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Review Article

BILATERAL DENTIGEROUS CYST OF MANDIBLE-A RARE ENTITY, CASE REPORT AND REVIEW OF LITERATURE

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ABSTRACT

Dentigerous cysts are developmental cysts that are usually associated with impacted mandibular third molar teeth. Bilateral occurrence of these cysts is very rare, the bilateral appearance is mostly seen in syndromic patients such as maroteaux-lamy (mucopolysaccharidosis type VI) syndrome and cleidocranial dysplasia. Their appearance in non syndromic patients is unusual. So far only 29 cases of bilateral dentigerous cysts are reported in literature with majority including pediatric patients. We report here a unusual case of bilateral dentigerous cyst involving impacted mandibular third molar on right side and impacted mandibular second molar on left side in a seventeen year old patient.

Key Words:

Bilateral, Dentigerous Cyst.

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INTRODUCTION

Dentigerous cysts are odontogenic cysts that are associated with unerupted teeth. These cysts enclose the crowns of unerupted teeth and are associated with cemento-enamel junction. The most commonly involved teeth are mandibular third molars. These cysts are usually asymptomatic and are noticed during routine radiographic examination. Their frequency in general population is 1.44¹. These cysts when enlarge sometimes expand the cortical plates. Bilateral occurrences of dentigerous cysts are rare and are usually associated with syndromes such as cleidocranial dysplasia and mucopolysaccharidosis.²

Case Report

A Seventeen year old male patient presented to the department of Oral and maxillofacial surgery, K L R Lenora institute of dental sciences with a chief complaint of swelling in the left side of mandible since two years. The swelling was 3x3 cm in size on left side. Firm in consistency, there was no pain associated with swelling. The swelling was non mobile, non pulsative, no signs of any discharge or sinus. There are no symptoms on right side. Intra oral examination reveals

unerupted 37 and 38 on left side of mandible and unerupted 48 on right side. The patient medical history was non-significant and showed no associated syndromes.

Radiographic examination of OPG (fig1)



OPG showing bilateral periapical radiolucency in mandible irt 36,37, 38 & 46, 47, 48. revealed a well defined radiolucency seen on right side with a horizontally impacted tooth attached laterally to the medial aspect and extending anteriorly from distal root of first molar (46), second molar 47 and impacted 48 posteriorly and superio- inferiorly from alveolar crest to lower

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border of the mandible, measuring more than 2.5 cm (approx) with a sclerotic border.

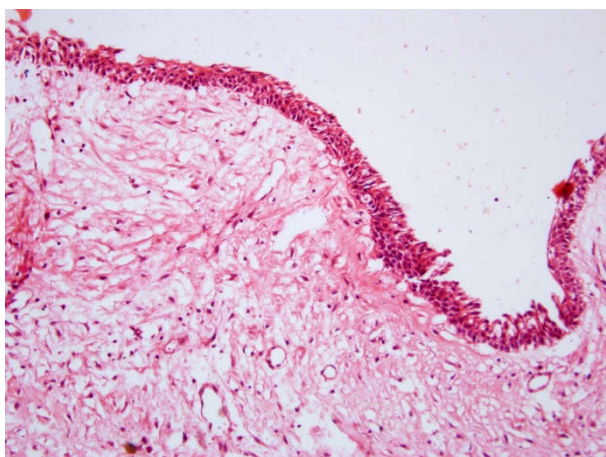
Another well defined radiolucency associated with submerged tooth 37, attached at the level of Cemento enamel junction (CEJ), extending anteriorly from medial root apex of 36 posterior to impacted third molar region 38. Superior inferiorly from alveolar crest to approximately 3 mm above lower border of the mandible measuring 2.5-3 cm, loss of lamina dura adjacent to 36, and involved tooth 37, surrounded by sclerotic border.

Treatment

Under general anesthesia incision placed along the ramus of mandible and buccal mucoperiosteal flap were raised and the cyst was carefully dissected out using blunt forceps and preserved on either side. Then the bone surrounding the impacted teeth was gutted with bur under copious saline irrigation. The impacted teeth were gently elevated from the socket and other teeth were removed normally with forceps. Extraction of 36, 37, 38 and 46, 47 and 48 were done. After teeth removal the follicles were enucleated. The entire surgical area was irrigated and closed with 3-0 silk sutures. The healing was uneventful. There was mild paresthesia on the left side of lower lip which healed in a month.

Histopathology Features

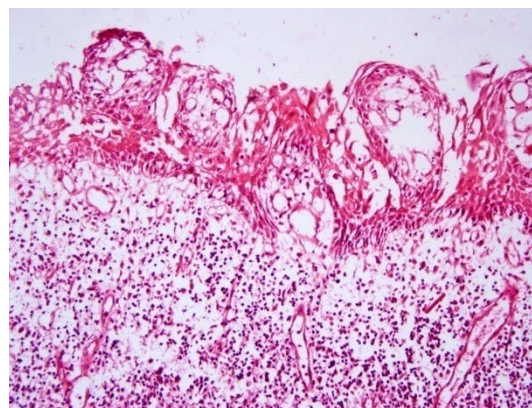
The H&E stained soft tissue section on left side (fig 2)



Photomicrograph of the lesion shows features suggestive of a dentigerous cyst (H&E, 20x magnification) on the left side. Shows epithelium of variable thickness and flat epithelial connective interface. Odontogenic epithelium is 2-4 layered seen lining the cystic cavity. Basal cells are cuboidal with centrally placed nucleus and squamous cells are seen in the superficial layer along with few mucous cells. The underlying connective tissue is dense fibrocellular with increased dilated blood vessels and cell rests were also observed. Diffuse minimal inflammatory cell infiltrate predominantly consisting of lymphocytes suggesting a dentigerous cyst.

The H&E stained soft tissue section on the right side (fig 3)

Photomicrograph of the lesion shows features suggestive of an inflamed dentigerous cyst (H&E, 20x magnification) on the right side. Shows non-keratinized stratified squamous epithelium with an arched pattern with intracellular edema.



The underlying connective tissue is loose cellular with increased dilated blood vessels and proliferating endothelial cells. Dense chronic inflammatory cell infiltrate predominantly of lymphocytes, plasma cells and few mast cells were evident suggesting an inflamed dentigerous cyst.

DISCUSSION

Dentigerous cysts are developmental cysts that occur in association with an unerupted tooth, most commonly mandibular third molars.² They also occur with maxillary third molars, canines and mandibular second premolars. They are defined as true developmental pathologies of jaws commonly seen in age group ranging from 10 yrs to 30 yrs.

There is a slight male predilection with prevalence in whites. Many of the lesions are asymptomatic and are usually identified during routine radiographic observation for missing teeth.⁵ The lesions are usually painless until secondary infection occurs.

The dentigerous cyst develops around the crown of the unerupted teeth by expansion of the follicle when the fluid collects or a space occurs between the reduced enamel epithelium and the enamel of the impacted teeth. Swelling, pain, tooth displacement, mobility and sensitivity may be present if the cyst reaches the size larger than 2cm.³

Bilateral dentigerous cysts are a very rare entity. They are usually associated with Maroteaux-Lamy (mucopolysaccharidosis type VI) syndrome and cleidocranial dysplasia, Basal cell nevus syndrome.⁴ Bilateral mandibular dentigerous cysts have also been reported after prolonged concurrent use of cyclosporine A and calcium channel blockers.⁴ Maroteaux-Lamy syndrome is a genetic disease resulting in the degradation of specific mucopolysaccharides. There is a deficiency of N-acetyl-4-sulphatase that results in impaired degradation of dermatan sulphate, which accumulates in tissues. Dental features include unerupted dentition, dentigerous cysts, malocclusions, condylar defects, and gingival hyperplasia.⁵ Cleidocranial dysplasia is an autosomal dominant disorder. The clinical features include absence of clavicles, frontal bossing, Multiple dentigerous cysts, maxillary micrognathia, prolonged retention of the primary dentition, delayed eruption of the permanent dentition, and unerupted supernumerary teeth.⁶ A dentigerous cyst rarely expands so rapidly that it presses on a sensory nerve and causes pain. When this occurs, the pain may be referred to any part of the face and is frequently described as a headache. Paresthesia, anesthesia, or mobile teeth are almost never produced.

Table List of Cases Reported In Literature

Author	Year	Age/sex	Cyst association	Treatment
Shiva Shirazian	2011	10/M	Second premolars	Marsupialization
Manoranjan Reddy	2011	11/F	Md primary molars	Surgical removal
Prabhakar	2011	10/M	Central incisor and canine	Enucleation
Harshaminder Kaur Grewal	2010	11/M	Mx primary molars	Enucleation
Sergio EV et al	2009	5/M	Md molars	Enucleation
Cury	2009	5/M	Md molars	Enucleation
Fregnani ER, et al	2008	5/M	Md first molars	Marsupialization
Yamalik	2007	51/M	Md third molars	Enucleation
DQ Frietas, et al	2006	13/M	Mx third molars and Md second molar	Enucleation
Garcia	2005	28/M	Md. Third molars	Extraction molars
Batra, Roychoudhury, Balakrishan, Parkash	2004	15/M	Md. third molars and second premolar	Enucleation
Ustuner, Fitoz, Atasoy, Erden, Akyar	2003	6/M	Mx. Canines	Enucleation
Shah, Thuau, Beale	2002	39/M	Md. third molars	No treatment
De Biase, Ottolenghi, Polimeni, Benvenuto, Lubrano, Magliocca	2001	8/M	Md. first molars	Enucleation
Ko, Dover, Jordan	1999	42/M	Md. third molars	Enucleation
Sands and Tocchio	1998	3/F	Md. central incisors and	Enucleation
Carr	1996	7/M	Md. first molar	Enucleation
Banderas and others	1996	38/M	Md. third molars	Enucleation
O'Neil and others	1989	5/M	Md. first molars	Enucleation
Eidinger	1989	15/M	Md. first molars	Enucleation
McDonnel	1988	15/M	Md. second premolar & second molar	Enucleation
Crinzi	1982	15/F	Md. third molars	Enucleation
Swerdloff and others	1980	7/M	Md. first molars	Enucleation
Burton and others	1980	57/F	Md. third molars	Enucleation
Callaghan	1973	38/M	Md. third molars	Enucleation
Stanback	1970	9/M	Md. first molars	Enucleation
Henerfer	1964	52/M	Mx. third molars	Enucleation
Tam	1955	7/M	Md. First molars	Enucleation
Myers	1943	19/F	Md. third molars	Enucleation

Radio graphically the dentigerous cyst presents as a well defined unilocular radiolucency often with sclerotic border. The radiolucency encircles the crown of tooth. Large dentigerous cyst present a multilocular appearance with the presence of trabeculae within radiolucency. Differential diagnosis include radicular cysts, odontogenic keratocysts, and odontogenic tumors such as ameloblastoma, Pindborg tumor, odontoma, odontogenic fibroma, and cementomas.

There are three variants of dentigerous cysts reported radio graphically. The central variety which surrounds the crown. Second is the lateral variety which develops laterally along the tooth root and third is the circumferential variety which involves the entire tooth.

The cysts vary greatly in size, from less than 2 cm in diameter to massive expansions of the jaws.⁷ The expansion may in turn produce gross deformity of the region involved. Although a slowly expanding cyst may markedly thin the cortical plates, it seldom erodes them. When the cortical plates are eroded, palpation reveals a rubbery, fluctuant, nonemptiable mass. Dentigerous cysts cause resorption of adjacent tooth roots in 55% of cases.⁸

The cysts may cause pathologic fracture and may become secondarily infected. Metaplastic and dysplastic changes may occur. An ameloblastoma, mucoepidermoid carcinoma, or squamous cell carcinoma may develop from the lining epithelium of a dentigerous cyst. Associated aneurismal bone cysts and hemangiomas have been reported in rare instances.⁹

Multiple dentigerous cysts appear as one of its kind. If multiple dentigerous cysts are found, the patient should be examined for basal cell nevus syndrome or cleidocranial dysplasia.

The bilateral dentigerous cyst involving mandible is very rare and with extensive research of the literature showed there are only twenty nine cases reported earlier and involvement of dentigerous cyst involving third molar on right side and involving second molar on left side is first reported here.

Most dentigerous cysts are treated by surgical enucleation and the removal of associated tooth. Large dentigerous cysts are also treated by marsupialization. A pathologic fracture is an inherent danger associated with a large cyst that has destroyed an extensive segment of the jaw. The jaw may become so weakened that it must be splinted before surgery to prevent fracturing during the procedure.¹⁰

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