of the basaloid cells, increased mitotic activity, transluminal hyperosmolality, synthesis of IL-1 and IL-6 by keratinocytes, etc.,[1,5,7,9] .Studies have shown that most cases of recurrence are usually seen during the first 5 years of surgery hence long term follow up wise [2,3,8,10]. Peripheral ostectomy and chemical cauterization and removal of overlying mucosa decrease the chances of recurrence following treatment[2].

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Answer to : **A Case of Global ST Depression**

ECG Discussion

ECG Shows Sinus rhythm,Normal axis,rate of 100/min, normal PR interval, widespread ST depression in leads I,II,III,aVF,V3-V6, ST elevation in aVR(3mm), V1(4mm), V2(2mm)

D/D of global ST depression with ST elevation in a VR

- LMCA occlusion (usually ST elevation in aVR > V1)
- Left main equivalent
- Proximal left anterior descending artery (LAD) occlusion (usually ST elevation in V1 > aVR)
- Severe triple-vessel disease
- Diffuse subendocardial ischaemia eg. due to O2 supply/demand mismatch

CAG done Showed proximal LAD 90% lesion before the origin of major septal and proximal LCX 75% before the origin of major obtuse marginal branch without left main disease which is considered to be left main equivalent.

- Dr.G.Ashok, Consultant Cardiologist, CSSH.