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Case Study

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AN UNUSUAL PRESENTATION OF ODONTOGENIC KERATOCYST IN MAXILLA

Dr. Joseph Johny[1]*, Dr. Sudeep C. B.[2], Dr. Nikhil Chandran[3] and Dr. Sufain S.[4]

[1]Associate Professor, Dept. of Oral Medicine, Sree Anjaneya Institute of Dental Sciences, Kozhikode, Kerala, India.
[2]Associate Professor & Head, Dept. Public Health Dentistry, Sree Anjaneya Institute of Dental Sciences, Kozhikode, Kerala, India.
[3]Assistant Professor, Department of Pedodontics, Sree Anjaneya Institute of Dental Sciences, Kozhikode, Kerala, India.
[4]Former Intern, Sree Anjaneya Institute of Dental Sciences, Kozhikode, Kerala, India.

***Corresponding Author: Dr. Joseph Johny**

Associate Professor, Dept. of Oral Medicine, Sree Anjaneya Institute of Dental Sciences, Kozhikode, Kerala, India.

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ABSTRACT

The odontogenic keratocyst (OKC) is an odontogenic developmental cyst of oral and maxillofacial region which has gained very special attention since last two decades due to its dilemmatic nature. It has specific histopathological and clinical findings but still the cyst is very special due to its aggressive behavior and high recurrence rate. Commonest site is mandible (66.8%), however different regions of maxilla can be involved. We report a rare case of an OKC involving maxilla.

KEYWORDS: Odontogenic keratocyst (OKC), Keratocystic odontogenic tumor (KCOT), Maxilla, Dentistry, Oral diseases.

INTRODUCTION

Odontogenic keratocyst (OKC) is known for their characteristic behavior, varied origin, debated development, unique tendency to recur, and disputed treatment modalities. The word Odontogenic keratocyst (OKC) was coined by Phillipsen[1] in 1956. Pinborg and Hansen[2] suggested the histopathological importance for the diagnosis of OKC in 1962. WHO proposed the terminology as keratocystic odontogenic tumor (KCOT) as it shows its neoplastic nature. But recently it has been reclassified back into the cystic category. It was initially called as primordial cyst as tooth primordium was thought to be the origin of the lesion. The origin of OKC comes from dental lamina remnants in maxilla and mandible[3,4] or from basal cells of oral epithelium overlying it.[5,6] Odontogenic keratocyst (OKC) is so named because keratin is produced by the cystic lining. It is a cyst-like lesion with parakeratin lining formed within bone. OKC is the one of the rare and distinctive developmental odontogenic cyst which form from the dental lamina, containing clear fluid and a cheesy material similar to keratin debris. Epidemiologically OKC accounts for approx. 7.8% of all cysts of the jaw and incidence vary from 4-16.5%. It occurs at all ages with peak incidence in 2nd and 4th decade of life. It predominantly occurs in white population with male: female ratio of 1.6:1. The most common site is mandibular ramus and angle region, maxilla is an uncommon site with the lesion crossing the midline. Patients with KCOTs complain of pain, bone expansion, drainage and neurological manifestations such as paresthesia of the lip. The enlarging lesions may lead to displacement of the adjacent teeth. The radiographic appearance is a unilocular or a multilocular radiolucency with scalloped margins.

CASE REPORT

A 54year old male patient reported to the Department of Oral Medicine & Radiology with a chief complaint of pain in the upper right back region of the jaw for the past 1 week. The pain was gradual in onset, dull throbbing type, diffuse in nature, intermittent in character, aggravates on lying down & is associated with a swelling. The swelling was noticed 2years ago which was small in size & gradually increased to the present size. It was not continuous in nature but with periods of remissions & exacerbations. He gives a medical history of diabetes & is under medication for the past 5 years & in his personal history, he stopped smoking cigarettes 20 years before. On extra oral examination, a diffuse swelling is seen in the right malar region 1 cm below the infra-orbital margin to the ala tragus line and anterioposteriorly from the ala of the nose to the line dropped from the outer canthus of the eyes [Figure 1] with no apparent color change of the surrounding skin & it was hard in consistency with mild tenderness and parasthesia. On intra oral examination, [Figure 2] a swelling is seen in the right side of the palate roughly oval in shape measuring 3x1.5cm extending anteroposteriorly from the mesial aspect of 11 to mesial aspect of 16, crossing the midline with well defined margins and no apparent color change in the surrounding

mucosa & on palpation, it was slightly tender and soft in consistency. Based on the history & clinical examination, the lesion was provisionally diagnosed as Dentoalveolar abscess. The patient was subjected for chair side investigations. On electrical pulp tester, 11, 12, 13, 14, 15 & 16 were non vital. Then, fine needle aspiration cytology was done which showed a white creamy fluid with keratin sediments [Figure 3]. General physical examination did not reveal any syndromic features. Correlating, with the history, clinical findings and chair side investigations, this case was differentially diagnosed as odontogenic keratocyst, ameloblastoma and infected radicular cyst. Patient was subjected for further investigations blood hematology & biochemistry, which were under normal limits. Then the patient was subjected for various radiographic imaging like intraoral periapical radiograph, maxillary occlusal view & CT scan. Intraoral periapical radiograph [Figures 4 & 5] showed well defined multilocular radiolucency with scalloped margins extending from 11, 12, 13 region to 16 region and maxillary occlusal view crossectional [Figure 6] shows mediolateral extension crossing the midline. CT scan [Figure 7] showed soft tissue mass in the right hard palate with destruction of hard palate more than 1 cm, destruction of pterygoid plates, destruction of lateral wall of nose and floor of maxillary sinus. As a part of management, cyst enucleation with extraction of 11, 12, 13, 14, 15 & 16 was done followed by prosthetic rehabilitation. The removed soft tissue section was sent for histopathological evaluation, which showed [Figure 8] lumen filled with straw colored fluid with cyst lining epithelium with 3-4 cell layer thickness with palisaded basal cell layer and corrugated surface parakeratin. The underlying connective tissue is fibrous with diffuse chronic inflammatory cell infiltrate, bands of cyst lining epithelium and daughter cyst. Correlating the clinical features, radiographic & histopathological findings, a final diagnosis of ODONTOGENIC KERATOCYST of right hard palate was made. The patient was followed up for 6 years & showed no recurrence.

CLINICAL PHOTOS

Figure 1: Extra oral view shows a diffuse swelling in the right malar region below the infraorbital margin.

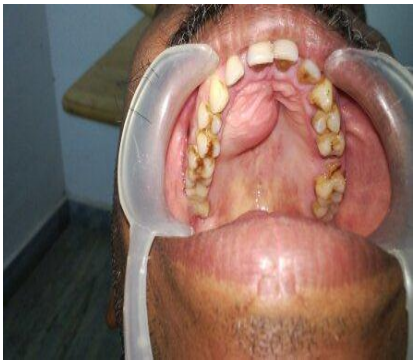


Figure 2: Intra oral view shows swelling in the right side of hard palate roughly oval in shape extending from 11 to 16 crossing the midline with well defined margins.

INVESTIGATIONS

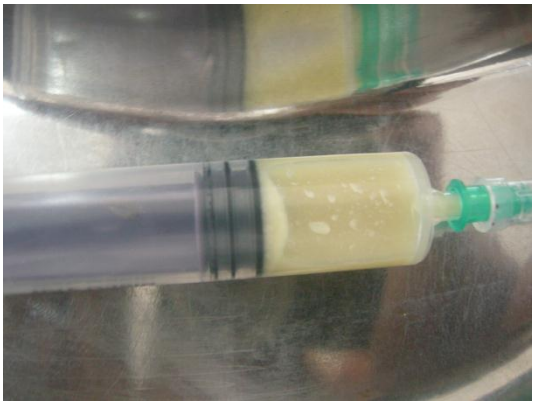
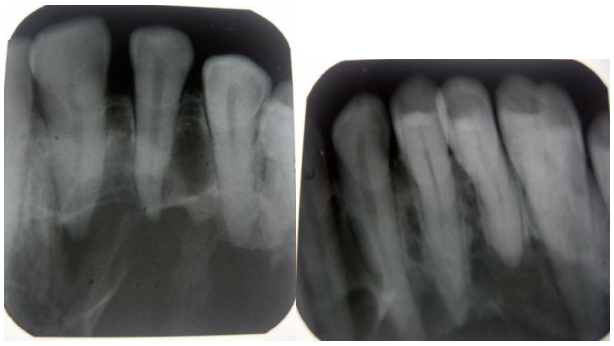


Figure 3: FNAC showed a white creamy fluid with keratin sediments.



Figures 4 & 5: IOPA showed a well defined multilocular radiolucency with scalloped margins extending from 11, 12, 13 region to 16 region.

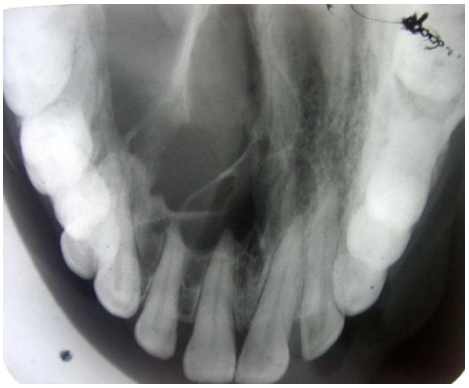


Figure 6: Maxillary occlusal view showed mediolateral extension crossing the midline.

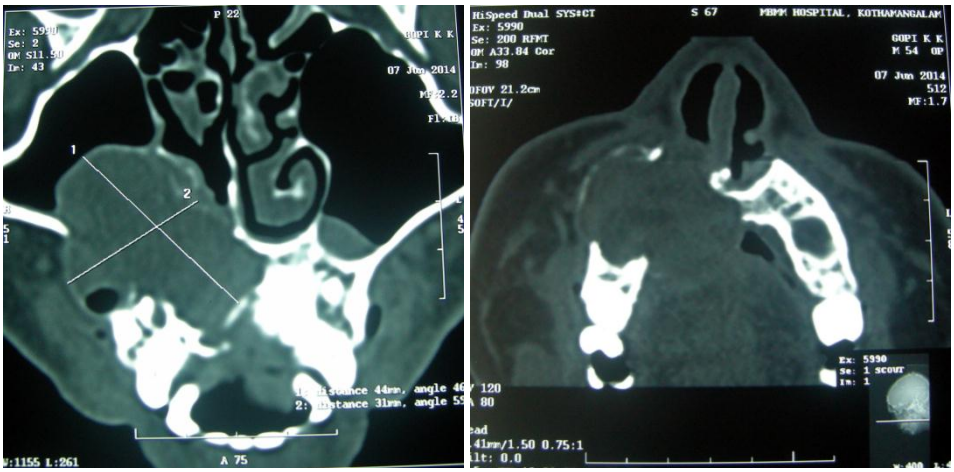


Figure 7: CT scan showed soft tissue mass in the right hard palate with destruction of hard palate, pterygoid plates, lateral wall of nose and floor of maxillary sinus.

HISTOPATHOLOGY

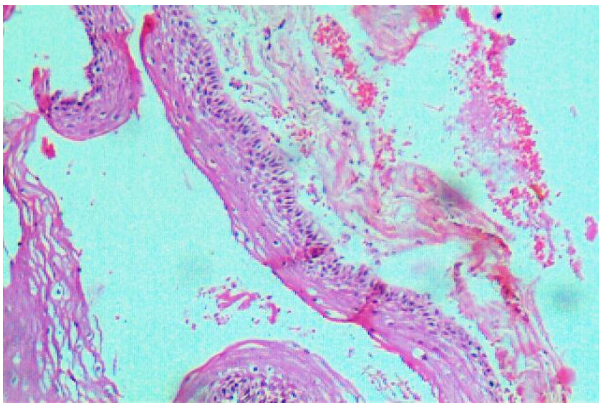
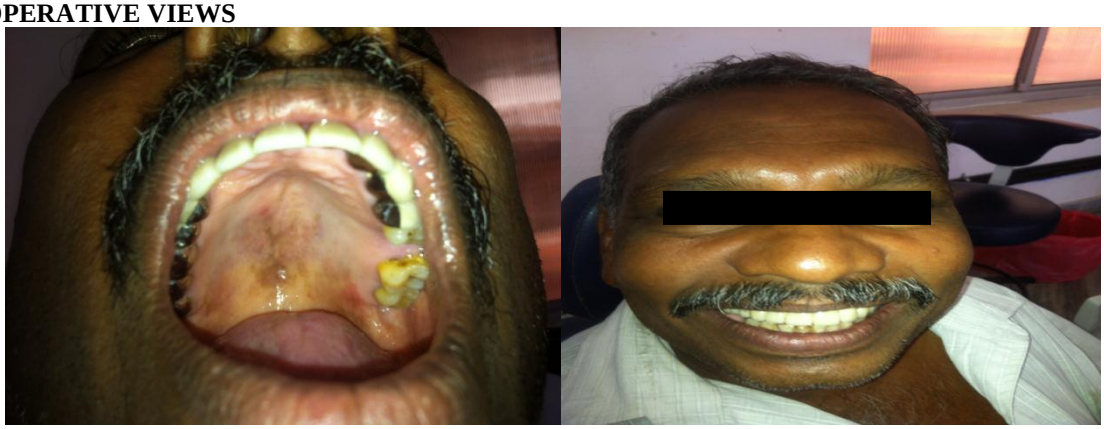


Figure 8: Histopathology showed lumen filled with straw colored fluid with cyst lining epithelium with 3-4 cell layer thickness with palisaded basal cell layer and corrugated surface parakeratin. The underlying connective tissue is fibrous with diffuse chronic inflammatory cell infiltrate, bands of cyst lining epithelium and daughter cyst.

POSTOPERATIVE VIEWS



INTRAORAL **EXTRAORAL**

DISCUSSION

The development of KCOT comes from remnants of dental lamina or from basal cell layer of oral mucosal epithelium. It accounts for approx. 7.8% of all cysts of the jaw and incidence vary from 4-16.5%. The most characteristic clinical feature of OKC is the high recurrence[9] rate (0%-60%), the parakeratinized have higher recurrence rate as compared to the orthokeratinized type. Location wise it is most commonly seen twice in mandible as compared to maxilla[7] with the predilection for angle-ascending ramus region (69-83%), but in our case it is present in right maxilla crossing midline. It occurs more frequently in males, the male: female ratio was 1.27:1 but in present case, KCOT was in male patient which is consistent with the review. It occurs at all ages with peak incidence in 2nd and 4th decade of life, but in present case the patient's age was 56years which is consistent with the review. OKC is mostly intraosseous lesion though peripheral counterpart also have been reported but our case was an intraosseous lesion. Clinically patients with KCOTs complain of swelling with or without pain, discharge, displacement of teeth, ocassionaly paresthesia of lower lip. The expansion of the cyst is very minimal in the initial stage and it is due to the classical characteristic of the cyst to grow in anteroposterior direction in the medullary space of the bone. Expansion of buccal cortex in 30% of maxillary and 50% of mandibular regions. In present case patient had pain which was gradual in onset, dull throbbing type, intermittent in character, diffuse in nature & he also had diffused swelling. The expansion of buccal & lingual cortical plates were present but neurological symptoms were not present in our case. CT scan showed the present case was an expansile, osteolytic & destructive bony lesion with destruction involving pterygoid plates, lateral wall of nose & floor of maxillary sinus. Distinctive clinical features include a tendency for multiplicity when it is associated with syndromes like Nevoid Basal cell carcinoma syndrome (NBCCS), Gorlingoltz syndrome, Marfans syndrome, Ehlers danlos syndrome, Noonans syndrome, Orofacial digital syndrome, Simpsongolabi-behmel syndrome. But in present case syndromic features were not presnet. Radiographically OKC presents as well defined unilocular or multilocular (25-40%) radiolucent lesion with smooth margin (corticated margin in secondarily infected cases), enlarging lesion may lead to displacement of adjacent teeth without root resorption. In the present case, a well defined multilocular radiolucency extending from 11, 12, 13 region to 16 region & crossing the midline was seen radiographically, which is consistent with the review. The differential diagnosis of Odontogenic keratocyst, Ameloblastoma & Infected radicular cyst was given. Ameloblastoma presents as a slow-growing, painless lesion which leads to migration and loosening of teeth with expansion of the cortical plates and paresthesia of the lip. It is most common in mandible than maxilla, five times in mandible posterior region. It occurs mostly in second to fifth decade of life. In cystic type of lesion, the aspirate could be negative or straw-colored fluid may be present. In present case, multilocular radiolucency was seen which is consistent with ameloblastoma but the site of radiolucency was right hard palate crossing midline with white creamy fluid with keratin sediments on aspiration which is rare and not consistent with ameloblastoma. Radicular cyst is the inflammatory odontogenic cyst associated with non-vital teeth. Expansion of the cortical plates is seen with bony hard swelling but later it produces crackling sound. The aspiration shows straw-colored fluid with cholesterol crystals. In present case, 11, 12, 13, 14, 15 & 16 were non vital but cholesterol crystals were not observed on aspiration, which is not consistent with radicular cyst. KCOTs are lined by stratified squamous epithelium which may be parakeratinized (80-90%) or orthokeratinized. The epithelium is corrugated, with a regular thickness of about 5-8 layer with lack of rete ridge formation, well defined often palisaded, hyperchromatic, basal layer consisting of columnar or cuboidal cells or mixture of both the cells superficial to basal layer are polyhedral and often exhibit intercellular oedema. KCOTs have a high recurrence rate of 25% to 60%. It is believed that the parakeratinized variant (previously called primordial cyst) is thought to have a higher recurrence rate than the orthokeratinized variant. But in present case, lumen was filled with straw coloured fluid with cyst lining epithelium with 3-4 cell layer thickness with palisaded

basal cell layer and corrugated surface parakeratin. The underlying connective tissue is fibrous with diffuse chronic inflammatory cell infiltrate, bands of cyst lining epithelium and daughter cyst. An unusual site of the tumour can mislead to the diagnosis and management. Treatments are usually classified as conservative like Enucleation with or without curettage and marsupialization and aggressive like peripheral ostectomy and chemical curettage with Carnoy's solution, cryotherapy, or electrocautery and resection. In present case, cyst enucleation was done with extraction of 11, 12, 13, 14, 15 & 16 followed by prosthetic rehabilitation. The goals of treatment should involve eliminating the potential for recurrence while also minimizing the surgical morbidity. Right maxilla is an uncommon site, with the lesion crossing the midline being a unique occurrence.

CONCLUSION

The nature of Odontogenic Keratocyst (OKC) either cystic or tumor, has been a matter of discussion since decades and few researchers classified the OKC as a benign tumor. Odontogenic keratocyst is renamed as keratocystic odontogenic tumor (KCOT) and is defined by World Health Organization (WHO) as benign unicystic or multicystic intraosseous tumor of odontogenic origin, aggressive in nature and lined by parakeratinized stratified squamous epithelium. The maxillary OKC is a benign lesion, which commonly involves the canine region, symptomatically may resemble with apical inflammatory lesions or periodontal cysts and maxillary sinus lesions such as sinusitis and antral polyps. So the accurate histological diagnosis is necessary for proper treatment and most importantly, follow ups for future recurrences and malignant transformation to squamous cell carcinoma. The treatment outcome of OKC depends upon age, radiographic and clinical extent of lesion, unilocular or multilocular appearance, presence of daughter cysts, recurrence rate. The aim should be to prevent recurrence, restore cosmetic and functional restoration of the patient. The recurrence rate of OKC is very higher than any other cysts of the jaws. Postoperative follow-up with clinical and radiological examination is necessary for at least 5 to 10 years.

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