Cone Beam Computed Tomography in Diagnosis and Conservative Management of Ohlers Type III Dens Invaginatus associated with Dentigerous Cyst: A Case Report

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Abstract

Dens in dente, also known as dens invaginatus, dilated composite odontoma, or deep foramen caecum, is a developmental malformation that usually affects maxillary incisor teeth, particularly lateral incisors. It may occur in teeth anywhere within the jaws, other locations are comparatively rare. It can occur within both the crown and the root, although crown invaginations are more common. The use of cone beam computed tomography (CBCT) is very helpful in diagnosis of complex anatomic variations. We present a rare case of Type III dens in dente or Dens Invaginatus (Oehler's Type III, Dilated Odontoma) in association with dentigerous cyst in a 16 year old female diagnosed by CBCT and treated by conservative surgical approach.

Keywords: apical surgery, cone beam computed tomography, conservative management, dens invaginatus, dilated odontoma, dentigerous cyst,

Introduction

Dilated odontoma a variant of dens invaginatus is a developmental tooth anomaly that shows wide morphological variations resulting from infolding of the enamel organ into the dental papilla before the mineralization phase. Prevalence of dilated odontoma ranges from 0.25% to 7.74%. Even though it occurs in both the deciduous and permanent dentitions, it affects mostly the maxillary permanent incisors, particularly the lateral incisors and very rarely the posterior teeth. Dilated odontoma currently do not feature in the most recent classifications of odontogenic tumors as an independent entity. They have been documented to occur with other developmental anomalies such as microdontia, oligodontia, macrodontia, taurodontism, etc or in association with cystic lesions especially dentigerous cyst. Dilated odontoma shows a completely inverted structure of hard tissue due to the severe invagination of the enamel organ into the developing dental papilla, presenting radiographically as a shell-like structure with an outer radiopacity and a central core of radioluclency. Here, we present extremely rare case of Type III dens in dente or Dens
Invaginatus (Oehler's Type III, Dilated Odontoma) in association with dentigerous cyst in a 16 year old female diagnosed by diagnostic modalities; Cone Beam Computed Tomography (CBCT) and treated by conservative surgical approach.

Case Report
A 16-year-old female patient came to the department of oral medicine and radiology with a complaint of pain in upper front tooth region since 2 months (Figure 1).

![Figure 1: Front profile of patient](image)

Patient was apparently normal 2 month back, later she felt dull and continuous pain which was sudden in onset, gradually progressed, aggravated while chewing and got relieved by its own. Patient's medical, family and past dental history was non contributory. General Physical examination revealed that she was moderately build and nourished and all the vital signs were within normal range. On extraoral examination no abnormality was detected. On intraoral examination, all the teeth were present except 23, fusion in relation to (i.r.t) 21, and peg shaped 22 was seen (Figure 2).

![Figure 2: Shows Developmental anomaly, fusion i.r.t. 21](image)

On palpation, vestibular tenderness i.r.t 22, 24 and bulge with crackling was present and teeth responded positively to pulp vitality testing. Patient was subjected to radiological investigations; intra oral periapical
radiograph and (IOPA) maxillary occlusal view (Figure 3 a, b) revealed bulbous root of 21 associated with open apex, well circumscribed round periapical radiolucency measuring approximately 1x1.5 cm i.r.t 21, 22.

Figure 3 a) Intraoral Periapical Radiograph

Figure 3b) Maxillary Occlusal view reveals well defined periapical radiolucency i.r.t 21,22 tooth within a tooth extending upto root apex i.r.t 21 with wide open apex.

Tooth within a tooth extending upto root apex i.r.t 21 was clearly evident. Peg shaped 22 and mesioproximal radiolucency involving enamel, dentin approaching pulp was seen i.r.t 12 on occlusal view. Panoramic radiograph revealed well defined pericoronal radiolucency i.r.t impacted 23 (Figure 4). Based on the clinical findings and radiological investigations, diagnosis of Type II dens in dente (dens invaginatus, dilated odontoma) i.r.t 21 with open apex and dentigerous cyst i.r.t. 23 was given. Radicular cyst i.r.t. 21,22 was considered under differential diagnosis.
Fine needle aspiration cytology (FNAC) was performed and mixed fluid was aspirated. Smear showed cells were discrete in pattern, majority of the cells were lymphocytes, macrophages and plasma cells in haemorrhagic background, findings were indicative of chronic inflammatory lesion. CBCT imaging was performed for more comprehensive understanding of the crown – root morphology; coronal view revealed well circumscribed, pericoronal hypodense lytic lesion surrounded by circular hyperdense rim in the left anterior maxilla i.r.t. impacted 23, below the level of roots of 22 and 24 extending mediolaterally from lateral aspect of apical 1/3 rd root of 22 till mesial aspect of apical 1/3 rd root of 24 and superiorly till the floor of nasal fossa measuring approximately 23x13 mm. Bulbous root was seen i.r.t 21, invagination started at central middle third of crown 21, divided it into two radiolucent pulpal canals that opened at the apex as wide radiolucent foramen (Figure 5).

3 Dimensional (3D) CBCT images showed well defined expansile pericoronal lytic lesion i.r.t 23 that resulted in thinning and perforation of labial cortical plate, this suggested that 23 was erupted close to the labial cortical plate and near the nasal floor. 21 was seen as deformed, had roughly oval shaped crown and wide open apex (Figure 6, 7). The treatment options were explained to the patient and informed consent was
obtained. After routine blood investigations that were within normal limits, enucleation of the cyst followed by surgical extraction of 23 was done under general anestheisia (Figure 8).

Figure 6: CBCT 3D view reveals expansile pericoronal lytic lesion i.r.t impacted 23 perforating the labial cortical plate

Figure 7: CBCT 3D view shows 21 as severely deformed with roughly oval shape crown

Figure 8: Enucleation of cyst followed by surgical extraction of 23

Complete curettage of the soft tissue and removal of the granulation tissue was done. Histopathological examination, Haematoxylin and Eosin (H&E, 10 x) stained section showed hyperplastic epithelial lining
with cystic space and exocytosis, the underlying connective tissue showed diffuse infiltration of chronic inflammatory cells (Figure 9).

Based on the CBCT and histopathological findings, we arrived at a diagnosis of chronic inflammatory cyst i.r.t. 23 and dilated invaginated odontoma i.e. 21. At a subsequent appointment, 21 and 22 was prepared for endodontic treatment. Apical surgery was planned for 21 followed by retrograde mineral trioxide aggregate (MTA) placement with thermoplastisized gutta-percha obturation. Patient was kept under regular follow up for 3 months and no recurrence of signs of cystic lesion was observed (Figure 10).

**Discussion**

Several authors have investigated the occurrence of dens invaginatus and found maxillary lateral incisors are the most commonly involved teeth with a prevalence of 38.5%–75%. Among Indians, the prevalence of dens invaginatus is as low as 0.4%. The invagination of the inner enamel epithelium may occur in more than one point within the tooth resulting in double or even triple dens invaginatus. Usually, the invagination originates as a pit on the coronal aspect that predisposes to caries and is separated from the pulpal tissue by only a thin layer of enamel and dentine. The invagination may occur within the root canal system or may exist separately, it may also present with intercommunications with the main canal which explains the rapid onset of pulpal necrosis.

According to World Health Organization (WHO) classification, odontomas can be divided into three groups: i) complex odontoma in which the calcified dental tissues are simply arranged in an irregular mass bearing no morphologic similarity to rudimentary teeth; ii) compound odontoma is composed of an odontogenic tissues arranged in an orderly pattern, result in many teeth-like structures, but without morphologic resemblance to normal teeth; and iii) ameloblastic fibro-odontoma consists of varying amounts of calcified dental tissue and dental papilla-like tissue, the later component resembles an ameloblastic fibroma. The ameloblastic-fibro odontoma is considered as an
immature precursor of complex odontoma. H M Worth et al described dilated odontoma as the lesion in which the crown or root part of tooth shows marked enlargement. The most widely accepted classification of this anomaly has been proposed by Oehler in 1957 who categorized it based on radiographic observation according to the depth of enamel invagination. Type I represents enamel lined invagination confined to the coronal part of the tooth; Type II extension of the invagination beyond the cementoenamel junction ending as a blind sac. Type III includes permeation of the root by the invagination to form an additional canal opening on lateral side of root.

Etiology of dens invaginatus is debatable, still many theories have been proposed towards occurrence of this anomaly. Stavrou et al reported a case of dilated odontoma in the root of maxillary third molar, proliferation and ingrowth of Hertwig's epithelial root sheet may have occurred into the dental papilla at an earlier stage of morpho differentiation, therefore crown of the tooth was normal in shape and size but marked dilation was seen in the root. Genetic etiology could not be excluded, it has been documented that dens invaginatus most commonly occurs in the siblings of the patient or is inherited from the parents and may accompany other developmental abnormalities such as microdontia, macrodontia, oligodontia, fusion, taurodontism etc. Literature revealed cases of dilated odontoma categorized between dens invaginatus and complex odontoma, with bizarre malformation of tooth structure. Crinicoli et al reported dilated odontoma in the second molar region of the maxilla that showed a pumpkin-like morphology with reduced ratio of both crown and root. In another case study, Joubert et al reported dilated odontoma in the third molar of the mandible with bizarre malformation of the crown and upper half of the root, features resembled complex odontoma.

Another interesting finding in the present case was radiographic diagnostic dilemma, well circumscribed periapical radiolucent lesion i.r.t. 21, 22 was appreciable on intraoral radiographs but further panoramic and CBCT images revealed pericoronal radiolucency surrounded by radiopaque rim i.e. impacted left maxillary canine, 23 that led to the diagnosis of dentigerous cyst. CBCT in this case helped us in proper understanding of the complex anatomy of the tooth and its association with dentigerous cyst. Dentigerous cyst encloses the crown of an unerupted tooth by expansion of the follicle and is attached to its neck. They are commonly associated with an unerupted tooth, impacted mandibular third molars, maxillary canines, mandibular second premolars and maxillary third molars, and rarely involve supernumerary and deciduous tooth. In our case, pericoronal radiolucency i.r.t. impacted left maxillary canine was highly suggestive of dentigerous cyst. In majority of cases dentigerous cysts are treated by enucleation followed by extraction of the involved tooth, large dentigerous cysts require marsupialization when enucleation and curettage might result in neurosensory deficit. The prognosis for most histopathologically diagnosed dentigerous cysts is excellent, recurrence being a rare finding. Occasionally, dentigerous cysts may transform into squamous cell carcinoma, mucoepidermoid carcinoma or an ameloblastoma.

Clinical management of dens invaginatus is influenced by the degree of complexity of anatomy of the tooth. Treatment strategies range from conservative restoration of the opening to non-surgical endodontic treatment, endodontic surgery, intentional reimplantation, and finally extraction. Large and extremely dilated invaginations often have abnormal crowns and need to be extracted. Similarly in the present case, conservative surgical treatment was planned for 21, intracanal retrograde MTA placement was followed by thermo plasticized gutta-percha obturation. Enucleation of the cyst was done followed by surgical extraction of impacted 23 under general anesthesia and no recurrence was reported at 3 months regular follow up visits.

Conclusion
Dilated Odontoma can present in diverse ways as seen in our case in which it was associated with dentigerous cyst i.r.t impacated canine. Well defined radiolucent lesion was a diagnostic challenge, it was seen in periapical location of a tooth with dilated odontoma on intraoral radiographs, but was seen surrounding the crown of impacted left maxillary canine on panoramic and CBCT images. Earlier treatment for tooth with dilated odontoma was limited to extraction, successful surgical management of this case was possible with the use of 3D imaging modality; CBCT that enabled us to understand the aberrant canal morphology better, and the cystic lesion associated with impacted canine. Thorough history, clinical examination and the striking radiographic features well appreciable on CBCT images will help the clinicians to arrive at accurate diagnosis and tailor better treatment plan of this rare developmental anomaly.
References

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