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Radicular dens invaginatus associated with radicular cyst in maxillary third molar – Rare case report



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ABSTRACT

Dens invaginatus is a developmental tooth anomaly showing broad spectrum of morphological variations. Most extreme form of the invagination results in a bulbous expansion of the affected crown and/or root has been termed as dilated odontome. Radicular dens invaginatus is a rare dental anomaly formed by infolding of Hertwig's epithelial root sheath and in few forms invagination is lined by enamel. Various techniques of treatment including conservative treatment, nonsurgical root canal treatment, and endodontic surgery usually are unsuccessful because of their complex morphology and are associated with periapical lesions. We report a case of radicular dens in dente with dilated root and was associated with radicular cyst in the right maxillary third molar in a patient aged 38 years. Histological examination showed a dilated bulbous root with enamel formation within root invagination and the associated radicular cyst was lined by pseudostratified ciliated columnar epithelium.

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1. Introduction

Dens invaginatus is also termed as “Dens in dente”, “dilated composite odontome”, “dents telescopes”, and “gestant odontome”. The most severe form of the dilated type of dens invaginatus is called a dilated odontoma. The frequency of occurrence is 0.04–10% and most commonly affected teeth are maxillary lateral incisors followed by central incisors, premolars, canines, and molars. The mandibular occurrence is rare and bilateral occurrence is not uncommon. There is no specific sex predilection but the condition exhibits a high degree of inheritability.[1–3]

Two variations of Dens Invaginatus are described – a coronal type and a radicular type. The coronal type is caused by invagination of all layers of the enamel organ into the dental papilla. Oehlers described dens invaginatus as occurring in three forms depending on the extension of enamel lined cavity, Type I – minor form occurring within the confines of the crown of the tooth and not extending beyond the amelocemental junction, Type II – which invades the roots but remains within its confines and Type III – severe form which

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penetrates through the root and shows a second foramen in the apical or periodontal area.[4–6]

The radicular type of Dens Invaginatus occurs due to infolding of Hertwig's sheath into the developing root. There are two distinct types of radicular dens invaginatus – first type shows cementum lined invagination and second type regarded as true form is represented as enamel lined invagination within root.[4,7] Very few cases of radicular dens invaginatus are reported and due to their complex morphology pulpal necrosis, apical lesions are often associated.[8,9] The radicular cyst or periapical cyst are usually lined by stratified squamous epithelium, however rarely they have been found to be lined partially or predominantly by ciliated columnar epithelium or muco-secretory cells.[10,11] Here we present a rare case report of radicular dens invaginatus with dilated root in right maxillary third molar which was associated with radicular cyst.

2. Case report

A 38-year-old male patient reported to the Institution, with severe continuous pain in the region of upper right third molar area since 3 months which increased in severity in last 3 days. The patient reported that one year back, he got restoration for the same in a private dental hospital. The patient was in good general health and revealed no significant extra oral findings. Intraoral examination revealed improperly done crown on 18 which was sensitive to palpation and percussion. No soft tissue findings were evident except for tenderness on palpation in periapical area in the region of 18. Periapical radiograph showed an abnormal 18 with a bulbous root with an apical crescent shaped invagination associated with periapical radiolucency. The periapical radiolucency measured approximately 1 × 2 mm with ill defined border. A clinical diagnosis of dens invaginatus with apical periodontitis was given.

A complete blood and urine analysis was done and all indices were found within normal limits. The tooth was extracted under local anesthesia after reflection of the muco-periosteal flap and was sent for histopathological examination. Morphological features revealed an abnormal tooth with improperly prepared crown and an enlarged bulbous dilated root with periapical soft tissue. The tooth measured 2 cm occluso-apically and 1.5 cm mesio-distally at the highest circumference near bulbous root with an abnormal crown to root proportion (Figs. 1 and 2). The soft tissue attached to the tooth and soft tissue enclosed within the dilated root was removed and sent for histopathological examination. The tooth was sectioned into two halves which showed an invagination near root apex extending towards pulp chamber. The inner surface of the invagination was irregular and was lined by white area morphologically resembling enamel (Fig. 3). One half was made into ground section which showed a well formed coronal enamel, dentin but the pulp chamber was crescent shaped with irregular pulpal canals. The radicular portion showed disorderly arrangement of dentin, enamel and inter radicular cementum but all the structures were well formed. The enamel was present between the root dentin and cementum within invagination. This radicular enamel was well-formed with enamel rod structure (Figs. 4 and 5). The developmental anomaly was diagnosed as true form of



Fig. 1 – Gross specimen of extracted dilated dens invaginatus showing abnormal morphology of root with periapical soft tissue.

radicular dens invaginatus with severe malformation resembling dilated odontoma. The soft tissue pathology revealed cystic space predominantly lined by pseudostratified ciliated columnar epithelium with plenty of mucous cells, clear vacuolated cells. The underlying connective tissue capsule was delicate, edematous with foam cells, mast cells, inflammatory cells, blood vessels; extravasated RBC's suggesting it to be radicular cyst with mucous metaplasia (Fig. 6).

3. Discussion

Dens invaginatus is a variation in the development of a tooth, which is thought to occur when there is an infolding of the surface of the crown before calcification has begun. Radicular dens invaginatus is a rare dental anomaly with bizarre radiographic appearance. Very few cases with enamel lined invagination within root and a rare form with inter-radicular crown have been reported.[9,12,13]

Radicular variety is further distinguished into two types, the first type originally described by Cohen who suggested the



Fig. 2 – Gross specimen showing the abnormal proportion of root to crown.

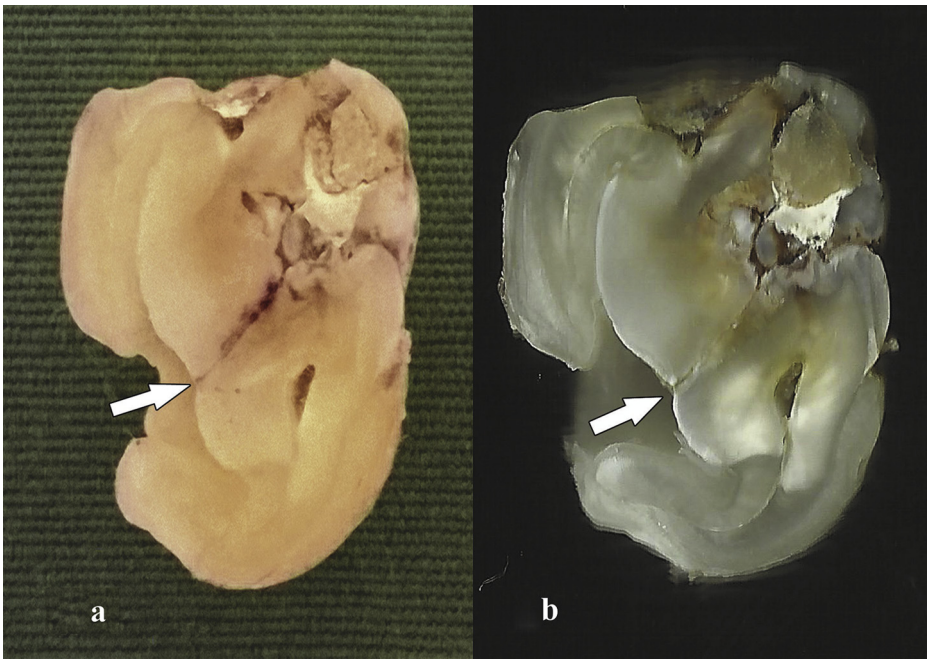


Fig. 3 – (a) Sectioned tooth showing an invagination near root apex which extended towards pulp chamber. (b) Scanned image of sectioned tooth showing inner surface of the invagination lined by enamel.

abnormality was the result of an incomplete attempt at root bifurcation with the depth of the infolding consists of cementum. Oehler considered this as a variation of root morphology which occurs more frequently. The second distinct type which conforms with concept of true "dens invaginatus", invagination arises from a foramen on root surface and instead of cementum, enamel is found to line its internal surface. The possible mode of formation is with infolding and proliferation of Hertwig's epithelial root sheath and differentiation of epithelial components into ameloblasts which subsequently result in enamel lined invagination within root. The invagination may contain connective tissue continuous with periodontal membrane and metaplasia may form osseous deposits.[4]

Clinically the extent of invagination and degree of dilatation may cause correspondingly distorted and dilated root.



Fig. 4 – Macroscopic scanned ground section showing inverted arrangement of root dentin, root enamel and cementum. (CE – Coronal enamel, D – Dentin, RE – Root enamel, C – Cementum). Square corresponds to the area shown in Fig. 5.

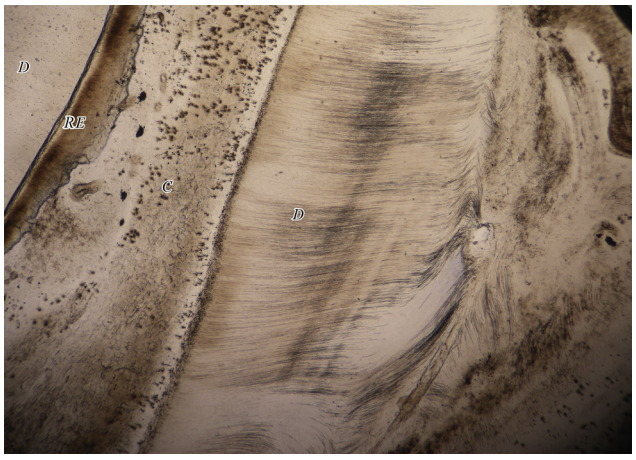


Fig. 5 – Photomicrograph of ground section (40×). The root enamel (RE) present between the dentin (D) and the interradicular cementum (C) and showed well formed enamel structure.

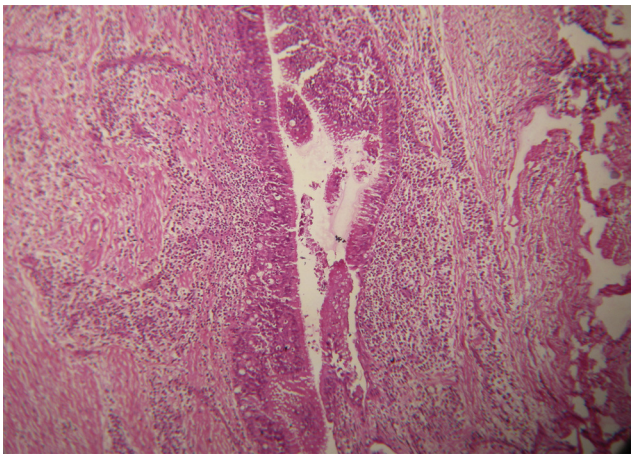


Fig. 6 – Photomicrograph (10×) Cystic lining consisting of pseudostratified ciliated columnar epithelium and connective tissue capsule showing inflammatory cells, foam cells.

From clinical point of view there remains no communication between the invagination through its opening on the root surface and the oral cavity but gingival recession and passage of infection from sulcus may cause infection and necrosis of tissues within the core of invagination and restorative management difficult.[4,9]

A dens invaginatus also has a variant showing dilatation of the crown and/or root of the affected tooth. The most severe form of the dilated type of dens invaginatus is called a dilated odontoma, although it is not clearly defined as an independent entity. A dilated odontoma has a circular or oval shape with radiolucent interior and presents a single structure often accompanied with central soft tissue and/or hard tissue.[14,15]

The present case showed a well formed crown with the invagination at the apex of a dilated, round, bulbous root. The interior of the root invagination was visible after sectioning and was lined by fine strip of well formed enamel which was extending between dentin and cementum with inverted hard tissue structure. The central area was filled with fibrous tissue and cystic lining extended from this area. The pulpal space was crescent shaped with irregular pulp canals. Similarly, Matsusue et al showed a completely inverted hard tissue structure due to severe invagination in incompletely formed dilated odontoma. The outer hard tissue was composed of dentin with dentinal tubules, a thin layer of immature enamel within the invagination and the central area was filled with fibrous tissue and small nest of bone and cementum.[14]

The radicular or periapical cyst arises from the epithelial residues in the periodontal ligament as a result of inflammation caused by irritation from a tooth with a necrotic pulp. They are usually lined by stratified squamous epithelium and the epithelial linings may have various forms of metaplasia and degeneration, for example, mucous cells, ciliated cells, para- and/or ortho-hyperkeratinization and formation of hyaline bodies.[10,11] The incidence of ciliated cells which are accompanied by mucous cells, in different studies varied from 0.8 % to 10%.The origin of mucous secreting cells in the lining of odontogenic cysts has been attributed to the grafting of such cells from contiguous epithelium normally containing mucous cells, i.e. the lining of the antral cavity, to presence of embryological pluripotential cells in the epithelial residues from which the cysts arise or to metaplasia of the cells in response to their altered environment.[10,16]

In conclusion, the present case is a rare form of true radicular dens in dente with formation of enamel within the root and had dilated root resembling dilated odontoma. It occurred in maxillary third molar area with radicular cyst lined by ciliated columnar epithelium which in itself is rare. Hence recognition of such entities is important and since such anomalies cannot be treated conservatively, tooth removal is the appropriate treatment modality.

Conflicts of interest

All authors have none to declare.

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