Concurrence of Dens Invaginatus and Evaginatus Involving All Maxillary Anteriors: A Rare Case with Comprehensive Review

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Abstract: Developmental malformations of tooth can occur due to various environmental and genetic factors playing role while tooth development. It is not unusual to find two or more such malformations in a dentition and in a single tooth. Simultaneous occurrence of dens invaginatus and dens evaginatus (talon cusp) is rare and complex. Hence, proper diagnosis and treatment plan is important so as to achieve favourable prognosis. This paper will discuss about such a rare combination in light of review on its various aspects.

Keywords: Developmental malformation,; Dens Invaginatus, Dens Evaginatus, Talon cusp

I. INTRODUCTION

Developing tooth germs are extremely sensitive to external stimuli, and any disturbance during development of tooth, whether systemic or local may result into various developmental malformations of tooth. Identification of these malformations becomes important in clinical practice because of various complications associated with them for patient as well as for clinician while making diagnosis and treatment. Dens invaginatus and dens evaginatus are two such malformations of tooth of exactly opposite nature and etiology, just like the two sides of a coin.

Dens invaginatus is a developmental anomaly resulting in a deepening or invagination of the enamel organ into the dental papilla prior to calcification of the dental tissues. (Hulsmann 1997). This kind of malformation was first described by 'Ploquet' in 1794 in whale's tooth, however, in a human tooth it was first described by a dentist named 'Socrates' in 1856 (Schulze 1970). In 1887 Tomes described the dens invaginatus in his textbook A System of Dental Surgery. There are number of terms to describe dens invaginatus like Dens in Dente (Busch 1897), dilated composite odontome (Hunter1951), dents telescope, gestant anomaly (Colby 1956). However, of the various terms, dens invaginatus (Hallet 1953) appears to be the most appropriate as it reflects the infolding of the outer portion (enamel) into the inner portion (dentine) with the formation of a pocket or dead space.

It is not uncommon to find dens invaginatus in clinical practice and the reported incidence is in a range of 0.04% and 10%, with the permanent maxillary lateral incisors most commonly involved. Isolated cases have been reported in the mandibular region and in the deciduous dentition.⁷ The anomaly usually present as a deep pit on the lingual surface of anterior teeth that may present with or without clinical symptoms thus can be overlooked easily at times. But, early diagnosis is important because of increased risk of developing caries, pulpal and periodontal inflammation in affected tooth.

Dens evaginatus or evaginated odontoma is a developmental anomaly that occurs more frequently in mandibular premolars. In canines and incisors, dens evaginatus originates in the palatal cingulus, often being bilateral and is known as "talon cusp". It occurs in both primary and permanent dentitions and results from the abnormal proliferation of enamel epithelium from the interior of the stellate reticulum of the enamel organ. So, the talon cusp is described as an anomalous hyperplasia of the cingulum of maxillary and mandibular incisors resulting in the formation of a supernumerary cusp resembling an eagle's talon. It was first described by Mitchell in 1892 as a process in the form of a "horn", curving from an inclined base to a cutting edge on the lingual surface of central incisor. However, it was named as "Talon cusp" by Mellor and Ripa in 1970.

Talon cusp consists of enamel, dentin with varying extensions of pulp. ¹¹ Clinically it varies widely in shape, size, structure, from that of a prominent cingulum to that of a cuplike structure extending to the level of the incisal edge. ¹² Early diagnosis and management is necessary to avoid complications like caries, periapical lesions, irritation of tongue during speech and mastication and occlusal interference which may lead to

accidental cusp fracture, displacement of the affected tooth, temporomandibular joint pain and periodontal problems because of excessive occlusal force. ¹³

Three fourths of all reported cases of talon cusp are located in the permanent dentition. The cusps predominantly occur on permanent maxillary lateral (55%) or central (33%) incisors and less frequently on mandibular incisors (6%) and maxillary canine (4%). ¹³

This article will discuss a case of unusual concurrent presence of dens invaginatus and dens evaginatus involving all the six permanent maxillary anterior teeth in a 21 years old male patient, along with a thorough review.

Case Report

A 21 years old male patient reported to the Outpatient Department of Purvanchal Institute of Dental Sciences, Gorakhpur (U.P) for routine dental check-up and treatment. On clinical examination there was an accidental finding of morphological alteration on lingual aspect of all maxillary anterior teeth. The right and left maxillary central and lateral incisors showed the presence of prominent and bifid cingulum along with a marked depression or pit in the groove present between the prominent bifid cingulum and lingual surface of teeth. Both right and left maxillary canines also showed the presence of cusp like prominence projecting from cingulum, measuring 1 mm with a deep groove present between the projection and lingual surface of teeth.

While examining the occlusal relationships, there was no interceptive occlusal contact in vertical overlapping of maxillary and mandibular anterior teeth in maximum intercuspation, excursive and protrusive movements of mandible and there was Angle's class I molar relation bilaterally.

Mild stains were present on lingual surfaces of teeth but there was no plaque and calculus deposits present in the grooves, depressions or pits of affected teeth. The involved teeth were caries free and hence patient was completely asymptomatic.

On the basis of clinical findings the provisional diagnosis of talon cusp (dens evaginatus) in relation to all six maxillary anterior teeth was made and that of dens invaginatus in relation to maxillary central and lateral incisors (Fig. 1).

Keeping the ethical rules in consideration and due to unwillingness of patient, no further radiographic investigations were made since, the patient was asymptomatic due to above mentioned clinical findings. However, preventive measures were undertaken using pit and fissure restorative sealant to seal the grooves and depressions or pits present, so as prevent caries development and pulpal pathosis.

II. DISCUSSION

From the 1920s to the 1990s numerous reports on cases of dens invaginatus and talon cusp were published in the dental literature, ^{3,9} however, very few [De Sousa et al (1999) and Attur M et al (2011)] have reported the simultaneous occurrence of both in a dentition and in a single tooth.^{2,8} The present case is another contribution of such a rare co-occurrence of dens invaginatus and talon cusp (dens evaginatus) to the dental literature.

Oehlers (1957) classification of invaginations is the most widely recognized and is as follows¹⁴:

Type 1 is an enamel-lined minor invagination occurring within the confines of the crown, not extending beyond the amelo-cemental junction.

Type 2 consists of an enamel-lined form that invades the root but remains as a blind sac; it may or may not connect with the dental pulp.

Type 3 invaginations penetrate through the root, perforating the apical area and forming a second foramen in the apical or periodontal area; there is no immediate connection with the pulp. Invaginations may be completely lined by enamel, although cementum is also frequently found. Also he described different crown forms (normal with a deep lingual or palatal pit; conical, barrel shaped or peg-shaped with an incisal pit) relating to the three groups mentioned. In 1958 he also presented radicular invaginations. It is thought to be the result of an invagination of Hertwig's epithelial root sheath, resulting in an accentuation of the normal longitudinal root groove. In contrast to the coronal type where it is lined with enamel, the radicular type defect is lined with cementum. Is

In the present case due to absence of any radiographic record it was difficult to classify dens invaginatus involving right and left maxillary lateral incisors merely on the basis of clinical appearance. On the other hand the talon cusp have been classified on the basis of degree of formation and extent by Hattab F N et al (1996) into following three types 11 -

- (1) True talon (an 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 cemento-enamel junction to the incisal edge)
- (2) Semitalon (an additional cusp of 1 mm or more but extending less than the distance from the cementoenamel junction to the incisal edge)

(3) Trace talon (enlarged or prominent cingulum and variations, i.e., conical, bifid, or tubercle-like).

The present case describes the presence of trace talon (bifid) in relation to right and left maxillary and lateral incisors, and that of semitalon in relation to right and left maxillary canines. Etiology of dens invaginatus is controversial and remains unclear till now. Several theories have been put forward to explain this phenomenon in past, such as growth pressure of the dental arch [Euler 1939, Atkinson 1943], focal failure of growth of the internal enamel epithelium [Kronfeld 1934], rapid and aggressive proliferation of a part of the internal enamel epithelium invading the dental papilla [Rushton 1937], distortion of the enamel organ during tooth development and subsequent protrusion of a part of the enamel organ [Oehlers 1957], 'twin-theorie' suggesting fusion of two tooth-germs [Bruszt 1950], infection [Fischer 1936 and Sprawson 1937], and trauma [Gustafson & Sundberg 1950]. Genetic factors cannot be excluded [Grahnen 1962, Casamassimo et al. 1978, Ireland et al. 1987, Hosey & Bedi 1996]. Support for this possible cause comes from a reported case of an individual lacking chromosome 7q32 who presented with dens invaginatus in addition to other dental abnormalities such as hypodontia (Pokala & Acs 1994). 3.6

Although the exact etiology is not known, Hattab et al (1995) believed that talon cusp occurs early in odontogenesis during the morphodifferentiation stage as a result of outward folding of inner enamel epithelial cells and a transient focal hyperplasia of the mesenchymal dental papilla. ¹⁶ Increased localized external pressure on a tooth germ during morphodifferentiation stage may result in either outfolding of the dental lamina as in the case of talon cusp and shovelling or infolding of the lamina as in dens invaginatus. ⁹ Other theories explaining etiology of talon cusp are endocrine disturbance during morphodifferentiation, hyperproductivity of dental lamina, and genetic factors. ^{9,10,11,13}

Talon cusp can occur as isolated condition or associated with other dental anomalies including bifid cingula, peg-shaped incisors, dens invaginatus, shovel-shaped incisors and exaggerated cusp of Carabelli. Also it has been seen in patients with Mohr, Rubinstein Taybi, Sturge – Weber, and Ellis van Creveld syndromes. In most cases a dens invaginatus is detected by chance as it was in the present case. Clinically, an unusual crown morphology (dilated, peg-shaped, barrel-shaped) or a deep foramen coecum may be important hints, but affected teeth also may show no clinical signs of the malformation. Dens invaginatus is clinically significant, as it allows entry of irritants and hence, possibility of the pulp being affected. Due to the tortuous lingual anatomy, it is possible for caries to develop inside the dens invagination without any clinically detectable lesion. Since the enamel lining is thin and in close proximity to the pulp chamber, a carious lesion could easily perforate the pulp chamber. In some cases the epithelial lining is incomplete and channels may also exist between the invagination and the pulp. As a result the possible sequelae could be pulpal necrosis, abscess and cyst formation, internal resorption. Hence, upon radiographic evidence of a dens invaginatus, the apical periodontium should be examined. Another than the pulp chamber.

Several treatment modalities have been described for dens invaginatus depending upon the degree of complexity of its anatomy. They include nonsurgical endodontic treatment, endodontic surgery, intentional replantation, and finally extraction. In cases in which there is an immature apex, the use of calcium hydroxide inside the root canal has been proposed to stimulate apexification.⁷

It is clinically difficult to establish an accurate diagnosis of talon cusp without radiographic examination. The suspicion arises from atypical morphology of the tooth crown or eruption difficulties of the suspected tooth. Treatment may differ, depending on each case. Small talon cusps are usually asymptomatic, necessitating no treatment. However, large prominent talon cusps necessitate definitive treatment with respect to esthetics, occlusion, periodontal and caries problem.

Treatment may be conservative or radical, depending on the accessory cusp like shape, location, size, and tooth affected. Periodic and gradual reduction of the cusp, with application of a desensitizing agent or, reduction of cusp with or without endodontic therapy, sealant application on the grooves, and esthetic restorations are options of treatment. Extraction being the last treatment option. 9,18

Although, patient was completely asymptomatic in the present case, but necessitates preventive and prophylactic care.



Fig 1: Intra-oral photograph of maxillary arch showing Dens Invaginatus and Talon cusp (Dens Evaginatus) in maxillary anterior teeth.

IV. CONCLUSION

Till dens invaginatus and dens evaginatus are asymptomatic seems to be uncomplicated, but just like an iceberg in ocean we cannot judge the complexity of the problem by its mere clinical inspection. So, complete clinical and radiographic investigations are mandatory followed by its comprehensive treatment. As much as can, efforts should be directed in order to preserve the dens with proper follow-up of the condition. This unusual combination of dens invaginatus and dens evaginatus is another addition to dental literature of such rare cases.

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