

Inflamed Dentigerous Cyst: A Case Report and Review

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ABSTRACT

Context: Developmental odontogenic cysts account for 25% of all odontogenic cysts of the jaw. They are commonly associated with impacted or embedded teeth like mandibular/maxillary third molar and maxillary canines.

Dentigerous cyst (DC) is a common developmental odontogenic cyst, but its inflammatory variant is quite rare.

Settings and design: The representative tissue received was 10% formalin fixed and was 3×2 cm in diameter.

Materials and methods: Sections of 5 µm thickness were obtained from paraffin-embedded tissues that had been processed and stained with routine hematoxylin and eosin (H&E) stain. These stained sections were then reviewed.

Conclusion: Examination of paraffin-embedded sections showed hyperplastic epithelial lining with rete ridges and the connective tissue component with infiltration of chronic inflammatory cells. The treatment of choice for inflamed DC is enucleation, which shows good prognosis.

Keywords: Infiltrating lipoma, Intramuscular lipoma, Skeletal muscles, Tongue.

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INTRODUCTION

Jaws anchor a wide variety of cysts and neoplasms, due in large part to the tissues involved in tooth formation.¹ Odontogenic cysts represent the most common form of cystic lesions affecting the maxillofacial region. Dentigerous cyst (DC) constitutes the most common developmental odontogenic cyst and accounts for approximately 25% of all odontogenic cysts of the jaws.

Their frequency estimated in general population has been at 1.44 for every 100 unerupted teeth.² Usually, no symptoms are found to be associated with DCs unless there is an infection, when it is followed by a painful swelling. A late, non-eruption of tooth could suggest the possibility of an underlying cyst. A DC can expand causing facial asymmetry, bony expansion, tooth malpositioning, and sensitivity.³ As with other cysts, DC can also bring about cortical plate expansion and may cause involvement of teeth and subsequent destruction of the tissues as it expands.⁴ Third molars, canines, and second premolars are the teeth that are most involved in their descending order of occurrence.^{4,5} Radiographically, DCs show typically unilocular radiolucency with a well-defined sclerotic border associated with the crown of an unerupted tooth, but an infected cyst will show ill-defined borders.^{6,7} Histopathologic observations have shown that the lining of DC has the potential to develop into an aggressive ameloblastoma; therefore, early detection and removal of the cyst is required to prevent the foreboding associated with the lesion as the prognosis is excellent and recurrence is rare if completely removed.⁸

CASE REPORT

An 8-year-old boy was referred to the Department of Pedodontics at the Royal Dental College, Chalissery, Palakkad, Kerala with a chief complaint of swelling in the upper left posterior teeth. Extraorally, a mild swelling that was slightly tender and hard on palpation was present (Fig. 1). Intraoral examination revealed dentoalveolar abscess in relation to maxillary left primary first molar.



Fig. 1: Extraoral swelling involving the left side of the face

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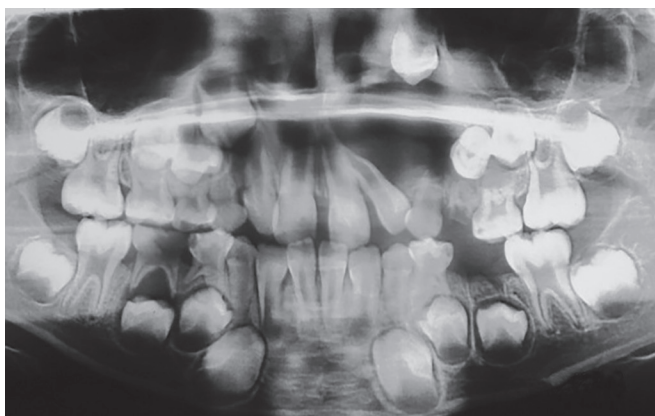


Fig. 2: Preoperative orthopantomogram showing enlarged follicular space in relation to first premolar on the same side extending toward the permanent canine

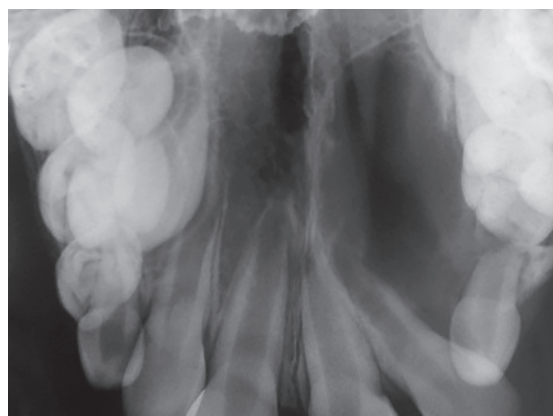


Fig. 3: Maxillary occlusal radiograph showing extension of radiolucent lesion up to maxillary first molar and midline of palate

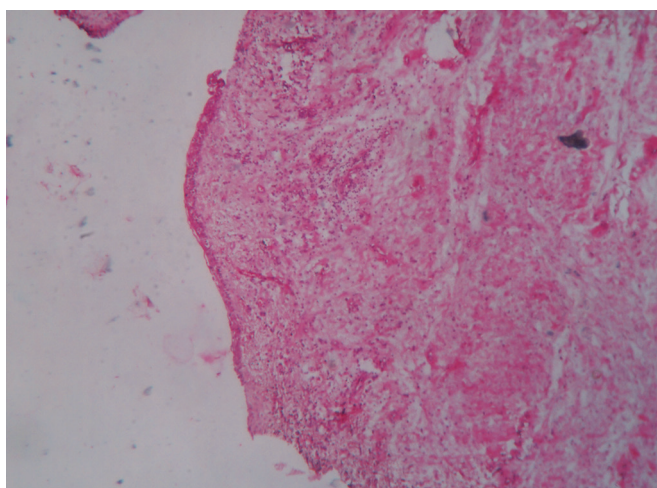


Fig. 4: Thin, nonkeratinized epithelial lining of 2 to 3 cell layer thickness (H&E 4x)

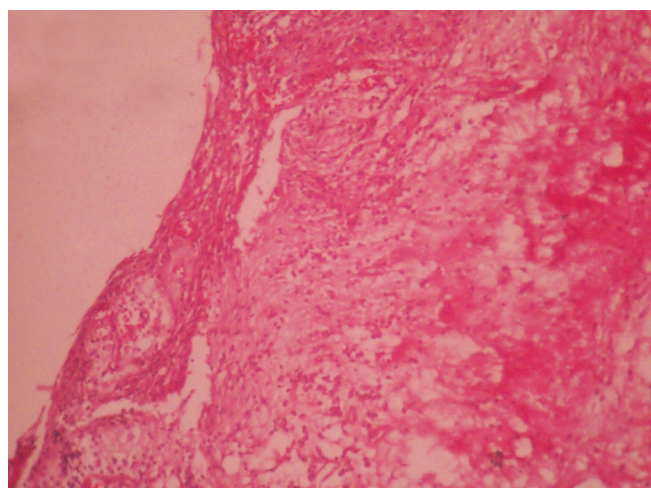


Fig. 5: Hyperplastic epithelial lining with cystic spaces and exocytosis. The underlying connective tissue shows diffuse infiltration of chronic inflammatory cells (H&E 10x)

A panoramic radiograph was taken to further assess the condition and it revealed an enlarged follicular space in relation to the first premolar on the same side, which was extending toward the permanent canine (Fig. 2). To get a clearer and more specific view of the maxillary area involved, an occlusal radiograph was taken, which revealed a huge radiolucent area extending superiorly up to the permanent first molar and medially up to the midline of the palate (Fig. 3). Fine needle aspiration cytology was done, and the aspirate was blood-tinged yellowish fluid. It was reported as an inflamed cyst.

As the cystic lesion was large in diameter, an attempt to save the premolar tooth by marsupialization of the lesion followed by insertion of an acrylic plug was planned. A part of the lining epithelium was incised for histopathologic evaluation. Since the biopsy report came as radicular cyst, surgical enucleation of the cystic lesion under general anesthesia was planned after 2 weeks.

Prior to surgery, routine blood, serology, and urine examinations were performed. The results were within

the normal limits. Surgical enucleation of the cyst was done and since the lesion was attached to the cement-enamel junction of maxillary left first premolar tooth, it had to be sacrificed. Microscopic examination of the lesion under low power showed a cystic lumen lined by thin nonkeratinized epithelium of 2 to 3 cell layer thickness (Fig. 4). Few areas within the lesion showed hyperplastic epithelial lining with cystic spaces and exocytosis (Fig. 5). Diffuse infiltration of chronic inflammatory cells was noted within the underlying connective tissue. The histopathological findings of the jaw lesion were suggestive of an inflamed dentigerous cyst. Then, the patient was recalled after 1 week for suture removal and follow up was done clinically and radiographically for 6 months.

DISCUSSION

A DC or follicular cyst is one of the most common type of developmental odontogenic cysts. As the term dentigerous literally means "tooth bearing," they are associated with the crown of impacted, embedded, or partially erupted

tooth.⁹ Dentigerous cyst is commonly associated with male population in the second or third decade of life, and about 70% of cases are noted involving the mandible and 30% the maxilla.⁷ The frequency of DCs cited in children has been low in the dental literature. Shear¹⁰ has estimated about 9% while Donath¹¹ about 4% of DCs to occur in the first decade of life. Though the pathogenesis of DC appears to be acceptable widely as developmental, two types of DCs are reported, namely, developmental and inflammatory in origin. Developmental type of cyst develops in a mature permanent tooth as a result of fluid accumulation, whereas the inflammatory counterpart develops in an immature permanent tooth.¹² The developmental histopathogenesis of DC is constructed on the bases of intrafollicular and extrafollicular theories. The extrafollicular theory of origin of DC does not hold good as the evidence reported for this origin is more inclined to be envelopmental or follicular odontogenic keratocyst. The intrafollicular theory postulates the possibility of cyst formation due to accumulation of fluid between the layers of inner and outer enamel epithelia after crown formation or that it can be attributed to the degeneration of stellate reticulum at an early stage of tooth development resulting in the cyst formation associated with enamel hypoplasia. Main's intrafollicular theory contributed to the same theory of developmental origin explaining that the pressure exerted by the impacted tooth on the follicle obstructs the venous outflow and induces rapid transudation of serum across the capillary walls, which in turn can increase the hydrostatic pressure thus causing the separation of crown from the follicle with or without reduced enamel epithelium. However, in addition to these views on the developmental origin, periapical inflammation of nonvital deciduous teeth has also been suggested as a factor triggering the formation of inflammatory DC (IDC) of the unerupted permanent successors.¹³ The inflammation present at the root apex of a nonvital primary tooth spreads to involve the follicle of the unerupted immature permanent successor. Therefore, the appearance of IDC is most commonly found involving the mixed dentition stage.¹² Consequently, the findings in our case reveal the cyst as an inflamed DC and can postulate that the necrotic pulpal inflammation of the primary maxillary first molar might have stimulated the development of a DC of the successor tooth.

Benn and Altini¹⁴ considered three possible mechanisms in the histogenesis of IDCs:

1. Intrafollicular developmental cysts formed around the crowns of permanent tooth that become secondarily inflamed, as a result of periapical inflammation spreading from nonvital deciduous predecessors.
2. Radicular cysts at apices of nonvital deciduous teeth that fuse with the follicles of unerupted permanent

successors. "Eruption" of successor teeth into the cystic cavity results in the formation of extrafollicular DC.

3. Periapical inflammation from any source, but usually from nonvital deciduous teeth spreading to involve follicles of unerupted permanent successors.

Dentigerous cysts are usually small asymptomatic lesions that are an incidental finding on routine radiographs; hence, when the cyst is smaller in size, it would be difficult to differentiate it from a larger but normal dental follicle. A working definition to rule out this radiographic confusion is that, a DC exists only when the distance between the crown and dental follicle is >2.5 to 3.0 mm.⁷ However, some DC may grow to considerable size causing painless bony expansion until secondary infected.¹³ Radiographs alone may not be sufficient to show the full extent of the lesion, and computed tomography (CT) imaging may also be necessary to avail the exact information about the lesion's size, content, and origin. Koca et al¹⁵ preferred panoramic radiographs for imaging in all cases. In this case, an additional CT scan was also required to assess the extent of the cystic lesion, and panoramic radiographs were preferred for the periodic follow-up visits.

Differential diagnosis of an odontogenic keratocyst, unicystic ameloblastoma, and radicular cyst must be considered in such cases corresponding with the radiographic details, but the incidence of all of the above lesions is rare in the first decade of life. Since radiographs alone cannot differentiate the abovementioned lesions, a histopathologic examination should be performed wherever possible.¹² However, as proposed by Kozelj and Sotosek¹⁶ leaking out of cystic fluid during an extraction of a primary tooth or during a decompression respectively, confirms the clinical impression of the cyst, but to ascertain the type of cyst for proper management and to prevent morbidity, histopathologic confirmation is mandatory.

The histopathology of DCs varies greatly depending on whether the cyst is inflamed or not. A noninflamed DC is lined by cuboidal or low columnar 2 to 4 cell layer thick epithelial lining, which is derived from reduced enamel epithelium and associated with loosely arranged fibrous connective tissue wall derived from the dental follicle of developing enamel organ which is rich in acid mucopolysaccharides. In contrast, the epithelium of inflamed DC demonstrates hyperplastic epithelium with rete ridges and the fibrous cystic capsule with inflammatory infiltrate. Metaplastic changes are occasionally noted within the epithelial lining in the form of mucous-producing cells or secretory cells, such as goblet cells. Pseudostratified ciliated columnar epithelium has also been reported.⁸

Treatment of DC depends on size, location, and disfigurement and often requires bone removal to ensure total removal of cyst especially in case of large ones.¹⁷ These cysts are frequently treated surgically, either by enucleation or by marsupialization. Marsupialization or decompression technique has been advocated widely for the treatment of DC in young patients.¹²

Marsupialization of cystic lining creates an accessory cavity to relieve intracystic pressure and accelerate the healing of the cystic lesion.¹² In this case too, marsupialization was the initial treatment of choice, which would reduce the size of the lesion to bring about repositioning of canine and to save 24. Following the treatment, the patient was reviewed for 6 months and considerable reduction in size of the lesion was noticed.

CONCLUSION

When a child patient presents with bony swelling associated with any tooth, the differential diagnosis should be DC, radicular cyst, odontogenic keratocyst, ameloblastomas, odontogenic fibromyxoma, odontomas, and cementoma. Ameloblastomatic transformation within the cystic space is the most important factor to be considered during the treatment planning of these cases.

Marsupialization is a preferred treatment modality especially in young children and long-term follow-up of cases treated with marsupialization usually reveals reduction in size of the lesion and normal eruption of involved teeth. But, in case of infected cysts, enucleation is a better choice of treatment especially in growing children to minimize disfigurement and to avoid complications.

CONSENT

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

AUTHORS' CONTRIBUTIONS

The team of surgeons from the Departments of Pediatric and Preventive Dentistry excised the cyst and Dr. Anuradha Sunil, Dr. Archana Nair, and Dr. Dafniya have contributed

in analyzing, reading, compiling, and writing this article. Dr. Nidhu Thambi has done the research for the article.

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