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Maxillary Odontogenic Keratocyst : A Rare Presentation In A Paediatric Patient

Dr.Amritha.V.S, Dr. Kannan Vadakkepurayil,
Dr.T.V.Anupam Kumar

(Junior Resident,Department Of Pedodontics,Govt. Dental College ,Kozhikode,Kerala,India.)
(Professor and Head ,Department of Pedodontics,Govt. Dental College ,Kozhikode, Kerala, India.)
(Additional Professor,Department of Pedodontics,Govt. Dental College ,Kozhikode, Kerala, India.)
**Corresponding author: Dr.Amritha.V.S*

Abstract: *Among jaw cysts, OKC accounts for third most common jaw cysts after periapical and dentigerous cysts. Most common location of OKC is mandible and is mostly found in second to fourth decade of life. Occurence of OKC in maxilla is rare. If it occurs ,it is commonly seen in older age group.OKC is known for its aggressive and infiltrative behaviour and its extreme reccurence rate. This case report describes the occurrence of this lesion in maxilla especially in a paediatric patient.*

Keywords*: OKC, maxilla, Young patient*

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I. Introduction

Due to the presence of odontogenic epithelial remnants ,jaw cysts are very common .Among the cystic lesions ,periapical cyst is the most common followed by dentigerous cyst and odontogenic keratocyst.[1] Odontogenic keratocyst (OKC) was first described in 1956 by Philipsen [2].It is now designated by World Health Organisation (WHO) as Keratocystic Odontogenic Tumour(KCOT) which reflects its neoplastic nature .[3] [4] OKC is defined by WHO as "benign uni-or multicystic ,intrasseous tumour of odontogenic origin,with a characteristic lining of parakeratinized stratified squamous epithelium and potential for aggressive ,infiltrative behaviour. [3] OKC is a developmental odontogenic cyst with distinctive histopathological characteristics.[5]OKC is known for its aggressive behavior and tendency to invade adjacent tissues .It is also known for its high rates of recurrence of 0 % -62%[6].OKC arises from the cell rests of dental lamina.[7]Multiple OKC s are seen in patients with nevoid basal cell carcinoma syndrome. Most common site of occurrence is posterior mandible .[6] There are inconsistencies in prominent location of OKC in maxilla.OKC is commonly seen in second and third decades of life. Occurence of OKC in maxilla especially in paediatric age group is extremely rare. Very few studies have reported OKCs crossing maxillary midline. If do occur they are in older individuals. [5]

II. Case Report

A 7-year-old boy reported to the Department of Pedodontics with the complaint of swelling on upper left side of the face since two months. Patient gave the history of mild continuous non radiating pain in the upper left primary molar region since two months . Pain was aggravated by chewing and relieved by medications. On extra oral examination a diffused swelling was evident extending from left ala of nose to left zygomatic prominence mediolaterally. Intraorally, swelling was seen extending from 62 till the maxillary tuberosity causing obliteration of buccal vestibule and extending till the midpalatine raphe medially. Erupted 25 was seen which showed Grade III mobility. Arrested caries of 51,53,54,55,61,62,64 was noted. On palpation the swelling was firm in consistency. No signs of any pus discharge or exudates was seen.

Panoramic radiograph showed a unilocular radiolucency extending from root apex of upper left lateral incisor(22) to maxillary tuberosity causing displacement of tooth bud of maxillary left second molar(26) .There was resorption of roots of premolars and first molar. Surgical enucleation of the lesion was done under General Anasthesia and chemical cauterization was done with Carnoy's solution .Histopathological report showed stratified squamous epithelial lining, typical 'tomb stone' pattern of basal epithelium, numerous hyaline bodies surrounded by lymphocytes, plasma cells and macrophages which were densely dispersed in stroma along with satellite cysts and hemorrhagic areas. These findings were suggestive of odontogenic keratocyst.

III. Discussion

Odontogenic keratocyst is the third most common among jaw tumours , first two being periapical cyst and dentigerous cyst. [5] OKC are derived from the epithelial remnants of dental lamina or the basal cell layer of the surface epithelium.It is a developmental odontogenic cyst with similar characteristics as a benign neoplasm.[7]Therefore the WHO has termed it as Keratcystic Odontgenic Tumour(KCOT) indicating its aggressive nature. OKC is most commonly seen in second and third decades of life with a slight male prediliction. Mandible is more commonly involved (60 to 80%) than maxilla with a mandible to maxillary ratio of 2:1.[1]The most common site of occurence is posterior body and ascending ramus of mandible. But there are inconsistencies regarding the predominant location of OKC in maxilla. In a study conducted by Mohammed Ali et al in 2003 the canine region was the most common site of maxillary OKC. But there is a general disagreement in the most common location of OKC in maxilla. Payne reported that there is equal rate of occurence of OKC in anterior maxilla and third molar tuberosity area. Panders and Hadders found that there were more OKC in anterior maxilla .Brannn ,Browne Myung and collegues said that posterior region of maxilla is the predominant site. Neville et al studied 18 cases of OKC of maxillary midline concluded that OKC should be included in the differential diagnosis of maxillary midline radiolucencies especially in older individuals.In this case report we have seen the occurrence of OKC in a very young child in maxillary posterior region extending till the tuberosity. Approximately 20 to 40% of OKC are associated with an impacted /unerupted tooth.[8] Larger OKC may show pain swelling or discharge whereas small OKCs are often asymptomatic.

Multiple OKC is often seen in nevoid basal cell carcinoma syndrome(NBCCS),orofacial digital syndrome,Ehler Danlos syndrome ,Noonan syndrome etc .NBCCS is characterized by multiple basal cell carcinoma ,bifid ribs ,frontal bossing ,calcification of falx cerebri,multiple epidermid cyst etc.[5]Radiologically OKC present as well defined radiolucent lesions with corticated margins and can be unilocular or multilocular. The diagnosis of OKC is made based on histopathological feactures.[7]

Histopathologically OKC are classified as orthokeratinized and parakeratinized .Parakeratinized was associated with a high rate of recurrence.[9] Histopathological report often shows stratified squamous epithelial lining with typical 'tomb stone'pattern of basal epithelium,numerous hyaline bodies surrounded by lymphocytes , macrophages and plasma cells densely dispersed in stroma along with satellite cells.[5]

OKCs are having a high rate of recurrence .The recurrence occurs within 5-7 years after treatment and some have reported recurrence after 10 years .Hence long term follow up is essential in case of OKC. The presence of daughter cysts ,thin friable cystic lining ,incomplete removal of remnants of dental lamina are said to be the cause of high recurrence rates .The percentage of carcinomatous transformation in odontogenic cysts is as low as 0.01% t 0.02%[6]

The treatment of OKC remains controversial. Treatment can be either conservative or aggressive. Conservative can be simple enucleation with or without curettage. Aggressive treatment includes peripheral ostectomy , chemical curettage with carnoys solution and resection.[10]

IV. Conclusion

Occurrence of OKC in maxilla is a rare occurence, especially in a very young patient. Eventhough OKC has a wide age range of occurrence ,lesions in maxilla is extremely rare in paediatric age group. Accurate diagnosis of OKC is extremely important due to its aggressive nature and high rate of reccurence.Only histopathologic evaluation can provide an accurate diagnosis.

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FIGURES

FIG 1: INTRAORAL VIEW OF MAXILLA

FIG:2:LESION CAUSING OBLITERATION OF BUCCAL VESTIBULE

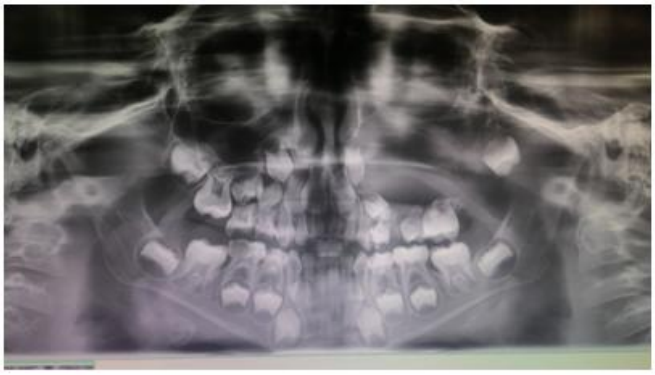

FIG 3: PANORAMIC VIEW SHOWING LESION ON LEFT SIDE OF MAXILLA

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