IOSR Journal of Dental and Medical Sciences (IOSR-JDMS)
e-ISSN: 2279-0853, p-ISSN: 2279-0861.Volume 23, Issue 6 Ser. 5 (June. 2024), PP 01-05
www.iosrjournals.org

Dentigerous Cyst With Dilated Odontoma: A Rare Combination

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Abstract
Dentigerous cysts are common odontogenic cysts typically associated with the crown of an unerupted tooth. However, the presence of dilated odontoma within dentigerous cysts is relatively rare. This case report highlights a unique occurrence of a dentigerous cyst associated with a dilated odontoma in an 11-year-old male patient who initially presented with localized swelling on the left side of the lower jaw. CBCT imaging revealed a complex lesion. Odontoma and cyst were surgically enucleated under general anaesthesia and sent for histopathological analysis. Histologically, the findings were indicative of a dentigerous cyst linked with a dilated odontome.. Early detection and comprehensive treatment planning through multidisciplinary collaboration among oral surgeons, radiologists, and pathologists are pivotal for achieving favourable outcomes in patients with such rare conditions. The unusual site of occurrence, age and manner of presentation and its association with a dentigerous cyst makes it interesting.
Key Word: *Dilated, odontoma, Dentigerous, Dens invaginatus*

Date of Submission: 03-06-2024 Date of Acceptance: 13-06-2024

I. Introduction

"Dens invaginatus," also known as dens in dente or dilated odontoma, represents a rare developmental anomaly of the teeth characterized by the invagination of the enamel organ into the dental papilla before mineralization. Historically, this condition was described as "dens in dente," reflecting the appearance of a tooth within a tooth, although current understanding suggests that it typically arises from the invagination of a single enamel organ. Although frequently and erroneously called ‘dilated odontoma’, dens invaginatus must be differentiated from an odontoma

The incidence of this anomaly varies widely, ranging from 0.25% to 10%, with a predilection for affecting the maxillary lateral incisors.(1) The severity of dens invaginatus can range from minor pitting to extensive invagination involving both the crown and root of the affected tooth.

Classification of dens invaginatus, particularly based on the depth of invagination, aids in understanding its severity and guides treatment decisions. Oehlers classified dens invaginatus into three types, with type III representing the most extreme form, often termed as Dilated Odontome (DO). DO is characterized by a complete inversion of the tooth's hard tissue structure due to profound invagination, sometimes accompanied by soft tissue abnormalities, leading to abnormal tooth development.

Diagnosis of DO primarily relies on radiographic evaluation, with advanced imaging modalities such as computed tomography (CBCT) providing detailed visualization of the internal anatomy, facilitating accurate diagnosis and treatment planning. However, differential diagnosis is essential, considering other conditions such as cemento-ossifying fibroma and osteoma.

This paper presents an extremely rare case of dilated odontoma associated with a dentigerous cyst, highlighting multidisciplinary management involving oral surgeons, radiologists, and oral pathologists for accurate diagnosis and appropriate treatment.

II. Case Presentation

An 11-year-old boy was referred to the Department of Oral and Maxillofacial Surgery at Goa Dental College Bambolim for detailed evaluation of swelling on the left side of his lower jaw. The patient noticed the occurrence and progressive increase in size swelling over past one month.

On extraoral examination, mild facial asymmetry with diffuse localized swelling was noted in the left parasymphysis region of mandible. The swelling was mildly tender and bony hard on palpation, with normal temperature of overlying skin. Intraoral examination revealed a single, localized, mildly tender, bony hard swelling measuring approximately 2 × 1 cm in the left lower buccal vestibule, adjacent to the canine and premolar teeth. No inflammatory signs were noted in the gingiva. The affected teeth were non-vital and asymptomatic, with no evidence of caries, periapical pathology, or periodontal involvement. The patient had no significant medical history. No abnormalities were found on physical examination.

Radiographic Findings

CBCT showed a well-defined ovoid hypodense lesion measuring approximately 24.2 mm × 18.2 mm × 20.2 mm(figure 1). It extended from root apex of #3-3 up to root apex of #3-5. Superior-inferiorly, it was approximately 4 mm superior to the inferior border of mandible. Bucco-lingually, it extended from the buccal to lingual cortex with cortical expansion and well-defined, corticated borders (figure 2). Two focal hyperdense areas were noted: one resembling enamel and dentin located superiorly, and another oval-shaped hyper density with a central hypodensity located inferiorly.

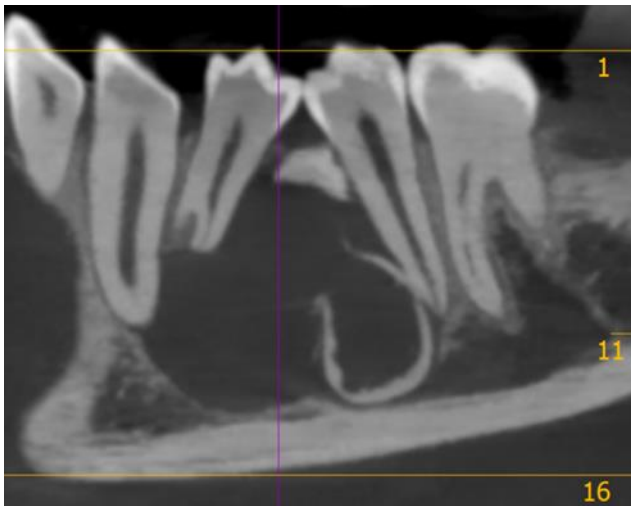

Figure 1:CBCT showing well-defined ovoid hypodense lesion with 2 focal hyperdense areas between the left mandibular premolar. Roots of premolar appeared to be diverged with resorption of root of I premolar, cortical outline of inferior alveolar nerve canal.

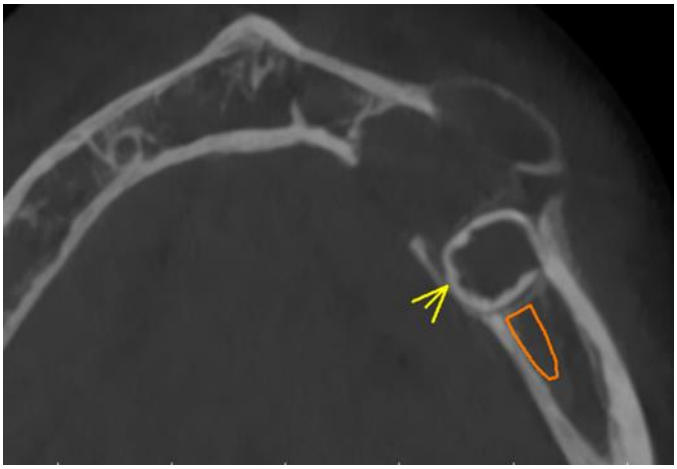

Figure 2:CBCT showing expansion thinning and loss of buccal and lingual cortices.

Surgical procedure

Surgical removal of dilated odontoma and the cyst was accomplished under general anaesthesia. On aspiration , straw coloured fluid was seen(figure 3a). A full thickness mucoperiosteal flap was reflected buccally between left mandibular central incisor to mandibular first molar. A thin layer of bone over the buccal surface was removed and tooth like structure along with cystic lining was exposed(figure 3b). Tooth like structure along with cystic lining was enucleated and the bony cavity was curreted, irrigated and packed with antibiotic sacked ribbon

gauze(figure 3c). The flap was replaced and secured with sutures (figure 4a). Tooth like structure along with cystic lining was sent for histopathological examination(figure 4b). Ribbon gauze was removed after 48 hours.

Figure 3 (a): A clear or straw- coloured fluid aspirated from lesion during diagnostic evaluation. (b):Intraoral view showing the exposed tooth-like structure along with the cystic lining (c): Surgical site following enucleation

Figure 4 (a): Surgical cavity packed with antibiotic-soaked ribbon gauze and mucoperiosteal flap replaced and secured with sutures. (b): Specimen of the tooth-like structure along with the cystic lining sent for histopathological examination.

Histopathological Findings

Microscopic examination of the cystic lining revealed stratified squamous non-keratinized reduced enamel epithelium-like lining with underlying fibrocellular connective tissue stroma(figure 5a). Microscopic examination of the tooth-like structure showed dentinal tubules, enamel rods, and entrapped pulpal tissue consisting of collagen fibers, fibroblasts, and blood vessels(figure 5b). Tall columnar cells with polarized nuclei were observed in some areas, suggestive of odontoblasts and ameloblasts associated with developing dental hard tissues. Histopathological features were suggestive of dentigerous cyst associated with dilated odontome.

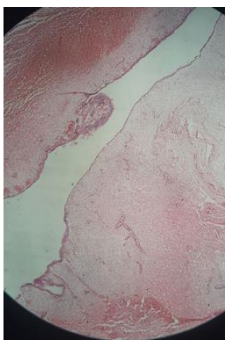

Figure 5 (a): Microscopic view of the cystic lining, demonstrating a stratified squamous non-keratinized epithelium resembling reduced enamel epithelium, supported by a fibrocellular connective tissue stroma. (b):Microscopic examination of the tooth-like structure reveals dentinal tubules, enamel rods, and entrapped pulpal tissue composed of collagen fibers, fibroblasts, and blood vessels.

III. Discussion

Dilated odontoma is considered the most severe form of dens invaginatus, characterized by a dilatation of both the crown and root of the affected tooth. Historically, the journey of DI and DO in dental literature can be traced back to the late 18th and early 19th centuries. One of the earliest observations of invagination within dental structures was by Ploquet in 1794 in a whale's tooth. Subsequent reports by Salter and Socrates in the 19th century further delineated the characteristics of these anomalies in human teeth. Swanson and McCarthy first presented a report of bilateral dens invaginatus. Ruston described the characteristics of dilated odontoma. Crincoli et al. reported a case in the second molar region of the maxilla, showing a pumpkin-like morphology and a C-shaped pulp cavity. Stavrou et al. reported a dilated odontoma in the root of the third molar of the maxilla. Conklin reported multiple dens in dente involving all four maxillary incisors and bilateral dens in dente in the mandibular incisor region associated with apparent congenital absence of two mandibular incisors. Although this anomaly in mandibular teeth is rare, Banner reported bilateral dens in dente in invaginations together with paradontal radiolucencies in mandibular premolars. Joubert et al. reported a dilated odontoma in the third molar of the mandible. A case of multiple dens in dente involving both maxillary first premolars, a maxillary lateral incisor and three mandibular premolars has been reported by Bottomley and Johnson.(2,3)

There remains variability in the terminology used in the literature, which can sometimes lead to confusion in diagnosis and classification. (4) Over the years, the term dilated odontoma (DO) has been used by some as part of the classification of odontomas, in which compound and complex odontomas are included, adding dilated odontoma as an additional variant; however, simultaneously, the denomination began to be used as a synonym to refer to the most complex variant of Dens invaginatus.(3,5)

Etiologically, several theories have been proposed to explain the development of DI and DO, including alterations in the internal epithelium of the enamel organ, trauma, genetic factors, and localized infections. There are theories proposed by different authors, such as Kronfel (1939), who proposes that it results from the delay in the growth of a portion of the internal enamel epithelium, while the remaining tissue continues to proliferate in a peripheral direction, while Bruszt (1950) mentions, in his Twin Theory, that the DI is a consequence of the fusion of two dental germs. Localized infection (Fischer, 1936; Sprawson, 1937) is considered a causal factor, as well as growth pressure causing dental germ instability (Euler, 1939; Atkinson, 1943), and traumas. (6,7)

Clinically, pain or swelling related to periapical infection are often the primary symptoms leading to the discovery of DO, mirroring observations in our case. While conventional radiographs like panoramic X-rays and intra-oral radiographs were employed initially, they lacked the necessary detail for a definitive diagnosis of the mineralized structure. Recognizing this limitation, we turned to cone beam computed tomography (CBCT) as a complementary imaging modality. CBCT proved instrumental in confirming the diagnosis of DO and provided invaluable insights into its relationship with adjacent teeth, as well as the extent of associated cystic lesions. By offering detailed internal anatomy in three dimensions, CBCT empowered us to tailor treatment plans accurately and anticipate potential procedural challenges with confidence.

The treatment of Dens Invaginatus (DI) varies depending on the severity of the condition. For Type I DI, which involves minimal invagination confined to the crown, prophylactic measures like fissure sealing may suffice. In Type II DI, where the invagination extends into the root but remains within it, options include restoration or endodontic treatment to address any pulp involvement. However, Type III DI, the most severe form, often requires surgical intervention due to complex root anatomy.(1) In our case, involving a severe form of DI, the decision was made to surgically remove the dilated odontoma and associated cyst under general anesthesia. A mucoperiosteal flap was reflected, and the affected tooth-like structure along with the cystic lining was enucleated. The bony cavity was then curretted, irrigated, and packed with antibiotic-sacked ribbon gauze. Histopathological examination of the excised tissue was performed, and the flap was replaced and secured with sutures.

Histologically, it is observed that the coronal invagination is lined with enamel within the dental papilla; the exposed pulp usually produces inflammation and necrosis, while the root part is lined with cementum and there is a fold of Hertwig's sheath within the developing root, which is associated with periapical lesions; therefore, it can be tubular or dilated below an initial bulbous shape like a large cavity, which modifies this pathology in a complex way, as all hard tissues produce macro and microstructured alterations. Collaboration among oral and maxillofacial surgeons, radiologists, and pathologists is paramount in delivering patient-centered care and achieving favorable treatment outcomes in individuals afflicted with this condition.

IV. Conclusion

Dentigerous cysts arising from dilated odontomes in the mandible represent a unique clinical entity that requires optimal management. Through a comprehensive understanding of the underlying pathology, meticulous diagnostic evaluation, and judicious surgical intervention, clinicians can effectively address these lesions and mitigate associated morbidity.

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