

Dens Invaginatus Revisited

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Abstract

The current study is aimed at investigating the intricacies of the dens invaginatus (DI) anomaly in the course of independent dental practice. The attempt was done to get a deeper insight of the mentioned dental problem, systemize and eventually work out independent better approaches to dealing with the dens invaginatus.

This paper deals with literature concerning the etiology, classification and prevalence of DI. The article presents four different cases of successful DI treatment. Case 1 refers to 13-year-old boy with symptomatic periodontitis of tooth #12; Case 2 applies to the female patient, aged X, with hidden carious cavities on the proximal surfaces of teeth #12 and #22; Case 3 belongs to 7-year-old girl with a tentative diagnosis of DI in tooth #31; Case 4 describes a routine treatment of symptomatic apical periodontitis in tooth #22 of the female patient, 22 years old.

The presented cases demonstrate a resultative way of DI medical dental care. DI is a malformation that can give rise to a number of pathological conditions, spreading through enamel and dentine to contaminate the pulp and cause soft tissue necrosis. Nowadays, DI, however complicated it may seem, can be efficaciously managed for protracted periods of time without causing disturbances to its sufferers. Timely diagnosis and treatment make surgery either considerably delayed and necessary in exclusive cases.

DI is now generally acknowledged as a meaningful anomaly with serious consequences. A ponderable number of DI cases occur in vital lateral incisors. Due to their uneven surface most such incisors are preventably treated to seal tooth irregularities that are likely to develop caries. Pediatric cases are extremely underreported but dealt with in routine dental care, especially owing to sealants. The X-rayed examination might not always be carried out. Nevertheless, provided appropriate follow-up is ensured, this condition may cause no life-long inconveniences to their hosts. Most of the cases are subject to treatment and preservation.

The approaches may be found useful by pediatric dentist and endodontists in their decision-making in regard of analogous cases. A number of issues are to be discussed in the second part of the paper.

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Introduction

Dens invaginatus (DI) is a tooth maldevelopment with bizarre dental hard tissue arrangement due to the enamel organ invasion into the tooth pulp chamber before the dental tissues have become mineralized that begins at the crown and sometimes extends into the root

with the formation of a pocket or dead space or it is an accentuation of the lingual pit of an incisor before calcification sets in Hegde et al.¹⁻³ It ranges in severity from superficial, where only the crown is affected, to deep, where both the crown and the root are involved.⁴ A minor DI is also known as foramen caecum (a blind fossa). A very deep foramen caecum is called a tooth in tooth as the radiographic appearance suggests presence of a tooth within a tooth⁵ where either enamel or cementum lies inside of dentin.⁶

This article was conceived as endodontists' fascinating journey into the intricacies of the anomaly in the course of our

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independent practice where we are trying to get a deeper insight and with a view of systemizing and eventually working out independent and, hopefully, better approaches to dealing with it. We present here our four cases.

Though numerous authors mention that the DI phenomenon per se has been known since the late 18th century (1794) and was described in humans about 60 years later⁷, they virtually invariably quote it as a rare condition; the thesis, however, needs to be updated in the light of contemporary data to be discussed below. Furthermore, it may seem ironic for a rarity, but we have counted over 10 synonyms for this allegedly infrequent malformation: coronalodontomas, deep foramen caecum, dens in dente (Latin), dent telescopée (French) = a telescoped or telescopic tooth,⁶ dentoid in dente, odontomedilated gestant odontome, dilated composite odontome, gestant anomaly, gestant odontoma, invaginated odontoma(e), odontopagus parasiticus in cretus, a tooth within a tooth, dilated odontoma, radix in radice, odontopagus (parasiticus) in cretus,⁸⁻⁹ tooth inclusion, and a warty tooth¹⁰. The list, however, may not be complete. The term dens in dente, first suggested by Busch in 1897^{10,11} is still widely used along with dens invaginatus. "Odontome", introduced in 1867 by Broca, is rather a developmental anomaly than a true neoplasm¹² and can essentially be applied only to a certain group of these malformations. We may suppose that this synonymy actually is due to the failure to reach consensus on the causation and clinical manifestations, on the one hand, and different approaches to classifying – either clinical or radiological, on the other hand, with dens invaginatus being the most frequent term reflecting specific variations that are fundamental for the condition.^{13-14,7}

Materials and methods

The study is organized in the way of combining theoretical and experimental parts that is presented in the form of successful cases of treatment DI avoiding surgery. Theoretical part is based on studying relevant literature concerning DI. Partial attention is paid on versatility of manifestations, localization and prevalence of DI anomaly.

Experimental part contains four cases of DI treatment. Case 1 refers to 13-year-old boy

with symptomatic periodontitis of tooth #12; Case 2 applies to the female patient, aged X, with hidden carious cavities on the proximal surfaces of teeth #12 and #22; Case 3 belongs to 7-year-old girl with a tentative diagnosis of DI in tooth #31; Case 4 describes a routine treatment of symptomatic apical periodontitis in tooth #22 of the female patient, 22 years old.

For conducting the study and obtaining experimental data methods of visual dental examination, instrumental examination, X-ray, microscope and CBCT were used. A detailed step-by-step therapy for each of four represented cases is described in the results part of the present article.

Results

Case 1

A 13-year-old boy presented with symptomatic periodontitis of tooth #12. Fortunately, his general dentist had made a pre-op and found "a strange anatomy" of the tooth. So, he referred the patient to us as endodontists. That was when we were first introduced to a tooth in a tooth at our clinic.

The patient was in good general health; and his medical history was non-contributory. The extraoral examination did not reveal any significant findings. The tooth was tender on percussion; however, it did not respond to thermal or electric stimuli. The periodontal probing was within normal limits.



Figure 1. A periapical lesion in the 12 tooth with Type II Invagination. Post op X-ray.

The CBCT of the tooth (Figure 1) showed a periapical lesion in the 12 tooth with Type II Invagination with a diameter of approximately 3-4 mm. After the coronal access was cleaned, orifices were located under magnification on a microscope. The tooth was instrumented and the invagination dissected using carbide burrs Komet EndoTracer for root opening and ultrasound. Then followed the irrigation protocol, which included 5.25% NaOCl for 20 minutes completed

with 3-fold sonication. Exposure to 15% EDTA lasted for 1 minute to remove the smear layer and C2H5OH was used at the end of the irrigation. Due to the large apical diameter of the root canal, instrumental treatment was very insignificant. For the mechanical cleaning of the peripheral walls, Sani Taper files were used.

After apical application of Gelatamp Roeko sponge, the root canal was obturated with mineral trioxide aggregate (MTA) matrix. The white MTA Rootdent was introduced using an MTA carrier and sealed under indirect ultrasound activation. After thorough debridement followed the restoration of the crown. Since the patient was 13 years old, we opted for a direct restoration with a glass fiber post.

The symptoms of periodontitis disappeared the day after treatment.

The patient noted a decrease in pain and edema. X-ray control a year later showed a decrease in the inflammatory process (Figure 2).



Figure 2. Recall 1 year

Case 2

The female patient, aged X, was referred to us by an orthodontist who found some hidden carious cavities on the proximal surfaces of teeth #12 and #22. The orthodontist also emphasized that the area of the blind fossae revealed somewhat confusing anatomy.

It was a bilateral Type I DI. The cold test results did not differ from that of the neighboring teeth and indicated to the normal pulp condition. Probing did not reveal any pathological pockets. The percussion test was negative. In order to preserve the tooth vitality, it was decided to preventively seal/close the in invaginated teeth. The procedures performed were the same on

both teeth: blind fossa opening, carious tissue removing, and filling the root according to the protocol of applying adhesive for total etching OptiBond FL with the material Filtek Ultimate.

On the day of the follow-up visit the teeth responded normally to the cold stimuli, complaints were absent (Figure 3).



Figure 3. X-ray films and CBCT scans of the 12 and 22 dens invaginatus.

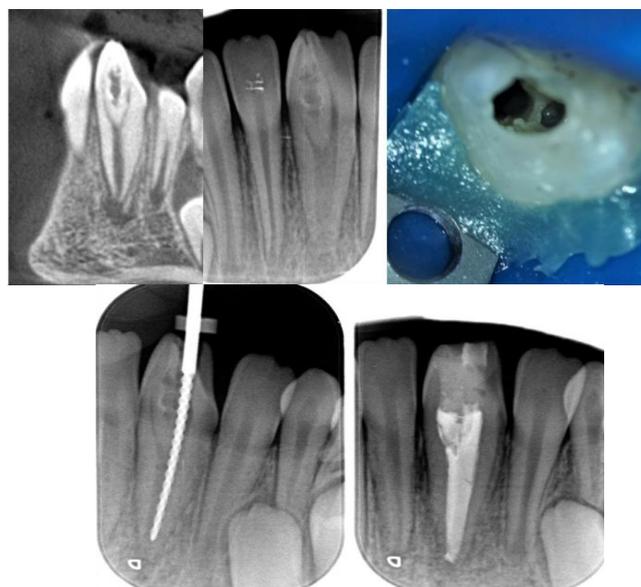


Figure 4. A periapical lesion in the 31 tooth with Type II Invagination. X-ray of the WL measurement and post op X-ray.

Case 3

On September 23, 2019, A 7-year-old girl was referred by a dentist surgeon from a pediatric dental clinic with a tentative diagnosis of DI in tooth #31. The dentist surgeon two days earlier had made a relief incision to alleviate the pain due to the exacerbated apical periodontitis

with an abscess. The patient presented with a feeling of fear, so the first visit was spent to get to know each other better, and an X-ray examination was carried out.

During the second visit, after opening the pulp horn the dissection was continued in the vestibular direction; and the scalpel fell into the invagination, the bottom which had to be removed using an ultrasound U- file size 40.

Having merged the lingual pulp horn with the invagination, we determined the working length under X-ray control. Then the vestibular pulp horn was opened and subsequently obturated by a hybrid method using master pin 90-02 with AH PLUS root canal sealer. The intervention was completed with X-ray control (Figure 4).

Case 4

It was a routine treatment of symptomatic apical periodontitis in tooth #22. The female patient, 22 years old, presented with the main complaint of pain when biting on the tooth; the palpation in the apex area was painful. Some time ago the patient began to notice greater mobility of the tooth when compared to the neighboring ones. The clinical examination established painful percussion, and negative cold test. The periodontal sulcus probing was within the normal range. The computed tomography showed signs of invagination, which usually complicates the treatment to a significant extent.

At the first visit, under magnification, the invagination was mechanically destroyed with the use of ultrasonic tips U-file and a bur for endodontic access called accordingly - Endo Access. Abundant purulent contents were obtained; calcium hydroxide was left in the canal for 2 weeks. Two weeks later, on the second visit, the symptoms significantly improved, but the exudation from the periapical zone continued. The final cleansing of the root canal was carried out, and the apical zone was modified with hand steel files up to size 110. The second visit also ended with adding calcium hydroxide for another 2 weeks. On the third visit, the canal was practically dry, though there was a slight leakage of exudate from the lateral side. There were no symptoms. So it was decided to obturate the canals. The apical zone was filled with white MTA Rootdent with a subsequent ultrasonic seal. The extensive process in the periradicular zone called into question the quality of lateral MTA

packing, and the decision was made to insert the apical matrix Gelatamp (Roeko); it took 2 complete blocks, with obturation carried out with an individualized gutta-percha pin. The pin was thermally softened and shaped laterally by several pre-softened applications. The entire root canal was filled with gutta-percha with AH+ sealer using the Coxo C Fill Obturation System.

The tooth was restored with 3M Filtek photopolymer material. Follow-up examinations were carried out in a year and in 3-years' time. Symptoms and complaints are absent, the tooth sits fast (Figure 5).

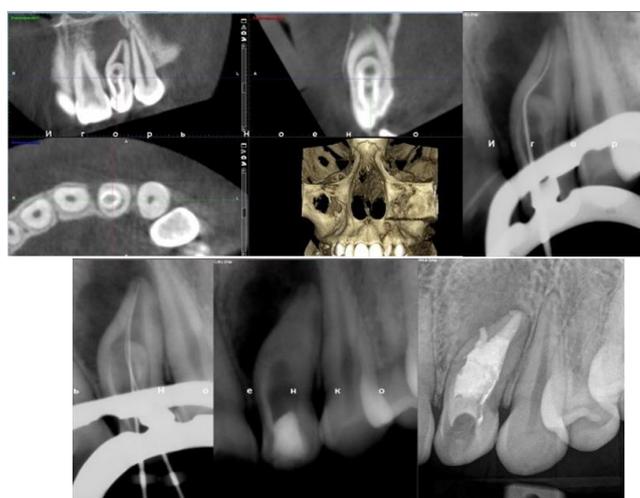


Figure 5. A periapical lesion in the 22 tooth with Type III Invagination. X-ray with files and recall 1 year.

Discussion

Histologic and Clinical Features

Microscopic, ultrastructural and microradiographic investigations of invaginated teeth reproduce the macroscopic variety of this anomaly.² The dentine below the invagination may be either hypomineralized or irregularly structured, or may look intact. Furthermore, it may contain strains of vital connective tissue or fine canals communicating with the dental pulp.^{1,14,15} The enamel in the invagination may also vary, be irregularly structured. It may be either hypomineralized or contain to eight times more phosphate and calcium as compared with the outer enamel. The internal enamel may have atypical and more complex rod shapes, with surface presenting the typical honeycomb pattern but no perikymata, which, however, were observed on the outer surface of the tooth.²

Clinically, the morphology of the DI crown can look normal; it can also have unusual forms such as an increased labiolingual and mesiodistal dimension, conical/peg-shaped/barrel-shaped/pear shaped, and have a talon cusp. A deep foramen cecum or an incisal notching should raise a suspicion as they may be suggestive of a coronal alteration, thus, being the sign of DI.^{16, 17} Sometimes DI is described as a defect with a prominent lingual cusp and centrally located fossa.¹⁸

Classification

A few classifications have been put forward so far, but, being simple and easy to use, Oehlers' one is almost ubiquitous.¹³ The recently suggested new classification of tooth anomalies also relies on Oehlers' three types, suggested by him in 1957.¹⁹

Based on the radiographic appearance of invagination, Oehlers²⁰ singled out the following types of dens invaginatus. Type I is characterized by minimal invagination, is enamel-lined, confined within the crown of the tooth, and does not extend beyond the level of the external amelocemental junction. Type II is enamel-lined, extending into the pulp chamber, but it remains within the root canal and does not communicate with the periodontal ligament. As a matter of fact, it is Type II that falls within the category of invaginated or dilated odontomes where the invagination extends the whole length of a tooth and sometimes through a widely dilated apex. Often the invagination has incomplete walls allowing the exterior to communicate with the pulp and devitalize it. Alternatively, food debris lodges in the cavity to cause caries which rapidly penetrates the superficially located pulp chamber.²¹

In Type III the invagination penetrates through the root, perforating the apical area and having a second foramen in the apical or periodontal area; it does not however immediately communicate with the pulp.^{22,23} So, occasionally infolded enamel and dentine reach the root apex.⁷ A year later, in 1957, Oehlers further extended the classification²⁰ to imply the presence of enlarged roots on radiograms. These fall into two distinct types. One is represented as an axial infolding of a root wall, indicating to an incomplete root bifurcation. The other one presents as an enamel-lined invagination within the root originating at an opening on the root itself.

A much broader classification was proposed by Schulze and Brand,²⁴ which includes 12 variations. They offer more extended analysis of the morphology of the invagination,⁷ but their classification has not gained such currency as Oehlers'.²⁰

Etiology

Numerous theories have been put forward to explain the underlying causes of this malformation – from external forces such as growth pressure of the dental arch that exert²⁵ an effect on the tooth germ during development and causing a trauma of the tooth germ, also causing focal growth retardation, infection, neoplasia, retarded growth, etc.,²⁴ – to genetic ones.¹ Since Grahnen et al.²⁶ published their results in 1959, showing the importance of absence of signaling molecules for morphogenesis, the scientific community has been leaning slowly towards the genetic causation. First, cautious 'may' was added to such recognition⁷, which was followed by "they seem more likely to be determined genetically".²⁷ In 2012, Kallianpur et al.²⁸ mentioned that syndromic association of dens invaginatus was reported in Ekman-Westborg-Julin syndrome, Williams syndrome and Nance Hulan syndrome, which is indicative of family inheritance. After 2015 more often than not we find unequivocal statements that the genetic theory is most probable.²⁹ A salient example of X-linked recessive inheritance was given by Casamassimo et al.³⁰ in 1978 in 5 generations of a family with concomitant generalized microdontia, taurodontism of the first permanent molars, and multiple teeth affected with one or more dens invaginatus, where the pedigree contained 5 affected males. The disorder appeared to be distinctly characterized. Recently, an international team working in the field of molecular genetics & genomic medicine. Gowans et al.,³¹ published a case of two families of European ancestry (at the time of the publication residing in New Zealand and the USA), where males related to each other via unaffected females had taurodontism, microdontia and DI. A linkage analysis identified two independent missense variants in KIF4A gene that segregate in affected males and female carriers. The gene is expressed in the developing tooth bud during development; and the p.Arg771Lys variant influences cell migration in an in vitro assay. These data implicate missense variations in

KIF4A in a pathogenic mechanism causing taurodontism, microdontia and DI phenotypes.

Nevertheless, while it is genes that pass instructions to DNA, from parents to their offspring, bacteria and viruses also can influence offspring's development. As those environmental factors can influence immunity more than our genes do, so may they influence immunity and ontogeny when people age, and the environmental impact grows stronger. So, Hedge et al.³ quote Omnell et al.²⁵ who consider that the fact that maxillary lateral incisor is most commonly affected can be attributed to the external forces applied on the lateral incisor tooth bud by the developing central incisor or canine which develops 6 months prior.

Thus, further investigations, perhaps in the allied science fields, will allow for a final definitive settlement of the DI etiology issue.

Prevalence, Localization and Comorbidities

While the first known DI specimen was obtained from a whale to be thoroughly described by Ploquet as early as 1794, it was only 62 years later, in 1856, that the American dentist Socrates identified such a tooth in humans.⁷ In 1934 Kronfeld indicated that somewhat more than 10 cases of DI or similar abnormalities were known to him from medical literature, which he painstakingly enumerated in a chronological order.⁶

The latter half of the 20th and early 21st centuries witnessed a close attention to DI, and it amassed numerous up-to-date individual case descriptions and discussions, with literature reviews either dealing exclusively or inter alia with the prevalence, which vary in many aspects thereby answering and asking further questions which will be further discussed below.

According to the literature, maxillary lateral incisors are most commonly affected.^{22,27,32,33} Bilateral occurrence with a prevalence of 43% was first reported by Swanson and McCarthy in 1947.³⁴

This predisposition of maxillary lateral incisors to DI development is corroborated by Chinese paleodontists who found a meaningful sample of 25 affected (4.83%) out of 517 old permanent teeth identified from 67 ancient adults who lived circa 2,000 years ago, with bilateral incidence of 19.05%.³⁵ Neither Type II or III DI were found in the study. When comparing their

results to those of a retrospective survey of DI from 1873 to present, S. Shi et al.³⁵ suppose that a higher rate of DI occurred more often than nowadays, which they attribute to the use of the advanced micro-CT technique and ethnic origin.

Different authors^{13,22,27,33} who have summarized data on teeth with DI in table form, mostly agree that figures vary from 0.04% to approximately 10, with lateral incisors accounting for the major part of them. Their tables indicate that different populations have different rates of invaginations, as supported by Colak et al.³⁶ Furthermore, the criteria differ, ranging from only right maxillary incisors, or just lateral incisors (in vivo and in vitro) to full mouth evaluations, on the one hand, and X-ray examinations (panoramic tomographs), on the other hand, mostly in mature adults.

Meanwhile, contrary to some popular belief³⁷, a genuine DI can be diagnosed very early, at the age of 12 months in one of four erupting incisors. Though minute, the DI sitting on the edge of a boy's left maxillary central incisor did not pass by his mother attention. After being confirmed by periapical radiograph the treated tooth revealed continuing normal root development eight months later³⁸. The further follow-up information has been unavailable so far.

Being rather distinguishable among other anomalies, DI provoked considerable attention of Turkish researchers in respect of its prevalence among their compatriots in the early 2000s, yielding at least 5 reviews known to us; regrettably, the information given there is rather discordant due to the diverse screening criteria, population numbers, methods and loci of investigation, etc. Furthermore, the studies largely dealt with adult population; carious, restored, and fractured teeth, incomplete apical foramen formation, undetectable furcation, and fused roots were not always included. Even CBCT examination can show either 5.90%³⁹ or 10.7%⁴⁰ prevalence in the examined populations. The latter article emphasizes the importance of careful clinical examination along with radiographic one. While not meant to be an anthropologic or forensic medicine tool in the first place, this paper discontinues here our comparative statistic endeavor.

As for localization, DI is also found in the mandible, either unilaterally or bilaterally (or in both jaws), albeit by far less frequently. The morphology of the crown of a DI tooth can either

be normal or abnormally versatile: peg-/ shovel-/barrel-/pear-shaped, or conical.

Also, the following conditions have been found to be reportedly associated with DI: dens evaginatus, macro- micro-/hypo-/oligodontia, taurodontism, gemination, fusion, supernumerary teeth, dentinogenesis imperfecta, coronal agenesis, mesiodens, obliterated pulp chambers, C-shaped canal configuration, palatoradicular groove defect, short root anomaly, dilacerations, albinism, periodontal abscess, multiple root canals (a peg-like lateral incisor with as many as five root canals in invagination was recently described),⁴¹ cranial suture syndromes, unicystic ameloblastoma, and coronal fractures. Moreover, multiple dental abnormalities, such as multiple dens invaginatus, mulberry molar and conical teeth were detected more frequently in patients with chromosomal disparities.⁴¹⁻⁴⁴

Thus, DI demonstrates significant versatility in its manifestations, localization and prevalence.⁴⁵

Conclusions

Since DI is now generally acknowledged as a meaningful textbook anomaly with serious consequences, bearing in mind that as far as a ponderable number of DI cases occur in vital lateral incisors, due to their uneven surface most such incisors are preventably treated to seal tooth irregularities that are likely to develop caries. We may dare to presume that pediatric cases are extremely underreported but dealt with in routine dental care, especially owing to sealants. (So can other DI teeth be). At this, the X-rayed examination might not always be carried out. Nevertheless, provided appropriate follow-up is ensured, this condition may cause no life-long inconveniences to their hosts. The rest of the cases, whatever their manifestations, are subject to treatment and preservation, the discussion of which is intended as a second part of this paper.

Furthermore, digital technologies may have far-reaching consequences establishing better rapport owing to dental homes, or with Bluetooth brushes that rouse patients' interest in their dentition and help patients establish better rapport with their dentists, thus ensuring the early DI diagnosis that so many times has been upheld in specialist literature.

Clinical Significance

The described cases' approaches may be found useful by pediatric dentist and endodontists in their decision-making in regard of analogous cases. The study may be also useful for further theoretization, generalization of the obtained clinical case studies on the dens invaginatus anomaly.

Patient Consent

The patients have given their written consent for representation of the clinical information. The procedures were carried out with a proper consent from the patients by explaining the treatment.

Declaration of Interest

The authors report no conflict of interest.

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