Odontogenic Keratocyst in Maxilla- A Distinctive Conundrum

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ABSTRACT- **Odontogenic keratocyst a benign tumour into a cystic lesion in the year 2017. OKCs represent 2-11% of all odontogenic cysts, affect all ages with a peak from the 2nd–4th decade and have a male to female ratio of 2:1. Benign lesions of the jaws are quite uncommon (17%) of which odontogenic keratocyst holds the most uniqueness due to its specific clinical characteristics, histological features and aggressive biological behaviour. OKC in mandible is quite common than maxilla. Moreover, OKC requires aggressive surgical management as compared to other odontogenic cysts especially in maxilla. Hence it is important to accurately diagnose this lesion. Here we present a case of OKC in maxilla which was provisionally diagnosed as infected cyst, treated by complete enucleation and after histological examination it is diagnosed as Odontogenic keratocyst**.

Keywords: Odontogenic keratocyst, OKC, Infected cyst, Enucleation

1.INTRODUCTION:

The Odontogenic Keratocyst (OKC) is one of the most aggressive odontogenic cysts owing to its high recurrence rate and tendency to invade adjacent tissues. OKC's arise from the remnants of the dental lamina. The World Health Organization (WHO) however reclassified the OKC as Keratocystic odontogenic tumour (KCOT) in 2005.As of 2017,the WHO again switched KCOT back to OKC as there was a lack of support, for their justification as a tumour entity [2].

The term Odontogenic Keratocyst (OKC) was first coined by Philisen in 1956 (Eryilmaz et al., 2009) and its characteristic features was first described by Pindborg and Hansen in 1963 (Pindborg and Hansen, 1963) [5]. OKC makes up to 3–11% of all jaw cysts and primarily affects individuals between 30 and 50 years of age group. Occurrence is more common in the posterior region of the dental arch involving the angle or the ramus of the mandible (66 77%) than in the maxilla [3]. It can occur in isolation or as a multitude of cysts which are linked to syndromes such as Gorlin–Goltz. OKC's may be regarded as a benign neoplasm rather than a conventional cyst [2]. The most customary clinical presentations are The most customary clinical presentations are swelling, discharge, and pain, but in some cases OKC can be asymptomatic and thus the frequency of a casual diagnosis ranged between 5.5% and 42.5%.[6]

2.PATIENT INFORMATION

A 32 year old male patient reported with a complaint of swelling and discomfort in the maxillary vestibular region in relation to 14, 15 for past one month. History of presenting illness revealed patient was apparently normal before one month, following which he developed a swelling which gradually increased to attain its current size. Timeline of history presented in [Fig 4]. Medical history was non-contributory. Personal history revealed no adverse oral habits.

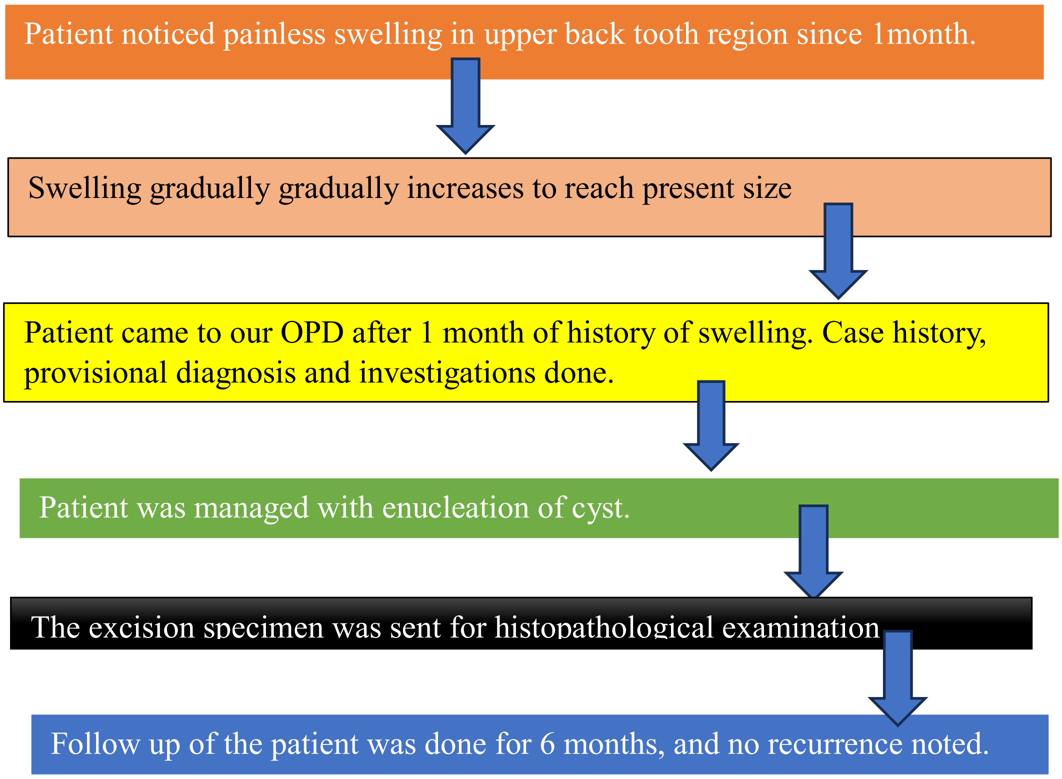

FIGURE 4 : TIMELINE OF HISTORY

3.CLINICAL DESCRIPTION

The patient's general examination revealed no abnormality. Vital signs were normal, moderately built, and nourished. Intraoral examination reveals, a swelling of size 0.5 cm approximately in the buccal vestibular region in relation to 14, 15 which was associated with sinus tract opening [Fig.1] On palpation, the swelling was painful and soft in consistency and it had smooth surface along with pus discharge. The swelling was not associated with parathesia. Pulp vitality test revealed immediate response in relation to 14, 15, 16.

FIGURE 1 - Swelling of size 0.5 cm approximately in the buccal vestibular region in relation to 14, 15 which was associated with sinus tract opening.

4.DIAGNOSTIC ASSESSMENT

Intra oral periapical and panoramic radiograph disclosed a homogeneous periapical radiolucency with ill-defined border extending from 14 to mesial root of 17 which involving the maxillary sinus with interdental bone loss [Fig 2 a and b]. MRI- face and neck revealed the lytic destruction of alveolar and buccal cortex of right maxillary alveolus (around the roots of first and second molars), floor of right maxillary sinus with extension to maxillary sinus cavity. All these findings lead to a provisional diagnosis of infected cyst with draining sinus in the right maxillary region. Differential diagnosis includes chronic periapical abscess, periapical cyst, lateral periodontal cyst and odontogenic keratocyst.

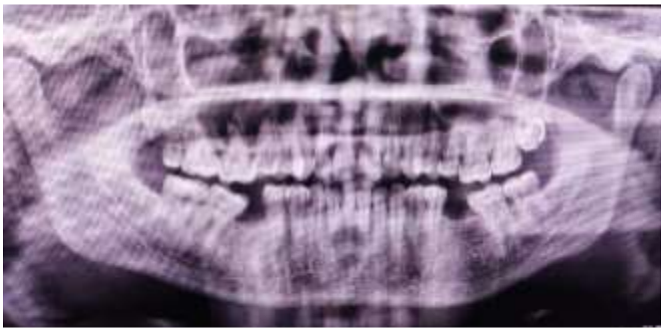

FIGURE 2 a *(Left)*- Intraoral periapical radiograph and **2 b**(*Right*)- Panaromic radiograph-Homogeneous periapical radiolucency with ill-defined border extending from 14 to mesial root of 17 which involving the maxillary sinus with interdental bone loss.

5.THERAPEUTIC INTERVENTION

After all routine preoperative investigations & under aseptic conditions, surgical enucleation of cyst with primary closure was done under local anaesthesia[Fig 3]. Tissue retrieved under aseptic conditions and sent for histopathological examination. Histopathological features revealed a cystic lining epithelium exhibiting corrugated surface parakeratin, uniform thickness and basal cell palisading. The findings were confirmatory for an Odontogenic keratocyst.

FIGURE 3 surgical enucleation of cyst

6. FOLLOW-UP AND OUTCOME

The patient is under follow-up for 6 months, and there is no recurrence reported to date, and the prognosis has been good.

7.DISCUSSION

OKC's arise from the remnants of the dental lamina. The most occurrence site of OKC is the mandible (73%) compared with 27% in the maxilla. Maxillary odontogenic keratocyst is rare and accounts less than 1% of the total OKC cases reported in the literature [1]. It is more common in canine region. It occurs most commonly in second and third decades of life. Multiple OKCs are frequently associated with Nevoid basal cell carcinoma syndrome. It is a benign developmental odontogenic tumour with many distinguishing clinical and histopathological features[2]. OKC may be derived from the dental lamina or its remnants, the primordium of developing tooth germ, or from the basal cell layer of the oral epithelium. It is mostly believed that OKC develops due to cystic degeneration of the stellate reticulum in developing tooth. Etiopathogenesis of OKC involves a high proliferation rate, which shows a significantly greater expression of proliferating cell nuclear antigen, Ki-67, overexpression of (the antiapoptotic protein) Bcl-2, MMP-2, and MMP 9[7]. OKC is believed to grow due to active cellular proliferation and has a destructive growth manner through bone trabeculae in anteroposterior orientation. OKC always expands through cancellous bone, so noticeable bone expansion is seen only in later stages; this distinguishes OKCs from other jaw cysts The rate of growth of keratocysts varies 2–14 mm a year, with an average of 7 mm[7]. It involves a wide range of age groups, with a peak occurrence in the second and fourth decades of life. The white population are commonly involved with a male: female ratio of 1.6:1. OKCs are found twice in the lower jaw than the upper jaw. These cysts usually occur in the angle-ascending ramus area (69%–83%). While the upper jaw OKCs may extend to the maxillary sinus, nasal floor, premaxilla, and maxillary third molar area[7].

Clinically, OKC can easily be misdiagnosed as inflammatory cysts because majority of the cases presents with common inflammatory signs such as pain, swelling and pus discharge. Diagnosis based on clinical information alone will be problematic [2]. According to histopathology, OKC is distinguished by a cystic lumen lined with six to eight uniformly thick layers of corrugated, Parakeratinized stratified squamous epithelium. Tall columnar basal cells with palisading nuclei and polarity reversal are characteristics of lining epithelium [3]. In our present case, a swelling with pain and pus discharge from the sinus tract opening. The tooth involved are vital. Periapical radiograph, OPG and MRI provides more detailed information for evaluation of the borders of the lesion and the relation of the lesion with adjacent anatomical structures. Both clinical and radiological appearance of OKC mimics other pathologies and hence patient had been provisionally diagnosed of infected cyst. Differential diagnosis of this case includes periapical cyst, odontogenic keratocyst, Ameloblatoma and it was treated by enucleation with primary closure. But, histopathology of the specimen confirmed the lesion as OKC. Treatment options for OKC are enucleation with primary closure, enucleation with secondary closure, enucleation with cryotherapy, enucleation with chemical cauterisation using Carnoy's solution and En bloc resection. Brannon stated that the recurrence rate of OKC, which was treated with enucleation alone, was 12 %. Recurrence rate reported for resection is 0-2%. However, resection can have high morbidity. Prognosis of OKC becomes poor when treated with enucleation alone due to its thin friable nature of the cyst lining and presence of daughter cysts. Special attention should be given to the dentate area if the enucleation is chosen as treatment, due to higher rates of recurrence found in OKC associated with teeth [4].

Prognosis of our case is fair and chance of recurrence becomes high as chemical cauterisation was not been performed since the lesion was provisionally diagnosed as infected cyst. Regular follow up with radiographic examination is necessary with OKCs because of the potential for recurrence. OKCs usually recur within five years after surgery but they can recur more than 15 years also[4]. Since our case of OKC was in Maxilla, chance of recurrence is even high. We followed up our case at one month, 3 month, 6 month interval prognosis is good and no recurrence yet.

8.CONCLUSION

OKC is an aggressive lesion with high recurrence rate. There have always been controversies regarding the cystic or the lesion's neoplastic behaviour. The use of fine-needle aspiration and incisional biopsy may help in the early diagnosis of OKCs [7]. It is rare to have OKC in the maxillary region, it typically does not exhibit the same clinical and radiographic characteristics as its other counterparts within the jaw bone. Because of the aggressive nature and high recurrence nature of odontogenic keratocyst, early diagnosis and appropriate treatment is essential to prevent complications and recurrence [8].

9.DECLARATION OF PATIENT CONSENT:

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient (s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

10. FINANCIAL SUPPORT AND SPONSORSHIP

Nil.

11. CONFLICTS OF INTEREST

There are no conflicts of interest.

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