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Bilateral Maxillary Dentigerous Cyst

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Introduction

A dentigerous cyst can be defined as a cyst that encloses the crown of an unerupted tooth, expands the follicle and is attached to the cementoenamel junction of the unerupted tooth. The substantial majority of dentigerous cysts involve the mandibular third molar and the maxillary permanent canine, followed by the mandibular premolars, maxillary third molars and rarely the maxillary premolars[1].

Although dentigerous cysts are common developmental cysts, bilateral dentigerous cysts and that too of maxillary origin are extremely rare in the absence of an underlying syndrome or systemic disease and hardly reported. Bilateral or multiple dentigerous cysts are usually associated with the Maroteaux-Lamy syndrome, mucopolysaccharidosis type VI, cleidocranial dysplasia, Basal cell nevus syndrome[2]. An extensive search of the English language literature has identified only 22 cases[3]. Although this finding may reflect the true rarity of the condition, it is conceivable that bilateral dentigerous cysts are either under recognized or under-reported as sometimes they are known to regress spontaneously[4].

Case Report

This is the case of a 13 year old male patient with bilateral maxillary dentigerous cyst reported here for the rarity of its occurrence. The patient presented with swelling of the left cheek with obstruction of the left nasal cavity for 4 months. There was history of dental pain in the right upper molar area. There was no other associated feature of any syndromic morbidity.

Figure 1: Clinical picture showing swelling of the left cheek

The patient had diffuse bony hard non tender swelling of the left maxillary area 5x6 cm in diameter (Fig 1). There was bulge of the left gingivobuccal sulcus and left side of the hard palate inferiorly (Fig 2, 3). Nasal endoscopy

revealed bulge of the lateral wall of the nasal cavity medially (Fig 4).

Thorough physical examination and relevant investigations were carried out to rule out any syndromic associations like mucopolysacchiridosis.

Figure 2 and 3: Clinical picture showing bulge in left palate and left gingivobuccal sulcus

Non Contrast CT scan showed a cystic lesion arising at the root of impacted tooth extending into to the left maxillary sinus causing remodelling of the sinus with expansion of the its walls. Similar cystic lesion was also present in the right maxillary sinus arising from the root of an impacted tooth (Fig 5).

Figure 4: Endoscopic picture, bulged lateral wall of nose

Figure 5: NCCT showing bilateral maxillary cystic expansile lesion associated with impacted teeth

Complete enucleation of the bilateral cyst was done with bilateral intranasal endoscopic and canine fossa approach in view of the left sided cyst filling and expanding the maxillary sinus (Fig 6, 7).

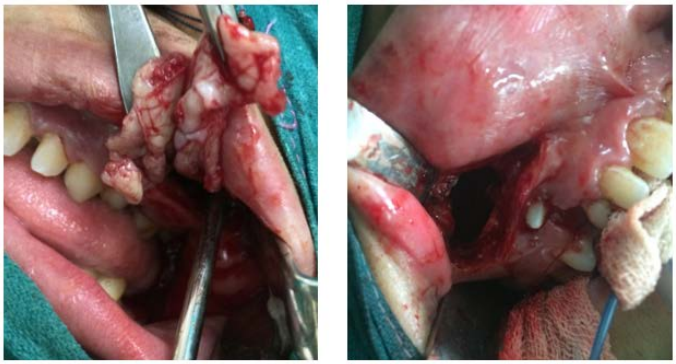

Figure 6 and 7: Intraoperative picture showing bilateral enucleation of the cyst through canine fossa approach

No intracavity teeth was found. Bilateral impacted teeth were removed.Histopathological examination of the specimen confirmed the nature of the lesion as dentigerous cyst.

Discussion

Dentigerous cysts are the second-most common true cysts of the jaws following radicular cysts.[5] Majority of the dentigerous cysts are associated with the permanent teeth during the second decade of life.[6] Most dentigerous cysts

are solitary. Bilateral and multiple cysts are usually found in association with a number of syndromes including cleidocranial dysplasia and Maroteaux-Lamy syndrome.[7] In the absence of these syndromes, bilateral dentigerous are extremely rare and hardly reported. Occurence in the mandibular third molar region is more common compared to maxillary third molar region.[8] The dentigerous cyst is usually associated with impacted third molars and canines.[9,10]

The exact histogenesis of the dentigerous cyst is not known. It is stated that the dentigerous cyst develops around the crown of an unerupted tooth by accumulation of fluid either between the reduced enamel epithelium and enamel or in between the layers of the enamel organ[11]. This fluid accumulation occurs as a result of the pressure exerted by an erupting tooth on an impacted follicle, which obstructs the venous outflow and thereby induces rapid transudation of serum across the capillary wall. Toller stated that the likely origin of the dentigerous cyst is the breakdown of proliferating cells of the follicle after impeded eruption. These breakdown products result in increased osmotic tension and hence cyst formation. Bloch suggested that the origin of the dentigerous cyst is from the overlying necrotic deciduous tooth. The resultant periapical inflammation will spread to involve the follicle of the unerupted permanent successor; an inflammatory exudate ensues and results in dentigerous cyst formation[12]. Most of the authors have reported the presence of carious or discolored deciduous teeth in relation to the development of dentigerous cysts. This suggests that the periapical inflammatory exudates from the deciduous teeth might be one of the risk factor for the occurrence of dentigerous cysts[12].

Dentigerous cysts are usually asymptomatic[13] and are diagnosed incidentally in routine radiographs. Unerupted teeth could indicate the possibility of a dentigerous cyst. A dentigerous cyst can expand causing facial asymmetry.

It is important to perform radiological examination for unerupted teeth in which panoramic radiograph may be used primarily. However, in extensive lesions, CT scan is considered as the gold standard. Thus in this case a CT scan was chosen as a radiological investigation of choice. Radiographic evaluations of dentigerous cysts are classically seen as radiolucent shadows associated with unerupted teeth and seemingly attached to the cementoenamel junction.[14] CT scan show well-defined, unilocular, well-corticated, hypodense lesions that are often associated with the crowns of impacted teeth.[15] The hypodensity is attached at an acute angle to the cervical area of an unerupted tooth. The radiographic differentiation between a dentigerous cyst and a normal dental follicle is based merely on size. A normal dental follicular space is 3-4 mm whereas a dentigerous cyst can be suspected when the space is more than 5 mm.[16]

The treatment of choice for dentigerous cyst is enucleation along with extraction of the impacted teeth.[17] Although in pediatric patients marsupialization has been considered to save the impacted tooth and developing tooth bud. It has been seen that tooth eruption potential is more in children who have open apices in the involved teeth.[18,19] In this case endoscopic enucleation was done through intranasal and canine fossa approach. Primary closure of the maxillary defects was done using resorbable synthetic suture material (vicryl 4-0), and the bone defects were packed with povidone Iodine-soaked gauze and was left open for secondary healing. Postoperatively the wound had healed satisfactorily and there was no complications.

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