

The presence of dens invaginatus (DI) and dens evaginatus (DE) on same tooth is a rare phenomenon. However, when these dental anomalies occur on a double tooth, it becomes an extremely rare phenomenon. The authors report a rare case of DI and DE on fused permanent maxillary central incisor with supernumerary tooth in a 40-year-old male. The present article also focuses on the differentiating fusion from gemination and also reviews preventive and management strategies for tooth with complex dental anatomy.

Keywords: Dens evaginatus, dens invaginatus, double tooth

How to cite this article:

Sharma G, Mutneja AR, Nagpal A, Mutneja P. Dens evaginatus and dens invaginatus in a double tooth: A rare case report. Indian J Dent Res 2015;26:545-9

How to cite this URL:

Sharma G, Mutneja AR, Nagpal A, Mutneja P. Dens evaginatus and dens invaginatus in a double tooth: A rare case report. Indian J Dent Res [serial online] 2015 [cited 2021 Sep 19];26:545-9. Available from: <https://www.ijdr.in/text.asp?2015/26/5/545/172085>

Double tooth is a rare developmental anomaly of morphology which occurs more commonly in deciduous dentition than permanent dentition with a low prevalence of 0.5% and 0.1%, respectively. [1] Various terminologies such as "double tooth," joined teeth, or connoted teeth have been used to describe fusion or gemination and concrescence. [2] The etiology of double tooth remains unknown. Various factors such as pressure or physical force, local metabolic disturbances, viral infection during pregnancy, hypervitaminosis A, and genetic factors have often been implicated as a causative agent for resulting in double tooth. Fusion is conjoining of two tooth germs during odontogenesis. The pulp chambers and canal may or may not be joined depending on the stage of development. [1] Fusion can occur between teeth of same dentition and between normal and supernumerary teeth. In comparison, gemination implies the division of a single tooth germ into a single large tooth with bifid crown with incisal notch or groove and usually common root and canal. [2] Clinical appearances of fusion and gemination are similar. A patient usually has a normal complement of teeth in gemination, whereas in fusion the number is reduced by one unit.

Dens evaginatus (DE) is an uncommon dental anomaly with a well-defined morphologically altered cusp-like structure projecting from the occlusal area of premolars or cingulum area of the anterior teeth. It is composed of normal enamel and dentin, has varying extensions of the pulp tissue, or maybe devoid of the pulp tissue. The characteristic appearance of DE (also termed as talon's cusp in anterior teeth) is conical and resembles an eagle's talon. [3] Dens invaginatus (DI) is a deep surface invagination of crown or root that is lined by enamel and dentin. DI arises from an invagination of dental follicle during morpho-differentiation stage. The prevalence of this dental anomaly has been found to be 0.25-10.0% of the population. [4] The diagnosis of DI is mainly based on the radiographic presentation of radiopaque ribbon or pear shape-like structure with equal density as enamel. The teeth most commonly affected with DI are permanent lateral incisors, central incisors, premolars, canines, and molars. [4]

Very few cases of DE and DI existing on same tooth have been documented. [5],[6] However, when these two concurrent dental anomalies occur on a double tooth, it becomes an extremely rare phenomenon. To the best of our knowledge, this is the first-ever reported case in which a myriad of dental anomalies, i.e., double tooth, DE, and DI, are coexisting together on the single tooth (permanent maxillary central incisor). The present article also focuses on the differentiating fusion from gemination and also reviews preventive and management strategies for a tooth with complex dental anatomy.

Case report



A 40-year-old male reported to the Department of Oral Medicine and Radiology with the chief complaint of pain in the upper right posterior region of jaw since 1 week. There was no history of swelling or pus discharge in the same area. No history of trauma could be elicited. There was no significant dental, medical, and family history.

Patient's extra-oral examination revealed no abnormalities. On intraoral examination, a double tooth with an altered morphology was observed in the left maxillary central incisor which was malpositioned partially [Figure 1]. On closer observation, a partial cleft could be observed labially in the same tooth. The groove was noncarious and stained. The palatal aspect of morphologically altered tooth revealed a talon's cusp extending approximately 7 mm [Figure 2]. Maxillary lateral incisor was also observed palatally and a dental floss could be passed through it and the anomalous tooth. A full complement of teeth was however present clinically. There were no occlusal interferences. An intraoral radiograph of the double tooth revealed a single pulp chamber with a single canal [Figure 3]. However, on closer inspection, a root outline of supernumerary tooth over the lateral incisor could be visualized. There was also the presence of radiopaque structure present in the coronal surface of same tooth. A pulpal extension was evident in the anomalous structure. An invagination was also evident in double tooth extending till cemento-enamel junction (CEJ). No altered root morphology and periapical pathology was evident. On an occlusal radiograph, the two coronal halves of the double tooth were evident with two roots [Figure 4]. The root outline was superimposed over the lateral incisor. A panoramic radiograph revealed no other dental abnormalities. Vitality testing revealed the double tooth to be vital [Figure 5]. The impression of the maxillary teeth was made and casts were poured. The approximate mesial-distal diameter at midcoronal level of the double tooth was 13 mm, and cervico-incisal length was 15 mm. Talon's cusp could easily be discerned on the cast. Based on these findings, the permanent maxillary central incisor was diagnosed with multiple dental anomalies like fused maxillary central incisor with supernumerary tooth associated with DE and DI. Since the patient was asymptomatic (for this tooth) and not concerned about esthetics, he was not willing for any interventional treatment for this tooth. The patient was advised endodontic treatment for maxillary left second molar. The patient has been kept on regular follow-ups and the patient is asymptomatic.



Figure 1: Labial view of a macrodont maxillary central incisor with uneven coronal halves

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Figure 2: Palatal view showing the presence of dens evaginatus on maxillary central incisor and palatally placed lateral incisor

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Figure 3: Intraoral periapical radiograph showing the presence of dens invaginatus and dens evaginatus on double tooth (maxillary central incisor)

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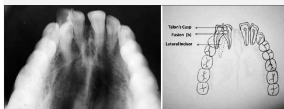


Figure 4: Anterior maxillary occlusal view showing the presence of fused central incisor with a supernumerary tooth and an illustration showing the presence of fusion of permanent central incisor and supernumerary

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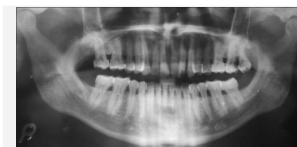


Figure 5: Panoramic radiograph showing the presence of double tooth (maxillary central incisor)

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Discussion



Fusion of normal and supernumerary tooth is unusual. [7] The distinction between fusion and gemination becomes difficult and challenging in this situation as the number of teeth in both these conditions remains normal. Supernumerary teeth are most commonly seen in anterior maxilla and usually have a conical form. The fusion of normal tooth and a conical-shaped supernumerary tooth will result in asymmetrical coronal shape of tooth, giving a crooked appearance. [8] In comparison, there is generally symmetry of the two halves of crown in gemination with both halves appearing as mirror images [Figure 6]. The characteristics of double tooth have been explained in [Table 1]. There were also well-defined gingival margins that could be evident clinically in different halves of tooth. If fusion occurs very late, it may lead to a broad clinical crown with an incisal notch or groove. The presence of groove can be observed both in gemination and fusion and radiographically, the presence of single pulp canal can be evident both in complete fusion and gemination.

Characteristics	Gemination	Fusion between two normal set of teeth	Fusion between normal tooth and supernumerary tooth
Common location	Anterior maxilla	Anterior mandible	-
Denition	Normal	Absence of adjacent teeth	Normal complement of teeth
Quadrant involvement	Unilateral	Unilateral	Unilateral
Clinical appearance	Bifid crown, each half symmetrical appearance	Unsymmetrical, crooked appearance	Unsymmetrical, crooked appearance
Radiographic appearance	Single root with one canal	Mostly two separate pulp chambers and root canals	Variable

Table 1: Depicts the differences in characteristics of gemination and fusion

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The visualization of two roots in this case required a very careful radiographic examination that was helped by the use of anterior occlusal radiograph. Utility of cone beam computed tomography could have been useful in differentiating this case but due to patient's refusal for radiation exposure, reluctance for any interventional treatment, and cost factor, it was not performed. The involved tooth here is more likely to be fusion between maxillary central incisor and supernumerary tooth than gemination because of asymmetrical halves of crown and visualization of two roots with pulp canals. Thus, the above case is an example of partial fusion involving only the crowns of teeth. Based on various case reports involving fusion and gemination, the authors have attempted a depicted illustration of various radiographic appearances of fusion, gemination, and twinning [Figure 6].

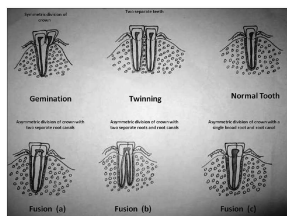


Figure 6: An illustrative diagram showing the presence of varied radiographic appearances of gemination, twinning, and fusion

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The etiology of DE is still unknown, though genetic and environmental factors have been implicated. The prevalence is reported to be only 0.06-7.7% with a predilection for maxillary lateral incisor. [9] DE has been found to coexist with mesiodens, shovel-shaped incisors, DI, double tooth, and odontomas. [9] Hattab et al. had classified talon cusp into three types according to the degree of the cusp formation and extension. [3] Type 1 is a morphologically well-delineated additional cusp that projects from the palatal surface to at least half the distance between CEJ and incisal edge. Type 2 refers to an additional cusp (≤ 1 mm) that may blend with the palatal surface or stand away from the rest of crown. It extends less than halfway between CEJ and incisal edge. Type 3 is an enlarged cingula that may have a conical, bifid, or tubercle-like appearance. Our case presented as Type 1 talon cusp variety presenting clinically as well-delineated additional cusp. Radiographic evidence of pulp extending in talon cusp was also evident.

The clinical appearance of DI, also known as dens in dente, varies considerably. DI has a female predilection and primarily occurs in permanent maxillary incisors. [4] The commonly used classification of DI was suggested by Oehlers. [10] Type 1 DI is characterized by invagination that is confined till crown, whereas Type 2 DI appears as deeper invagination extending apically beyond CEJ. In Type 3, DI presents as an invagination also extending beyond CEJ but opening either laterally in the periodontium or apically as a second apical foramen. In our case, Type 1 DI was evident radiographically.

Double tooth, similar to DE and DI, originates during morpho-differentiation stage of tooth development. It is remarkable that in this patient all these dental anomalies co-occurred together on the same tooth. Chondroectodermal dysplasia, focal dermal hypoplasia, and oro-facial-digital syndrome have been associated with fusion, [11] whereas DE has been documented in Rubinstein-Taybi syndrome, Mohr syndrome, and Sturge-Weber syndrome. [1] DI has been reported in Nance-Horan syndrome, William's syndrome, and Ekman-Westborg-Julien syndrome. [4] Our patient had none of any features suggestive of above syndromes and conditions. The etiology in our case could have been multifactorial with trauma most likely cause of the trigger for above malformation.

There is generally no intervention done for asymptomatic primary anterior double tooth until there are complications with esthetics, spacing, and caries. [12] However, the management of a fused permanent tooth with complex dental anatomy would serve as a challenge to a clinician. Various management procedures have been reported for a fusion between normal anterior tooth and supernumerary tooth. Fused teeth have been hemisectioned with removal of morphologically altered supernumerary tooth followed by endodontic treatment of normal tooth. [13] Other management strategies such as orthodontic therapy, recontouring of crown shape, and extraction of fused tooth followed by prosthetic management have been tried with varying success. [12] Our case is further complicated by the presence of coexisting dental anomalies such as DE and DI. Use of aqueous intracanal medicaments and 56 thermoplasticizing techniques has been successfully accomplished in cases of DI. [14] Various treatment modalities such as intentional reimplantation, removal of invaginated part, and even extraction for mobile necrotic teeth have been suggested. [15] However, our patient was asymptomatic and also not concerned about esthetics and thus refused any kind of treatment.

In a symptomatic complex morphologically altered tooth, there would be various therapeutic problems such as the placement of rubber dam, access opening (due to the presence of pulpal extension in DE), determining working length, and even prosthetic rehabilitation due to the presence of DE and DI in a double tooth. Patients should be properly informed of the anomalous condition and its potential sequelae. An early diagnosis can prevent the expensive therapeutic interventions and possibly reduce any associated physical and psychological discomfort due to compromised esthetics and function.

The main clinical problem associated with double tooth is their unsightly abnormally large crown leading to compromised esthetics. [12] Higher predisposition to caries, periodontal disease, and spacing problems has also been documented in double tooth. [2] The incisal groove can act a harbinger of bacterial plaque accumulation leading to periodontal disease. DE would present as a challenge to endodontic therapy in our case as a pulpal extension was radiographically evident. The other complications associated with DE are attrition, compromised esthetics, accidental cusp fracture, occlusal interference, irritation of tongue during speech, and susceptibility to caries. [9] The risk of pulpal necrosis and pulpitis is also high in patients with DI. [9] None of these complications except compromised esthetics were observed in our patient. The patient was advised sequela of condition and was advised regular follow-ups.

A good oral hygiene is imperative to maintain periodontal health. Simple composite restorations can be used to camouflage the double tooth and prevent caries developing in the fissures. [1] Selective grinding of palatal projection can be done to remove any premature contact and sealants can be placed to prevent caries both in incisal groove as well as palatally. An interdisciplinary management of oral and maxillofacial radiologist, endodontist, prosthodontist, periodontist, and orthodontist would be required in cases like these, which is as complex morphologically as it could possibly become, to achieve functional and esthetic success.

The rarity of all these dental anomalies occurring together on same tooth makes it a unique case. A detailed and an accurate radiographic examination is required for diagnosis and subsequently management.

Acknowledgement

The authors would like to thank Dr. Prachi Kathuria for contribution to preparation of the illustration of [Figure 6].

Financial support and sponsorship

Nil.

Conflicts of interest

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