

Multiple Dentigerous Cysts: A Case Report and Review

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Abstract Dentigerous cysts are one of the common cysts of the jaws. Bilateral dentigerous cysts generally occur in association with a developmental syndrome or systemic diseases. Bilateral dentigerous cyst in absence of a syndrome is rare and to date only 18 cases have been reported. After reviewing the literature, no case reports of dentigerous cysts involving all the quadrants of the jaws were reported. As far as our knowledge, this is the first case to be reported.

Keywords Dentigerous cyst · Unerupted tooth · Unilocular · Panoramic radiography

Introduction

A dentigerous cyst is an epithelial—lined developmental cavity that encloses the crown of an unerupted tooth at the cemento-enamel junction [1]. It was reported in 1847, and was known as distended capsule, osseous cyst or serous cyst. These cysts are the second most common odontogenic cysts after radicular cysts [1–4]. They account for approximately 24% of all true cysts in the jaws [1].

Dentigerous cyst develops by accumulation of fluid between reduced enamel epithelium and enamel or within the enamel organ. Pressure exerted by an erupting tooth on an impacted follicle obstructs the venous outflow. This leads to rapid transudation of serum across the capillary walls. Increase in the hydrostatic pressure of the pooling fluid

occurs, leading to separation of follicle from the crown with or without reduced enamel epithelium. An intrafollicular spread of periapical inflammation from a deciduous tooth may also result in the development of dentigerous cyst [2].

Dentigerous cysts are frequently discovered when radiographs are taken to investigate a failure of tooth eruption, a missing tooth or malalignment. There is 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. While normal follicular space is 3–4 mm, a dentigerous cyst can be suspected when the space is more than 5 mm [1]. Bilateral and multiple cysts have been reported in patients with syndromes or systemic diseases. As noted in Table 1, there have been only 18 cases of multiple non-syndromic cysts reported in literature from 1943 to 2006 [5]. An unusual case of multiple dentigerous cysts is described here.

Case Report

A 17 year old male patient reported to the Department of Oral Medicine & Radiology with a chief complaint of displeasing appearance of upper front teeth, which he noticed since 5 years. He gave a history of retained maxillary anterior deciduous teeth. There was no relevant past dental, medical or family history and there was no associated syndromes or systemic diseases present.

Intraoral examination revealed that his maxillary central and lateral incisors, maxillary and mandibular canines and mandibular premolars were unerupted (Fig. 1). There were retained maxillary central incisors, lateral incisors and right canine and also mandibular canines, first and second

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Table 1 Review of literature regarding bilateral multiple dentigerous cysts [5]

| Authors/Year | Sex | Age (years) | Location | Treatment |
|--|-----|-------------|---------------------------------------|--------------|
| Myers/1943 | F | 19 | Md. third molars | Enucleation |
| Henefer/1964 | F | 52 | Mx. third molars | Enucleation |
| Stanback/1970 | M | 9 | Md. first molars | Enucleation |
| Callaghan/1973 | M | 38 | Md. third molars | Enucleation |
| Burton, Scheffer/1980 | F | 57 | Md. third molars | Enucleation |
| Swerdloff, Alexander, Ceen, Ferguson/1980 | F | 7 | Md. first molars | Enucleation |
| Crinzi/1982 | F | 15 | Md. third molars | Enucleation |
| McDonnell/1988 | M | 15 | Md. second premolar and second molar | Enucleation |
| Eidinger/1989 | M | 15 | Md. first molars | Enucleation |
| O'Neil, Mosby, Lowe/1989 | M | 5 | Md. first molars | Enucleation |
| Bandaras, Gonzalez, Ramirez, Arroyo/1996 | M | 38 | Md. third molars | Enucleation |
| Sands, Tocchio/1998 | F | 3 | Md. central incisors and first molars | Enucleation |
| Ko, Dover, Jordan/1999 | M | 42 | Md. third molars | Enucleation |
| De Biase, Ottolenghi, Polimeni, Benvenuto, Lubrano, Magliocca/2001 | M | 8 | Md. first molars | Enucleation |
| Shah, Thuau, Beale/2002 | M | 39 | Md. third molars | No treatment |
| Ustuner, Fitoz, Atasoy, Erden, Akyar/2003 | M | 6 | Mx. canines | Enucleation |
| Batra, Roychoudhury, Balakrishnan, Parkash/2004 | F | 15 | Md. third molars and second premolar | Enucleation |
| Frietas, Tempest/2006 | M | 14 | Mx third molars and Md second molar | Enucleation |

M Male, *F* Female, *Md* mandibular, *Mx* maxillary

molars on the right side and second molar on left side. There was a diffuse swelling involving the attached gingiva and vestibular mucosa, present in relation to the distal aspect of right lower lateral incisor till the mesial aspect of right lower deciduous first molar, measuring approximately 2×1 cm. The swelling was soft to firm in consistency and there was expansion of the buccal cortical bone in relation to right lower deciduous canine and second molar (Fig. 2).

Patient was advised for intra oral periapical radiographs in relation to 51, 52, 61, 62, 73, 83, 84 and panoramic radiograph, which revealed a well defined unilocular

radiolucency with sclerotic borders associated with the crowns of right maxillary central and lateral incisors, measuring approximately 2×2 cm. In the second quadrant, radiolucency was associated with left maxillary central, lateral incisors and canine, measuring 3×2 cm. Bilateral heart-shaped radiolucencies were present in the third and fourth quadrants associated with canine and second premolar in the third quadrant and first and second premolars in fourth quadrant measuring 3×2 cm and



Fig. 1 Intraoral photograph showing retained upper anterior deciduous teeth



Fig. 2 Intraoral photograph showing expansion of the buccal cortical bone in relation to right lower deciduous canine and second molar

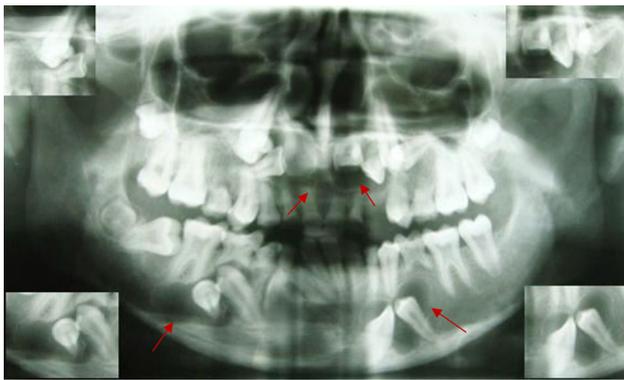


Fig. 3 Panoramic radiograph is showing bilateral radiolucencies in maxilla and mandible

4 × 2 cm respectively. There was radicular resorption of the retained deciduous maxillary and mandibular teeth (Fig. 3). Computed Tomography revealed, crowding and multiple unerupted teeth and 1.5 × 1 cm sized, cyst in right and left halves of maxilla and 2 × 1 cm sized cysts in right and left halves of mandible. CT imaging also showed the integrity and expansion of the buccal cortex (Figs. 4, 5, 6). The clinical and radiographic findings were suggestive of dentigerous cysts. Under general anaesthesia the dentigerous cysts in maxilla and mandible were enucleated and the unerupted permanent teeth were removed along with the cystic lining and the specimen was sent for histopathological evaluation. Microscopic examination of all the sections revealed that odontogenic epithelium surrounding



Fig. 5 3D volume of mandible showing multiple unerupted teeth

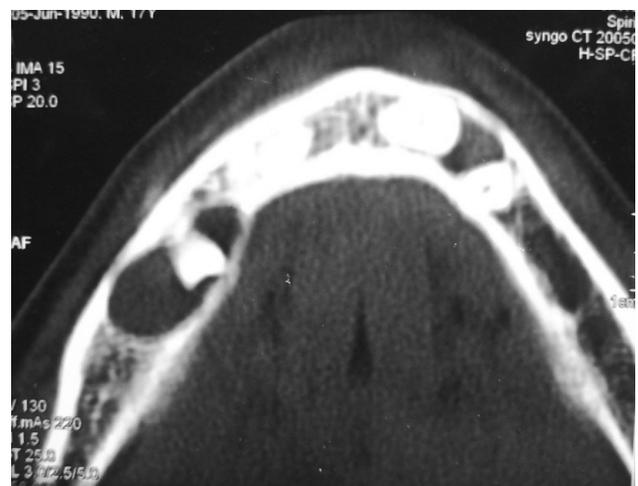


Fig. 6 Axial CT image shows the buccal and lingual cortical plates of the mandible to be intact



Fig. 4 Coronal CT image shows multiple unerupted teeth and radiolucencies in maxilla

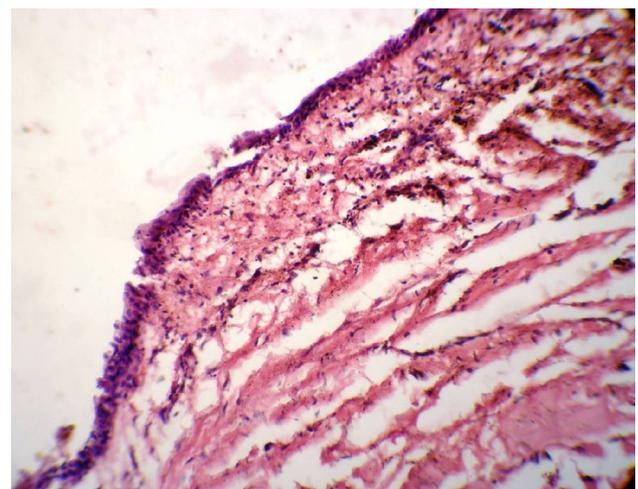


Fig. 7 Photomicrograph of the lesion showing that the odontogenic epithelium composed of flattened cells (haematoxylin and eosin, magnification 40×)

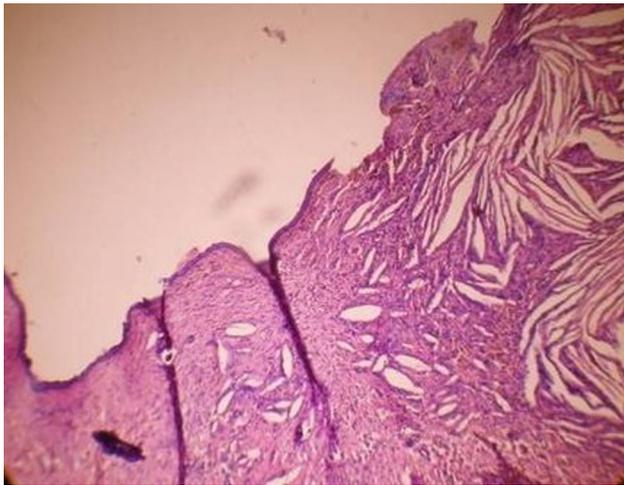


Fig. 8 Connective tissue stroma is showing severe chronic inflammatory cells and cholesterol clefts (haematoxylin and eosin, magnification 40×)

the cystic cavity to be composed of flattened cells and measured about three to five cells in thickness. Connective tissue stroma showed interlacing collagen fibres, fibroblasts and vascularity. Severe inflammatory cell infiltration and cholesterol clefts were seen in the stroma, suggestive of infected dentigerous cysts (Figs. 7, 8).

Discussion

Dentigerous cysts are benign odontogenic cysts associated with the crowns of permanent teeth. It may involve impacted, unerupted permanent teeth, supernumerary teeth, odontomas and rarely deciduous teeth [4]. They are usually present in the second or third decades of life and are rarely seen during childhood. The mandibular third molar and maxillary canines are involved most frequently [4, 5].

The review of literature (Table 1) reveals the involvement of mandibular third molars in eight cases and involvement of mandibular second premolar in two cases [6, 7]. Only one case has been reported to occur in association with lower central incisor [8]. In the present case, there is rare involvement of upper right and left central and lateral incisors and lower right first premolar tooth. The age range for reported cases varies widely, from 3 to 57 years [5]. In the present case patient was 17 year old, with no abnormal physical or laboratory findings suggesting any syndrome and there was occurrence of bilateral dentigerous cysts in both the jaws. In all reported cases, including the present case, radiographic examination showed a unilocular radiolucent lesion associated with the crown of an unerupted tooth and well defined sclerotic margins.

The earliest case of multiple dentigerous cysts was recorded by Glaswald in 1844 [9]. Bilateral and multiple cysts have been reported in Basal cell nevus syndrome, Mucopolysaccharidosis, Cleidocranial dysplasia and after prolonged concurrent use of cyclosporine A and calcium channel blockers [4]. Multiple Dentigerous Cysts have also been reported with Klippel Feil Syndrome [10].

Dentigerous cyst may cause displacement of adjacent teeth and resorption of teeth roots [11]. In the present case resorption of roots of the deciduous maxillary and mandibular teeth was present. It may displace or obliterate the maxillary sinus and nasal cavity, and cause paraesthesia of inferior alveolar nerve. The cyst's lining may contain areas of ortho-keratinization, ciliated cells, or mucin secreting cells. Because of this inherent ability for metaplastic change, some dentigerous cyst appear to progress to more aggressive lesions such as an odontogenic keratocyst, ameloblastoma, mucoepidermoid carcinoma or squamous cell carcinoma [2]. It is important to perform radiographic examinations in case of unerupted teeth. CT imaging gives information about origin, size, content, expansion of cortical plates and relationship of the lesion to adjacent anatomical structures [5]. Enucleation was the line of treatment in 17 of the 18 reported cases, although larger lesions may be surgically drained and marsupialized to relieve the pressure within the cysts and to prevent damage to the involved permanent teeth.

This case report emphasizes the importance of radiographic examination of all unerupted teeth, using panoramic radiography supplemented with computer tomography for a better delineation of the extent of the lesion and its relationship to adjacent anatomical structures.

Conflict of interest The authors would like to state that there is no area of conflict and consent was duly obtained from patient for publication.

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