

## Case Report

### Dentigerous Cyst associated with impacted permanent maxillary canine

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#### Abstract:

Odontogenic developmental cysts of jaws usually presents as asymptomatic lesions. Dentigerous cyst is one of the variety of these cysts which may grow to a large size intraosseously within jaws before it manifest clinically. Numerous immunohistochemical and molecular marker studies have been carried out for this lesion to understand its etiopathogenesis and aggressive behavior. Another reason being its potential to transform to ameloblastoma and squamous cell carcinoma. We present a case of dentigerous cyst associated with impacted maxillary canine manifesting as a unilateral swelling of midface region.

**Key Words:** Dentigerous cyst, unerupted, dental follicle, marsupialization.

#### Introduction:

Cysts of the jaw usually present as asymptomatic swellings of the mandible and midface region. Different varieties of the odontogenic cysts are the ones that develop from odontogenic epithelium or the epithelial remnants of the odontogenic apparatus. The second most common odontogenic cyst is the dentigerous cyst. A dentigerous cyst is one that encloses the crown of an unerupted tooth by expansion of its follicle, and is attached to its neck. (Shear & Speight, 2007) The diagnostic feature of this cyst is the presence of the unerupted tooth in its cavity (Johnson et al,1994). It is most frequently associated with mandibular third molar, maxillary canine, mandibular premolar and maxillary third molar in decreasing order of frequency (Jones et al,2006). Most dentigerous cysts are asymptomatic, and their discovery is usually an incidental finding on radiography. The cyst being asymptomatic may attain a large size with resorption of the roots of teeth till it manifest clinically or become evident radiographically. It is now documented that the dentigerous cyst lining has the potential to develop into an aggressive ameloblastoma. Treatment includes enucleation of the cyst with the removal of the unerupted tooth. Marsupialization is occasionally done with very large cyst to decompress the cyst. Prognosis is excellent and recurrence is rare if completely removed.

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#### Case Report:

A 11year-old female patient presented with a progressively increasing swelling in the left maxillary midface region for last one month. Intraoral examination revealed a hard swelling with cortical plate expansion in relation to 63 to 25 (Fig.I).



Fig. I: intraoral view showing swelling extending from 22 distally upto 25

Distally tilted 22, grade II mobility in relation to 63 and grade I mobility with 24 was observed. Diagnostic orthopantomograph (Fig.II) showed a unilocular radiolucent lesion associated with 63, 24 and 25, 23 was seen impacted and enclosed within the lesion. Displacement of crown and root of 24, distal dilaceration

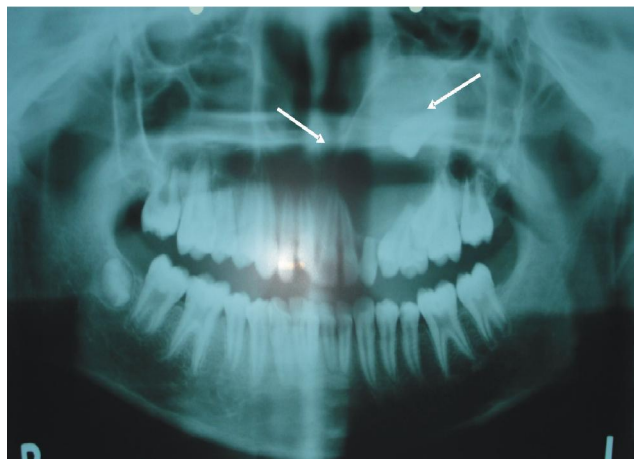


Fig. II: Orthopantomograph revealing unilocular well defined radiolucency surrounding impacted permanent left maxillary canine

at middle third root of 25 and resorption of the root of 63 was observed. There was discontinuity of lamina dura of 25 and in mesiobuccal root of 26. Routine laboratory parameters were normal.

Aspiration of the lesion was done and 2ml clear yellowish cystic fluid was obtained. Cystic enucleation was done and histopathologically examined.

Histological section showed characteristic thin cystic lining resembling reduced enamel epithelium and connective tissue stroma showed features of a primitive type ectomesenchyme. (Fig.III). Radiographic findings, surgical resection and histopathological examination confirmed the diagnosis of dentigerous cyst associated with the impacted canine. The patient remained under follow up for six months and no complications were observed.

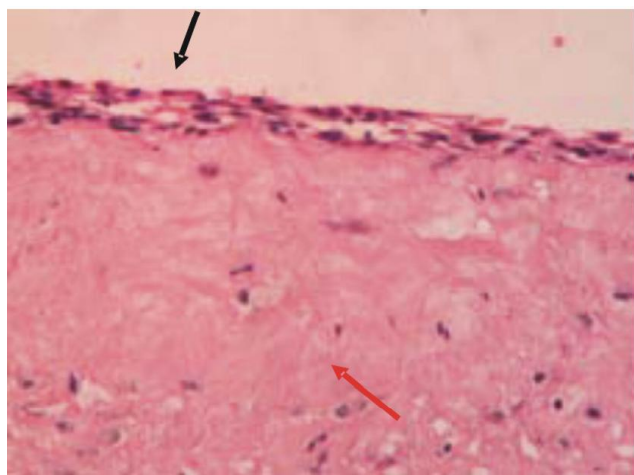


Fig. III : Dentigerous cyst with thin lining resembling reduced enamel epithelium( Black arrow) with no rete pegs the connective tissue is delicate as of primitive type (Red Arrow, H & E, 40X)

## Discussion:

Dentigerous cyst is one that encloses the crown of an unerupted tooth by expansion of the follicle and is attached to its neck.

Dentigerous cyst was earlier termed as 'Follicular Cyst' but as the latter implies to be derived from the tooth follicle which is mesodermal structure, the term was discontinued. Another reason suggested for discontinuation was that follicular cyst also refers to follicular cyst of the ovary, and also to hair follicle cyst. The term dentigerous is preferred, the literal meaning being 'tooth bearing' (Browne & Smith, 1991)

Dentigerous cysts are associated with an unerupted tooth. In order of its frequency they are associated mandibular third molars, maxillary canines, mandibular second premolars and maxillary third molars (Neville et al, 2006). They may also occur around supernumerary teeth, however, they are only rarely associated with primary teeth (Neville et al, 2002; Kusakawa et al, 1992). Our case was related with permanent maxillary canine.

These cysts can grow to very large size and can cause displacement of teeth, or in few cases it may remain relatively small. The age range varies widely, from 5 to 57 years (Shear 2007). Many dentigerous cysts are small asymptomatic lesions that are discovered serendipitously on routine radiographs, although some may grow to considerable size causing bony expansion that is usually painless until secondary infection occurs. Since cysts can attain considerable size with minimal or no symptoms, early detection and removal of the cysts is important to reduce morbidity. Moreover, almost all of the reported cases (Swerdlhoff et al, 1980) including the present case, present without pain and discovered during investigation of asymptomatic slowly-growing swellings.

Radiographically, the dentigerous cyst presents as a well-defined unilocular radiolucency, often with a sclerotic border. Since the epithelial lining is derived from the reduced enamel epithelium, this radiolucency typically and preferentially surrounds the crown of the tooth. A large dentigerous cyst may give the impression of a multilocular process because of the persistence of bone trabeculae within the radiolucency. However, dentigerous cysts are grossly and histopathologically unilocular processes and probably are never truly multilocular lesions (Shear 2007). Three types of dentigerous cyst have been described radiographically: The central variety, in which the radiolucency surrounds

just the crown of the tooth, with the crown projecting into the cyst lumen. In the lateral variety, the cyst develops laterally along the tooth root and partially surrounds the crown, the circumferential variant exists when the cyst surrounds the crown but also extends down along the root surface, as if the entire tooth is located within the cyst. Our case was radiographically a classic presentation of the circumferential variety.

The histological features of dentigerous cysts may vary greatly depending mainly on whether or not the cyst is inflamed. In the non-inflamed dentigerous cyst, a thin epithelial lining may be present with the fibrous connective tissue wall loosely arranged. As the lining is derived from reduced enamel epithelium it is 2-4 cell layer thick primitive type. The cells are cuboidal or low columnar. Rete pegs formation is absent except in cases that are secondarily infected. As the connective tissue wall is derived from the dental follicle of developing enamel organ, it is a loose connective tissue stroma which is rich in acid mucopolysaccharides.

In the inflamed dentigerous cyst, the epithelium commonly demonstrates hyperplastic rete ridges, and the fibrous cyst wall shows an inflammatory infiltrate. Young fibroblasts are present in the stroma. The cell lining may show metaplastic changes in the form of mucous producing cells or secretory cells such as goblet cells. Pseudostratified ciliated columnar epithelium has also been reported. Rarely sebaceous glands in the walls are observed. The content of the cystic lumen is usually thin watery yellow fluid and is occasionally blood tinged.

Histopathogenesis of dentigerous cyst is based on intrafollicular and extrafollicular theories. There can be no good reason for the extrafollicular theory of origin of dentigerous cysts, as the evidence is that those reported as arising in this manner all appear to be envelopmental or follicular odontogenic keratocyst (Shear, 2007). Intrafollicular theory postulates the possibility of cyst formation due to fluid accumulation between the layers of inner and outer enamel epithelium after the formation of crown. Another possibility is due to degeneration of stellate reticulum at an early stage of tooth development resulting into cyst formation associated with enamel hypoplasia. (Al-Tabani & Smith, 1980).

*Main's theory (1970):* The impacted tooth exerts pressure on the follicle which obstructs the venous outflow and induces rapid transudation of serum across the capillary walls. The increased hydrostatic pressure exerted by this pooling of fluid

causes separation of crown from the follicle with or without reduced enamel epithelium.

The osmolality of the cyst fluid is modified by increased permeability to glycosaminoglycans like hyaluronic acid, heparin & chondroitin sulphate which causes expansile growth rapidly (Browne & Smith et al, 1980).

Edamtsu et al (2005) examined the expression of Fas, bcl-2 & single stranded DNA (ss-DNA) in dental follicles to classify the possible role of these apoptosis related factors in the pathogenesis of dentigerous cyst. Fas is a cell surface glycoprotein that transmits apoptotic signals from the cell surface to the cytoplasm, while bcl-2 proto-oncogene encodes a protein that inhibits apoptosis. ss-DNA antibody recognizes DNA fragmentation in the nuclei during programmed cell death as well as Deoxy-Nucleotidyl transferase mediated dUTP-biotin nick end labeling (TUNEL).

These apoptosis related factors are detected in tooth germ tissues and several types of epithelial odontogenic cysts & tumors. Expression of Fas & single stranded-DNA was detected in superficial epithelial cells of both follicles & cyst. Expression of Ki 67 and bcl-2 was also found positive in basal cells.

Most dentigerous cysts are treated with enucleation of the cyst and removal of the associated tooth. Large dentigerous cysts may be treated with marsupialization when enucleation and curettage might otherwise result in neurosensory dysfunction or predispose the patient to an increased chance of pathological fracture. Occasionally it transforms to squamous cell carcinoma, mucoepidermoid carcinoma, or ameloblastoma from or in association with a dentigerous cyst (Banderas et al, 1996, Johnson et al, 1994; Eversole et al, 1975; Leider et al, 1985).

The prognosis for most histopathologically diagnosed dentigerous cysts is excellent, recurrence being a rare finding. In all dentigerous cyst, the microscopic features must be determined, to rule out its transformation in an ameloblastoma or, to squamous-cell carcinoma.

## Bibliography:

1. Al-Talabani NG, Smith CJ: Experimental dentigerous cysts and enamel hypoplasia: their possible significance in explaining the pathogenesis of human dentigerous cysts. *Journal of Oral Pathology*, 1980; 9: 82-91.

2. Banderas JA, Gonzalez MA, Ramirez F, Arroyo A: Bilateral mucous cell containing dentigerous cysts of mandibular third molars: Report of an unusual case. *Archives of Medical Research*, 1996; 27:327-329.
3. Browne RM: The pathogenesis of odontogenic cysts: a review. *Journal of Oral Pathology*, 1975; 4 (1): 31-46.
4. Browne RM, Smith AJ: Pathogenesis of odontogenic cysts. In: *Investigative Pathology of the Odontogenic Cyst*. CRC Press Boca Raton, 1991; pp. 88-109.
6. Edamatsu M, Kumamoto H, Ooya K, Echigo S: Apoptosis-related factors in the epithelial components of dental follicles and dentigerous cysts associated with impacted third molars of the mandible. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology and Endodontics*, 2005; 99: 17-23.
5. Eversole LR, Sabes WR, Rovin S: Aggressive growth and neoplastic potential of odontogenic cysts. With special reference to central epidermoid and mucoepidermoid carcinomas. *Cancer*, 1975; 35:270-282.
7. Johnson LM, Sapp JP, McIntire DN: Squamous cell carcinoma arising in a dentigerous cyst. *Journal of Oral & Maxillofacial Surgery*, 1994; 52:987-90.
8. Jones AV, Craig GT, Franklin CD: Range and demographics of odontogenic cysts diagnosed in a UK population over a 30-year period. *Journal of Oral & Pathology and Medicine*, 2006, 35: 500-507.
9. Kusakawa J, Irie K, Morimatsu M, Koyanagi S, Kameyama T: Dentigerous cyst associated with a deciduous tooth: A case report. *Oral Surgery, Oral Medicine Oral Pathology*, 1992; 73:415-418.
10. Leider AS, Eversole LR, Barkin ME. Cystic ameloblastoma. *Oral Surgery Oral Medicine Oral Pathology*, 1985; 60:624-630.
11. Main DMG: The enlargement of epithelial jaw cysts. *Odontologisk Revy*, 1970(b); 21: 29-49. as cited in Shear M, Speight P, (2007) *vide infra*.
12. Neville BW, Damm DD, Allen CM, Bouquot JE: Odontogenic cysts and tumors. In: *Oral and maxillofacial pathology*. BW Neville, DD Damm, CM Allen, JE Bouquot, (Eds.) 2<sup>nd</sup> Edn.; WB Saunders. Philadelphia, 2002. p. 589-642.
13. Shear M, Speight P: Cysts of the Oral and Maxillofacial Regions. 4th Edn.; Blackwell Publishing Ltd., 2007; pp 59-78.
14. Swerdloff M, Alexander SA, Ceen RF, Ferguson FS. Bilateral mandibular dentigerous cysts in a seven-year-old child. *Journal of Pedodontics*, 1980; 5:77-84.