Co-Existence of Two Dens Invaginations with One Dens Evagination in a Maxillary Lateral Incisor: A Case Report

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Abstract
A case with two simultaneous dens invaginations (DIs) and one dens evagination (DE) in a permanent maxillary lateral incisor is reported for the first time in a 21-year-old girl.
DE known as talon cusp of the anterior teeth is a rare entity and its co-existence with DI has been reported scarcely in the literature. Simultaneous occurrence of two DIs with one DE has not been reported elsewhere. Undoubtedly, familiarity with the internal anatomy of such a rare condition can help prevent pulpal disease while performing restorative procedures.

Key Words: Dente; Incisor; Tooth Abnormalities

INTRODUCTION
Dens invaginatus (DI) is referred to as a developmental anomaly that results from an infolding within the crown prior to calcification [1]. According to the 0.04 to 10% frequency of dens invagination in the currently available literature, this condition is regarded as a rarity [2]. Several hypotheses have been proposed concerning the etiology of invaginated teeth, including constriction of the dental arch on the enamel organ, decreased or increased growth rate of the internal enamel epithelium, distortion of the enamel organ during dental development or insufficient nutrition of a portion of a single tooth germ [3]. The epithelium forms a lining of enamel in a channel inside the dentin of the affected tooth. Following calcification of the enamel, it radiographically appears as parallel radiopaque lines within the less radiopaque dentin [4]. The enamel lining is sometimes imperfect and develops caries, which can lead to dentin or pulpal exposure and eventual pulpal pathosis [5]. Maxillary lateral incisors are most commonly affected and bilateral occurrence is not uncommon, involving 43% of all cases [6, 7].
Dens evagination (DE) (also known as talon cusp) is a relatively infrequent developmental abnormality characterized by the existence of an accessory cusp-like structure projecting from the cingulum area or cemento-enamel junction (CEJ) of the maxillary or mandibular anterior teeth both in the primary and permanent dentition [8-10].
This evagination is composed of enamel, dentin and varying amounts of pulpal tissue [11]. DE can occur in both sexes and the most commonly affected teeth are the permanent maxillary incisors, particularly the lateral. It has also been reported, although less commonly, in the mandibular teeth [12]. The most commonly affected posterior teeth are the premolars [8-10]. Although both DI and DE have been reported frequently in the literature, simultaneous occurrence of these two entities within a single tooth has been noticed only limitedly [13-15]. To the best of our knowledge, co-existence of a DE with two DIs within a single tooth is considered a rarity and has not been encountered in the literature.

CASE REPORT
A 21-year-old girl came to the School of Dentistry, Kerman University of Medical Sciences for her regular dental follow-up visits and was advised to receive a full-mouth periapical radiographic evaluation. The patient did not have any contributory medical history. Extra-oral examination did not indicate any significant finding. Intra-oral examination revealed several carious lesions, restorations and a mild periodontal disease. Presence of a DE was noticeable in the patient’s permanent right maxillary lateral incisor (Fig 1). There was no associated swelling or sinus tract. The patient’s maxillary left lateral incisor did not have any problem based on clinical and radiographic evaluations. Periapical radiographic evaluation showed co-existence of two DIs with the aforementioned DE (Fig 2). The periapical region had a normal radiographic appearance. The tooth responded as vital on electrical and thermal pulp sensitivity tests and no periapical lesion was detected radiographically. Pits mesial and distal to the DE were conservatively removed and restored by bonded restorations (3M, ESPE, St Paul, MN). The tooth did not have any occlusal interference. The patient did not express any esthetic or functional problem. Regular clinical and radiographic recall visits were contemplated for the patient.

DISCUSSION
Dens invaginatus is a relatively common dental anomaly. It has been stated that DI affects maxillary lateral incisors in 0.25% to 5.1% of the population [16, 17]. However, double DI in a single tooth has infrequently been reported [14, 18, 19]. DI is clinically important because of the resultant aberration in anatomy. It is possible for dental caries to easily reach the pulp chamber in DI cases. The patient is usually detected incidentally by intraoral periapical radiographs. The reported patient did not have any complaint about the considered tooth and the problem was accidentally diagnosed on full-mouth radiographic examination. According to Oehlers’ description of invagination [20], the case was categorized as “type I” because the invaginated cavities did not extend beyond the cemento-enamel junction. It has been stated that the morphology of the tooth with DI may undergo changes such as increased labio-lingual or mesio-distal dimension, incisal notching associated with a labial groove, a peg-shaped or conical morphology or the presence of an exaggerated palatal cingulum or ‘talon cusp’ [21, 22]. Simultaneous occurrence of two DIs with one DE in a single tooth has not been previously reported but separate conditions in different teeth have been reported exclusively in the literature.
In other words we are reporting a case with three simultaneous malformations in a single tooth. This actually makes our case report unique. Presence of canals and cracks that reach the pulpal cavity are numerous in DE. They may result in contamination of the pulp and progressive loss of pulp vitality. Therefore, such teeth should be regularly followed for development of pulpal pathosis.

DE is a rare dental anomaly occurring commonly on the lingual surface of primary or permanent teeth [23, 24]. Usually, grooves or fissures can be found at the junction of the evagination with the lingual tooth surface, which often allows plaque retention, thereby causing caries and endodontic and periodontal inflammation [25]. The etiology of this condition is multifactorial with genetic and environmental factors playing significant roles [26]. It has also been reported that this anomaly is related to Rubinstein-Taybi, Mohr, and Sturge-Weber syndromes and to anomalies including odontome, dens invaginatus, double tooth, supernumerary tooth, peg-shaped lateral incisors, agenesis of canines, mesiodens, megadont, and shovel-shaped incisors [8, 12, 24, 25, 27, 28].

Although there is no strong correlation between the aforementioned syndromes and talon cusp, in a study by Hennekam and Van Doorne [29] on 45 patients with Rubinstein-Taybi syndrome, 92% of these patients had talon cusps. The authors concluded that there was a positive correlation between these two entities.

Other signs of this syndrome such as mental and developmental retardation, thin upper lip, retrognathia, micrognathia, and cleft palate were not evident in our patient, therefore the presence of this syndrome was ruled out.

Talon cusp has a prevalence of 0.06-7.7 percent with a predominance in the male gender [12, 23, 30, 31]. Although many authors have reported DE in the primary dentition [8, 25], it is seen more frequently in permanent dentition. Talon cusp can cause problems for the patient including poor esthetics, caries, occlusal trauma, displacement of the involved tooth, irritation of the tongue during speech and mastication, periodontal problems, accidental cuspal fracture and attrition causing pulpal exposure or periapical pathosis [11, 12, 32-34]. The radiographic appearance of talon cusp resembles that of a supernumerary tooth.
or compound odontoma, consequently increasing the likelihood of misdiagnosis [11, 12, 32]. DE can either be treated conservatively or radically, depending on the shape or size of the affected tooth [11, 28, 33, 35-37]. Treatment remedies may consist of gradual periodic cuspal reduction with fluoride as a desensitizing agent, one-visit reduction with or without endodontic therapy, application of sealants for developmental grooves and placement of esthetic restorations. In this case, simultaneous occurrence of two DIs and one DE was encountered in a permanent maxillary lateral incisor. This patient represented positive responses to vitality tests without any radiographic signs of apical periodontitis. In such cases, it is recommended that the susceptible parts should be sealed prophylactically using restorative materials in order to prevent formation of any communication pathways between the oral environment and pulpal space.

This was carried out in this patient using a composite resin restoration. It has to be taken into consideration that Ohlers' classification is based on the presence of a single dens invagination; whereas, this patient represented two invaginations simultaneously. According to similar reports in the literature [18, 38], this classification should be modified based on the presence of multiple defects.

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