Non Syndromic Bilateral Dentigerous Cysts- A Case Report
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Abstract
Bilateral dentigerous cysts, when present, occur in association with syndromes like mucopolysaccharidosis or cleidocranial dysplasia. Very few non-syndromic bilateral dentigerous cysts have been reported and almost all have involved the mandibular molars. Bilateral involvement of maxillary canines with dentigerous cyst is rare. Only one such case has been reported in our knowledge. We report a rare case showing bilateral dentigerous cysts in association with impacted maxillary canines.

Key words: Dentigerous Cysts; Bilateral; Non-Syndromic; Maxilla.

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Introduction
Dentigerous cysts are the second most common odontogenic cysts after radicular cysts. Their frequency in general population is about 24% of all true cysts of the jaws(1). A dentigerous cyst is one that encloses crown of an unerupted tooth by expansion of its follicle, and is attached to its neck.

Some unerupted teeth have a slightly dilated follicle in the pre-eruptive phase but this does not signify a cyst unless the pericoronal width is more than 3-4mm. Most dentigerous cysts are solitary. Bilateral or multiple dentigerous cysts occur usually in association with syndromes like Maroteaux-Lamy syndrome(2). It is rare to find non syndrome associated bilateral dentigerous cysts. In our knowledge, 18 cases of non syndromic bilateral dentigerous cysts have been reported till date out of which only one was associated with maxillary cuspids bilaterally. We report a rare case of non syndromic bilateral dentigerous cysts in an 11 year old child involving the maxillary canines.

Case report
An 11 year old boy reported in with the chief complaint of swelling on the left side of the face. He had noticed the swelling 2 months ago. The swelling was associated with intermittent pain and had slightly increased in size in the past 2 months. Extra oral examination revealed a diffuse swelling on the left side of the face. The swelling extended superiorly 1cm below the infra orbital margin, inferiorly 0.5 cm above the upper lip, mesially up to the lateral aspect of the nose and laterally 4cms anterior to the posterior border of mandible.

Intra oral examination revealed obliteration of the labial vestibule in the right and left anterior region. On palpation, egg shell crackling was felt in the region of the left maxillary 2nd deciduous molar. The patient had no associated syndrome or systemic disease present.

Panoramic radiograph (Fig.1) revealed bilateral radiolucencies in the maxillary anterior region with associated impacted tooth on the left side and obliteration of the left maxillary sinus.

Figure 1 Panoramic radiograph

Coronal soft tissue imaging (Fig.2) reveals bilateral, soft tissue mass with well-defined radio opaque borders.

Figure 2 Coronal soft tissue imaging
On the left side there is complete obliteration of the maxillary sinus and displacement of infra orbital margin, while the
soft tissue mass on the right side shows superior displacement of the maxillary sinus. The axial view shows cystic lesion on the right side associated with a tooth like radio opaque structure at the inferior border of the lesion. On the left side the cystic lesion is seen with a radio opaque structure at the superior border, suggestive of an associated impacted tooth.

Surgical removal of both the lesions was performed under general anesthesia. Left and right crestal incisions were given and the flap was raised. Labial and buccal obliteration was evident. Bony window was created and cystic lining removed from the intact bone. Thorough curettage was done and procedure repeated on the right side. Iodoform gauze pack was placed in both cavities, sutures placed and the curetted material along with the associated teeth were sent for histopathological analysis in two different bottles marked right(R) and left(L).

Grossly, the soft tissue specimens measured 12 x 10 x 8 mm (L) and 9 x 8 x 6mm(R). Both the specimens had a maxillary canine attached to each of them at the cementoenamel junction of the teeth. Histopathologically, the specimen marked (R) showed 2-3 layer thin non keratinized stratified squamous epithelium of flattened cells, with underlying connective tissue capsule showing parallel bundles of collagen fibers. The second specimen marked (L) also showed non keratinized stratified squamous epithelium (3-4 cell thick) of flattened cells, resembling reduced enamel epithelium, with underlying connective tissue capsule showing mild inflammatory infiltrate. The connective tissue capsule showed a few odontogenic islands. Both the lesions were diagnosed as dentigerous cysts.

Discussion
Dentigerous cysts are the most common developmental cysts with a frequency of 1.44 per 100 unerupted teeth(2). The anatomic environment of an unerupted tooth is probably of some significance in determining the development of a cyst. Other factors, possibly innate may also play a role in cyst development. Al Talabani and Smith(3) suggested the possibility of two types of dentigerous cysts. One arising by the degeneration of stellate reticulum at an early stage of development and thus being associated with hypoplastic teeth and the second developing after the completion of crown by accumulation of fluid between the layers of reduced enamel epithelium. Batra P et al(4) in 2004 reported a case of bilateral dentigerous cysts associated with mandibular third molars with polymorphism in chromosome 1qh.

Bilateral dentigerous cysts are usually seen associated with syndromes like cleidocranial dysplasia or mucopolysaccharidosis (5). De Biase A et al, in 2001 reported bilateral mandibular dentigerous cysts to occur with prolonged, concurrent use of cyclosporine A and calcium channel blockers (6). Non syndromic bilateral dentigerous cysts are not frequent. To our knowledge, only eighteen cases of non-syndromic bilateral dentigerous cysts have been reported till date (4, 5, 7, 8). Out of these, sixteen cases were associated with mandibular teeth. Fregnani et al(9) reported a case of bilateral cysts involving the maxillary molar region and the mandibular molar region(9) and Utsner E et al(5) in 2003 reported a case of bilateral dentigerous cysts associated with the maxillary canines. The present case shows the presence of bilateral dentigerous cysts involving the maxillary canines, the left cyst being larger than the right one. The age of the patients ranged from 3 to 57 years with mean age being 17.92 yrs. Most dentigerous cysts occur in the 2nd and 3rd decades of life. Out of these 18 cases, 12 were males and 6 females (Table I). Thus, the male female ratio was 2:1. The male female ratio for solitary dentigerous cysts has been reported to be 1.8:1. The present case was of an 11 year old male.

Usually dentigerous cysts occur as an asymptomatic swelling but in the present case, the patient complained of mild intermittent pain on the left side. The lesion on the left side showed egg shell crackling on palpation. This could be due to the associated inflammation in the cyst. Only in one previously reported case by Swerdloff M et al,(10) 1980, the patient reported with associated pain(11).

In all reported cases, radiographic examination revealed radiolucent lesions associated with impacted teeth. However, in this case, the CT imaging was done as the lesion on the left side was extensive, as it gives information about the size, extent, cortical plates and relationship of the lesion to adjacent structures and helps to rule out solid and fibro osseous lesions(11). It also helps to see the encroachment of the lesion onto the maxillary sinus, nasal cavities and the orbit(12).

In the present case, CT imaging provided with important information regarding the association of the cystic lesions with
impacted teeth and also showed the obliteration of the left maxillary sinus and encroachment of the inferior border of the orbit on the left side, and the displacement of the right maxillary sinus superiorly. CT imaging also confirmed that despite the expansion of the bone, the cortical plates were intact.

Histopathological analysis is essential in the diagnosis especially with such extensive lesions. In the present case, the gross specimen showed attachment of the cyst lining at the neck of the canine in both the lesions. The lining epithelium in both the lesions resembled reduced enamel epithelium confirming the diagnosis of dentigerous cyst. The underlying connective tissue showed the presence of a few odontogenic islands. Chronic inflammatory infiltrate was present in the dentigerous cyst of the left side suggestive of secondary infection. This was correlated with the patient’s complain of intermittent pain in the lesion involving the left side. As egg shell cracking was observed on the left side, serial sections were taken to rule out transformation into a unicystic ameloblastoma or other neoplasms but no such changes were seen histopathologically.

Since dentigerous cyst can attain considerable size with minimal or no symptoms, early detection and removal of the lesion is important to reduce morbidity. It is therefore important to perform radiographic examination of all unerupted teeth. Although multiple dentigerous cysts are rare, once a cystic lesion is recognized, the patient must be examined carefully to rule out other possible dentigerous cysts. Complete removal of the lesion is essential given that the dentigerous cyst is essential or the remnants have been documented to differentiate into ameloblastoma, squamous cell carcinoma, AOT, complex odontomas.

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