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# A Rare Case of Dentigerous Cyst in a One Year Old Child: The Earliest Known Reported Occurrence

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**Abstract** Dentigerous cysts are developmental odontogenic jaw cysts, commonly manifesting in the second and third decades of life. Very few of these cysts have been reported in children younger than 10 years of age. This article describes a rare case of dentigerous cyst in a 1-year-old boy, the youngest case to be documented. The clinical, radiographic and histopathologic features are discussed; the increased possibility of occurrence of these cysts at a very young age and the importance of timely diagnosis of such cysts to avoid future complications is emphasized.

**Keywords** Cyst · Developmental cyst · First decade · Dentigerous cyst · Odontogenic

## Introduction

Dentigerous cysts comprise the second most common type of odontogenic cysts, after radicular cysts, representing nearly 20% of all the true jaw cysts [1, 2]. The WHO classification of jaw cysts refers to the dentigerous cysts as epithelial developmental odontogenic cysts [3]. They are attached to or enclose the crown of the unerupted tooth at the cemento-enamel junction [4]. Though dentigerous cysts may be seen in patients across a wide age range, they are most frequently discovered in patients between 10 and

30 years of age. Their frequency in the first decade of life is reported to be lower than in the second and third decades. Majority of them are associated with impacted or unerupted mandibular third molars followed by maxillary canines and maxillary third molars [5]. This case report presents the rare occurrence of dentigerous cyst in a 1-year old boy, the earliest age of occurrence of these cysts than those already documented. This implies that very young patients with developing teeth have the potential to develop dentigerous cysts. Their early diagnosis and timely treatment is extremely important to prevent further growth of the cyst and its complications.

## Case Report

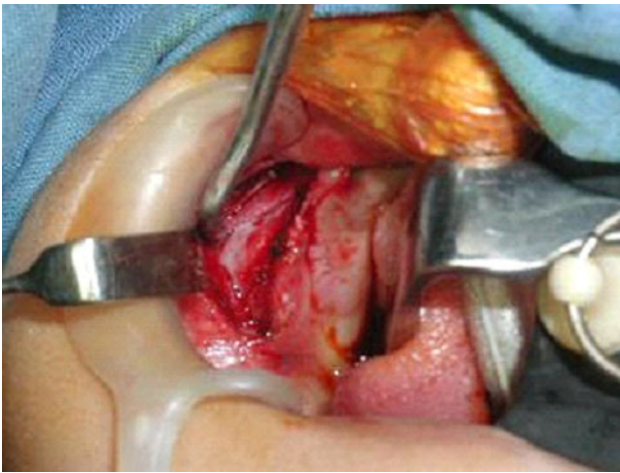
A male child patient, aged 1 year, was brought to the Department of Head & Neck Surgery, Amrita Institute of Medical Sciences and Research Centre, Kochi with a complaint of a swelling on the left side of the face, of 3 months duration. There were occasional episodes of increase in size and tenderness which were relieved temporarily on antibiotics. Associated with these episodes, the patient had difficulty during mastication. Extra-oral examination showed a diffuse swelling on the left side of the face at the angle of the mandible, hard and non tender on palpation at the time of clinical examination.

Intra-orally, a swelling was seen in the mandibular left posterior region obliterating the buccal sulcus (Fig. 1). An orthopantomogram taken revealed a well circumscribed radiolucency extending from the left angle of the mandible posteriorly to the primary second molar region anteriorly. A developing molar tooth was noticed inside the lesion.

A CT scan showed a well-defined cystic lesion involving the left angle of the mandible with a forming molar

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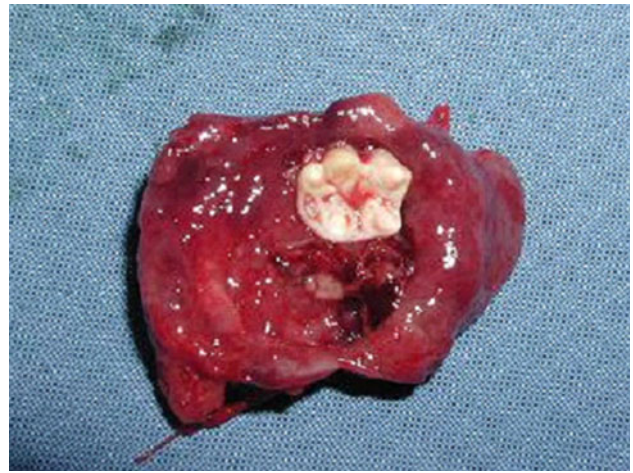
**Fig. 1** Intra oral photograph showing obliteration of buccal sulcus in the mandibular left posterior region

tooth inside (Fig. 2). Provisional diagnosis of dentigerous cyst was made correlating the clinical and radiographic findings. The clinical history of intermittent pain in relation to the swelling which was relieved with antibiotics was suggestive of a secondary infection in the cyst.

Enucleation of the cyst was performed under general anesthesia and the entire specimen along with the tooth bud of the left mandibular first permanent molar sent to the Department of Oral Pathology and Microbiology, Amrita School of Dentistry, for histopathologic evaluation. Gross examination of the specimen showed a brown cystic sac  $3.5 \times 2.5$  cm, with a corrugated surface, attached to the crown of partially formed mandibular first permanent molar (Fig. 3). Sections were taken after processing and stained with Hematoxylin and Eosin for histologic evaluation.



**Fig. 2** Photograph of the CT reconstituted image showing the cyst in the left ramus of mandible



**Fig. 3** Photograph of gross specimen showing the relation of the cyst to the crown of the forming tooth

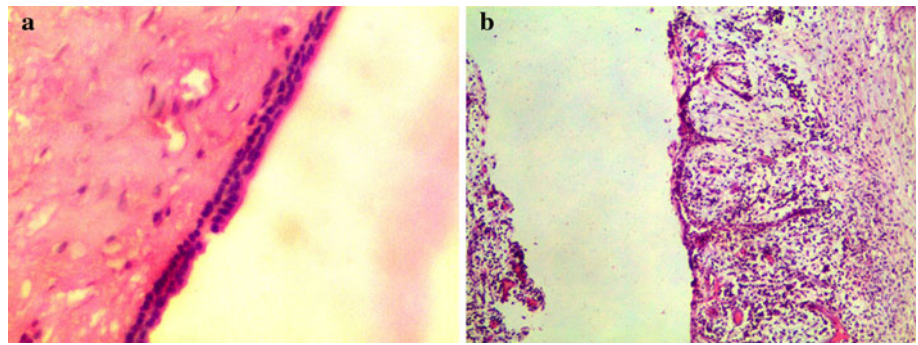
Microscopic examination of the sections showed a cystic lumen lined by reduced enamel-like epithelium (non-keratinized), 3–4 cell layers thick in most areas (Fig. 4a). The underlying fibrous capsule consisted of collagen fibers. A dense chronic inflammatory cell infiltrate consisting of lymphocytes and plasma cells was noticed in certain areas of the connective tissue capsule and the epithelial lining overlying the areas of inflammation was hyperplastic (Fig. 4b). Based on these features, the final diagnosis of inflamed dentigerous cyst was given. A follow up was done 6 months later, during which the clinical and radiographic evaluation showed no signs of recurrence.

## Discussion

Dentigerous cysts are developmental odontogenic cysts, believed to develop around the crown of an unerupted or impacted tooth by accumulation of fluid between the reduced enamel epithelium (REE) and the tooth enamel. The increased hydrostatic pressure of this pooling fluid is thought to separate the follicle from the crown, with or without the REE [6]. Toller [7] suggested that the likely origin of this cyst could be the breakdown of proliferating cells of the dental follicle after impeded eruption. Though most dentigerous cysts are developmental in origin, there are a few examples of these cysts with inflammatory pathogenesis. Benn and Altini [8] have proposed a possible mechanism for inflamed dentigerous cysts—they may develop from dental follicle and become secondarily inflamed and the source of inflammation is usually a non-vital tooth.

A review of related literature shows that though the age range for the reported cases of dentigerous cyst varies

**Fig. 4 a** Photomicrograph showing the lining epithelium which is thin and non-keratinized, covering the cyst wall. (H&E, 40x)  
**b** Photomicrograph showing areas of inflammation in the cyst wall and increase in thickness of the lining epithelium. (H&E, 20x)



widely from 5 to 57 years, they most commonly manifest in the second and third decades of life [4]. Their frequency in the first decade is reportedly lower than in the second and third decades [9]. Bhayya and Shyagali [10] have documented a case of dentigerous cyst in association with a developing mandibular second premolar in a 10 year old girl, while Hayasaki et al. [11] have reported a case of asymptomatic dentigerous cyst in a 4 year old girl in relation to the primary mandibular second molar. Dentigerous cysts have also been reported by Richard [12] in a 4 year old patient, Aguilo and Gandia [13] in a 5 year old and by Kalaskar et al. [14] in a 7 year old child. To date, of the cases of dentigerous cysts reported in the first decade of life, the youngest patient was a 3-year old child who presented with multiple dentigerous cysts [15]. In our case the age of the child at the time of manifestation of the cyst was 1 year, which makes this the youngest case of dentigerous cyst to be documented.

A vast majority of the dentigerous cysts are associated with impacted mandibular third molars, followed by the permanent maxillary canines and the maxillary third molars [5, 9]. A few cases have also been reported to have occurred in association with the permanent maxillary central incisors, permanent mandibular first and second molars [13, 14, 16]. Dentigerous cysts have also been reported to occur in association with primary teeth [11]. In this case, the dentigerous cyst was associated with the developing permanent mandibular first molar.

The cyst in a growing child is thought to enlarge more rapidly than in an adult, though the patient may only give history of a slowly enlarging painless swelling [17]. Dentigerous cyst usually presents as a painless expansion of bone unless it is secondarily infected. It typically shows a unilocular radiolucency which is associated with the crown of an unerupted tooth, on the radiograph. Most commonly, the cyst surrounds the crown of the tooth and the crown projects into the cyst, as seen in our case. The lateral form of dentigerous cyst usually grows laterally along the root surface and partially surrounds the crown, while in the circumferential variety the cyst surrounds the crown and also extends along a portion of the root [5, 9, 18]. In this

case, the patient's parents gave history of a gradually expanding swelling for 3 months, in the child, with intermittent pain in between. This was probably due to the secondary infection, which may be a reason for the microscopic finding of inflammatory infiltrate in the capsule. In the absence of pain, these cysts can attain considerable size with minimal or no symptoms [19]. Failure to identify and treat such cysts may allow their expansion to produce impingement on surrounding structures like inferior alveolar nerve and roots of adjacent teeth, resulting in paresthesia, root resorption etc. [20]. Hence early detection and removal of such cysts is extremely important.

Microscopically dentigerous cysts in most instances show a lumen lined by stratified squamous epithelium, which is usually non-keratinized, 2–3 cell layers thick. This may show variable thickness as in the case of inflamed dentigerous cysts, where the lining epithelium exhibits hyperplasia and increased thickness. The underlying fibrous capsule is collagenized and may show foci of chronic inflammatory cells in case of 'inflamed dentigerous cysts' [5, 18]. In our case, the histopathology revealed thin and non-keratinized lining epithelium, resembling reduced enamel epithelium overlying a fibrous connective tissue capsule in most of the areas. The capsule showed foci of chronic inflammatory cells and the lining epithelium corresponding to these areas showed increase in thickness. Attachment of the cyst wall to the neck of the associated tooth and the cyst lining in most cases demonstrating a thin flattened stratified squamous epithelium histologically help in confirming the diagnosis of dentigerous cyst [5, 9].

As per the guidelines of the American Academy of Pediatric Dentistry, the first panoramic radiographic examination should be performed following the eruption of the first permanent tooth. Radiographic examination of all unerupted teeth must be done to detect any associated pathology at an early stage [21]. In our case, the cyst was detected because of the clinical swelling and intermittent pain associated with it. Ideally, an orthopantomogram at 1 year of age may be recommended to diagnose any lesion in association with the developing permanent teeth so that timely treatment can be initiated.

Treatment of dentigerous cyst depends on its size and location. The options available for their treatment in children include enucleation of the cyst with primary closure and marsupialization and healing by secondary union [22]. In this case, the cyst was enucleated in toto. Since the tooth-germ of the left mandibular first permanent molar was inside the cystic lining, it was removed along with the cyst.

Complete removal of the cyst is extremely important given that, recurrent cysts, ameloblastoma, squamous cell carcinoma and mucoepidermoid carcinoma have been reported to result from long-standing dentigerous cyst or its remnants [18].

## Conclusion

Dentigerous cysts are rare in the first decade of life, and when they occur, they manifest with minimal symptoms. But they can attain considerable size over a period of time. A thorough review of a child's initial radiographs is important to detect the presence of these cysts in very young children. Our case, which is a rare presentation of dentigerous cyst in a 1-year old boy—the earliest case to be documented—points to the fact that the cyst may be manifested even before the eruption of the first permanent tooth, and can be actually associated with it. Hence an early panoramic radiographic examination preferably at the age of crown completion of the first permanent molar would be of immense diagnostic significance.

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