Triple dens invaginatus in a single tooth: Rarest of rare case report

Abstract
Dens invaginatus is a rare malformation affecting the teeth. The teeth that are most commonly affected are the permanent maxillary lateral incisors. The mandibular posterior teeth are rarely affected by dens invaginatus. In most of the cases, a single dens invaginatus is seen affecting the tooth. Cases of double dens invaginatus affecting a single tooth are rare, and those of triple dens invaginatus in a single tooth are even rarer. Here, we present a case of a 14-year-old where three dens invaginatus were seen in the mandibular second premolar. This is the fourth case of three dens invaginatus in a single tooth as only three have been reported previously.

Keywords: Dens, dente, invaginatus

Introduction
Dens invaginatus arises as a result of anomalous development during morphodifferentiation and numerous different terms have been used to describe this condition, such as dens in dente, dilated composite odontoma, gestant anomaly, tooth inclusion, dentoid in dente, invaginated odontoma, and dens telescope.[1-4] It was first described in human tooth by Socrates in 1856.[5] This abnormality in the tooth morphology affects most commonly the permanent maxillary lateral incisors, followed by maxillary central incisors, premolars, canines, and less frequently the molars.[6] Dens invaginatus is rarely seen in mandibular teeth.[7-11] In most of the cases, teeth are affected by a single dens invaginatus whereas the occurrence of three dens invaginatus in a single tooth is very rare.[6,12,13] Dens invaginatus may be easily overlooked due to the lack of any clinical signs of the anomaly.[5] However, the early identification of the tooth affected by dens invaginatus is very important.[5] Due to the irregular shape of the root canal system, root canal treatment in cases of dens invaginatus presents with several problems because proper cleaning and shaping of root canals are very difficult.[1]

Hence, early diagnosis of such cases is very important so that the teeth can be treated or restored before the pulp involvement.

Case Report
A 14-year-old boy reported with a chief complaint of pain in the lower back tooth for 1 month. On intraoral examination, it was found that the pain was due to grossly carious mandibular left second molar for which root canal treatment was advised. A thorough intraoral examination revealed that mandibular right second premolar was abnormal in anatomical morphology, rotated, and was out of occlusion as is shown in Figures 1 and 2. However, the second premolar on the opposite side was normal in morphology and occlusion as shown in Figure 3. A routine intraoral periapical radiograph was taken which revealed that the right mandibular second premolar had a bulbous root. The root was associated with three radio-opaque masses equal in density to enamel as shown in Figure 4. Out of these, the mesial radiopacity was flame shaped and resembled a small well-formed tooth, the distal radiopacity was leaf shaped, and the third radiopacity was present in between these two radiopacities and was overlapped by the distal radiopacity as shown in Figure 4. The radiographs were taken using different angulations, and it was found that all these radiopacities were present in both the coronal and radicular portions that extended beyond the cementoenamel junction. Hence, a final diagnosis of dens invaginatus type II was given based on the above-mentioned findings.
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Discussion

Dens invaginatus is a rare malformation of teeth, and according to Hulsmann, dens invaginatus is a developmental anomaly that arises due to an invagination or deepening of the enamel organ into the dental papilla before the calcification of dental tissues takes place. It usually begins at the crown and often extends to the root. It was first described in 1794 by Ploquet in a whale's tooth and first described in human tooth by Socrates in 1856. Hallet, who first introduced the term dens invaginatus and gave the first classification it.

A number of other classifications have also been described, but the classification given by Oehlers in 1957 is the most commonly used, in which he classified dens in dente into three types depending on its extent into the crown, root, and root apex. It is based on two-dimensional radiographic images and on the depth of penetration and communication with the periodontal ligament or periapical tissue.

Type I: it is a minor form, lined by enamel and occurs within the confines of the crown and does not extend beyond the cementoenamel junction and this is the most common type. Type II: these are those cases, in which the invagination extends beyond the cementoenamel junction. It is lined by enamel, invades the root but is confined as a blind sac. It may or may not communicate with the dental pulp. It has no communication with periodontal ligament. Similar finding was seen in this case report also as shown in Figure 4. Type IIIa: In this type of dens in dente, the invagination extends through the root and communicates laterally with periodontal ligament space through a pseudo foramen. However, there is no communication with the pulp which lies compressed within the root. Type IIIb: This type may be lined by enamel, but sometimes cementum may also be found lining this invagination. Here, the invagination extends through the root and communicates with periodontal ligament at the apical foramen through a pseudo foramen with no communication with the pulp.

The etiology of dens invaginatus is controversial and is still unclear and numerous theories have been proposed for its etiology. The various factors that may be
responsible for its formation are the fusion of the two tooth germs, constriction of the dental arch, focal retardation in the growth of the tooth bud, invagination of the crown that occurs before the calcification of teeth, genetic factors, infection, altered external pressure, or forces on the tooth germ during development from surrounding tissues such as adjacent tooth germs, trauma, localized discrepancy in cellular hyperplasia.\(^{[4,8,9,11]}\) Focal inadequate nutrition of the odontogenic epithelium due to retardation in growth of a portion of the tooth germ or relative retardation of ameloblastic activity has also been stated as one of the factors responsible for the formation of dens invaginatus.\(^{[1]}\) Nevertheless, the more significant issue is the correct diagnosis because dens invaginatus can present with a broad spectrum of variations, and in some cases, the diagnosis can be extremely difficult.\(^{[9]}\)

The incidence of dens invaginatus ranges from 0.04% to 10%.\(^{[7]}\) The most commonly involved tooth is the permanent maxillary lateral incisor followed by maxillary central incisors.\(^{[2,7,10,13]}\) This might be due to the unfavorable position of the maxillary lateral incisor during the formative stages, and it is also the last anterior tooth to calcify.\(^{[8]}\) According to the findings of Grahnén et al., in their study, of 3020 lateral incisors in the year 1959, bilateral occurrence of dens in dente is seen in 43% of the patients.\(^{[5]}\) Hence, if one tooth in a patient is affected by dens invaginatus, then, the contralateral tooth should also be investigated.\(^{[4,8,17]}\) The posterior teeth are less likely to be affected by dens invaginatus, and this was supported by the findings of Hamasha and Al-Omari, in which out of 14,090 teeth affected by dens invaginatus only 6.5% were posterior teeth with no involvement of mandibular teeth.\(^{[5]}\) In another study conducted by More and Patel in the year 2012, it was found that involvement of the mandibular teeth was rare and none was found affecting mandibular premolars.\(^{[8]}\) Dens invaginatus is rarely seen in mandibular teeth.\(^{[4,7,11]}\) However, the finding of this case is contrary to the above-mentioned studies as the tooth affected here was a mandibular premolar tooth. This condition is in most of the cases seen in a single form.\(^{[1]}\) Double dens invaginatus in a single tooth is an extremely rare dental anomaly as very few cases have been reported, and according to Hulsmann, only 14 cases of double dens invaginatus have been reported.\(^{[5,11,13]}\) Triple occurrences of dens invaginatus in a single tooth are even rarer, and a few cases have also been reported.\(^{[4,5,11]}\) Only three cases of triple form of this condition have been described by Hitchin and McHugh in the year 1954, Mader in the year 1977, and by Serrano in the year 1991.\(^{[1,12]}\) This report of three dens invaginatus in a single tooth will add to the above-mentioned case reports and is only the forth case to be reported till date.\(^{[19]}\)

Most of the cases of dens invaginatus are diagnosed by chance on a radiograph, and most of the patients are asymptomatic and unaware of this condition as was seen in this case report where the patient was totally unaware of the condition and was asymptomatic.\(^{[5,4,8,13]}\) Clinically, the teeth that are affected usually have unusual crown morphology.\(^{[1]}\) Similar findings were observed clinically in this case report also where the affected tooth was rotated and had an unusual crown morphology. Clinical examination may reveal a deep pit or a fissure on an anterior tooth, but the radiographic analysis is the most realistic. Usually, dens invaginatus presents as a deep invagination in the occlusal or lingual pit area.\(^{[11]}\) This invaginated area is separated from the pulpal tissues by a thin layer of dentin and may also communicate with the oral cavity which allows the entry of the irritants and microorganisms. This may lead to dental caries or pulpal necrosis or periapical abscess or periodontal abscess.\(^{[4,11,16]}\) Sometimes clinically, the teeth may show no clinical signs of any malformation.\(^{[11]}\)

Radiographically, it shows a radiopaque invagination, equal in density to enamel, extending from the cingulum into the root canal. The defects may vary in size and shape from a loop like, pear-shaped or slightly radiolucent structure to a severe form resembling a tooth within a tooth as was seen in case report where one of the radiopacities resembled a well-formed tooth.\(^{[11,13]}\) Dens invaginatus may even be detected radiographically even before the tooth erupts into the oral cavity.\(^{[1]}\)

Salter in the year 1875 described a case of an upper lateral incisor which had several tooth-like structures projecting into the pulp chamber.\(^{[15]}\) The findings of this case report are similar to those of Kirk and Moral. In the year 1918, Kirk described a case of a cuspid in the root of which was a smaller tooth of similar shape. The crown of this inner tooth had normal enamel covering on the outside.\(^{[15,18]}\)

**Conclusion**

Dens invaginatus is a relatively rare malformation and in most of the cases a single dens invaginatus is seen affecting the tooth. The teeth most commonly affected are the maxillary teeth, and it rarely affects the mandibular teeth. Even though a quadruple dens invaginatus has also been reported, the occurrence of triple dens invaginatus in a single tooth is a very rare finding, and this case of triple dens invaginatus in mandibular second premolar is only the fourth such case to be reported till date.\(^{[19]}\)

**Financial support and sponsorship**

Nil.

**Conflicts of interest**

There are no conflicts of interest.

**References**

1. de Sousa SM, Bramante CM. Dens invaginatus: Treatment choices. Endod Dent Traumatol 1998;14:152-8.
2. Alani A, Bishop K. Dens invaginatus. Part 1: Classification, prevalence and aetiology. Int Endod J 2008;41:1123-36.
3. Munir B, Tirmazi SM, Majeed HB, Khan AM, Iqbalbangash N. Dens invaginatus: Aetiology, classification, prevalence, diagnosis and treatment considerations. Pak Oral Dent J 2011;31:191-8.
4. Kirzioglu Z, Ceyhan D. The prevalence of anterior teeth with dens invaginatus in the western Mediterranean region of Turkey. Int Endod J 2009;42:727-34.
5. Hulsmann M. Dens invaginatus: Aetiology, classification, prevalence, diagnosis, and treatment considerations. Int Endod J 1997;30:79-90.
6. Sedano HO, Ocampo-Acosta F, Naranjo-Corona RI, Torres-Arellano ME. Multiple dens invaginatus, mulberry molar and conical teeth. Case report and genetic considerations. Med Oral Patol Oral Cir Bucal 2009;14:E69-72.
7. Tavano SM, de Sousa SM, Bramante CM. Dens invaginatus in first mandibular premolar. Endod Dent Traumatol 1994;10:27-9.
8. More CB, Patel HJ. Dens invaginatus: A radiographic analysis. Open Access Sci Rep 2012;1:1-4.
9. Chaniotis AM, Tzanetakis GN, Kontakiotis EG, Tosios KI. Combined endodontic and surgical management of a mandibular lateral incisor with a rare type of dens invaginatus. J Endod 2008;34:1235-60.
10. Bansal M, Singh N, Singh AP. A rare presentation of dens in dente in the mandibular third molar with extra oral sinus. J Oral Maxillofac Pathol 2010;14:80-2.
11. Canger EM, Kayipmaz S, Celenk P. Bilateral dens invaginatus in the mandibular premolar region. Indian J Dent Res 2009;20:238-40.
12. Serrano J. Triple dens invaginatus in a mesiodens. Oral Surg Oral Med Oral Pathol 1991;71:648-9.
13. Zengin AZ, Sumer AP, Celenk P. Double dens invaginatus: Report of three cases. Eur J Dent 2009;3:67-70.
14. Aneegundi RT, Kaveri H, Patil SB, Punnya A. Double dens invaginatus in an impacted molariform supernumerary tooth: A unique case. J Indian Soc Pedod Prev Dent 2008;26 Suppl 1:S26-8.
15. Kronfeld R. Dens in dente. J Dent Res 1934;14:49-66.
16. Colak H, Tan E, Aylikiç BU, Uzgur R, Turkal M, Hamidi MM. Radiographic study of the prevalence of dens invaginatus in a sample set of Turkish dental patients. J Clin Imaging Sci 2012;2:34.
17. Bishop K, Alani A. Dens invaginatus. Part 2: Clinical, radiographic features and management options. Int Endod J 2008;41:1137-54.
18. Kitchin PC. Dens in dente. J Dent Res 1943;15:117-21.
19. Ather H, Athar A. Concomitant quadruple dens invaginatus and talon cusp. J Mass Dent Soc 2013;61:41.