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1. Concurrent Dentigerous Cysts Associated With Impacted Mandibular Premolars: A Rare Case Report

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Abstract

Background: *Dentigerous cysts are common odontogenic cysts associated with impacted teeth; however, their occurrence in children, particularly in association with premolars and as multiple lesions in non-syndromic patients, is rare.*
Case Presentation: *A 9-year-old female presented with a slowly enlarging, painless swelling of the left mandible. Clinical and radiographic examination, including panoramic radiography and CBCT, revealed two distinct cystic lesions associated with impacted mandibular premolars, causing significant cortical expansion and facial asymmetry. A provisional diagnosis of adjacent dentigerous cysts was made.*
Conclusion: *This case highlights the rare occurrence of multiple inflammatory dentigerous cysts in a non-syndromic paediatric patient. Early diagnosis through careful clinical and radiographic evaluation is essential to prevent extensive jaw destruction and preserve the developing dentition.*

Keywords: *Dentigerous cyst; Multiple dentigerous cysts; Non-syndromic multiple dentigerous cysts; Pediatric patient; Premolars; Mixed dentition; Cone-beam computed tomography (CBCT); Tooth impaction; Odontogenic cyst.*

Introduction

Dentigerous cysts are developmental odontogenic cysts that arise from the accumulation of fluid between the reduced enamel epithelium and the crown of an unerupted tooth. They are the second most common odontogenic cysts of the jaws and are most frequently associated with impacted mandibular third molars, maxillary canines, and mandibular premolars. Although dentigerous cysts are commonly encountered in young adults, their occurrence in

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children during the first decade of life is relatively uncommon. Inflammatory dentigerous cysts developing in association with premolars are particularly rare. Furthermore, the simultaneous occurrence of multiple dentigerous cysts in non-syndromic patients is exceptionally uncommon. This report describes a rare case of two adjacent inflammatory dentigerous cysts associated with impacted mandibular premolars in a non-syndromic 9-year-old female patient, resulting in significant mandibular expansion and facial asymmetry.

Case Report

A 9-year-old female presented with a significant swelling on the left side of the lower face that had been progressively increasing in size over the past 3 years. According to the patient's mother, the swelling was initially noticed intraorally in the left posterior mandibular region when it was small and asymptomatic. At that time, it was presumed to be associated with the eruption of the permanent mandibular left first molar (36), and the patient was taken to a local dentist. Medications were prescribed; however, no reduction in the swelling was observed. As the lesion remained painless, no further intervention was sought.

Over the ensuing years, the swelling gradually increased in size and eventually became conspicuous extra orally, prompting the parents to seek treatment at the Outpatient Department of Bokaro General Hospital. The patient had no relevant history of facial trauma, systemic illness, or allergies.

Extraoral examination revealed a non-tender, hard swelling measuring approximately 3 × 3 cm involving the left body of the mandible. The overlying skin appeared normal in color and texture and was freely movable over the swelling. Intraoral examination demonstrated an expansion of the buccal vestibule extending from the region of 73 to 46. The overlying mucosa appeared normal, except for mild inflammation of the free gingival margin. Grossly decayed retained deciduous molars were also noted.

Over the ensuing years, the swelling gradually increased in size and eventually became conspicuous extraorally, prompting the parents to seek treatment at the Outpatient Department of Bokaro General Hospital. The patient had no relevant history of facial trauma, systemic illness, or allergies.

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Fine-needle aspiration cytology (FNAC) was performed to assess the nature of the lesion and to rule out the presence of cystic fluid. Orthopantomograph (OPG) revealed two distinct radiolucent lesions in the left posterior mandible. A smaller radiolucency was observed beneath the grossly carious retained deciduous first molar, associated with a horizontally impacted mandibular left first premolar. A larger well-defined radiolucency was identified adjacent to the unerupted/impacted second premolar, extending posteriorly up to the mesial root of the mandibular left first permanent molar (36). (Figure 1) Based on these findings, a provisional diagnosis of two separate dentigerous cysts was made.

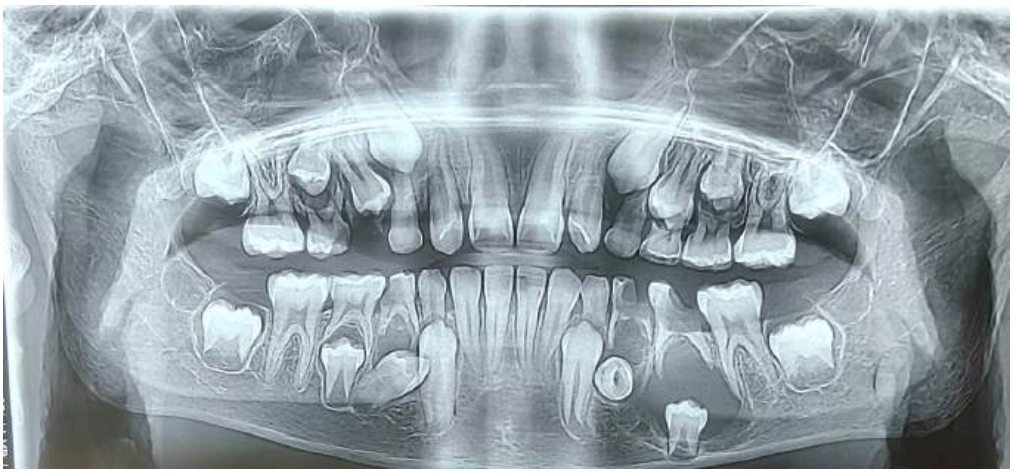

Figure 1: Orthopantomograph

Subsequently, cone-beam computed tomography (CBCT) was performed, which confirmed the presence of two well-defined radiolucent lesions in the left posterior mandible. The larger lesion measured approximately 33.7 mm anteroposteriorly, 23.1 mm superoinferiorly, and 24.7 mm buccolingually, (Figure 2a) whereas the smaller lesion measured approximately 11 mm × 12 mm. The cortical borders of both lesions were well-defined, with marked thinning and decortication of the buccal, lingual, and inferior cortical plates of the mandible. A distinct bony septum separating the two lesions was evident. (Figure 2b) The inferior alveolar nerve canal appeared displaced inferiorly, and its outline was indistinct, thereby limiting accurate tracing of the nerve course.

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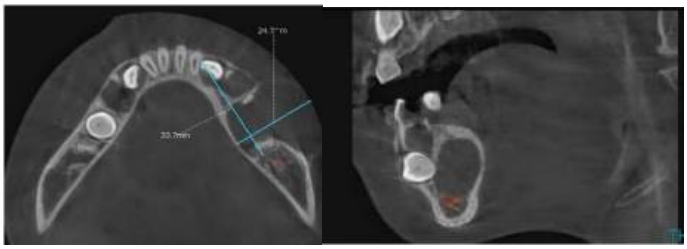

(a) *(b)*

Figure 2: (a):Larger cyst dimension (b) Ssmall cyst differentiated with clear bony septa

Both the cysts were enucleated via intraoral approach under general anaesthesia. Externally buccal bone was compressed to aesthetic position and then thin dead buccal plate was removed, extraction of deciduous molars was done and the cystic lining were sent for histopathological examination (HPE) and cystic fluid for FNAC.

Figure 3: Two cysts with impacted premolars and buccal plate

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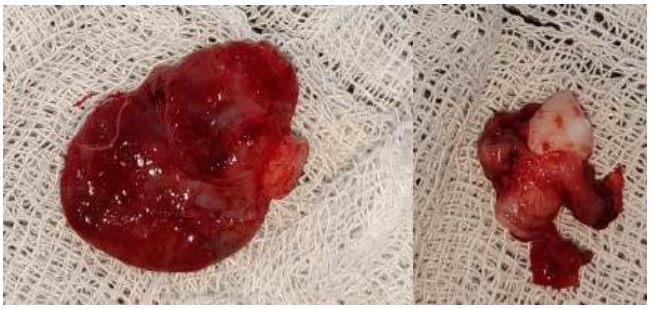

Figure 4: large cyst with 35 *Figure 5: small cyst with35*

FNAC showed chronic inflammatory infiltrate. HPE confirmed a cystic lining epithelium appears to be non-keratinized stratified squamous epithelium with an irregular or undulating surface. The underlying connective tissue wall is fibrous and relatively collagenous. Multiple gland-like or microcystic spaces can be seen within the epithelial lining, with some areas showing vacuolated or mucous cell-like changes. Mild chronic inflammatory infiltrate was present in the cyst wall. (Figure 6) These features suggestive of a odontogenic cyst, particularly because of the gland-like structures and microcystic spaces within the lining epithelium.

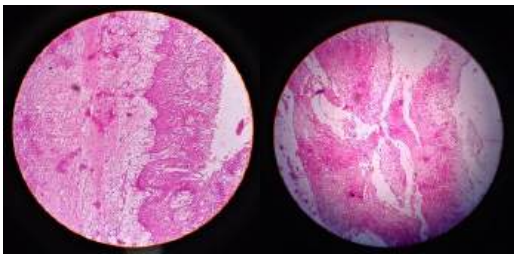

Figure 6: histo pathological image

Discussion

Dentigerous cysts, also called follicular cysts, are slow-growing benign and non-inflammatory odontogenic cysts that are thought to be developmental in origin. It originates from the accumulation of fluid between the reduced enamel epithelium and the crown of an unerupted tooth.

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Dentigerous cysts are the second most common odontogenic cysts of the jaws after radicular cysts and account for approximately 20–24% of all epithelium-lined cysts of the jaws.[1]

Dentigerous cysts most commonly involve impacted mandibular third molars, maxillary canines, and mandibular premolars and are typically diagnosed during the second and third decades of life. Their occurrence in children is relatively uncommon. In our present case report, it occurred in 9-year-old female and in relation to mandibular premolars.

Cases of Bilateral dentigerous cysts in non-syndromic patient have been reported by Shirazian S, Agha-Hosseini F. in 2011 and Jae-Yun Jeon et. Al. in the year 2016. Our case is also a non-syndromic patient where the cyst developed adjacent to each other as separate entity. Till date probably only one such case has been reported in the year 2025.[2,3]

The occurrence of multiple dentigerous cysts in non-syndromic patients is exceedingly rare and is usually associated with syndromes such as cleidocranial dysplasia, Maroteaux-Lamy syndrome, or basal cell nevus syndrome. The absence of any syndromic manifestations in our patient makes this presentation particularly unusual.[4]

On the contrary study done in 2024 states that, compared to some other rare multiple developmental odontogenic cysts, multiple non-syndromic dentigerous cyst are much more common.[5]

Clinically, dentigerous cysts are usually asymptomatic and are frequently discovered incidentally during routine radiographic examination. However, when the lesion enlarges considerably, patients may present with cortical expansion, facial asymmetry, delayed eruption, displacement of adjacent teeth, or rarely pain secondary to infection. The present case exhibited a slowly progressive, painless swelling over a period of three years resulting in significant facial asymmetry, which ultimately prompted the patient's parents to seek treatment.[6]

Radiographically, a dentigerous cyst typically appears as a well-circumscribed unilocular radiolucency surrounding the crown of an unerupted tooth and attached at the cemento-enamel junction. The unique feature of the present case was the presence of two distinct radiolucent lesions associated with impacted mandibular premolars separated by a well-defined bony septum.

The radiographic differential diagnosis included odontogenic keratocyst, unicystic ameloblastoma, radicular cyst involving deciduous teeth, and enlarged dental follicles. However, the intimate association of both lesions with the crowns of unerupted premolars, along with histopathological examination, established the diagnosis of dentigerous cysts.

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Conventional panoramic radiography was useful in identifying the lesions; however, CBCT provided additional information regarding the exact extent of the cysts, cortical plate expansion, decortication, displacement of the inferior alveolar canal, and the presence of a distinct septum separating the two lesions. Such information is invaluable in surgical planning, particularly in pediatric patients where preservation of adjacent structures and developing permanent teeth is of paramount importance.[7,8]

The management of dentigerous cysts in children depends on factors such as lesion size, age of the patient, relationship with adjacent anatomical structures, and the possibility of preserving the involved permanent teeth. Conservative approaches such as marsupialization or decompression are often preferred in pediatric patients because they facilitate spontaneous eruption of the associated permanent teeth while minimizing surgical morbidity.[9] However, complete enucleation may be indicated when the lesion is extensive, when the associated tooth is severely displaced, as in our present case, or when preservation of the tooth is not feasible.

The present case highlights the importance of careful clinical and radiographic evaluation of retained and grossly decayed deciduous teeth in children. Persistent facial swelling or delayed eruption should never be overlooked, as early diagnosis and intervention can prevent extensive bone destruction, displacement of developing permanent teeth, and associated functional and esthetic complications.

Pearls of this case:

- Age of occurrence approximately 6 years as patient reported to us at the age of 9 with present clinical symptoms. Approximately 4–9% of dentigerous cysts occur during the first decade of life, making them relatively uncommon in children.
- Involvement of dentigerous cysts in association of premolars are rare, accounting for less than 3% of cases.
- Adjacent or multiple dentigerous cysts in non-syndromic individuals are considered a rare clinical anomaly, which was seen in our case.

Conclusion

The present case highlights the rare occurrence of multiple inflammatory dentigerous cysts associated with impacted mandibular premolars in a non-syndromic pediatric patient. The asymptomatic nature and slow progression of these lesions may result in delayed diagnosis, leading to significant cortical expansion and facial asymmetry. Careful clinical examination, supplemented by panoramic radiography and CBCT, is essential for accurate diagnosis, assessment of lesion extent, and treatment

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planning. Early recognition and timely management of retained and grossly decayed deciduous teeth are crucial to prevent extensive jaw destruction and to preserve the developing permanent dentition. This case emphasizes the importance of considering dentigerous cysts in the differential diagnosis of persistent facial swelling and delayed tooth eruption in children.

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