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NON SYNDROMIC, BILATERAL, DENTIGEROUS CYSTS ASSOCIATED WITH INVERTED MANDIBULAR THIRD MOLARS: A CASE REPORT

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ABSTRACT

Dentigerous cyst is an epithelial-lined developmental crater that encloses the crown of an unerupted tooth at the cementoenamel junction. Bilateral or multiple dentigerous cysts are often associated with few syndromes. Even though dentigerous cysts are common developmental cysts, bilateral occurrence of dentigerous cysts are extremely uncommon and to date, only 22 cases have been reported in literature till 2011. In English based language literature review, we only found nine reports of non-syndromic bilateral dentigerous cyst associated with mandibular third molars. The present article report a case of bilateral non-syndromic, dentigerous cysts associated with inverted mandibular third molars in a 25 years old male patient with brief review of literature.

Keywords: Dentigerous cyst, Inverted teeth, Developmental cysts, Maroteaux-Lamy syndrome.

Contribution/ Originality

The paper contributes about the rare entity which is associated with various syndromes and systemic disorders, so careful examination and proper diagnosis is useful to rule out various systemic disorders & syndromes

1. INTRODUCTION

Dentigerous simply means having or containing teeth. Dentigerous cysts are developmental odontogenic cysts and are constantly associated with an unerupted or developing tooth bud. [1] They are the second most widespread odontogenic cysts subsequent to radicular cysts, accounts for approximately 24% of all true cysts in the jaws.[2] Dentigerous cysts are frequently discovered when radiographs are taken to investigate a failure of tooth eruption, a missing tooth

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or malalignment. There is usually no pain or discomfort associated with the cyst unless it becomes secondarily infected.[3]

Radiographs show a unilocular, radiolucent lesion characterized by well-defined sclerotic margins and associated with the crown of an unerupted tooth. While a normal follicular space is 3-4 mm, a dentigerous cyst can be suspected when the space is more than 5 mm.[4] Bilateral dentigerous cysts are unusual in the absence of an underlying syndrome or systemic disease. Bilateral and multiple cysts are usually found in association with a number of syndromes including cleidocranial dysplasia, Maroteaux-Lamy syndrome and in mucopolysaccharidosis.[5]

A widespread search of literature has been acknowledged very few reported cases. In English based language literature review, we only found only nine reports of nonsyndromic bilateral dentigerous cyst associated with mandibular third molars.[1] Even though this judgment may reflect the true rarity of the condition, it is plausible that bilateral dentigerous cysts are either under- recognized or under-reported.The present article report an unusual case of non-syndromic, bilateral dentigerous cysts associated with inverted mandibular third molars.

2. CASE REPORT

A 25 years old male reported to the department of Oral Medicine and Radiology with a complaint of pain in his left lower back tooth region since 2 months. History revealed pain in the left mandibular molars which was gradual in onset, throbbing type, intermittent in nature, aggravating during night time and relieved on taking medication. There was no history of systemic diseases or trauma. On general examination, the patient was apparently healthy and there were no other abnormalities were observed, suggestive of any syndromes. No extra-oral swellings or tenderness was elicited in relation to the left side of mandible. Bilateral sub-mandibular lymph nodes were palpable. The nodes were solitary, oval in shape, around 1x1 cm in diameter, freely mobile, firm in consistency and non tender on palpation. Intra oral examination revealed deep dental caries in relation to the occlussal surface of left mandibular second molar and clinically missing third molar.

A panoramic radiograph showed Bilateral, unilocular well-defined corticated radiolucencies of around 1x2 cm in diameter, surrounding both un-erupted mandibular third molars. The anterior border of both radiolucencies appeared to involve the distal root of the second molar (**Figure-1).**

Radiographic diagnosis of bilateral dentigerous cyst associated with inverted mandibular third molars was considered. The cysts were enucleated along with the associated third molars. The surgical sites showed good healing.

Histopathologically, microscopic sections of both specimens were similar, showing cystic wall composed of fibrous tissue and lined by 3-4 layers of stratified squamous, non-keratinized epithelium with Rushton bodies suggestive of dentigerous cyst (**Figure-2).**

3. DISCUSSION

A dentigerous cyst is one that encloses the crown of an unerupted tooth by expansion of its follicle and is attached to the neck of the impacted teeth. It is the second most widespread odontogenic cyst contributing about 16.6% to 21.3% of all odontogenic cysts. [2] The age range for the reported cases in the literature varies broadly, from 5 to 57 years of age.[3]

They most commonly involve the mandibular third molars, followed in order of frequency by the maxillary permanent canines, mandibular second premolars and maxillary third molars. Even though dentigerous cysts are common developmental cysts, bilateral dentigerous cysts are extremely uncommon [4].On data basis research (English language literature) only 23 cases of bilateral DC not associated with to syndromes were identified and just nine were associated to permanent mandibular right and left third molars [1]. We report the unusual occurrence of non-syndromic bilateral DC associated with mandibular impacted and inverted third molars.

The exact pathogenesis of the dentigerous cyst is not known. There are various theories on its pathogenesis. It is stated that the dentigerous cyst develops around the crown of an unerupted tooth due to accumulation of fluid either between the reduced enamel epithelium and enamel or in between the layers of the enamel organ. 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 thus 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. 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. [2]

Bilateral or multiple dentigerous cysts are usually associated with the Maroteaux- Lamy (mucopolysaccharidosis, type VI) syndrome and cleidocranial dysplasia and Gorlin-Goltz syndrome. [6] *Maroteaux-Lamy syndrome* is one of the mucopolysaccharidoses (MPS), a group of diseases ensuing from a genetic defect in the degradation of specific mucopolysaccharides. Dental features include unerupted dentition, dentigerous cysts, malocclusions, condylar defects, and gingival hyperplasia [6, 7].*Cleidocranial dysplasia* is an autosomal dominantly inherited disorder that results in a partial or complete lack of clavicles, short stature, frontal and parietal bossing, maxillary micrognathia, prolonged retention of the primary dentition, delayed eruption of the permanent dentition, and unerupted supernumerary teeth.[6] *Gorlin-Goltz syndrome* is autosomal dominant with a high penetrance and variable expressivity. It is caused by mutations in the PTCH gene, a human homologue of the Drosophila gene mapped to chromosome 9q21-23. It is

also known as basal cell nevus syndrome, an uncommon disorder, which is characterized by numerous basal cell carcinomas (seen in 50–97% of people with the syndrome), maxillary keratocysts (present in about 75% of patients) and musculoskeletal malformations.[8] In our case none of the above features of syndrome were present.

Dentigerous cysts are a benign condition and surgical removal of the lesion along with the tooth is the usual treatment for cyst-associated with impacted teeth. There are some cases of spontaneous regression if early diagnosed. However, removal of associated tooth and enucleation of soft-tissue components is definitive therapy in most instances. There are two main surgical procedures, marsupialization and enucleation. In our case, surgical enucleation of both cysts were performed under general anesthesia together with the extraction of the associated third molars. [7, 8]

In conclusion, multiple dentigerous cysts not associated with any systemic disease or syndrome is a rare condition. Thus, a detailed clinical and systematic examination should be done to rule out any associated syndrome. In some cases, the cystic lining may undergo transformation to mural ameloblatoma. Early diagnosis and enucleation of the cyst is important to reduce morbidity and avoid more aggressive surgical procedures.

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Figure Legends

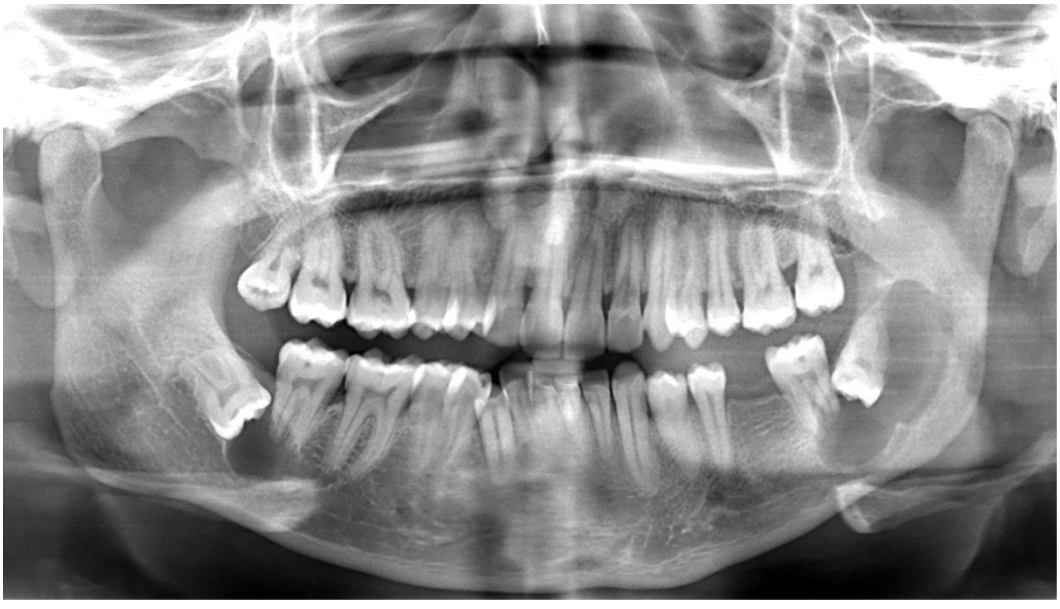

Figure-1.A. panoramic radiograph showing Bilateral, unilocular well-defined corticated radiolucencies surrounding unerupted, inverted mandibular third molars.

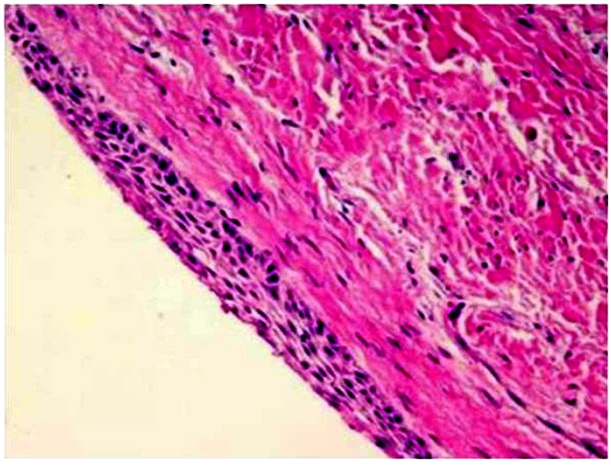

Figure-2. Histopathologically, microscopic sections of the specimens were showing cystic wall composed of fibrous tissue and lined by stratified squamous, non-keratinized epithelium with Rushton bodies.