

A Rare Case of Multiple Dentigerous Cysts in a non-syndromic patient: A Case Report and Literature Review

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Abstract

Multiple dentigerous cysts are rare and are typically associated with syndromes or systemic diseases. This article presents a case report of a non-syndromic patient with multiple dentigerous cysts, a presentation that is extremely uncommon. The case report is accompanied by a review of the relevant literature.

Keywords: Multiple Dentigerous Cysts; Enucleation.

Introduction

Dentigerous cysts are a type of odontogenic cyst that is typically associated with the crown of an unerupted or impacted mandibular third molar. They are relatively common and account for around 24% of all true cysts in the jaw [1]. Dentigerous cysts are usually asymptomatic, and they are often discovered incidentally during routine dental radiography [2]. However, multiple dentigerous cysts are typically associated with syndromes or

systemic diseases. This article presents a rare case of multiple dentigerous cysts in a non-syndromic patient and provides a review of the relevant literature.

Case Report

A 15-year-old female patient presented to the department of oral and maxillofacial surgery with complaints of pus discharge and pain in the lower left back teeth region and upper right jaw region. Upon evaluation, the patient was found to have cystic swelling in the bilateral mandibular third molar region, which extended up to the anterior border of the ramus. All third molars were impacted, and the patient's father had a history of a cystic lesion in the lower anterior region. General examination revealed no signs of systemic illness or associated syndromes.

A dental panoramic radiograph showed a well-defined unicystic radiolucent lesion surrounding the impacted third molars bilaterally, which extended up to the roots of the second molars. The impacted third molars were entirely enclosed within the lesion. The lesion in the maxilla was located between the right lateral incisor and the right first premolar region. A CECT face showed an expansile unilocular cystic lesion in the bilateral mandible around the crown of unerupted third molars, with evidence of dehiscence of the lateral wall of the left mandibular canal. Based on the clinical and radiological findings, enucleation under GA was planned. The patient was intubated nasally, and a crevicular incision was made with an anterior releasing incision in the mandible and maxilla. A mucoperiosteal flap was raised, and the cystic lining was identified. With the help of a curette, all the linings were detached, and the cyst was removed in its entirety. The associated mandibular impacted third molar was also extracted. Thorough irrigation of the cavity was performed using normal saline and povidone

iodine solution, and primary closure using silk 30 was done. The specimens were sent for histopathological examination after proper marking. Hematoxylin and Eosin-stained sections showed a cyst lined by non-keratinized squamous epithelium with chronic inflammatory infiltrates. The patient was followed up after 6 months, and an OPG showed satisfactory bone formation with no signs of recurrence.

Discussion

The present case involves a rare occurrence of multiple dentigerous cysts in a non-syndromic patient, affecting both impacted mandibular and maxillary canines. Dentigerous cysts are developmental cysts that usually arise from the follicular epithelium and are lined by nonkeratinized stratified squamous epithelium.[3] These cysts are commonly seen in mandibular third molars, maxillary canines, and mandibular premolars, and they rarely involve deciduous teeth. Bilateral presence of this cyst is very rare and usually occurs in association with syndromes such as Maroteaux-Lamy syndrome and cleidocranial dysplasia or long-term consumption of certain medications. [4-6]

Although multiple/bilateral dentigerous cysts are extremely rare in the absence of a syndrome or systemic disease, they can occur in patients of various ages and are most commonly seen in the second and third decades of life. Multiple dentigerous cysts in a non-syndromic patient are rare, with only few reported cases in literature.

A comprehensive search of English literature revealed only a few reported cases of multiple dentigerous cysts in non-syndromic patients, and most of them involved mandibular third molars[7-9]. Literature review by Freitas et al. has revealed only 17 cases of multiple dentigerous cysts in non-syndromic patients with most

of them involving the mandibular 3rd molars [10]. Saluja et al. reported a case of multiple dentigerous cysts involving multiple missing teeth in both maxillary and mandibular arches [11]. Tam-gadge et al. reported 21 cases of bilateral dentigerous cyst in their literature review [12]. However, in our case, the multiple dentigerous cysts involved impacted mandibular and maxillary canine and in a non-syndromic patient. These statistics point towards the true rarity of the condition. Reflecting the rarity of the condition, it is likely that multiple dentigerous cysts are either under-recognized or under-reported.

Comparing this case with previous similar cases published in literature, the present case is unique as it involves multiple dentigerous cysts affecting both impacted mandibular and maxillary canines in a non-syndromic patient, while previous cases mostly involved mandibular third molars. The rarity of multiple dentigerous cysts suggests that the condition is likely under-recognized or under-reported.

Radiographic examination showed a unilocular radiolucent lesion associated with the crown of an unerupted tooth with well-defined sclerotic margins. CT imaging may be necessary in cases of extensive lesions to determine origin, size, content, cortical plates, and relationship to adjacent anatomical structures.

The treatment of choice for dentigerous cyst is enucleation along with extraction of the impacted teeth. In pediatric patients, marsupialization may be considered to save the impacted tooth and developing tooth bud[13,14]. Tooth eruption potential is higher in children who have open apices in the involved teeth. Complete enucleation is important to ensure that no pathological lining is left behind.

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Legend Figures

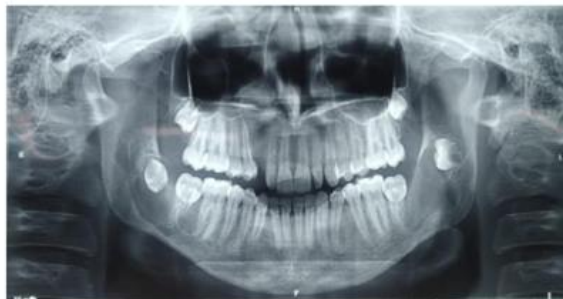


Image 1 : OPG showing unilocular radiolucency with respect to bilateral mandibular angle region and right maxillary anterior region



Image 2 : Axial section of CBCT face shows expansile unilocular cystic lesion in bilateral mandible around the crown of unerupted third molars

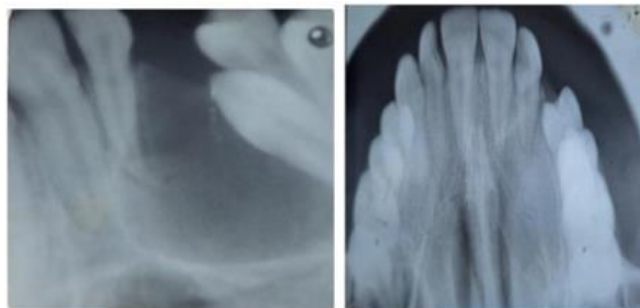


Image 3 : Occlusal and periapical view showing Maxillary Cyst in the Canine region previously operated three months back

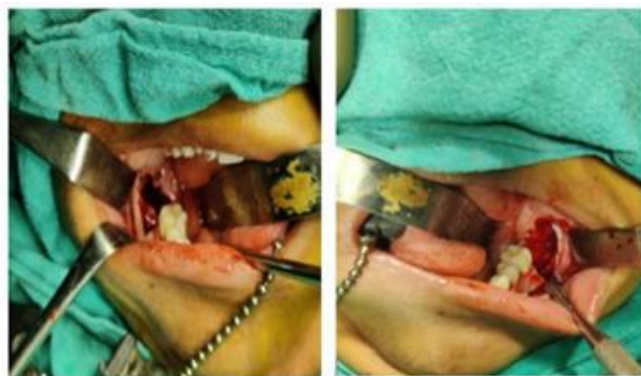


IMAGE 4 : Intra op photos showing enucleation of the cyst

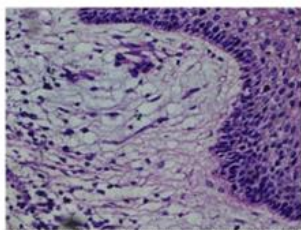


Image 5 : H&E section shows cyst lined by non keratinizing squamous epithelium with flattening of rete ridges. Subepithelium shows fibromyxoid stroma with chronic inflammatory cells.



Image 6: Post op radiograph showing formation of bone after enucleation at 6 month follow up period.