

Odontogenic Keratocyst- An Unusual Clinical Case Presentation with Review of Literature

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Abstract: Odontogenic keratocyst (OKC) characterized by a single lesion, with greater frequency in the mandibular jaw. The diagnostic approach is based on a combined analysis of the medical history, the clinical appearance and the radiographic appearance. The diagnosis may be confirmed by the anatomical pathology report. Finally, treatment consists of surgical excision and follow up is characterized by a high rate of recurrence. The authors report an unusual case of OKC of the upper jaw and review the various diagnoses and therapeutics.

Keywords: Radicular cyst, Odontogenic keratocyst, pathological migration

INTRODUCTION

The odontogenic keratocyst (OKC) is a cystic lesion of odontogenic origin, which is classified as a developmental cyst derived from the dental lamina. This lesion was first described in 1956 by Phillipsen [1]. OKC is known for its rapid growth and its tendency to invade the adjacent tissues including bone. Symptoms such as pain, swelling and drainage may be present, especially with larger lesions. However, at least half of all lesions are discovered as incidental radiographic findings [2]. Radiographically, it appears as a well-defined unilocular or multilocular osteolytic lesion. OKCs of the maxilla are smaller in size compared to those of the mandible. When they are large, they tend to expand bone [3]. Due to the propensity of OKCs to grow within the medullary bone, they have the potential to become extremely large without causing any clinical signs or symptoms [4].

This paper presents a case wherein the clinical presence of only bony hard swelling in left region of upper jaw later lead to the diagnosis of odontogenic keratocyst (OKC).

CASE REPORT

A 46 year old female reported to the Department of Oral & maxillofacial surgery, career P.G. institute of dental sciences & Hospital, Lucknow, Uttar Pradesh, India, with chief complaints of swelling in left side of face following trauma to left side of face two months back. She also had paresthesia over left side of face. On examination she had 3 X 1 cm bony hard swelling over left cheek and intraoral examination revealed oral cavity showed fullness in the left gingivo buccal sulcus from canine to second molar teeth [Fig.1]. Her medical history was non contributory. An occlusal radiograph of the area revealed a well defined

circumscribed radiolucent lesion [Fig. 2]. There were also signs of buccal cortical plate expansion. Vitality testing showed that teeth 22,23,24,25 and 26 were non vital but tender to percussion. On the basis of clinical and radiological feature, diagnosis of traumatic bone cyst was made. There was no evidence of mobility of teeth. The differential diagnosis of odontogenic keratocyst was considered and treatment plan made and patient was informed about the procedure. Aspiration of cyst revealed white cheesy material [Fig. 3].



Fig-1: Preoperative view



Fig-2: Pre-operative diagnostic occlusal radiograph



Fig-3: Aspiration of cyst revealed white cheesy material

Treatment

Endodontic treatment of the teeth 22,23,24 25 and 26 was done in single visit. Patient was recalled after 24-hrs for surgical management. On the day of surgery, after anesthesia was achieved by 2.0% lignocaine with 1:100000 epinephrine, a crevicular incision was made on the buccal marginal gingiva extending from the mesial aspect of tooth 22 to the distal aspect of tooth 26. A vertical releasing incision was given extending beyond the mucogingival junction on the mesial aspect of tooth 22. A full thickness mucoperiosteal flap was elevated to gain access to the defect. A well defined lesion, measuring approximately 4 cm × 1.0 cm was found [Fig. 4]. The lesion was enucleated without rupture and sent for histopathologic examination [Fig. 5]. The bony defect was thoroughly debrided until it bled [Fig.6]. The walls were decorticated using a half round bur. The defect was irrigated with Povidone-iodine and normal saline. The defect was filled with bone Graft which contained hydroxyapatite granules [Fig.7]. The flap was replaced and sutured using simple interrupted sutures of Mersilk 3-0, and primary closure was achieved [Fig.8]. The patient was prescribed amoxicillin with potassium clavulanate 625 mg TDS and ibuprofen 400mg BD for 5 days, along with written and oral instructions. Patient

was advised 0.12% chlorhexidine rinse twice a day for 7 days for maintaining oral hygiene. Sutures were removed after one week. Healing was uneventful at 1 and 3 months recall appointment, and results were deemed satisfactory.



Fig-4: Intraoperative view: Approx. 4.0 cm × 1.0 cm lesion seen after raising full thickness flap



Fig-5: Enucleated tissue



Fig-6: Defect after complete debridement



Fig-7: Defect after placement of bone graft (HP)



Fig-8: After repositioning the flap

The histopathologic report revealed uniform epithelial layer with an absence of rete ridges, a corrugated orthokeratinized luminal layer and a prominent basal cell layer. The height of the epithelial cells and the number of nuclei they contained were reduced. A few inflammatory cells were also seen in the lumen [Fig.9]. All the histopathological, radiographic and clinical features guided to the final diagnosis of odontogenic keratocyst.

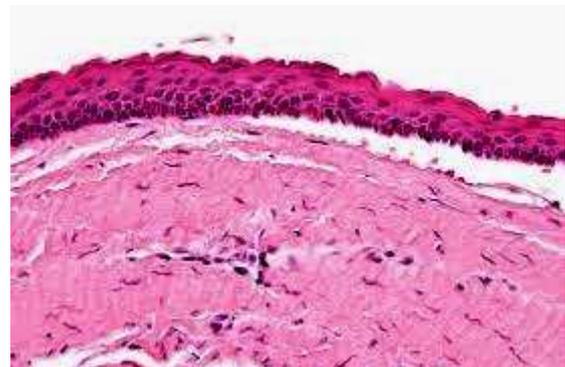


Fig-9: Histopathological view

DISCUSSION

OKCs of maxilla have diagnostic difficulties due to lack of specific clinical and radiographic characteristics. They are less common in maxilla than

mandible with only 31.3% in maxilla. But then they do occur, they are more common in the canine region which was the case in our patient also. Some cases are diagnosed clinically, but others histologically from biopsied specimens [5, 6]. In the adult, the most frequent site is between the mandibular canine and premolar, which is also a site for lateral periodontal cyst. Some cases are diagnosed clinically, but others histologically from biopsied specimens. They may include bone involvement and may give rise to gingival swelling. This can lead to confusion in the diagnosis, as in this case, as an OKC can present as a radicular cyst, lateral periodontal cyst or gingival cyst like lesion.

Radicular cyst is commonest of all odontogenic cysts. Palpation egg shell crackling may be felt in the anterior wall of the maxilla over the canine fossa whereas lateral periodontal cysts are associated with lateral canals within the tooth structure and traumatic Bone Cyst occurs following Bleeding within the bone caused by trauma. Defect in organisation of a blood clot which liquefactively necrotises. The bone is destroyed by enzymatic activity. The bone cavity enlarges by the increased pressure of its content. However, Dentigerous cyst is second commonest always associated with unerupted tooth, Primordial cyst develops in place of tooth, Residual cyst is caused by retained periapical cysts after the teeth is removed and gingival cysts are small cysts located in the alveolar ridges [6-10].

Maxillary OKC tended to exhibit a unilocular, smooth, round border while mandibular ones scalloped border [5]. The occurrence of OKC in maxilla is relatively rare and invasion of the maxillary sinus is unusual. In OKC, the extension is more in anteroposterior direction and the pressure of the fluid is quite low and grows by extension rather than by expansion. The extension here is due to reasons like fingerlike projections from the cyst wall into the marrow spaces, and enlarges slowly. So, not much of cortical expansion is seen in the initial stages and by the time it shows clinical swelling the lesion would have been quite huge [5, 11]. Histopathologically our case showed orthokeratinised lining. A diagnosis of OKC was made [5, 10]. Cawson *et al* [10] have considered OKCs as keratinising cysts and have divided it into para-keratinised and orthokeratinised linings. They have called para-keratinised cysts as odontogenic keratocysts or keratocystic odontogenic tumor (OKC/KCOT). Ortho-keratinised cysts have been called as orthokeratinised odontogenic cysts (OKOC). Philipsen and Riechert have suggested that OKC should be considered as a benign tumour and hence be called as KCOT or keratocystic odontogenic tumour.

Ideal treatment includes curettage with peripheral osteotomy. Curettage alone would suffice if the outer cortex of mandible is intact. If Carnoy solution

is used after curettage these daughter cysts can be destroyed [12]. Carnoy's solution composed of 1 g of ferric chloride [FeCl₃] dissolved in 6 ml of absolute alcohol, 3 ml of chloroform, and 1 ml of glacial acetic acid; the reformulated Carnoy's solution without chloroform is currently generally accepted because exposure to chloroform has been associated with cancer and reproductive toxicity [13]. The solution can be applied inside the cystic lesion or, more commonly, directly over the remaining bone bed after the lesion has been removed.

In cases of large cysts after marsupialization the cavity can be obliterated using bone graft. In cases with very large cysts the entire procedure can be staged. The decompression procedure can be performed under two steps. In the first step the cyst is opened and a drain is placed. Enucleation can be performed at a later date after the cyst has shrunk to a more manageable size. OKCs are considered to be benign cystic neoplasms and require modified surgical procedures, such as curettage of bony walls, peripheral osteotomy with a bone bur or, occasionally, jaw resection [14]. At present there is no consensus regarding the optimal management modality in treating these patients.

CONCLUSION

In conclusion, in any patient with OKCs, a complete clinical examination and histopathologic analysis must be performed. All methods of treatment are satisfactory which primarily close the bone cavity and which at the same time attempt to reduce the empty space and prevent infection of the blood clot. In this case, it was expected that minimal regeneration would take place without the use of regenerative techniques. A bone allograft improved the outcome of the treatment after total surgical removal with debridement and decortication. Finally, regardless of the surgical method used during the treatment of odontogenic keratocysts, we believe that recurrences will occur in all cases during which part of the cystic capsule remains in the cavity, or beside the basic cystic capsule are so called microcystic satellite changes which develop in the immediate vicinity. Therefore follow up for OKC's is once in a year for at least 5 years is recommended.

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