A Novel Combination of *dens evaginatus* and *dens invaginatus* in a Single Tooth – Review of the Literature and a Case Report

Robert P Anthonappa * / Cynthia KY Yiu ** / Nigel M King ***

Dens evaginatus (DE) and dens invaginatus (DI) are rare developmental dental anomalies affecting both the primary and permanent dentitions. Concurrence of DE and DI within the same tooth is extremely rare. We report a case of DE and DI in a maxillary right lateral incisor tooth. Comprehensive clinical and radiographic examinations are essential to identify such defects; the early diagnosis can then result in the appropriate prophylactic treatment being performed, thus preventing undesirable pulpal complications.

Keywords: *case report, dens evaginatus, dens invaginatus*
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INTRODUCTION

Dens evaginatus (DE) is a rare developmental anomaly of a tooth resulting in the formation of an accessory cusp[1] comprising of enamel, dentin and varying amounts of pulp tissue.[2] This uncommon anomaly was first reported by Windel[3] where he mentioned two cases of supernumerary cusps on the palatal surface of permanent maxillary incisors and canines. Later, Mitchell[4] in her letter described a curved horn-like process extending from the palatal surface to the incisal edge of a permanent maxillary central incisor tooth. Since then numerous case reports have been published in the English literature,[1] and it is usually reported to be found on the occlusal surface of premolars or projecting from the lingual surface of anterior teeth in both the primary and permanent dentitions. Talon cusp occurs at the morphodifferentiation stage of tooth development, as a result of an out-folding of the enamel organ, or hyperactivity of the dental lamina.[5] It has a strong predilection for permanent maxillary incisors and may present unilaterally or bilaterally.

Dens invaginatus (DI), which is a rare malformation of the tooth, probably arises from an infolding of the dental papilla during tooth development,[6] it has a broad spectrum of morphological variations. It has been reported that DI was first described in a human tooth by a dentist named 'Socrates' in 1856.[7] It frequently affects the permanent maxillary lateral incisors, usually bilaterally[8, 9] and the morphology varies from a short pit confined to the crown of the tooth to a deep invagination into the root. Radiographically, it presents with an infolding of enamel and dentin (enamel located in the center which is covered by dentin peripherally due to the invagination), which may extend into the pulp cavity, the root, and sometimes even to the root apex.[6] Several classifications of invaginated teeth have been reported in the literature[6]; the most commonly used being the one proposed by Oehlers.[8]

The numerous reports of DE in the literature was recently reviewed by Levitan and Himel.[1] DE predominantly occurs in people of Asian descent (including Chinese, Malay, Thai, Japanese, Filipino, and Indian populations) with varying estimates reported at 0.5% to 4.3%, depending upon the population group studied.[10] Conversely, the prevalence of DI varies between 0.25% and 10% in different studies.[11] This variation is probably due to geographical differences and variations in diagnostic criteria and/or investigation methods. Talon cusp, which is a variation of DE, is found in the primary and permanent maxillary incisors with variations in size, shape, length and mode of attachment to the crown.[12] DE has been reported to be associated with numerous developmental dental anomalies like odontome, double tooth, supernumerary and impacted teeth, as well as several syndromes such as Mohr syndrome, Rubinstein–Taybi syndrome and Sturge–Weber syndrome.[12-16] The concurrence of DE and DI within the same tooth is a rare finding and has only been reported twice in the literature.[17, 18] Here we report a case of DE and DI affecting a maxillary right lateral incisor.

* Robert P. Anthonappa, BDS, MDS, AdvDipDS, Postgraduate student in Pediatric Dentistry, Faculty of Dentistry, University of Hong Kong, Hong Kong SAR, China.
** Cynthia KY Yiu, BDS, MDS, PhD, Clinical Associate Professor, Faculty of Dentistry, University of Hong Kong, Hong Kong SAR, China.
*** Nigel M King, BDS, MSc, PhD, Professor, Faculty of Dentistry, University of Hong Kong, Hong Kong SAR, China.

Send all correspondence to: Dr. Cynthia Yiu, Paediatric Dentistry and Orthodontics, Faculty of Dentistry, The University of Hong Kong, Prince Philip Dental Hospital, 34 Hospital Road, Hong Kong SAR, CHINA.

Tel: 852-28590256
Fax: 852-25593803

E-mail: ckyyiu@hkucc.hku.hk

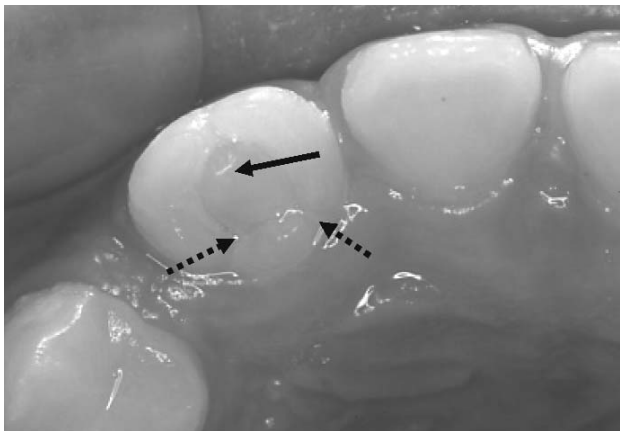

Figure 1. The palatal surface of the maxillary right lateral incisor with well defined grooves (dashed arrows) and dens evaginatus (arrow).

CASE REPORT

A 10-year-old Chinese boy attended for a routine dental examination. His medical and dental histories were unremarkable and the family history did not reveal any evidence of hereditary dental anomalies. His parents were dentulous and he was the only child in the family. The extra-oral examination showed a normal facial appearance. Intraorally he was in the mixed dentition stage with fair oral hygiene. The palatal surface of the maxillary right lateral incisor exhibited well defined developmental grooves with a projection (Figure 1), extending to more than half the height of the tooth crown; making this DE a type 1 talon cusp according to Hattab and his co-workers[5] see Table 1. The talon cusp neither irritated the tongue during speech and mastication nor interfered with the occlusion. A periapical radiograph of the maxillary right lateral incisor showed the talon cusp (DE) to be a typical inverted cone, with enamel, dentin and pulp extending only to the base of the cusp.

An invagination resembling a type II DI[8] [Table 2], involving the same tooth was found apical to the DE (Figure 2a). The invagination extended beyond the level of the cemento-enamel junction but was limited to the root of the tooth which had an open apex. The tooth was unaffected by caries or periodontal pathology and there was no evident restoration, or loss of vitality, but there was a notch in the incisal edge. Examination of the other teeth revealed carious lesions and restorations, also a fracture of the incisal edge of the maxillary left permanent central incisor confined to enamel. The molars on the left side were in a half-unit class II relationship, while the molars on the right side were in a class I relationship.

The patient has completed his restorative treatments and is currently under review. Composite restoration followed by fissure sealant has been placed on the developmental grooves of the maxillary right lateral incisor to reduce the accumulation of plaque and hence the risk of caries and pulpal pathology. Periapical radiographs of the same tooth (Figure 2b) at the time of one year review exhibited further root completion with no associated pathology. The tooth will be followed up clinically and radiographically at regular intervals.

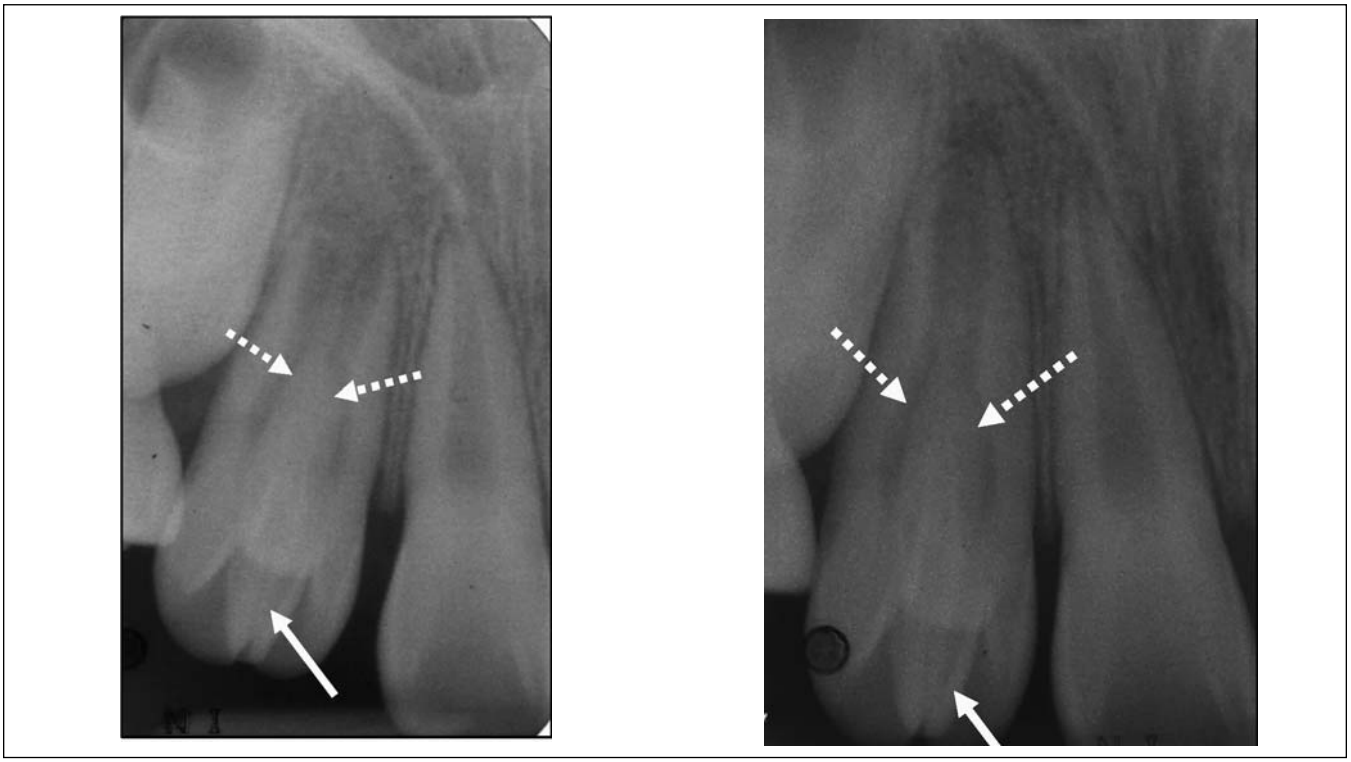

Figure 2. The periapical radiographs showing a DE (arrow) and a DI (dashed arrow) on the right maxillary lateral incisor.
a) at the time of diagnosis **b)** at the time of review a year later

Table 1. Classification of dens evaginatus according to Hattab *et al*. (1996)

Type	Name	Explanation
1	Talon	A morphologically well-delineated additional cusp that prominently projects from the palatal surface of a primary or permanent anterior tooth and extends at least half the distance from the cementoenamel junction to the incisal edge.
2	Semi talon	An additional cusp of a millimeter or more but extending less than half the distance from the cementoenamel junction to the incisal edge. It may blend with the palatal surface or stand away from the rest of the crown.
3	Trace talon	Enlarged or prominent cingula and their variations, i.e. conical, bifid, or tubercle-like. Radiographically it may appear typically as a V-shaped radiopaque structure, as for true talon and semi talon, or tubercle-like, originating from the cervical third of the root.

Table 2. Classification of dens invaginatus according to Oehlers (1957)

Type	Explanation
I	The invagination, which is enamel lined, is of the minor form. It is confined within the crown of the tooth and does not extend beyond the level of the external amelocemental junction.
II	The enamel-lined invagination invades the root but remains confined as a blind sac. There may be a communication with the pulp. The invagination may or may not be grossly dilated.
III	The invagination penetrates through the root and 'bursts' apically or laterally at a foramen. There is usually no communication with the pulp, which lies compressed within the wall around the invagination process.

DISCUSSION

The term *dens invaginatus* (DI) introduced by Hallet[19] is the preferred term to describe this anomaly, although dens in dente is still often used. Since its first description in human tooth, numerous theories have been proposed to explain the possible causes of DI; however, its etiology remains contentious and unclear. Nevertheless, most authors opine that DI occurs due to a deep infolding of the foramen coecum during tooth development, which in some cases, may even result in a second apical foramen.[6, 7] Furthermore, genetic factors cannot be excluded.[20]

Accessory cusps when observed on the lingual surface of anterior teeth are referred as talon cusp, because of its resemblance to an eagle's talon.[21] However, when present on posterior teeth, various terminologies such as, evaginatus odontoma (evaginated odontome), occlusal enamel pearl, occlusal tubercle, tuberculum anomalous, supernumerary cusp, interstitial cusp, tuberculated cusp, tuberculated premolar and Leong's premolar have been proposed.[1] Currently, DE[22] is the favored terminology used to describe this developmental abnormality, with a multi-factorial etiology involving both genetic and environmental factors. Several classifications have been proposed to describe the different types of DE, based on their shape,[23] location[24] and pulp contents.[22]

The present case report describes a type I talon cusp,[5] with a type II invagination[8] on the same tooth. Such a combination, in a single tooth, is extremely rare; however it was recently reported by Mupparappu and his co-workers,[18] who described DE and DI in the maxillary right lateral incisor of a 25-year-old male patient. Lorena and her co-workers[17] reported a similar condition of DE and DI in a maxillary left lateral incisor in a 21-year-old second-generation Japanese-Brazilian male, but the patient also had other dental anomalies including, two supernumerary teeth, carabelli's trait and shovel shaped incisors. The only other almost similar report described the occurrence of DI in a maxillary lateral incisor with a tubercle rather than a true DE.[25]

The definition of talon cusp has attracted a large variation ranging from descriptions as, a demarcated projection of a millimeter or more to a further prominent projection extending at least half the distance from the cementoenamel junction to the incisal edge on the lingual surface of anterior teeth.[26, 27] In the present case, periapical radiographs demonstrate the talon cusp as a V-shaped radiopaque structure superimposed on the image of the affected crown. It is outlined by two distinct white lines converging from the cervical area of the affected tooth towards the incisal edge. This clearly implies that the evagination in this case is a true talon based on Hattab's classification.[5] Furthermore, an invagination was clearly evident apical to the talon cusp, with the vector towards the root apex. It invaded the root but remained confined as a blind sac thus; resembling a type II invagination according to Oehlers' classification.[8] DE and DI are usually asymptomatic and are often incidental findings made during routine clinical or radiographic examinations as in the present case. Consequently, they have a higher chance of causing pulpal complications due to the close proximity of the pulp to the oral environment, especially if these conditions are not recognized early. Therefore, comprehensive clinical and radiographic examinations are beneficial to identifying such defects and to prevent their undesirable sequelae.

Yumikura and Yoshida[28] reported that 42% of DE has pulp tissue within them; for this reason fracture, or attrition of the tubercle may expose the pulp horn resulting in pulpal complications. Consequently, prophylactic grinding of the tuber-

cles followed by topical fluoride application, in an attempt to stimulate reparative dentin formation is often detrimental and unpredictable. Nevertheless slight grinding of the opposing tooth in case of traumatic occlusal interference is a viable option. The reduced cusp should have a topical fluoride application to enhance its caries resistance. Fukuta *et al*.[25] advocated prophylactic reconstruction of the DE to prevent intra-operative pulpal exposure. Plaque accumulation around the base of the projection predisposes the affected tooth to caries, while occlusal interferences and trauma to the tongue are also reported complications of DE. Flowable resin composites can be placed incrementally onto the tubercles and surrounding tooth surfaces to support the tubercle.[1] Application of resin materials on to the tubercle prior to reaching functional occlusion has an added advantage, especially in the posterior teeth, by allowing the cusps of the opposing teeth to move into a more favorable position within the occlusion. Various procedures of intentional assault of the normal pulp including direct pulp capping, shallow pulpotomy and modified Cvek's pulpotomy using MTA[29] and other medicaments have been described to avoid future pulpal complications. Nevertheless, such radical measures are not often prophylactic and are of debatable value. Furthermore, endodontic treatment is indicated in cases of pulpal pathology and when this is impractical due to the aberrant nature of the pulp chamber and root canal, extraction remains the only other option.

The management of DI ranges from the application of fissure sealant to a conservative restoration of the opening, to endodontic treatment or even extraction of the tooth. The thin enamel and inaccessibility of the orifice to cleansing increases the chance of pulpal involvement secondary to caries. In addition, thin canals may be present within the enamel of the DI, which directly communicate with the pulp resulting in pulpitis and necrosis. Furthermore, an immature root on the affected tooth presents special problems particularly in the presence of periapical pathology where endodontic therapy becomes difficult and unpredictable. Therefore, early detection followed by conservative management with composite resin and/or a fissure sealant can prevent future complications. In the presented case the DI was covered with composite restoration followed by fissure sealant. No problems have occurred in the presented case since the patient has been placed under regular review.

It is important for clinicians to be aware of the potential complications that may occur with DE and DI especially in a single tooth so that the appropriate treatment can be provided promptly following early diagnosis, thus preventing the development of untoward pulpal complications. In conclusion, concomitant occurrence of DE and DI in a single tooth is extremely rare, and a comprehensive clinical and radiographic examination is beneficial in identifying such defects.

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