A Very Rare Case Of Bilateral Dentigerous Cyst In A Nonsyndromicpatient

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ABSTRACT:

Dentigerous cyst, also known as follicular cyst represent the second most common odontogenic cysts of the jaws after radicular cysts and are usually associated with the crowns of unerupted permanent teeth, supernumerary teeth and rarely deciduous teeth. They are usually solitary in presentation. Multiple & bilateral dentigerous cysts are extremely rare in the absence of developmental syndromes or systemic disease or the use of certain drugs. We hereby present a case of bilateral dentigerous cysts of maxilla involving crowns of unerupted 2nd premolar teeth on (Rt) & canine teeth on (Lt) side. An effort is also made to review existing literature on this entity.

Keywords: dentigerous cyst, bilateral, nonsyndromic, maxilla, crowns

INTRODUCTION:

Dentigerous cyst also known as follicular cyst is the second most common form of benign developmental odontogenic cyst that accounts for approximately 24% of all true cysts in jaw and results from accumulation of fluid between reduced enamel epithelium and enamel of an unerupted tooth. The most frequently involved tooth is mandibular 3rd molar followed by maxillary canine. They generally progress very slowly and may pass unnoticed for several years. They are frequently discovered when radiographs are taken to investigate a failure of tooth eruption, a missing tooth or malignment. Most dentigerous cysts are solitary though bilateral and multiple cysts are usually found with a number of syndromes or concurrent use of certain drugs. In absence of these factors, occurrence of bilateral dentigerous cyst is very rare, so we are going to present the following case.

CASE REPORT:

A 38 years old male patient attended with the complain of facial pain over the upper jaw area Left side and slight fullness on that area from last 1 month. Patient also complain of nasal blockage both side (left> right) from last 2 yrs. He has no history of any recurrent nasal discharge, headache or epistaxis. He has no history of trauma or past surgical history. No known systemic illness, drug history or any genetic disorder.

On clinical examination we found that there is facial asymmetry on left side maxillary area, deviated nasal septum on left side & mild tenderness present on left maxillary sinus area.

Oral examination revealed a mixed dentition & clinically absent left side canine (upper) and right sided 2ndpremolar (upper). There is a swelling noticed on leftgingivobuccal sulcus area.

Endoscopic evaluation showed deviation of nasal septum towards left side.

On X ray PNS it was found that there is hazyness in both maxillary sinus area with two radioopaque density probably impacted tooth on bilateral maxillary antrum. On CT evaluation there are two well defined thin smooth wall unilocular cystic lesion measuring about 3cmx4cm and 3.5cmx5cm seen on right and left upper alveolus area respectively. There are unerupted teeth noted within the cystic lesion.

Then the patient was plan for surgical enucleation of cyst along with extraction of impacted tooth. We approach the cyst through sublabial incision on both side. Intraoperatively we found bilateral big cystic masses almost completely filling both maxillary sinuses. The cystic masses were dissected from the wall of both sinuses & impacted tooth removed & the whole specimen sent for histopathological examination. Haemostasis was achieved. Histopathological report confirmed the diagnosis of dentigerous cyst.

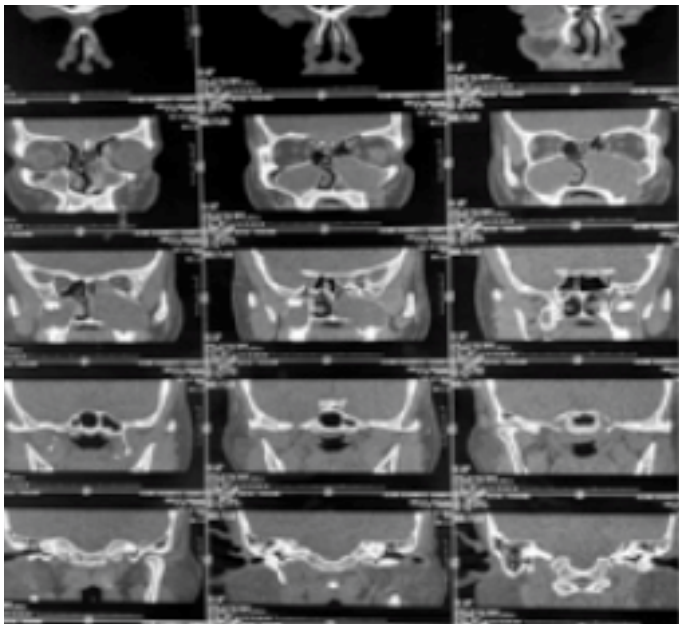

Fig 1 : Preoperative CT scan PNS showing disease

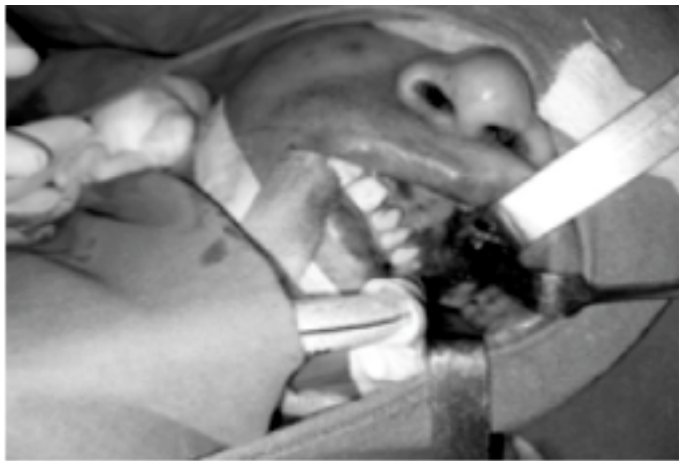

Fig 2 – Intraoperative photograph

Fig3 – Post operative photograph showing specimen

DISCUSSION:

Dentigerous cyst represent benign odontogenic cyst associated the crowns of either unerupted or impacted permanent teeth or supernumerary teeth but rarely deciduous teeth.[1,2] They mostly present in 2nd or 3rd decades of life and are rarely seen during childhood. The substantial majority of dentigerous cyst involve mandibular 3rdmolar(45.7%) followed by maxillary canine, mandibular 2nd premolar, maxillary 3rd molar & rarely maxillary premolar.[3]

The exact pathogenesis of this entity is not clearly understood. It is seen to develop by accumulation of fluid between he reduced enamel epithelium andenamel or within enamel organ.[4]The venous of flow is seen to get obstructed due the pressure exerted by the empty tooth on an impacted tooth follicle, leading to rapid transudation of serum across capillary walls. This leads to increase in hydrostatic pressure with resultant separation of folliclefrom to crown with or without the reduced enamel epithelium.

Dentigerous cyst usually occurs solitarily in most of the cases. Bilateral and multiple cases have been reported to occur in association with the number of syndromes including Basal cell nevus syndrome, Gardner's syndrome, Mucopolysacharidosis type IV, Cleidocranial dysplasia, and Klippel Feil syndrome.[5] Sometimes this entity is induced by some medications like combined use of Cyclosporin A & Calcium channel blockers.[6]Pleomrphism in chromosome 1qh+ has also been reported with this condition.[7] In our case syndromic association or medication usage was clearly ruled out.

A comprehensive search of Pubmed & English literature from 1943 to 2019 could locate a total 48 cases of nonsyndromic bilateral dentigerous cyst with mean age of presentation is 19.2 years, having a male predominance 1.8:1 and three quarters are associated with mandibular 3rd molar.[8] It reflects true rarity of condition. Daley et al reported an incidence rate of 0.1 – 0.6%, whereas Shear found the incidence to be 1.5%.[9]

Dentigerous cysts are usually painless unless secondarily infected & may cause facial swelling only when they have reached larger in size. Delayed teeth eruption is also seen. Therefore it is important to perform radiographic examination (panoramic view) in case of unerupted teeth. The classical picture is that of a unilocular radiolucent lesion of various sizes with well-defined sclerotic borders associated with crown of an unerupted tooth. A follicular space more than 5mm, displacement of adjacent teeth, resorption of roots can also be observed radiographically.[10,11]

Differential diagnosis of this lesion included periapical cyst, odontogenic keratocyst and unicysticameloblastomas as they share same radiologic pictures, so histopathological examination is paramount for definitive diagnosis. The cystic lining has an inherent ability for metastatic change largely due to areas oforthokeratinisatin, ciliated cells or mucin secreting cells present in the lining.[10]In our case, linings were devoid of metastatic or dysplastic change.

As far as treatment is concerned, most dentigerous cysts are treated with enucleation& removal of associated teeth. Large cyst can be marsupialized initially to decompress the cystic contents and enucleated more conservatively. The main drawback of marsupialzation is that the pathological tissue remains in situ. The prognosis is usually excellent & recurrence has been nonexistent withthis entity.

CONCLUSION:

Bilateral dentigerous cyst not associated with any syndrome or systemic disease is an extremely rare findings& therefore necessitates a special mention. A thorough clinical examination is paramount to rule out any associated syndrome or disease. Early diagnosis using both conventional (panoramic view) and advanced imaging modalities (CT Scan) is important to reduce morbidity and avoid more aggressive surgical procedures specially in case of person with missing or unerupted tooth or malalignment teeth.

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