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CASE REPORT

CLINICALLY MIMICKING DENTIGEROUS CYST IN MAXILLARY LEFT CUSPID REGION DIAGNOSED AS MURAL UNICYSTIC PLEXIFORM AMELOBLASTOMA IN A 17 YEAR OLD FEMALE: A RARE VARIANT - A CASE REPORT.

Shyamalendu Laskar [1], **Prasanta Majumder** [2*], **Anshika Suman** [3], **Manisha Koul** [4]

1. Assistant professor , Department of Oral and Maxillofacial surgery, Index Institute of Dental Sciences , Madhya Pradesh, India.
2. Post graduate student, Department of Public Health Dentistry, Coorg Institute of Dental Science, Karnataka, India.
3. Post graduate student, Department of Periodontology and implantology, Institute of Dental Sciences , Uttar pradesh, India.
4. Masters in Public Health Dentistry, Rajiv Gandhi University of Health Science, karntaka, India.

Corresponding author:
2* Dr Prasanta Majumder
Coorg Institute of dental science
Virajpet, Karnataka, India
Pin-571218
Email Id – drprasantamajumder94@gmail.com

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ABSTRACT

Ameloblastoma is a benign odontogenic tumor occuring in the jaw bones. It develops from the residual epithelium of the tooth germ, epithelium of odontogenic cysts stratified squamous epithelium and epithelium of the enamel organ. About 80% of ameloblastomas occur in the mandible mainly the molar - ramus region and the rest 20% in the maxilla. Ameloblastoma clinically appears as an aggressive odontogenic tumor, often asymptomatic and slow-growing, with no evidence of swelling which affects the treatment outcome. Since ameloblastoma is also associated with a high rate of recurrence when treated inadequately, might results in aggressive recurrence. The rate of recurrence accounted for 60.3% of all odontogenic tumors in Indian population which is significantly high. This case report aimed to add to the existing literature a variation of a clinically appearing dentigerous cyst in a 17 year old female in maxillary.

Keywords: Dentigerous cyst , Ameloblastoma, maxillary, odontogenic tumour

INTRODUCTION

Odontogenic cysts and tumors are common pathologic lesions occurring in the jaws and soft tissues of oral cavity. Maxillary swellings can occur due to varied etiology but other than odontomas , ameloblastomas is the most common lession which comprise between 10% and 50% of all odontogenic tumours [1,3].

Ameloblastoma usually painless, grows slowly, but it is locally invasive . Although origin of ameloblastoma is not clear with certainty, but it was suggested that it is the result of alterations of gene. Robinson defined it as " usually unicentric, non-functional, intermittent in nature. World Health Organization classified subtypes of ameloblastoma as : Classic Solid/ Multicystic Ameloblastoma (SMA), Unicystic Ameloblastoma (UA), Peripheral Ameloblastoma (PA), Desmoplastic Ameloblastoma (DA), including so-called hybrid lesions of ameloblastoma (HLA). Most commonly seen in the age between 30 and 40 years, and the majority of cases occur in the 30 to 60 years age group. The prevalence is equal in both gender and mostly it affects the mandible (80-85% of cases) [4,5,6,7].

In conventional radiograph, ameloblastoma usually appears as unilocular or multilocular corticated radiolucency. Conventional radiograph is sufficient for small lesions but for extensive lesions CT and MRI is mandatory to evaluate the extent of the lesion. Depending on size ameloblastomas management options varies from enucleation, marsupialization, resection- segmental and marginal, cryosurgery, electrocautery, sclerotherapy, and radiotherapy. Acccording to literatue the rate of recurrence are more for the conservative treatment therapy[8].

This case report aimed to add to the existing literature a variation of a clinically appearing dentigerous cyst in a 17 year old female in maxillary cuspid region as a case of Mural Unicystic ameloblastoma diagnosed histopathologically, which proves that histopathology is the gold standard compare to

various imaging modalities such as CT and CBCT.

CASE PRESENTATION

A 17-year-old female reported to our department of oral medicine and radiology from where she was refferred to the department of Oral and Maxillofacial surgery department for needful management. Patient chief complaint was of swelling and mild pain on upper left side of her face since 2-3 months.

History revealed that the patient was alright 3 months back when she noticed swelling on left upper side of her face and tenderness on the left maxilliary posterior tooth region. Patient then visited a local dentist who gave her medications as a result the swelling and tenderness subsided but patient again started noticing swelling after one month. Patient then reported to an another doctor who advised PNS X- ray for further evaluation. The PNS X-ray revealed the presence of a cystic lesion in the upper left cuspid region and also advised the need for surgery. Patient then reported to our institute for needful treatment.

No significant past/present medical and family history. On general examination found that patient was moderately built and moderately nourished, no signs of pallor, icterus, cyanosis, clubbing, lymphadenopathy, all vitals were also in normal limits.

Extra oral inspection revealed a presence of a diffused swelling on the upper left side of the face extending anterioposteriorly from ala of the nose to malar region and superioinferiorly from malar region to the corner of the mouth, overlying skin is normal in appearance. On palpation tenderness was present on the left side of the face and swelling was firm and hard in consistency.

Intra oral inspection revealed presence of a diffuse swelling on the left upper quadrant extending from distal of 22 to distal of 27, buccal vestibule is obliterated, absence of tooth irt 23,24. On palpation diffuse swelling is present on the left upper quadrant extending from 22 to 27 bucally as well as palatally, overlying mucosa appeared smooth with evidence of bony expansion [Fig- 1].

On the basis of clinical characteristics a provisional diagnosis of a dentigerous cyst of left maxillary region is made. Since she came already with radiographic examination such as PNS, OPG as well as CBCT. All of the radiographs showed presence of unerupted/ impacted permanent left canine as well as premolars and a well defined radiolucent lesion which gives the impression of a dentigerous cyst since it is involved with the crown of an unerupted tooth [Fig-2,3,4]

The patient was planned to be operated under general anesthesia with nasal intubation and the procedure planned was enucleation with delayed primary closure. Vestibular incision was placed in the left upper region from cuspid region to first molar region and flap eleveated and the lesion exposed. Deroofing of the bone done and the lesion was enucleated in a parts and it also contains the unerupted cuspid and bicuspid covered in the lesion, then the lesion was thoroughly inspected for any remanents of the cyst and chemical curettage with modified carnoy's solution done to prevent chances of recurrence since pre - operative incisional biopsy was not performed and the operated area was packed with gauge dipped in BIPP (bismuth iodoform and paraffin paste) dressings and closure done loosely leaving sufficient space for change of dressings post- operatively for a week, total closure done with 3-0 vicryl after a week. The histopathological report of the patient confirmed the diagnosis of Mural unicystic ameloblastoma-plexiform type.

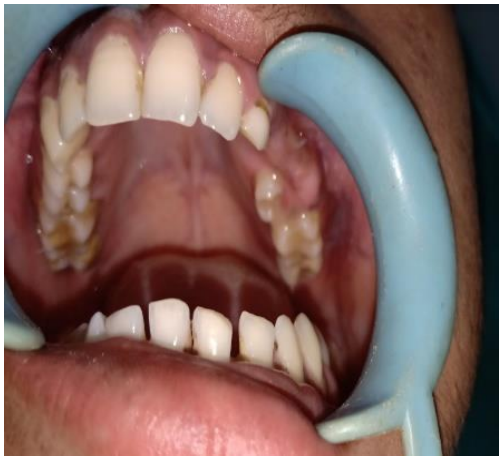

Fig 1: Intra oral view showing deciduous canine and missing permanent canine and first bicuspid.

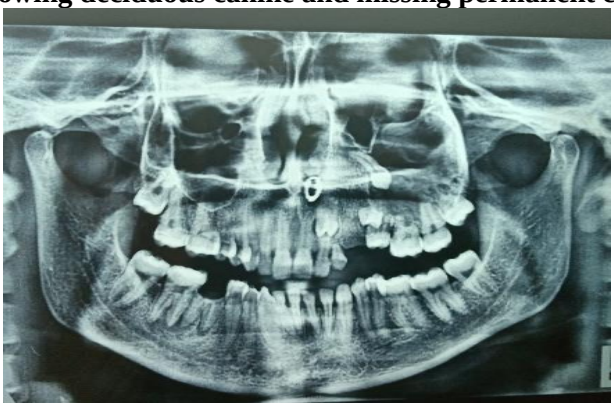

Fig 2: OPG showing the presence of unerupted permanent maxillary left canine and partially developed crown of left first bicuspid.

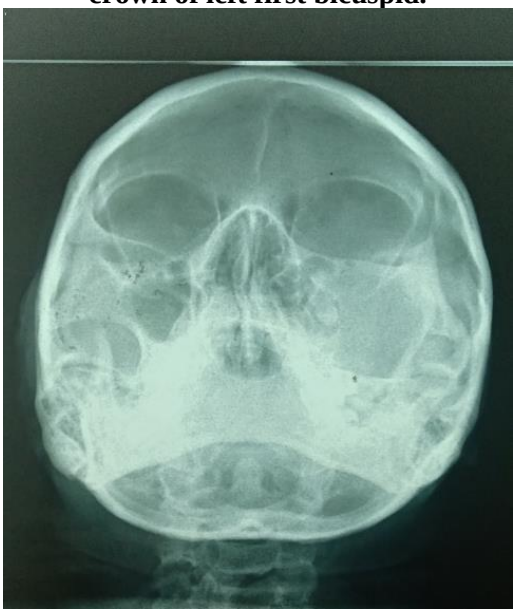

Fig 3: PNS showing the presence of unerupted maxillary left permanent canine.

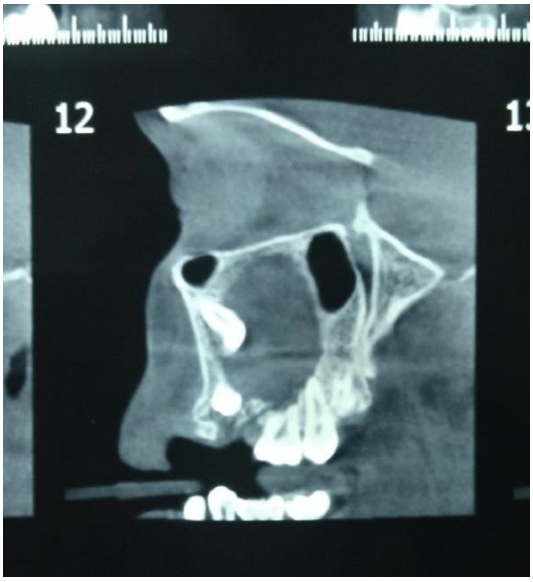

Fig4: CBCT showing the presence of unerupted maxillary canine.

DISCUSSION

Ameloblastoma is the most commonly occuring odontogenic neoplasm. Churchill first use the term ameloblastoma in 1934. There are four subtypes or variants of ameloblastomas which are classified as (i) the classic solid/multicystic ameloblastoma (SMA),(ii) the unicystic ameloblastoma (UA),(iii) the peripheral ameloblastoma (PA),(iv) the desmoplastic ameloblastoma (DA), which also includes the so-called lesions of mixed type [9].

The frequency of occurrence of unicystic ameloblastoma varies from 5% and 22%. Robinson and Martinez in 1977 were the first to describe unicystic ameloblastoma , Plexiform unicystic ameloblastoma is a relatively rare variant of unicystic ameloblastoma. Ameloblastomas are typically differentiated histologically into unicystic intraosseous, multicystic, solid intraosseous (80-90% of all ameloblastomas) or peripheral. The term unicystic ameloblastoma refers to those cystic lesions that show clinical, radiographic or gross features of a jaw cyst but on histologic examination show a typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor growth [10].

As evident from literature review that ameloblastoma is most commonly seen in younger age with equal frequency and sex, this present case patient was a 17 year old female which is accordance with the literature, but usually ameloblastoma are common in mandible but in this case ameloblasotoma was found in maxilliary area which is rare[1,2].

In Radiograph it is unilocular or multilocular, with a tendency for expansion, Unicystic lesions usually shows unilocular radiolucencies but can vary from total radiolucent to mixed radiolucent and radiopaque which appears like soap bubble / honey comb . This present case radiograph also shows well defined radiolucent lession [11].

According to literature treatment of UA includes both radical and conservative surgical excision, curettage, chemical and electrocautery, radiation therapy or combination of surgery and radiation. Sehdev et al and Shatkin and Hoffmeister et al reported high chances of recurrence in conservative procedure. In this present case enucleation with delayed primary closure management was adopted as the patient falls in to young age group . This present case shows clinically and radiographically similarities with dentigerous cysts, but true nature of the lesion may only become evident when the entire specimen is available for histologic examination which happened in the present case , the histologic examination revealed the lesion as Mural Unicystic plexiform ameloblastoma. UA is a prognostically distinct entity with a recurrence rate of 6.7-35.7%, and the average interval for recurrence is approximately 7 years [12,13,14].

CONCLUSION

Ameloblastoma has a high rate of local recurrence usually due to late diagnosis because of its poor symptoms and low prevalence and if it is not adequately removed. Radical surgical resection of ameloblastoma is the treatment of choice, especially in cases of large, expansive tumors a radical surgical protocol is a very good option to prevent relapse of the tumor on a long-term basis. The success factor associated with the treatment is the early diagnosis and to correlate the histopathologic findings with clinical and radiographic features which might alter the final treatment plan, in this case the final diagnosis came as Mural Unicystic plexiform ameloblastoma. So the patient is advised to be upon regular follow ups.

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Informed Consent: Written informed consent was obtained from the patient and parents.

Code of Ethics : The study was done according to The Code of Ethics of the World Medical Association (Declaration of Helsinki) for studies involving humans.

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