

Non-Syndromic Bilateral Maxillary Dentigerous Cysts: A Report of An Unusual Case

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ABSTRACT

Background: Dentigerous cysts are benign odontogenic cysts that arise from the dental follicle of an unerupted or developing tooth. They are the second most common odontogenic cysts after radicular cysts. Bilateral dentigerous cysts are rare and occur typically in association with a developmental syndrome or systemic disease, such as mucopolysaccharidosis, cleidocranial dysplasia and basal cell nevus syndrome. The reported occurrence of bilateral dentigerous cysts in the absence of a syndrome is rare. One previous report of non syndromic bilateral maxillary cysts involving the canines is available in the literature. The following is a report of a case of bilateral non- syndromic dentigerous cysts associated with maxillary canine in a 10 year old boy. The present case report describes the surgical enucleation of dentigerous cysts, involving the permanent maxillary canines.

Keywords: Dentigerous cyst, Enucleation, Non-syndromic.

INTRODUCTION

A dentigerous cyst is an epithelial – lined developmental cavity that encloses the crown of an unerupted tooth at the cemento-enamel junction.^[1] Dentigerous, or follicular cysts, are the second most common odontogenic cysts after radicular cysts, and make up an estimated 14-20% of all jaw cysts.^[2] It develops by expansion of follicle when fluid collects or a space occurs between the reduced enamel epithelium and the enamel of an impacted tooth. Dentigerous cysts are always associated with an unerupted or developing tooth bud and are found most frequently around the crown of the mandibular third molars, maxillary canines, maxillary third molars and, rarely, the maxillary central incisor.^[3] Here, we report the case of an unusual occurrence of nonsyndromic bilateral dentigerous cysts associated with maxillary canine.

CASE REPORT

A 10 year old boy reported for evaluation of asymptomatic facial swellings in the right and left side that had started one month ago. On general examination patient was apparently healthy. On extraoral examination a diffused swelling was present over maxilla bilaterally obliterating nasolabial fold. Intraoral examination revealed bilateral swelling present over the maxilla with obliteration of the buccal vestibule. Mucosa over the swelling was normal. There were no associated syndromes or diseases present. Aspiration of the cystic cavities yielded a straw colored fluid. A panoramic radiograph showed bilateral thin sclerotic border surrounding well defined unilocular radiolucency on maxilla that was associated with the crowns of unerupted permanent right and left canine which was displaced laterally. (Fig: 1.A)

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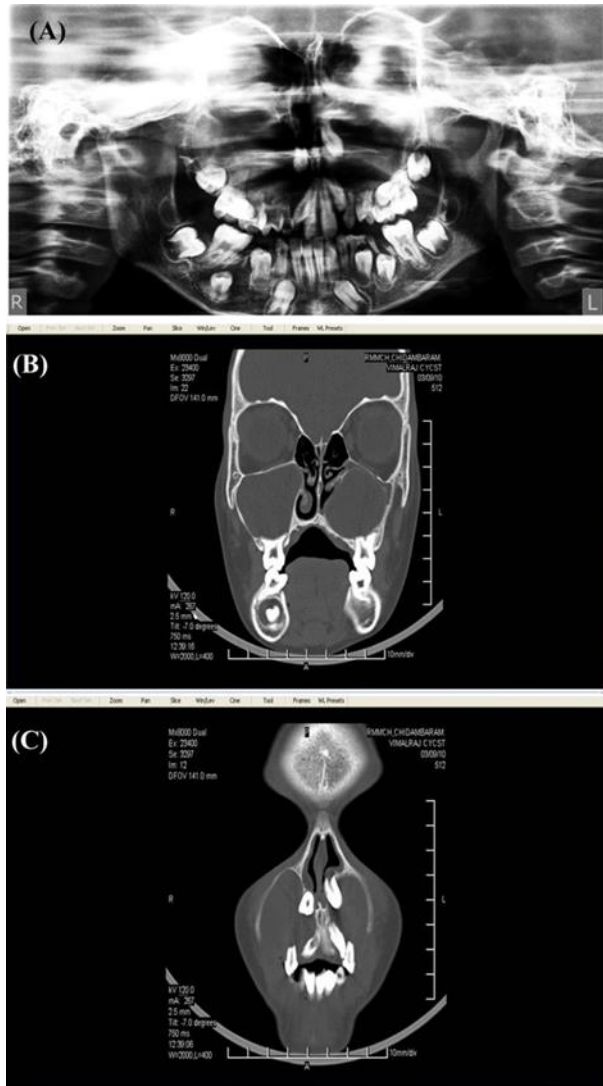


Fig 1: a. Pre- operative OPG showing well defined bilateral radiolucencies involving permanent canine. b & c. Coronal CT image showing well defined lesion with soft tissue attenuation containing impacted teeth.

Computed tomography (CT) showed well defined soft tissue masses displacing and obliterating both maxillary sinuses especially on the left maxillary sinus. The nasal turbinates were atrophied with left inferior turbinate pushed up. The bony walls were thinned and contained mural teeth structures. (Fig: 1. B&C) Both the cysts were enucleated along with the involved teeth, under general anesthesia with naso-endotracheal intubation. Crevicular incision was placed from first premolar of right side to first premolar of left side with vertical releasing incision placed in relation to right second premolar and left second premolar. Mucoperiosteal flap was elevated. Bone removal

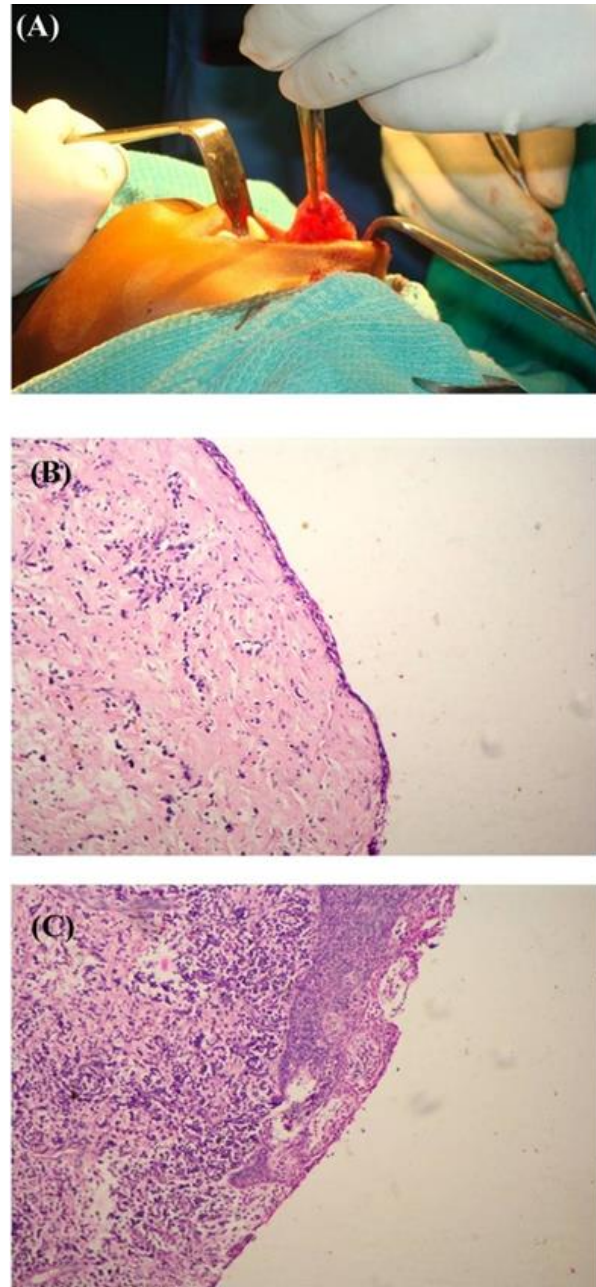


Fig 2: a. Intra operative photograph. b & c. Photomicrograph showing cystic lesions lined by the non-keratinized stratified squamous epithelium. Hematoxylin and eosin(H&E) stain with magnification 10X.

over the left and right side of the maxilla was done, window was created, cystic epithelial lining was removed along with the impacted teeth. Haemostasis was achieved using tincture benzoin soaked gauze. Patient was extubated uneventfully. (Fig: 2.A)

The specimens consisting of cystic sacs were submitted for histopathological examination measuring approximately 35x15x5 mm. Microscopic sections of both specimens were similar, showing cyst walls composed of fibrous tissue and lined by stratified squamous, non – keratinized epithelium. Connective tissue wall showed dense chronic inflammatory cell infiltration. (Fig: 2. B & C Hematoxylin and eosin(H&E) stain with magnification 10X)

The clinical, radiographic, intraoperative and histopathological findings lead to a final diagnosis of bilateral dentigerous cysts.

DISCUSSION

The key to the formation of a dentigerous cyst appears to be the accumulation of fluid either between the reduced enamel epithelium or between the layers of enamel organ. This fluid accumulation might be a result of pressure exerted by potentially erupting tooth on the follicle, which obstructs the venous outflow and induces serum transudation across the capillary wall.^[4] Dentigerous cysts most commonly occur in the second and third decades of life. These lesions can also be found in children and adolescents and show a male predilection.^[3] In the present case, the dentigerous cysts were associated with the permanent canines in a 10 year old boy. They are frequently discovered when radiographs are taken to investigate a failure of tooth eruption, a missing tooth or malalignment. 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 unerupted tooth. A dentigerous cyst can be suspected when the follicular space is more than 5 mm. Most dentigerous cysts are solitary. Bilateral and multiple cysts are usually found in association with a number of syndromes including cleidocranial dysplasia, mucopolysaccharidosis, basal cell nevus syndrome and Maroteaux-Lamy syndrome.^[1] In the present case report, patient was apparently healthy with no systemic diseases or syndromes.

Maxillary cysts may displace and obliterate the maxillary antrum and nasal cavities; mandibular cysts may cause paraesthesia of inferior alveolar nerve, or metaplastic and dysplastic changes.^[5] In

the present case there was obliteration of the maxillary antrum and nasal cavities as in CT imaging. Dentigerous cysts are frequently treated surgically, either by enucleation or marsupialisation. Following enucleation of the cyst and extraction of the unerupted tooth, prognosis is excellent and recurrence is rarely observed after complete removal.^[6] In the present case the cysts were completely enucleated along with the unerupted tooth.

A functional space maintainer later replaced by endosseous implants can lead to a successful functional and esthetic rehabilitation.^[7] In the present case after surgical enucleation complete oral rehabilitation followed by fixed orthodontic therapy was planned.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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