CASE REPORT

Infection's Legacy- How the Periapical Infection of Predecessor Shapes the Successor. Case Report of Inflammatory Dentigerous Cyst.

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Abstract

Introduction: Inflammatory Dentigerous Cyst (IDC), a type of dentigerous cyst, is reported to occur only in children, with low frequency during their mixed dentition period. The reason could be a carious, endodontically treated or non-vital deciduous tooth affecting the development of its successor leading to IDC.

Case Presentation: Herein we present a case of 12-year-old female patient with a chief complaint of pain and mild swelling in right lower back tooth region, with history of extracted carious deciduous tooth in the same region. Radiographic and gross features clued for the diagnosis of dentigerous cyst with unilocular well-defined, coronal radiolucency of successor crown, attaching at cemento-enamel-junction (CEJ). Histopathology revealed varied thickness of non-keratinized stratified squamous epithelium resembling reduced enamel epithelium.

Management and Prognosis: Surgical enucleation was done with six month follow up which was uneventful.

Conclusion: Infections of deciduous teeth and their consequences on permanent teeth should not be overlooked. Prompt diagnosis and early intervention is needed to save the permanent teeth and prevent further complications of tooth resorption, infections, displacements.

Keywords: Dentigerous cyst, Deciduous molars, Dental follicle, Enucleation, Inflammatory follicular cyst, Primary tooth infection.

Introduction

The deciduous teeth and their successors are interrelated topographically with each other, any alterations in the form of dental caries, trauma, or infections to the predecessors can alter the homeostasis between them and adversely affect the permanent teeth as well as their surrounding tissues.[1] The chronic inflammation constantly occurring in periapical region of primary dentition influences odontogenesis in permanent dentition after a period of six weeks approximately. Till six weeks a thin fibrous barrier between inflammation zone and enamel epithelium remains as a response to short-term infection. Disturbances due to overlying inflammatory process may manifest as developmental alterations of crown, root and rarely can cause odontogenic cysts. Cysts occurrence in primary teeth is considered infrequent due to dental exfoliation, decompression, resolution and unperformed biopsies or improper dental records.[2]

Dentigerous cysts, the second most common odontogenic cysts next to radicular cysts are seen to account for about 17.1% of all jaw cysts with about 9% occurrence in children according to Shear and about 4% as by Donath.[3,4] Their exact etiopathogenesis is still unclear but Benn and Altini had proposed two types; developmental type and inflammatory type.[5] Main designated Inflammatory dentigerous cyst as Inflammatory follicular cyst which was discerned to occur only during mixed dentition and with low frequency.[5,6,4] They occur usually in association with infected, non-vital, endodontically treated teeth, or endodontic failures, from which the inflammation can spread into the developing dental follicle

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of their successor teeth that may bring about degenerative or destructive changes in the follicle, in few cases that might lead to dentigerous cyst in an unerupted successor.[5,7,8,9] Most commonly involved are mandibular premolar successors with relation to primary second molar seen in age group of six to twelve years, with male predilection.[2,5,8] They can range from a small asymptomatic lesion with incidental finding in radiographs to large swellings at times causing bone expansion, springiness of bones, tooth displacement, root resorption.[4] They may appear as pericoronal radiolucencies attached at the CEJ as well can be seen in continuation with lamina dura of deciduous tooth.

Case Presentation

A 12-year-old female patient presented to a private dental clinic, complaining of pain and swelling in lower right back tooth region since 3-4 days. The patient gave a history of extraction of a carious deciduous posterior tooth two days back in the same region but was not relieved from symptoms. No contributory medical, personal or family history was recorded. No abnormalities were detected on extra-oral examination, and upon intraoral examination, mixed dentition was seen along with extracted socket i.r.t 85 region, surrounded by mild swelling which on palpation was soft to firm in consistency and non-tender. Orthopantomograph (OPG) analysis showed radiolucency in the right premolar region of mandible with missing 85. The cystic lesion was well defined, unilocular with sclerotic borders primarily located in the body of mandible having second premolar (45) crown protruding into the cavity. (Figure1) Thus, a provisional diagnosis of dentigerous cyst i.r.t 45 was given and differential diagnosis of hyperplastic dental follicle, Adenomatoid odontogenic tumor and Odontogenic fibroma were considered.

Management and Prognosis

Following administration of local anaesthesia, vertical incisions were made, mucoperiosteal flap was raised and a total enucleation of the lesion including removal of 45 was done. After wound irrigation, the mucoperiosteal flap was closed and specimen was sent to our Department of Oral and Maxillofacial Pathology for further evaluation. Postoperative medications included antibiotics and analgesics. The prognosis was good and no complaint was received during the follow up of 6 months.

Macroscopic examination of the specimen showed multiple bits of soft tissue with extracted 45 tooth enclosed within soft tissue, together measuring about 1.5 x 2.4 cm approximately. The soft tissue attachment was noticed at cemento-enamel junc-

Fig. 1: A. Extraction socket i.r.t 85 with surrounding mild swelling. B. Unilocular coronal radiolucency with attachment at CEJ i.r.t unerupted 45. C. Site after enucleation.

Fig. 2: A and B. Mixed tissue bits with 45 showing soft tissue attachment at CEJ.

tion (CEJ) of 45. (Figure 2).

Histopathological examination of HandE soft tissue sections revealed non-keratinized squamous cystic epithelial lining of varied thickness ranging from predominantly bilayered resembling reduced enamel epithelium to multilayered edematous epithelium with prominent intercellular spaces. The subjacent connective tissue capsule was edematous, consisting of delicate haphazardly arranged collagen bundles, spindle and stellate shaped fibroblasts, endothelial lined blood vessels, diffuse dense chronic inflammatory cell infiltrate chiefly plasma cells, lymphocytes, mast cells, macrophages, odontogenic rests at focal areas, confirming the diagnosis of dentigerous cyst. (Figure 3)

Discussion

Dentigerous or follicular cyst is the one that encloses the crown of an unerupted tooth by expansion of its follicle and is attached to its neck or CEJ, this definition is applied strictly and diagnosis is not made on radiographic evidence alone. Browne and Smith stressed the term dentigerous cyst meaning "tooth bearing" and is more appropriate than follicular.[10]

First perception towards inflammatory dentigerous cysts was given by Bloch-Jorgensen in 1928, that are different to developmental types which occur in an immature permanent tooth as a consequence of inflammation in the non-vital overlying primary tooth.[3,5,9]

Benn and Altini proposed three plausible mechanisms for their formation. First; Intrafollicular developmental dentigerous cysts formed around the crowns of permanent tooth that become secondarily inflamed, as a result of periapical inflammation spreading from nonvital deciduous predecessors. Second; the radicular cysts that are present at the apices of non-vital deciduous teeth may fuse with follicles of unerupted permanent successor teeth, eruption of these successor teeth into the cystic cavity can result in the formation of an extra-follicular dentigerous cyst. Shear and Speight regarded this mechanism to be exceptionally rare because radicular cysts that involve the deciduous dentition are uncommon[10] and considered that in such cases, the erupting tooth may indent rather than penetrate the wall of the radicular cyst. Third; Periapical inflammation that

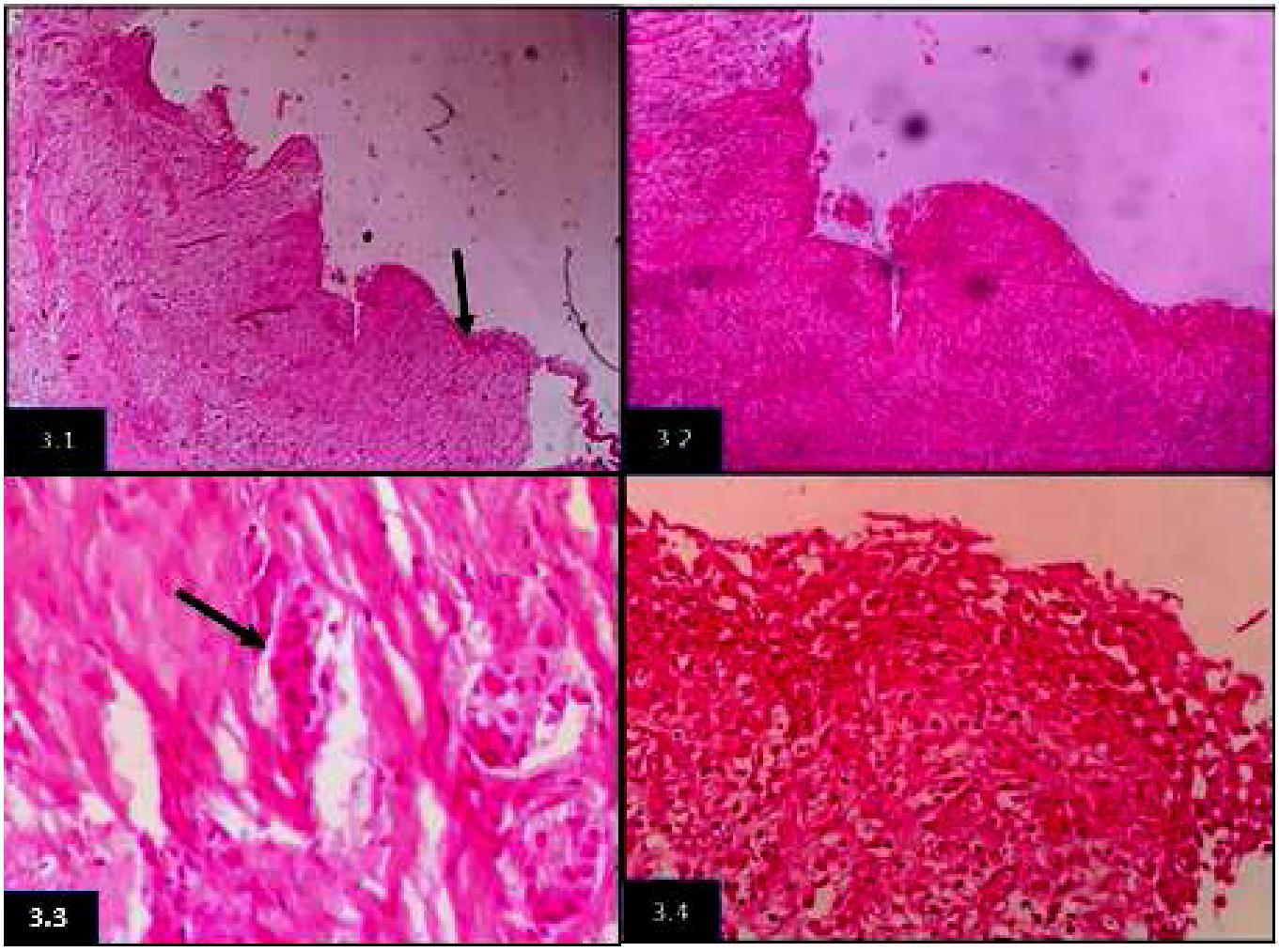

Fig. 3: A. Cystic lining with bilayered epithelium resembling reduced enamel epithelium. HandE- 4X, HandE- 10X B. Odontogenic rests at focal areas. HandE- 40X C. Moderate chronic inflammatory cell infiltrate, infiltrating into epithelium HandE- 40X.

can occur from any source but usually from the non-vital deciduous teeth spreads to the follicles of unerupted permanent successors.[6] The inflammatory exudate formed can cause separation of reduced enamel epithelium from enamel that result in the formation of cyst.[5] In our case the mechanism involved could be third, as the patient had given history of pain, infection and extraction of deciduous molar, which might have led to the cyst involving second premolar.

Clinical presence of an infected deciduous tooth i.e., 85, radiographic and gross features with cystic attachment at CEJ of its successor i.e., 45. on aspiration, light yellowish color fluid was observed (Reports by some authors mentioned about cystic aspiration and leaking out of the cyst's fluid during an extraction of a primary tooth or during a decompression technique).[9,4] All of which clued up for the diagnosis of IDC. Therefore, combining clinical, radiographic and histopathological findings will provide a precise diagnosis of "Inflammatory Dentigerous cyst".

Primary mandibular second molars are closer to the follicles of their permanent successors that may influence the spread of inflammation, which could be the reason for their common involvement.[2]

Radiographically it may overlap with other lesions such as unicystic ameloblastoma, envelopmental odontogenic keratocyst, adenomatoid odontogenic tumor.[1,9,11,12] Those can be differentiated with its peculiar feature of CEJ attachment. The enlarged dental follicles can be excluded by the diameter of pericoronal radiolucency which usually will be more than 2.5 mm in its greatest diameter for dentigerous cyst.[5]

Histologically it may show overlapping features with radicular cysts and enlarged dental follicles. It is usually lined by a hyperplastic non keratinized stratified squamous cystic epithelium with frequent anastomosing rete ridges mimicking the arcading pattern of radicular cysts likely due to the underlying moderate to intense inflammation, and few areas of epithelium may show two-three layered cuboidal epithelium with resemblance to reduced enamel epithelium, Odontogenic epithelial rests can also be evident in the cystic capsules.[5,13]. The developmental type usually consists of uniform thickness of cystic epithelium without any interconnecting reteridges and inflammatory cells unless it is secondarily infected.

Regarding treatment, the primary concern is to address the chief complaint and hence, surgical removal of offending primary tooth to reduce the spread of inflammation should be done.[4] Followed with options such as enucleation and marsupialization that is decided based on the size of lesion, condition of tooth, bone factors, age factors etc.

Marsupialization is the preferred method for children as they have high bone regenerating capacity and the teeth with open apices also shows great eruptive potential.[9,1,2] The disadvantage would be remnants of lining that may left behind, still this can be done as dentigerous cysts are known to recur rarely. Additions such as obturators or graft materials can be placed in relation to the cysts for enhancing the prognosis and also orthodontic treatment for proper eruption of the permanent teeth.[14]

Enucleation is recommended for those teeth showing arrested development, displacements and associated with extensive lesion.[7,4,11] In present case, Enucleation was carried out along with involved tooth because of considerably large size of lesion and apical displacement of tooth.

Conclusion

Prompt diagnosis and early intervention is needed so as to save the permanent tooth and to prevent further complications of tooth resorption, infections, displacements and chances of cystic lining transformation to ameloblastoma that may occur as in cases of Inflammatory dentigerous cysts.[15] The infections of deciduous teeth and their consequences on permanent teeth should not be overlooked.

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