Bilateral Dentigerous Cyst: An Unusual Case Report and Review of Literature

Shweta Tikekar, Shirish S Degwekar, Rahul R Bhowate

1Postgraduate Student, Department of Oral Medicine and Radiology, Sharad Pawar Dental College and Hospital, DMIMSU, Sawangi (M), Wardha, Maharashtra, India
2Professor and Head, Department of Oral Medicine and Radiology, Sharad Pawar Dental College and Hospital, DMIMSU, Sawangi (M), Wardha, Maharashtra, India
3Professor, Department of Oral Medicine and Radiology, Sharad Pawar Dental College and Hospital, DMIMSU, Sawangi (M), Wardha, Maharashtra, India

Correspondence: Shweta Tikekar, Postgraduate Student, Department of Oral Medicine and Radiology, Sharad Pawar Dental College and Hospital, DMIMSU, Sawangi (M), Wardha, Maharashtra, India, e-mail: sh_24@rediffmail.com

Abstract
Dentigerous cysts are the most common developmental cysts of the jaws, most frequently associated with impacted mandibular third molar teeth. Bilateral dentigerous cysts are rare and occur typically in association with a developmental syndrome. The reported occurrence of bilateral dentigerous cysts in the absence of a syndrome is rare and, to date, only 17 cases have been described. The following is a report of a case of unusual bilateral nonsyndromic dentigerous cysts associated with developing mandibular second premolars and a review of literature.

Keywords: Dentigerous cyst, mandibular second premolar, deciduous mandibular second molar.

INTRODUCTION
Dentigerous cysts are the most common developmental cysts of the jaws. They are one of the most prevalent types of odontogenic cysts associated with the crown of unerupted or developing permanent or deciduous tooth. These cysts are often asymptomatic unless there is an acute inflammatory exacerbation and therefore these lesions are usually diagnosed on routine radiographic examination. Swelling, tooth mobility, teeth displacement and sensitivity may be present if the cyst reaches a size larger than 2 cm in diameter.

Radiographically, the cyst appears as ovoid, well-demarcated, unicocular radiolucency with a sclerotic border surrounding the crown of an unerupted tooth. Histologically, the dentigerous cyst consist of a fibrous wall lined by nonstratified squamous epithelium consisting of myxoid tissue, odontogenic remnants and rarely, sebaceous cells.

Among the permanent teeth, most commonly the mandibular third molars are associated with dentigerous cyst, the other teeth that are commonly affected are in the order of frequency, the maxillary canines, maxillary third molars and rarely the central incisors.

The complications associated with dentigerous cyst include pathologic bone fracture, loss of permanent tooth, bone deformation and development of squamous cell carcinoma, mucoepidermoid carcinoma and ameloblastoma. Since the cyst may increase in size, the indicated treatment is surgical removal of lesion and involved teeth, or decompression to salvage the involved teeth.

Most dentigerous cysts are solitary. Single dentigerous cysts are the second most common odontogenic cysts after radicular cysts. Bilateral and multiple cysts have been reported in patients with syndromes or systemic diseases, such as mucopolysaccharidosis and cleidocranial dysplasia. There have been only 17 cases of multiple nonsyndromic cysts reported in the literature from 1943 to 2005, but none of these have been reported in developing mandibular premolars on both sides. This unusual case is one of the nonsyndromic bilateral dentigerous cysts associated with developing mandibular second premolars due to carious deciduous second molars on both sides.

CASE REPORT
An 11-year-old boy had visited the department of Oral Medicine and Radiology, SPDC, Wardha with a complaint of a diffuse swelling in lower right side of face since past 2 months. On general examination, patient was apparently alright. There was no relevant past medical history. Past dental history revealed severe caries with lower left primary second molar for which patient had undergone root canal treatment. Extraoral examination revealed a bony swelling which caused a bulging of cortical bone. Initially the swelling was small of approximately 0.5 cm × 0.5 cm and gradually increased in period of 2 months to attain the present size of approximately 1.5 cm × 1.5 cm. There was no history of pain or pus discharge or bleeding from the swelling.

Intraorally, there was grossly carious 85. A diffuse swelling was associated with 85 obliterating buccal vestibule (Fig. 1). There was no tenderness on palpation and the consistency was firm. There was no pus discharge or bleeding from the swelling. Pain on percussion with 85 was negative.
Bilateral Dentigerous Cyst: An Unusual Case Report and Review of Literature

On radiographic examination, panoramic view showed a well-defined radiolucent lesion in periapical region of 85 with hyperostotic borders extending from periapical region of 85 up to mesial root of 46 involving the crown of developing permanent second premolar. The lamina dura of 85 was resorbed. On left side, 75 showed radiographic evidence of inadequate radiopaque root canal filling and a similar well-defined radiolucent lesion in periapical region with 75 involving developing tooth bud with 35 of size approx 1 cm × 0.5 cm was also noticed (Fig. 2).

Occlusal radiograph showed expansion of buccal cortical plate in association with 85 with thin sclerotic borders (Fig. 3).

After clinical and radiological examination, a provisional diagnosis of periapical cyst with 85 and 75 was given. Among the differential diagnosis dentigerous cyst with 35 and 45 were included. Since the lesion was associated with carious deciduous primary second molars and was involving the developing tooth buds of permanent second premolars. Marsupialization of the cyst with extraction of primary second molars was planned. Prior to surgery routine blood investigations were performed, which were within normal limits. The surgery was done under local anesthesia and under antibiotic cover. The right cyst was surgically marsupialized first with the extraction of primary tooth and after the gap of 15 days the cyst of left side was treated in the same manner (marsupialized). The cysts were attached to cementoenamel junction of left and right second premolars. The specimen were sent for histopathologic examination. Biopsy report of both specimen were suggestive of dentigerous cyst.

DISCUSSION

A dentigerous cyst can be defined as a cyst that encloses the crown of an unerupted tooth, expands the follicle and is attached to cement enamel junction of the unerupted tooth. Dentigerous cysts account for more than 24% of jaw cysts. The substantial majority of dentigerous cyst involves the mandibular third molars and the maxillary permanent canines, followed by mandibular premolars, maxillary third molars and rarely central incisors. Mourshed stated that 1.44% of impacted teeth undergo dentigerous cyst transformation. Daley et al reported an incidence rate of 0.1 to 0.6%, whereas Shear found the incidence to be 1.5%.4

The exact histogenesis of the dentigerous cyst is not known. It is stated that the dentigerous cyst develops around the crown of an unerupted tooth by accumulation of fluid either between the reduced enamel epithelium or in between the layers of enamel organ. This fluid accumulation occurs as a result of pressure exerted by the erupting tooth on an impacted follicle, which obstructs the venous outflow and thereby induces rapid transudation of serum across the capillary wall.8 Toller9 stated that the likely origin of the dentigerous cyst is the breakdown of proliferating cells of follicle after impeded eruption. These breakdown products result in increased osmotic tension and hence cyst formation. Bloch10 suggested that the origin of 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 exudates ensues and dentigerous cyst forms. In the present case, there are dentigerous cysts with unerupted mandibular second premolars with overlying grossly carious deciduous second molars.
Although dentigerous cysts are common developmental cysts, reported bilateral dentigerous cysts are extremely rare. Bilateral or multiple dentigerous cysts are usually associated with the Maroteaux-Lamy (mucopolysaccharidosis, type VI) syndrome and cleidocranial dysplasia. Both are developmental conditions that are detected in young individuals with stigmata of the syndromes. Maroteaux-Lamy syndrome is one of the mucopolysaccharidoses (MPS), a group of diseases resulting from a genetic defect in the degradation of specific mucopolysaccharides. With this syndrome, there is a deficiency of N-acetyl-4-sulphatase that results in impaired degradation of dermatan sulphate, which accumulates in tissues and is excreted in the urine. Dental features include unerupted dentition, dentigerous cysts, malocclusions, condylar defects, and gingival hyperplasia. Cleidocranial dysplasia is an autosomal dominantly inherited disorder that results in a partial or complete absence 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. Multiple dentigerous cyst formation occurs in both conditions and can develop at any site in the upper or lower jaws.

Bilateral dentigerous cysts are extremely rare in the absence of a syndrome or systemic disease. After searching the literature, only 17 cases were identified from 1943 to 2005. Twelve of 17 cases have been associated with mandibular molar teeth, with seven of these associated with third molar teeth and five associated with first molar teeth. None of them presented in association with developing mandibular second premolar teeth as in the present case that had two cysts: one associated with right mandibular second premolar and one with left mandibular second premolar. Thus in the present case, two dental quadrants were involved.

The age range for reported cases varies widely, from 3 years to 57 years of age. The mean age of the 17 cases was 22.5 years. Ten of them occurred in children under the age of 15 years. This was so in the present case too, since the patient was 10 years old. They usually present at this age because of the tooth eruption chronology.

Patients frequently present unerupted teeth or asymptomatic slow-growing swellings. As in this case, there was asymptomatic slow growing swelling in right side and no clinical evidence of the cyst on left side which was evident when a panoramic radiograph was taken.

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. But a radiograph cannot differentiate whether the resorption of lamina dura is the result of periapical pathology related to nonvital tooth or is the result of pressure resorption from expanding pathology in adjacent region, hence pathological analysis of the lesion is essential for the definitive diagnosis. Other lesions may share the same radiological features as dentigerous cysts, such as periapical cyst (if overlying nonvital deciduous tooth is present), odontogenic keratocysts and unicystic ameloblastoma. Although involvement of the tooth, cortical expansion and radicular resorption are characteristics more related to dentigerous cysts, other lesions were not excluded until the results of the pathological analysis were known. Odontogenic keratocysts do not expand the bone to the same degree as dentigerous cysts and are less likely to produce teeth resorption. According Tsukamoto et al., the mean age of patients with odontogenic keratocyst was less than that of patients with dentigerous cyst; the mean area of the odontogenic keratocysts was larger than that of dentigerous cysts; and dentigerous cysts are more likely to have smooth periphery and odontogenic keratocysts are more likely to have scalloped periphery. It is not possible to differentiate unicystic ameloblastomas from dentigerous cysts with clinical and radiographic examinations.

Unlike other odontogenic cysts, the epithelial cells lining the lumen of dentigerous cyst possesses an unusual ability to undergo metaplastic transition. On occasion, some dentigerous cysts rarely transform into an odontogenic tumor, e.g. ameloblastoma or a malignancy like squamous cell carcinoma. Enucleation was the treatment in 16 of 17 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. In present case, marsupialization of both dentigerous cysts were performed with extraction of overlying nonvital deciduous second molars.

REFERENCES