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Review

Bi-maxillary dentigerous cyst in a non-syndromic child – review of literature with a case presentation

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ABSTRACT

Dentigerous cysts are the most common developmental odontogenic cysts of the jaw, which usually occur in the second and third decade of life. It is most frequently associated with impacted mandibular third molar teeth and impacted canines. Bilateral/multiple dentigerous cysts are rare and typically associated with developmental syndromes. Non-syndromic dentigerous cyst occurring bilaterally or involving both arches at the same time is very rare. Here, we discuss the review of literature with a case of unusual occurrence of non-syndromic bi-maxillary dentigerous cysts in a child.

1. Introduction

Dentigerous cyst is defined as an epithelial-lined developmental cyst formed by accumulation of fluid between the reduced enamel epithelium and crown of an un-erupted tooth. It is formed due to an alteration in the reduced enamel epithelium, and encloses the crown of an un-erupted tooth at the cemento-enamal junction [1]. Dentigerous cysts are the second most common odontogenic cysts after radicular cysts, accounting for approximately 24% of all true cysts in the jaws [2]. Their frequency in the general population has been estimated at 1.44 cysts for every 100 un-erupted teeth [3]. It is most commonly associated with an impacted mandibular third molar, followed by the maxillary canine and maxillary third molar [3]. Dentigerous cysts are usually incidental discovery when radiographs are taken to investigate a failure of tooth eruption, a missing tooth or mal-alignment [4]. There is usually no pain or discomfort associated with the cyst unless it becomes secondarily infected. Radiographs show a unilocular, radiolucent lesion characterized by well-defined sclerotic margins and associated with the crown of an un-erupted 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]. Most dentigerous cysts are solitary.

Bilateral and multiple cysts are usually found in association with a number of syndromes including cleidocranial dysplasia, Maroteaux-Lamy syndrome [3] and in mucopolysaccharidosis [6]. In the absence of these syndromes, the occurrence of multiple dentigerous cysts is rare [3,7,8].

The present case is a rare occurrence of dentigerous cyst simultaneously in both the jaws associated with right maxillary canine and left mandibular second premolar in a non-syndromic child patient.

2. Case report

An 8-year-old boy visited the department for the evaluation of an asymptomatic, cystic lesion in the left mandibular region. Intraoral examination revealed a mixed dentition and clinically absent premolars and there was definite swelling in association with un-erupted premolars i.e., 34 and 35. Insignificant extraoral swelling or tenderness in relation to the mandible on same side was noted. The patient's medical history was non-significant and no associated syndromes were present. On panaromic radiographic examination, a cystic lesion surrounding the crown of 35 suggestive of radicular/dentigerous cyst was found (Fig. 1a). Other incidental finding was another quiescent dentigerous cyst on the contralateral side of maxilla in relation to 13 and 14 (Fig. 1a). Fine needle aspiration cytology (FNAC) of both side showed cholesterol crystals with inflammatory cells

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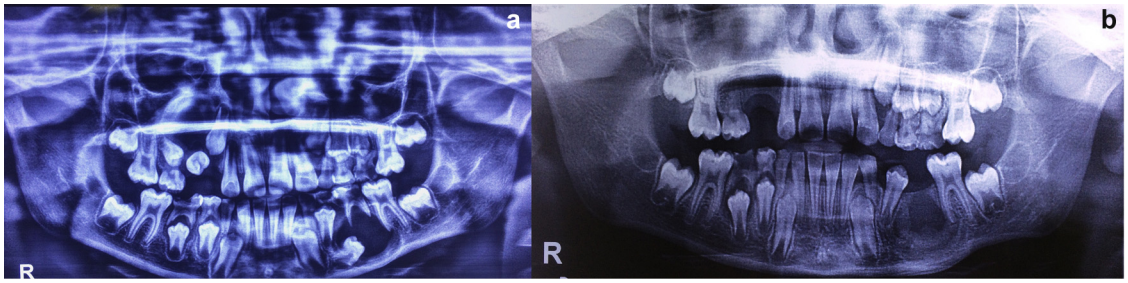

Fig. 1. Pre- (a) and postoperative (b) panoramic X-rays.

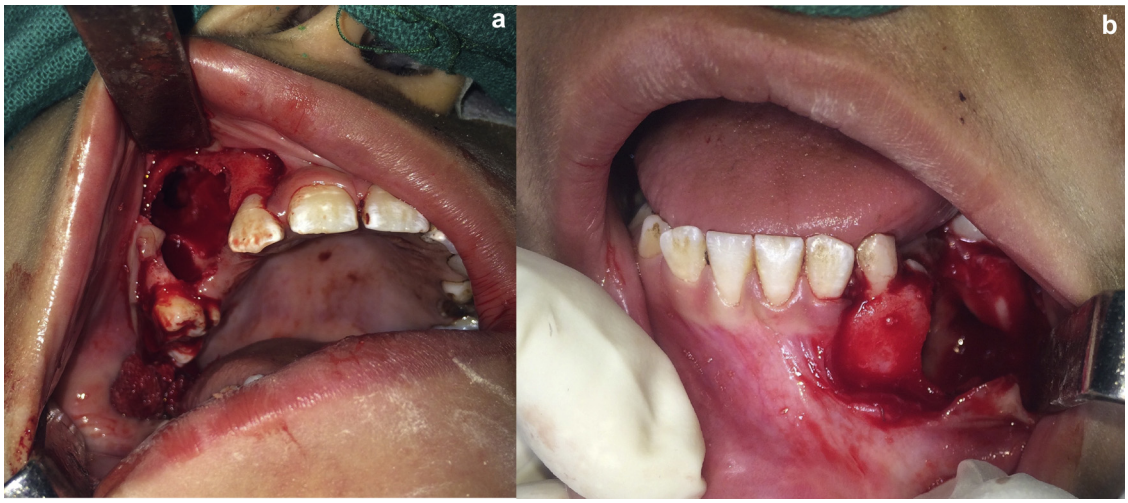

Fig. 2. Right maxillary (a) and left mandibular (b) surgical sites.

suggestive of cystic lesions. After clinical and radiological examination, a provisional diagnosis of bilateral dentigerous cyst was made; however, large periapical cyst, odontogenic keratocyst, adenomatoid odontogenic tumor and ameloblastic fibroma were also considered in the differential diagnosis.

Routine blood and urine investigations were within normal limits. Surgical enucleation of the cyst was planned as the treatment of choice and executed (Figs. 2 and 3). During surgical enucleation 75, 35, 13, 14, 15 were removed with the cystic lining and sent for histopathological analysis.

Histologically the two specimens were similar and showed a thin fibrous cystic wall lined by a 2–3 cell layers thick nonkeratinized stratified squamous epithelium. Rete pegs were absent and the connective tissue showed inflammatory cell infiltrate. Sub-epithelial layers showed parallel bundles of collagen fibers observed at periphery. These findings confirmed diagnosis of dentigerous cysts (Fig. 4). Follow-up period of one year showed no evidence of recurrence (Fig. 1b and Fig. 5).

3. Discussion

Dentigerous cysts are common developmental cysts, however, multiple dentigerous cyst in a non-syndromic patient is extremely rare. 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 [6]. Saluja et al. reported a case of multiple dentigerous cysts involving multiple missing teeth in both maxillary and mandibular arches [9]. Tamgadge et al. reported 21 cases of bilateral dentigerous cyst in their literature review [10]. Aher et al. [11] and Jae-Yun Jeon et al. [12]. reported cases of dentigerous cyst involving all four quadrants.

A comprehensive search of PubMed and English literature from 1943 to 2016 revealed that only 32 cases of bilateral dentigerous cysts in non-syndromic patients were reported; 24 cases occurred in the mandible, 3 in the maxilla, and 3 in both the mandible and maxilla [12–14]. Out of these 32 cases, we could trace only four

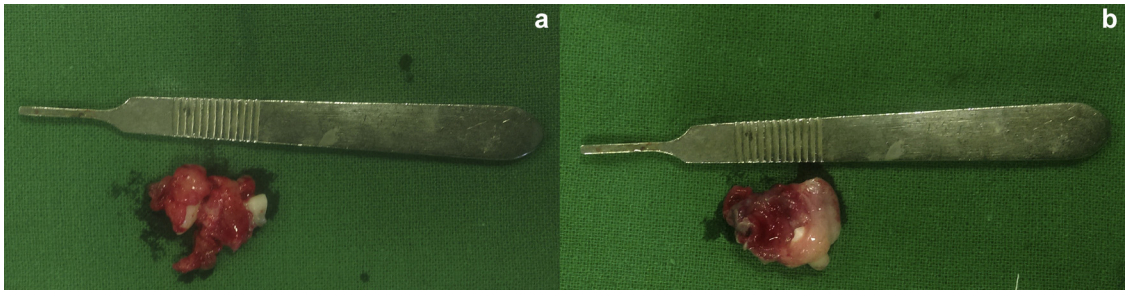

Fig. 3. Right maxillary (a) and left mandibular (b) specimens for histopathology.