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Dentigerous cyst with recurrent maxillary sinusitis; A case report with literature review

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Abstract

Introduction: Dentigerous cyst is an odontogenic cyst of the crown of impacted or unerupted teeth which remains completely asymptomatic unless when infected or incidentally found on routine dental radiographs.

Case report: Here, an 18 year-old male patient presented with a dentigerous cyst arising from left maxillary third molar extending to the maxillary sinus with recurrent maxillary sinusitis.

Keywords: Dentigerous cyst, maxillary sinus, marsupialization

1. Introduction

Dentigerous cyst (DC) is a common oral lesion formed by fluid accumulation between the fully formed tooth crown and the reduced enamel epithelium^[1]. It is considered as a developmental abnormality arising due to reduced enamel epithelium around the crown of an unerupted tooth^[1]. While a normal follicular space is 3 to 4 mm, a dentigerous cyst can be suspected when the space is more than 5 mm^[3].

DC is the most common developmental odontogenic cyst accounting for approximately 25% of all odontogenic cysts of the jaw^[2]. Their frequency in general population has been estimated at 1.44 for every 100 unerupted teeth. The cyst arises from the separation of follicle from the crown of an unerupted tooth. Though it may involve any tooth, mandibular third molars are most commonly affected^[3]. They are frequently noted as incidental findings on the radiographs because majority of these cysts are asymptomatic, unless they become secondarily infected^[1, 2]. Radiograph shows unilocular, radiolucent lesion characterized by well-defined sclerotic margins and associated with the crown of an unerupted tooth^[2, 3].

2. Case Report

An 18 year old male patient reported to the Department of Oral Medicine and Radiology, Bangalore Institute of Dental Sciences with chief complaint of pus discharge from the upper left back tooth region since one and half years. The patient had noticed recurrent pus discharge with discomfort and slight pain in the distal region of left second molar. It was accompanied with episodes of nasal mucous secretion and heaviness in the left anterior maxillary and left temporal region on bending the head down since past 6-8 months.

He visited a private dental clinic for the same 1 year back, where a paranasal sinus radiograph was suggested. After reviewing the radiograph, the patient was explained about a fluid filled sac of that region and referred for a CT scan of maxilla. Later an option of surgery along with removal of the involved tooth was offered as a mode of treatment. Patient did not visit due to his exams. On noticing that the pus draining had not regressed patient visited Bangalore Institute of Dental Sciences for further evaluation.

There was no relevant medical history and personal history. On general physical examination, the patient was moderately built and nourished, well oriented to time and place. Extra oral examination showed no significant findings (Fig1).

Intraoral examination showed no obvious swelling but revealed a shallow pocket in the distal aspect of 27 and tenderness in the mucobuccal sulcus in relation to 27 and 28.

An intra-oral periapical radiograph (IOPAR) w.r.t. 27 and 28 region was taken. It showed an impacted tooth apical to 26 and 27 extending into the maxillary sinus surrounded by a radiolucent area extending from the cemento-enamel junction of the impacted 28 coronally, measuring approx 1.5 by 1.2 cm surrounded by an oval cortical margin.

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Maxillary occlusal view showed buccally impacted 28 surrounded by a radiolucent area with a corticated margin expanding to the palatal side around the crown of 28. Panoramic radiograph was done and the radiograph showed an impacted 28 apical to the roots of 26 and 27 extending into the maxillary sinus from the floor, 0.5 cm medial to pterygomaxillary fissure. A small amount of radiolucent space within the corticated margin in the inferior part of crown of 28 was seen (Fig 2). Other findings include impacted 18, 38 and 48.

PA view of skull (Water's Position): revealed clouding of left maxillary sinus extending from the floor of the sinus up to 0.5 cm below the inferior margin of the orbit (Fig3).

Based on the history, clinical findings and radiographic features, a provisional diagnosis of dentigerous cyst was given with a differential diagnosis of radicular cyst, calcifying odontogenic cyst, ameloblastoma, ameloblastic fibroma, maxillary sinus mucocele and maxillary sinus antrolith. The cyst was surgically excised with the involved tooth (Fig4). Histopathological examination showed a cystic wall lined by non-stratified squamous epithelium. At places the epithelium was disrupted due to chronic inflammatory cell infiltrate of the underlying connective tissue. No cellular atypia were noted (Fig5). A final diagnosis of infected dentigerous cyst was confirmed based on the radiographic and histopathological reports.



Fig 1: Extra oral examination showed no significant findings.



Fig 2: Panoramic radiograph showed an impacted 28 apical to the roots of 26 and 27 extending into the maxillary sinus from the floor, 0.5 cm medial to pterygomaxillary fissure. Also, seen is a small amount of radiolucent space within the corticated margin in the inferior part of crown of 28. Other findings include impacted 18, 38 and 48.



Fig 3: Skulls PA view (Water's Position): it revealed clouding of left maxillary sinus extending from the floor of the sinus up to 0.5 cm below the inferior margin of the orbit.

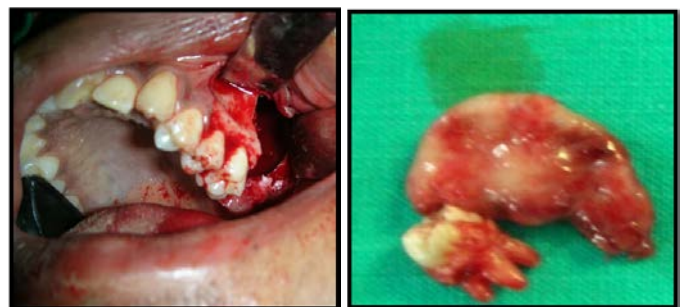


Fig 4: Surgically excised cyst with the involved tooth

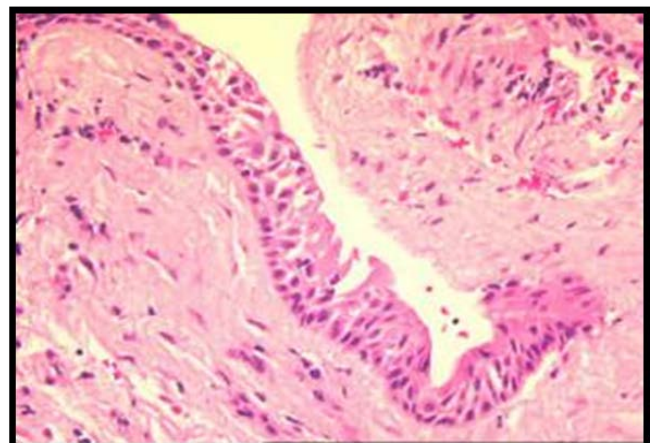


Fig 5: Photomicrograph showing a cystic wall lined by non-stratified squamous epithelium. At places the epithelium is disrupted due to chronic inflammatory cell infiltrate of the underlying connective tissue. No atypical features noted (H& E X 40)

3. Discussion

Dentigerous cyst is one of the most common lesions of the jaw. It is defined as a cyst that encloses the crown of an unerupted tooth by expansion of its follicle and is attached to the neck of the tooth^[5]. Clinically, it is asymptomatic but can cause cortical bone expansion.

Dentigerous cyst is always associated with an unerupted or developing tooth bud and found most frequently associated with crowns of mandibular third molar followed by maxillary canines and then maxillary molars.

Dentigerous cysts are the second most common odontogenic cysts after radicular cysts, accounting for approximately 24% of all true cysts in the jaws^[2, 4]. Dentigerous cysts around supernumerary teeth account for 5% of all dentigerous cysts, most developing around a mesiodens in the anterior maxilla^[3, 4]. Mesiodens, a kind of supernumerary tooth, located in the

maxillary central incisor region. It has an overall prevalence of 0.15-1.9%, can occur individually or in multiples and often does not erupt. More common in males than females [5]. Over 75% of all cases are located in the mandible [4].

A dentigerous cyst is formed by the hydrostatic force exerted by the accumulation of fluid between reduced enamel epithelium and the tooth crown of unerupted teeth [1, 3, 5]. It resembles as if the cyst encloses the crown and is attached to the neck at the cemento-enamel junction [5]. They almost exclusively occur in permanent dentition. The cyst is lined by stratified squamous non-keratinized epithelium [6].

Usually all dentigerous cysts arise from the enamel organ after completion of amelogenesis. They arise due to accumulation of fluid causing separation of enamel of the unerupted tooth. The fluid present inside the cyst is hyperosmolar due to the presence of albumin, immunoglobulin and squamous epithelial debris [1, 4, 9]. This hyperosmolar fluid causes influx of extracellular fluid into the cyst causing huge expansion of cyst. The epithelial lining of the cyst secretes collagenase and osteoclast activating factor which causes local bone resorption causing further increase in the size of the cyst. This enlarging cyst encloses the crown of the unerupted teeth and is attached to its cemento-enamel junction [1, 4].

Theories of dentigerous cyst formation [7]:

3.1 Intrafollicular theory: According to this theory cyst formation occurs due to fluid accumulation between the layers of inner and outer enamel epithelium after crown formation.

3.2 Enamel hypoplasia theory:

This theory suggests that dentigerous cyst formation occurs due to degeneration of stellate reticulum at a very early stage of tooth development. There is also an associated enamel hypoplasia.

3.3 Main's theory: This theory suggests that impacted tooth exerts pressure on the follicle with resulting obstruction of venous outflow. This induces rapid transudation of fluid across the capillary walls.

The DCs are mostly asymptomatic and may be found on routine dental radiographic check-up. They may also cause symptoms like pain or swelling with the enlargement of the cyst size. Several researchers reported the pathologic fracture of the mandible caused by huge DC [1, 3]. The outgrowth of the cyst may also cause the resorption of adjacent teeth. According to Eliasson's report, roughly 1% of impacted maxillary third molars will subsequently become involved with a DC [1].

Radiographically, the typical DC shows a well-defined radiolucency with sclerotic border associated with the crown of an unerupted tooth [1, 2]. Three varieties of the cyst-to-crown relationships can be seen on radiographic examination. They are central variety, lateral variety and circumferential variety [1, 4]. In the case presented here, the cyst-to-crown relationship was classified to a central variety.

Panoramic radiographs are used for evaluation of dentigerous cyst followed by an occlusal IOPAR. In present case the PNS view was more useful to see the involvement of maxillary sinus. The computed tomography may be useful for evaluating the extent of bony involvement. Since DCs may contain fluid, in the magnetic resonance imaging (MRI), the cyst fluid may be seen as low intensity on T1-weighted and high intensity on T2-weighted images [1].

Patients with DCs over maxillary sinus might present nasal symptoms such as sinusitis. In addition, ophthalmological symptoms such as proptosis, diplopia, ptosis, epiphora, rarely

loss of visual acuity may be present [1, 10]. Fracture of the orbital bone caused by DC has been reported. Spontaneous remission of the lesion without surgical removal may happen, but in very few cases [1]. In present case left side nasal discharge and stiffness with involvement of maxillary sinus was evident.

The differential diagnosis of DC includes odontogenic keratocyst (OKC), adenomatoid odontogenic tumor (AOT), calcifying epithelial odontogenic cyst (COC), calcifying epithelial odontogenic tumor (CEOT), and unicystic ameloblastoma (UAs) [6, 7].

In addition to the histopathologic differences between the feature of the epithelium of OKC and DC, the differential diagnosis can also include the development and the recurrence tendency of these cysts [1]. About 40% unilocular OKC contain impacted tooth. The OKC is more aggressive with higher recurrence risk than DC and may be associated with nevroid basal cell carcinoma syndrome [1].

Recently, researches showed mutation of PTCH gene and over activated of *Shh* signaling may be associated with the clinicopathological expression of OKCs. BMP-4 may be a useful biochemical marker to differentiation of OKC and DC [2, 3]. BMP-4 is expressed more intensive in OKC compared with DC, and is even more intensively expressed in the recurred cases [1].

Histopathological examination of cyst shows reduced enamel lining epithelium. Connective tissue stroma shows features of primitive type of ectomesenchyme [1, 7, 11]. Findings would depend on whether there is inflammatory component to the cyst is present or not. In non infected cysts the lining epithelium is 24 layers thick formed by primitive ectomesenchyme. These lining cells are low cuboidal to columnar. Retepegs could be seen only in cysts which are infected. The connective tissue stroma is loose and is rich in acid mucopolysaccharides. When the dentigerous cyst is inflamed then it is characterised by the presence of hyperplastic rete ridges and the cyst wall demonstrates inflammatory infiltrate [1, 11]. In present case showed a cystic wall lined by non-stratified squamous epithelium. At places the epithelium is disrupted due to chronic inflammatory cell infiltrate of the underlying connective tissue. No atypical features noted.

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 [3].

The standard treatment for a dentigerous cyst is enucleation and extraction of the cyst-associated impacted or unerupted tooth. However, large lesions can be marsupialised [4]. In present case the cyst was surgically excised with the involved tooth.

Occasionally, it transforms to squamous cell carcinoma, mucoepidermoid carcinoma or ameloblastoma. The prognosis for most histopathologically diagnosed dentigerous cysts is excellent, recurrence being a rare finding [4].

4. Conclusion

Dentigerous cyst associated with impacted permanent teeth is not uncommon, early diagnosis is important to prevent morbidity. Imaging studies helps to diagnose, know the extent of lesion and for treatment planning. Knowledge of histopathologic features helps to confirm the diagnosis. Management of dentigerous cyst arising from ectopic third molar in the maxillary sinus is usually enucleation or surgical removal of cyst with associated tooth.

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