Case Report

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INFLAMMATORY DENTIGEROUS CYST OR INFLAMMATORY CYSTIC LESIONS OF MIXED DENTITION?: A REPORT OF THREE CASES

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ABSTRACT:

A dentigerous cyst is one of the most common cystic lesions of the jaws, comprising approximately 20-24% of all jaw cysts. Most authors favour a developmental origin from tooth follicle, with fluid accumulation between layers of reduced enamel epithelium (REE) or between REE and crown. However, the relation between persistent and prolonged inflammation of a primary tooth and development of a dentigerous cyst involving the permanent successor has been extensively discussed by several investigators suggesting an inflammatory origin. Although dentigerous cyst is classified as developmental odontogenic cyst, some investigators proposed classifying its inflammatory variant as inflammatory cyst due to its pathogenetic mechanism. This article deals with reporting of 3 cases of dentigerous cysts of probable inflammatory origin and discusses the dilemma associated with terminologies and classification of inflammatory dentigerous cyst.

Key words: Dental follicle; Dentigerous cyst; Inflammation; Odontogenic cysts

INTRODUCTION

Dentigerous cysts are the most common developmental odontogenic cysts, comprising approximately 17-18 % of all jaw cysts.[1] They are classically defined as cystic lesions that are caused by separation of follicles from around the crown of unerupted teeth.[2] It had been suggested that dentigerous cysts may be of either intrafollicular or extrafollicular origin.[1] Those of intrafollicular origin may develop by accumulation of fluid either between the reduced enamel epithelium and the enamel, or within the enamel organ itself. Extrafollicular origin has been refuted as those reported to be arising in this manner were all appeared to be envelopmental or follicular OKCs.[2] However this was not the only ambiguity associated with dentigerous cyst. Benn and Altini (1996) described inflammatory variant of dentigerous cyst. They suggested that developmental type cyst occurs in mature teeth usually as a result of impaction, while inflammatory type cyst occurs in immature teeth as a result of inflammation.[2] Inflammation from predecessor primary was considered as initiating factor for dentigerous cysts; first suggested by Bloch-Jorgensen (1928) in their case series, followed by Azaz & Shteyer (1973) and Shaw et al (1980).[3] These so called inflammatory dentigerous cyst were reported in mixed dentition;

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associated with the roots of non-vital primary teeth and the crown of unerupted permanent successors.[2,4-6] This case reports deals with 3 such cases of dentigerous cysts of probable inflammatory origin.

CASE DETAIL

In all the 3 cases, patient reported to our institute with complain of pain and swelling. Informed consent was obtained from patient's relatives. The Clinical, radiographic, and histologic features of the 3 cases were tabulated and analysed. (Table 1) Based on clinical history of carious primary tooth and radiographic evidence of impacted permanent tooth with pericoronal radiolucency, provisional diagnosis of inflammatory dentigerous cyst was made. In differential diagnosis, odontogenic keratocyst and unilocular ameloblastoma were considered. Conservative approach of extraction of carious primary and decompression of cystic cavity was done in all 3 cases to allow normal eruption of permanent successors. On histopathologic examination, presence of nonkeratinised stratified squamous epithelium with inflammation confirmed the final diagnosis of inflammatory dentigerous cyst. Cases were kept under follow up.

DISCUSSION

Inflammatory dentigerous cyst is found in mixed dentition only and is ten times more likely to occur in the lower jaw than the upper jaw.[7] Most commonly involved tooth by the cyst in the first decade of life is mandibular second premolar. This may be because the deciduous mandibular second molar has a higher susceptibility to decay and its roots are more closely associated with the follicle of unerupted mandibular second premolar. This close physical relationship facilitates spread of infection in comparison with other primary teeth.[7,8] Cases reported here shared age range of 7-10 years, male gender, clinical history of carious primary molar and radiographic evidence of mandibular premolar involvement with a pericoronal radiolucency with previous reported cases.[2-7] Benn and Altini (1996) suggested that usually developmental type are lined by thin nonkeratinised stratified squamous epithelium while inflammatory type are lined mostly or entirely by hyperplastic nonkeratinising stratified sqaumous epithelium of varying thickness.[2] Histological presence of hyperplastic nonkeratinised stratified squamous epithelium with focal areas of arcading in all the 3 cases confirmed our diagnosis.

It is generally accepted that extraction of a non-vital primary tooth and marsupialisation will allow rapid healing of the lesion and eruption of the permanent tooth, provided that these procedures are performed at the normal time of eruption.[9-11] This conservative approach is preferred owing to greater bone regeneration capacity and eruption potential of immature teeth in younger patients.[9,10] Similar procedure was followed in case 2 & 3 and kept under follow up to monitor eruption of permanent tooth. However in case 1, enucleation of the cyst with removal of the involved tooth was done due to

extensive size of lesion and apical displacement of the unerupted second premolar.

There are many diagnostic ambiguities associated with inflammatory dentigerous cyst (IDC). Attachment of cyst wall to neck of tooth and histological presence of REE like cystic lining were two strict criteria suggested to diagnose dentigerous cyst.[1] However conservative approach of decompression makes it difficult to appreciate cyst wall attachment to impacted tooth in most of the cases. Also most of the cases reported as IDC histologically showed presence of hyperplastic epithelium as in inflammatory cysts.[2-7] Over the years, investigators failed to give conclusive evidence on whether inflammation may be the reason/coincidental finding in inflammatory dentigerous cysts. It is difficult to differentiate between inflammatory dentigerous cyst and infected dentigerous cyst, where inflammation is secondary process.[12] However in most of the previously reported cases as suggested by Benn & Altini (1996); there was no element of tooth impaction as tooth eruption had hardly commenced (as in case 2 & 3) for intrafollicular development of dentigerous cyst. They refuted the possibility of secondary inflammation of developmental dentigerous cyst.[2]

Although inflammatory dentigerous cysts are classified as a variant of dentigerous cyst which is a developmental odontogenic cyst, its pathogenetic mechanism suggests otherwise. In relevance to this, paradental cysts are classified as inflammatory cysts as inflammatory source from periodontal pocket or pericoronitis in partially erupted tooth causes unilateral expansion of follicle.[13] As pathogenesis of inflammatory dentigerous cyst is related to inflammation induced expansion of the follicle of the erupting tooth, we believe that same rule should be applied to them and they should be classified as inflammatory cystic lesions. Martinez B et al 2012 have proposed a new classification scheme for inflammatory cysts which is as follows[14]

- Those arising from periapical inflammation – radicular cyst, residual cyst
- Those arising from inflammation during eruption of teeth – paradental cyst, inflammatory dentigerous cyst

They suggested the latter group those associated with eruption of the tooth, are not clearly identified in the literature. This may be due to the confusing terminology that exists on these cysts and sometimes they are not treated properly.[14] Further insight is needed on confusion regarding terminology and classification of inflammatory dentigerous cyst whether as variant of dentigerous cyst or as inflammatory cystic odontogenic lesion.

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TABLE:

Table 1 - Demographic information, location, radiologic and histopathological features of 3 cases

Case no.	Clinical findings		Radiographic features	Histopathologic features
	Age/ sex	Location and Intraoral examination		
1	10 years/Male	Pain and Swelling in mandibular right posterior region, Carious 84,85,	On lateral cephalogram, large unilocular radiolucency surrounding 45 which is pushed downwards, root pieces of 85 (Figure 1A) On Occlusal radiograph, buccal cortical expansion seen with root piece of 84, 85 and impacted 45 (Figure 1B)	Cystic lining composed of nonkeratinised stratified squamous epithelium of 2-4 cell layer thickness with diffuse inflammation in cystic wall (Figure 1C)
2	9 years/Male	Pain and slight intraoral swelling in mandibular left posterior region, Carious 75	In Lateral cephalogram, unilocular radiolucency surrounding 35 in continuity with lamina dura of 75 (Figure 2A) In intraoral periapical radiograph, intracoronal radiolucency in 75 indicating carious lesion, pericoronal radiolucency surrounding 35 (Figure 2B)	Cystic cavity was lined by nonkeratinised stratified squamous epithelium with flat or cuboidal cells resembling REE. Intense inflammation juxtaepithelially (Figure 2C) Lining was hyperplastic in areas showing an arcading pattern. Moderately inflamed cystic capsule (Figure 2D)
3	7 years /Male	Pain in mandibular left posterior region, Carious 75, (Figure 3A)	In orthopantomogram, large unilocular radiolucency associated with CEJ of impacted 35 and in continuation of lamina dura of 75, root resorption of 75, 36, and displacement of 34, large radiolucent carious lesion in 75 was seen (Figure 3B)	Cystic cavity lined by nonkeratinised stratified squamous epithelium disrupted by inflammatory cells, fibrous connective tissue with moderate inflammation (Figure 3C)

FIGURES:

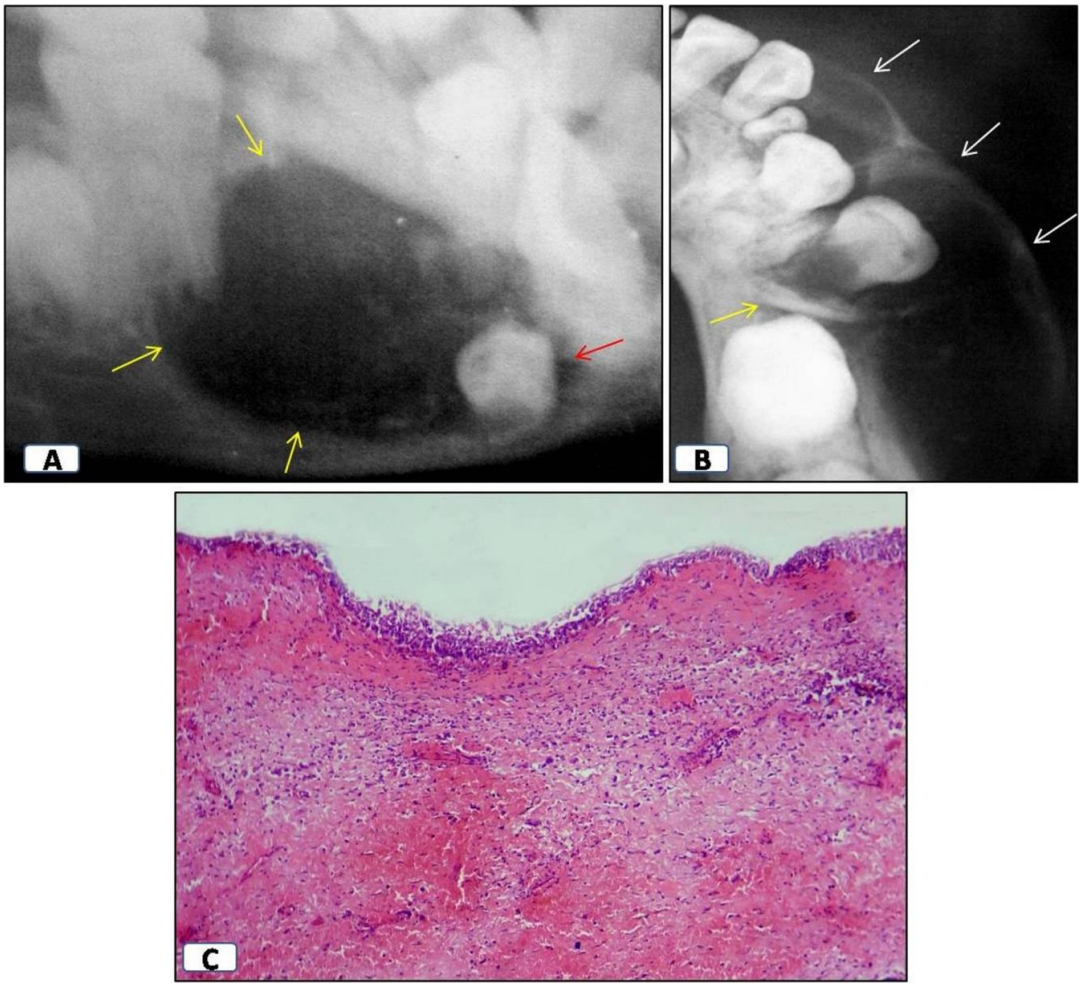

Figure 1. Case 1. A. Lateral cephalogram showing large pericoronal radiolucency (yellow arrows) surrounding impacted 45 pushed towards inferior border of mandible **(**red arrow), root pieces of 85 can be seen. **B.** Occlusal radiograph showing large radiolucency expanding buccal cortex (white arrows) with impacted 45 and root pieces of 85 (yellow arrow) **C.** Histopathology showing nonkeratinised stratified squamous epithelium of 2-4 cell layer thickness with moderately inflamed cystic wall **(**H & E staining, x200 magnification).

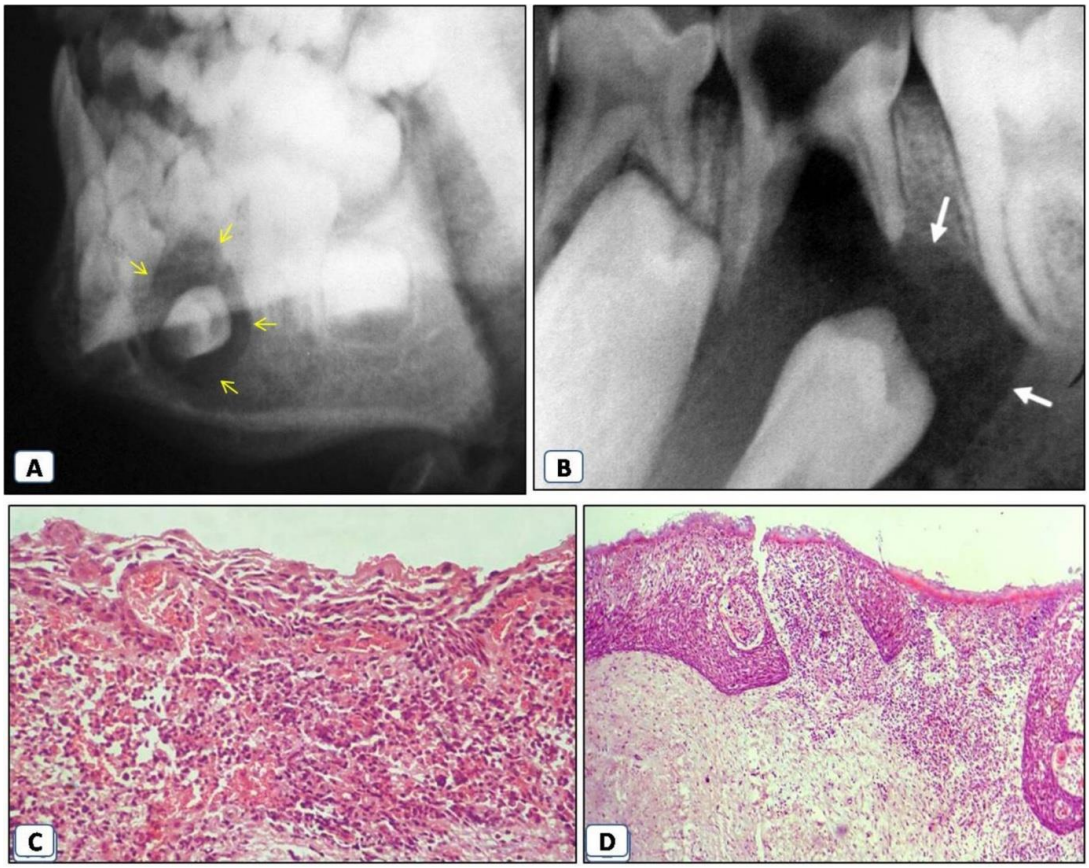

Figure 2. Case 2. A. Lateral cephalogram showing unilocular pericoronal radiolucency around 35 (arrows) **B.** Intraoral periapical radiograph showing pericoronal radiolucency around 35 (arrows) with carious 75. **C.** Histopathology showing nonkeratinised stratified epithelium with flat or cuboidal cells. Intense juxtaepithelial inflammation seen. (H & E staining, x400 magnification) **D.** Lining was hyperplastic in areas showing an arcading pattern. (H & E staining, x200 magnification).

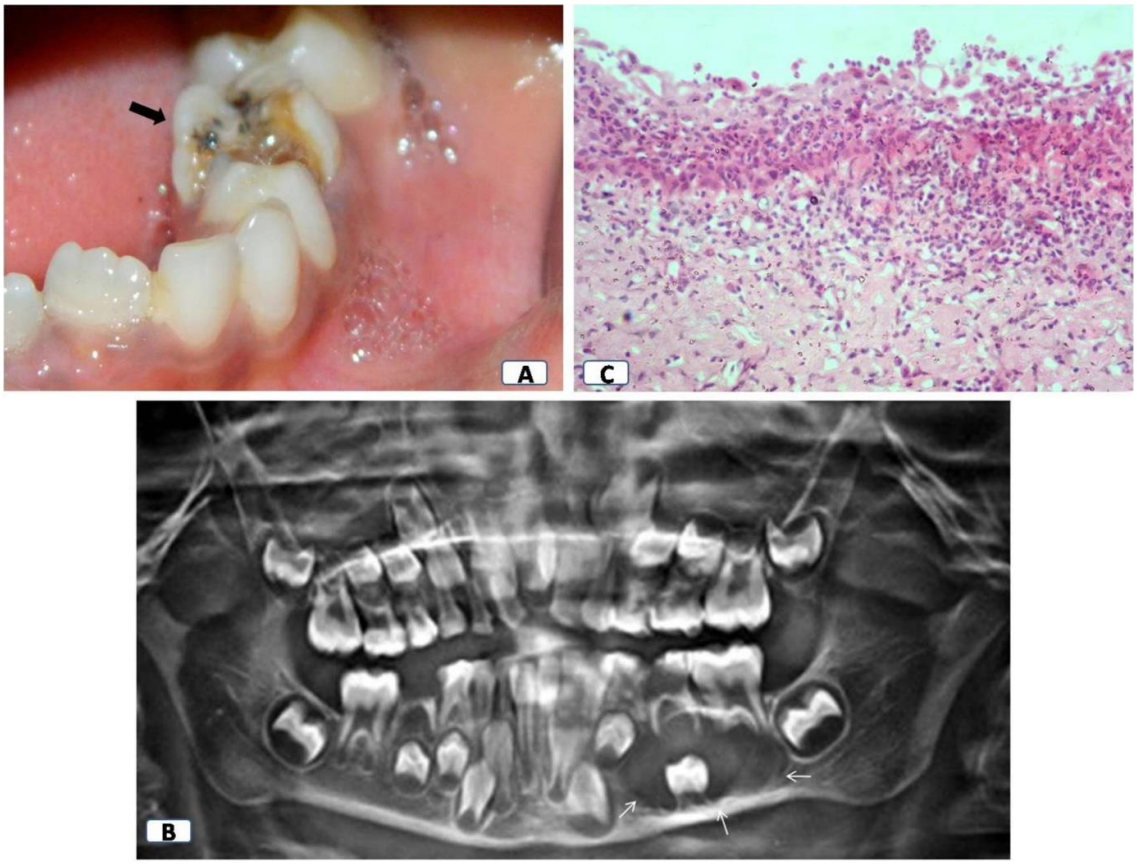

Figure 3. Case 3. A. Intraoral clinical picture showing carious tooth 75 (arrow) **B.** Orthopantomogram showing large pericoronal radiolucency around 35 (arrows). Root resorption of 75, 36, and displacement of 34 seen. **C.** Histopathology showing nonkeratinised stratified squamous epithelium disrupted by mixed inflammatory cells (H & E staining, x400 magnification).