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Up-to Date Review And Case Report

Dilated odontoma: an unusual case report and literature review

P. Galvez[1,*], E. Garot[2,3], A. Bres[1], A. Crelier[3], C. Castain[4], S. Catros[1], M. Fenelon[1,2]

[1] CHU Bordeaux, Oral Surgery Department, 33076 Bordeaux, France
[2] CHU Bordeaux, Centre de compétence des maladies rares orales et dentaires, O-RARES, 33076 Bordeaux, France
[3] CHU Bordeaux, Odontology and Oral Health Department, 33076 Bordeaux, France
[4] CHU Bordeaux, Pathology Department, 33076 Bordeaux, France

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Abstract – Introduction: Dilated odontoma is the most severe form of a dens invaginatus, which is a rare dental malformation resulting from an invagination of the enamel organ into the dental *papilla* before calcification occurs. **Observation:** A healthy 7-years-old girl was referred to the oral surgery department to remove an impacted odontoma causing a delayed dental eruption of the right mandibular lateral incisor (tooth 42). The patient was painless and a lingual osseous swelling was observed. A computed tomography and a 3D segmentation revealed a shell-shaped mass in the position of the right mandibular lateral incisor, showing a complex anatomy. Surgical excision was carried out under general anesthesia. Histological analysis confirmed the diagnosis of DO. **Discussion and conclusion:** We performed a literature review investigating 16 cases of severe forms of *dens invaginatus* which required extraction. It highlighted the atypical aspect of our case and the importance of 3D imaging and segmentation in contributing to the accurate diagnosis and treatment of this dental malformation.

Introduction

Dens invaginatus (DI) or *dens in dente* is a rare dental malformation which was first formalized by Baume in 1874. It is a developmental anomaly resulting from an invagination of the enamel organ into the dental *papilla* before mineralization occurs. The incidence of DI is reported to range from 0.25% to 10% and it affects either the primary or the permanent teeth. The maxillary permanent incisors are the most frequently affected teeth [1].

A DI shows a broad *spectrum* of morphological dilatation of the crown and/or the root of the affected tooth. Oehlers classified DI into three categories according to the invagination depth [2]. In type I Oehlers classification, the invagination is confined to the crown and is not ranging beyond the cemento-enamel junction (CEJ). In type II, the enamel-lined invagination extends beyond the CEJ. In rare type III Oehlers classification, the invagination extends apically beyond the CEJ and perforates the surface of the root to create a second lateral foramen (type IIIa) or apical foramen (type IIIb) [3] (Fig. 1). The incidence of type I is 79% compared to 16% for type II. Type III is the least frequently reported (5%) [4]. In some rare cases, the invagination is so dilated that it impairs the formation of the tooth resulting in abnormal tooth development, which is also called dilated odontoma (DO), although it is not clearly defined as an independent entity in the current classification of odontogenic tumors. In those most extreme forms of DI, the tooth has a circular or oval shape with a radiolucent interior and presents a single structure, often with a central soft tissue mass [5].

To the best of our knowledge, the occurrence of the most severe variant of DI in the mandibular incisor region has never been reported before. The following report presents the diagnosis and management of a rare case of an impacted DO on the tooth 42 in a 7-year-old female with 2-years follow-up and a literature review of severe forms of DI which required extraction.

Case report

A healthy 7-years-old girl was referred to the department of oral surgery by her general dental practitioner regarding a delayed dental eruption. The dental practitioner performed an orthopantomogram and suspected a delayed eruption of the right mandibular lateral incisor (tooth 42) caused by an impacted odontoma (Fig. 2). The patient was painless and intraoral examination showed a lingual osseous swelling near

* Correspondence: paul.valgez@gmail.com

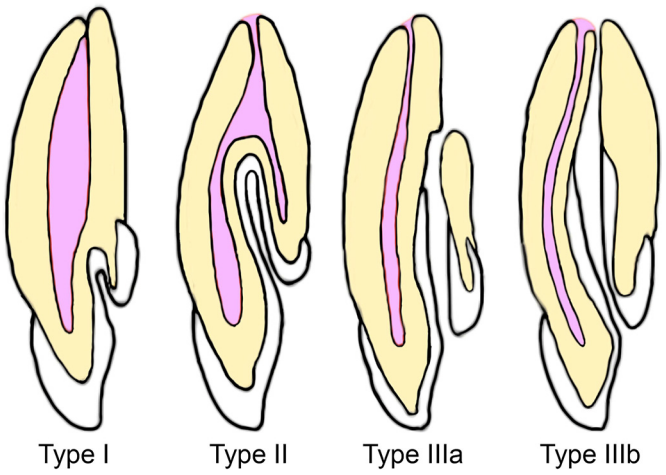

Fig. 1. The different types of dens invaginatus (Oehlers' classification). *According to Dr. Gambiez's diagram*.

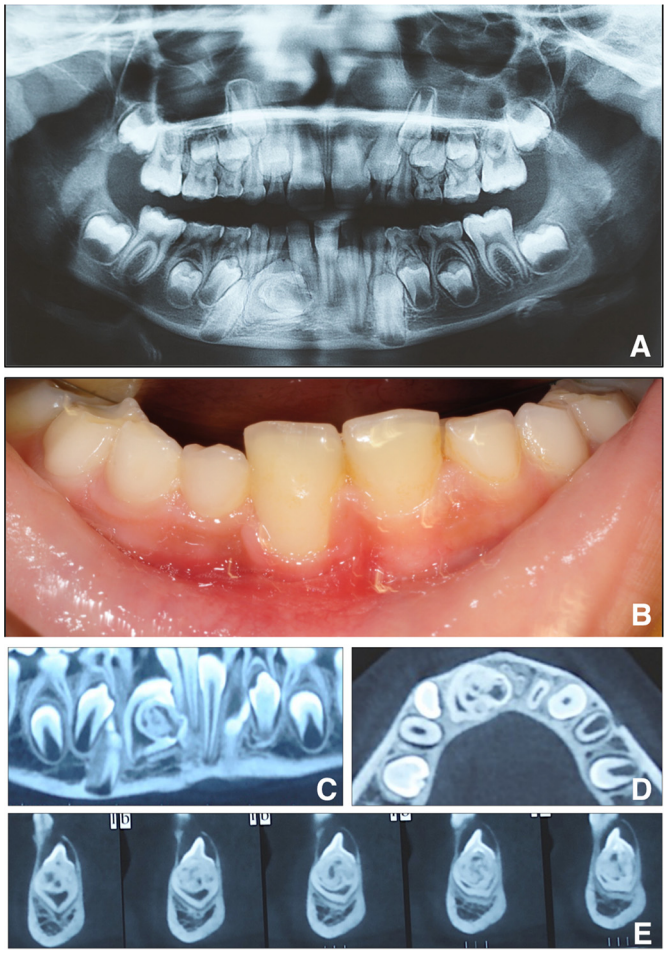

Fig. 2. (A) Panoramic X-ray findings at the first visit. (B) Intraoral clinical photograph at the first visit. (C) CBCT Panoramic view. (D) CBCT Axial view. (E) CBCT Coronal view. CBCT findings showing a well circumscribed shell shaped with a complex hypodense/hyperdense anatomy.

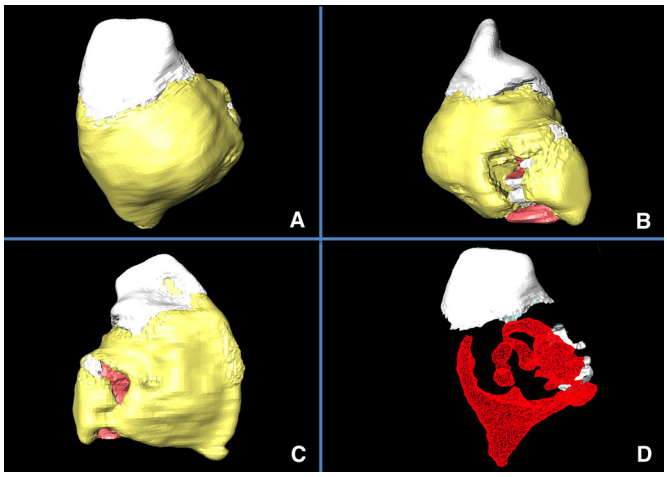

Fig. 3. Reconstructed 3D CBCT images of the final volume of the dilated odontoma in 16-bit. Each type of tissue was characterized by a color. The enamel-like mineralized tissue was colored in white, the cement-like structure was in yellow whereas the non-mineralized pulp-tissue was in red. (A) Labial view. (B) Mesial view. (C) Lingual view. (D) Internal view showing the complex anatomy of the pulpal canal and enamel invagination (in white).

the primary right mandibular lateral incisor (tooth 82) that was firm on palpation. A cone beam computed tomography (CBCT) was performed and showed a well-defined shell-shaped mass in the position of tooth 42, with hypodense and hyperdense components inside the mass (Fig. 2). The final volume of the lesion was reconstructed in 16-bit. The CBCT images were compiled with the 7.0.1. software Avizo® (Visualization Sciences Group, FEI® Company, OR, USA), enabling us to segment the various tissues composing this malformation. Each type of tissue was characterized by a particular color in order to further evaluate the lesion anatomy (Fig. 3). This 3D analysis revealed an enamel invagination and a complex pulp canal anatomy, contributing to the diagnosis of DO for the tooth 42. The inclusion of 42 was due to a significant increase of the tooth volume, preventing its eruption. Extraction under general anesthesia was planned. A lingual full-thickness mucoperiosteal flap was raised and the tooth 42 was extracted after osteotomy and luxation. Curettage of the extraction site was performed and the flap was closed using 4–0 absorbable sutures (Fig. 4). The macroscopic observation showed an oval hard mass of 12 × 11 × 10 mm (Fig. 5). The anatomopathological examination (AE) of the lesion revealed a disorganized dental structure (Fig. 6) and confirmed the diagnosis of DO. No complication occurred after the surgery. Two years after surgery, no alveolar ridge resorption was evidenced. Thanks to its mesio-version, the right mandibular canine (tooth 43) spontaneously erupted in position of the lateral incisor. 24 months later, an orthopantomogram revealed a complete healing of the lesion and an orthodontic treatment started to treat her overbite (Fig. 7).

Discussion

The DO is the most severe form of *dens invaginatus* (DI). Although the etiology and genesis of DI are still controversial,

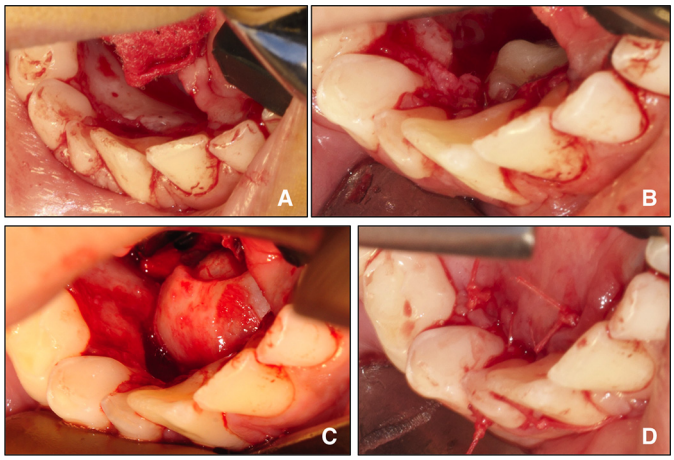

Fig. 4. Extraction of the dilated odontoma (DO). (A) Full-thickness mucoperiosteal lingual flap. (B) Osteotomy of the lingual cortical bone. (C) Luxation and extraction of the DO. (D) The wound was closed using absorbable sutures.

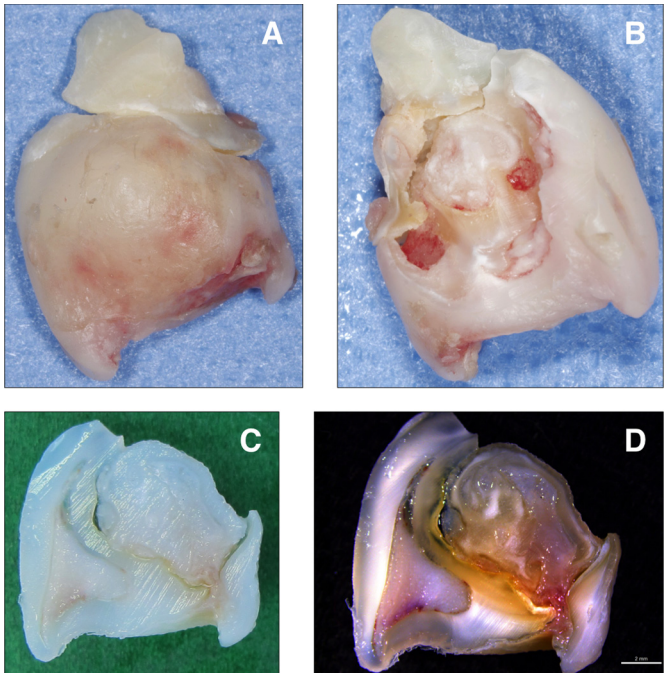

Fig. 5. Macroscopic view of the lesion. (A) Labial surface. (B) Lingual surface. (C) Sliced surface. (D) Sliced surface photographed in white light. A mass of enamel-like mineralized calcifications, dentin-like mineralized tissue and non-mineralized pulpal-type tissue is included in a better organized dental structure with enamel covering a dentine tissue and a pulp tissue. Scale bar: 2 mm.

several theories have been proposed: an aggressive proliferation of the inner enamel epithelium could occur into the dental *papilla*, or a delay of a focal group of cells and the pressure by an adjacent tooth germ could be involved. Genetic factors as well as infection or trauma have also been suggested as possible etiological factors [1].

We have found in the literature 16 cases of type III DI which required extraction (Tab. I). Among them, eight were

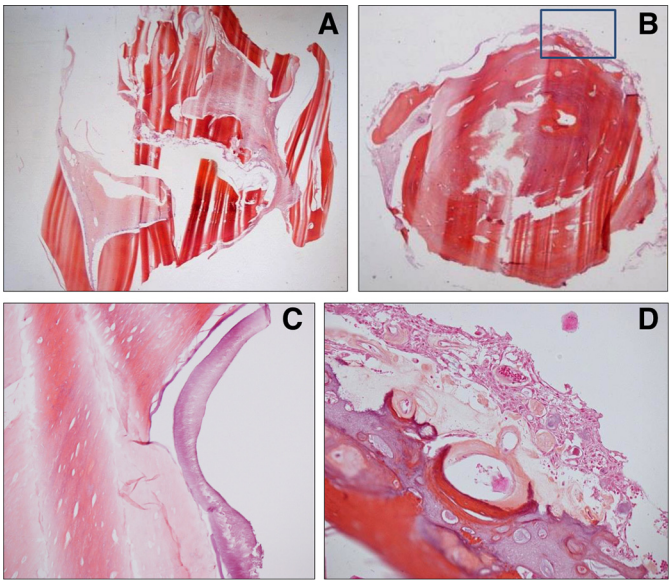

Fig. 6. Microscopic histological findings of the lesion. (A and B) HES, magnification ×10. A tubular dentin-type mineralized tissue associated with focal enamel-like mineralized tissue is surrounded by a non-mineralized pulpal-type tissue coated with its odontogenic epithelium. Blue square labeled corresponds to the area shown in Figure 6 (D). (C) HES, magnification ×200. An eosinophilic tubular dentin-type mineralized tissue associated with a more basophilic enamel-like mineralized tissue. (D) HES, magnification ×200. An odontogenic epithelium is found focally. Some ghost cells and dystrophic calcifications are present.

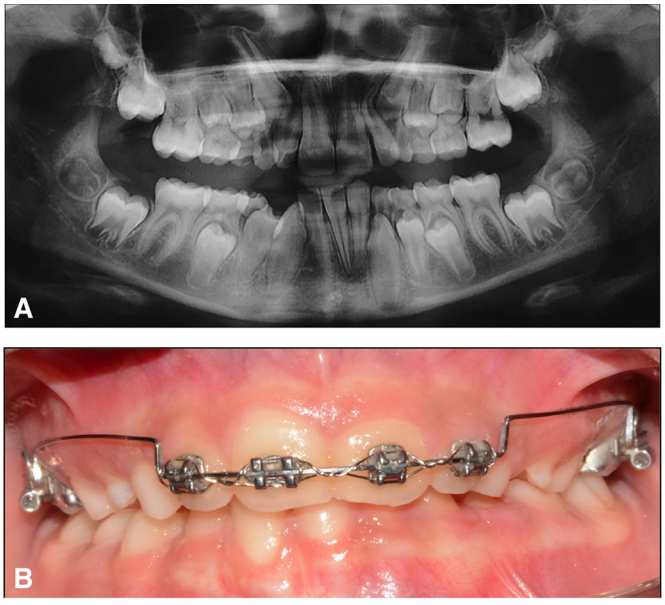

Fig. 7. (A) Panoramic X-ray 24 month after the surgery. (B) Intraoral clinical photograph.

categorized as DO [1,4–9]. Pain or swelling related to a local periapical infection are the main causes of DO discovery, as illustrated in 12 out of the 16 cases [3,4,6–14]. Indeed, one of the most significant clinical concern of DI is the risk of

Table I. Cases of type III DI which required extraction. DO: dilated odontoma; CBCT: cone beam computed tomography; DI: dens invaginatus; AE: anatomopathological examination.

Parameter Articles	Age / Sex	Circumstances of discoveries	Tooth number / eruption	Diagnosis	Additional examination(s) leading to diagnosis	Therapy
CASE N° 1 [1]	14 years / Female	Unerupted molar tooth	37 / Unerupted	DO	Panoramic X-ray, Computed tomography, AE	Extraction under general anesthesia
CASE N° 2 [4]	24 years / Male	Pain and swelling	21 / Erupted	DO	Panoramic X-ray, CBCT	Extraction under local anesthesia
CASE N° 3 [10]	30 years / Female	Swelling and cutaneous fistula	48 / Erupted	Type III DI	Panoramic X-ray	Extraction
CASE N° 4 [6]	16 years / Male	Swelling	44 bis / Unerupted	DO	CBCT	Extraction under local anesthesia
CASE N° 5 [6]	24 years / Male	Pain and swelling	17 / Unerupted	DO	CBCT, AE	Extraction under general anesthesia
CASE N° 6 [3]	45 years / Male	Pain and swelling and impacted canine (13)	12 / Erupted	Type IIIb DI	Panoramic X-ray	Extraction under local anesthesia
CASE N° 7 [16]	15 years / Male	Microdontic and discolored teeth	22 / Erupted	Type III DI	Panoramic X-ray, CBCT	Extraction
CASE N° 8 [11]	12 years / Female	Palatal swelling	22 /Erupted	Type III DI	Periapical radiograph, Panoramic X-ray	Extraction
CAS N° 9 [15]	12 years / Female	Subluxation injury	13 / Erupted	Type III DI	CBCT, AE	Extraction under general anesthesia
CASE N° 10 [7]	47 years /Female	Diffuse pain and discomfort	Mandibular left third molar region / Erupted	DO	Panoramic X-ray, Intra-oral X-ray, CBCT	Extraction under local anesthesia
CASE N° 11 [5]	28 years /Female	Asymptomatic. Radiological finding	48 / Unerupted	DO	Panoramic X-ray, CBCT, AE	Extraction under general anesthesia
CASE N° 12 [12]	10 years /Female	Cellulitis	12 / Unerupted	Type III DI	Panoramic X-ray, Intra-oral X-ray	Extraction under general anesthesia
CASE N° 13 [13]	42 years / Female	Pain and swelling	12 / Erupted	Type IIIb DI	Periapical radiograph, Computed tomography	Extraction
CASE N° 14 [8]	18 years / Male	Malformed tooth + intermittent mild pain	21 bis / Erupted	DO	Pariapical radiograph, Occlusal radiograph, Panoramic X-ray, CBCT	Extraction under local anesthesia
CASE N° 15 [14]	25 years / Female	Pain and luxation of all premolars	14 15 24 25 34 35 45 / Erupted	Type III DI	Periapical radiographs, AE	Extractions
CASE N° 16 [9]	14 years / Male	Swelling	23	DO	Pariapical radiograph, CBCT, AE	Extraction

developing pulpal necrosis. Invagination can communicate with the oral cavity, and the enamel is generally thin, of poor quality or even missing, allowing microorganisms to colonize the pulp. Some canals can also extend directly from the invagination to the pulpal tissue, leading to pulpal or periapical pathologies even in the absence of caries [11].

The maxillary teeth are more commonly affected than mandibular teeth. Permanent maxillary lateral incisors are more frequently affected, and they are followed by maxillary central incisors, premolars, canines and molars. The female to male ratio is 3:1 [3]. In our review, almost 60% of the cases involved women [1,5,7,10–15]. Eleven cases were related to maxillary teeth [3,4,6,8,9,11–16]. Among them, nine were located in the anterior maxillary region [3,4,8,9,11–13,15,16]. Our report presents an extremely rare case of DO localized in the anterior mandibule. Few cases of DO have been reported in the mandible, but none of them was located in the anterior region [1,5–7,10,14]. In most cases, teeth with DI can erupt, as demonstrated in our review. Here, the oversized shape of the tooth caused its impaction. Finally, this review showed that the mean age at diagnosis was 24 years, which highlights our early management.

The diagnosis of DI can be mentioned with an anatomic alteration of the crown and/or the root. A DO usually presents a circular or ovoid shape with a radiolucent interior corresponding to a central soft tissue mass, which can be accompanied by hard tissue [5]. The hard tissue structure can be completely inverted due to severe invagination. Thus, anatomopathological examination after extraction can help or confirm the diagnosis [5,6,15,17]. Here, the ovoid shape of the teeth without morphological appearance of a crown or root suggests that the invagination occurred in the early stage of tooth development [1].

DI can be detected after a radiographic evaluation with a panoramic X-ray and/or intra-oral radiographs [3,9–14]. However, conventional radiographs are not sufficient to ensure the diagnosis of the mineralized structure observed. In these cases, the use of computed tomography (CBCT) can be helpful. Among the 16 reported cases, computed tomography was used in eleven patients [1,4–9,13,15,16]. In this case report, the CBCT allowed to rule out the diagnosis of odontoma and to identify a DO on the tooth 42. Tomographic images can also clarify the relationship between the invaginated portion of the tooth with the chamber and/or the root canals, and determine whether the invagination is communicating with the periodontal ligament space [13]. Tomographic images provide details about the internal anatomy in numerous thin slices in different planes of section and allows the clinician to accurately adapt the treatment and assess difficulties [9,15]. This exam thus plays a key-role in the early diagnosis and management of these developmental anomalies.

The most common differential diagnosis of DI encountered is odontoma [18]. According to the 4th edition of the World Health Organization Classification of Head and Neck tumors, odontoma can be divided into two groups: complex odontoma and compound odontoma [19]. In the complex odontoma, the calcified dental tissues are simply arranged in an irregular mass bearing no morphologic similarity to rudimentary teeth. The compound odontoma is made of all odontogenic tissues in an orderly pattern, which result in many teeth-like structures, but without morphologic resemblance to normal teeth [6]. In this case, an odontoma was initially suspected and the differential diagnosis between a complex odontoma and a DO was performed using the CBCT and then confirmed by the anatomopathological examination. Cemento-ossifying fibroma and osteoma are also two frequently mentioned differential diagnoses [5,7].

The treatment of DI is related to the degree of complexity of the tooth anatomy. Therapeutic options range from prophylactic fissure sealing, as recommended for type I DI, restoration or endodontic treatment for type II DI [20], to a surgical approach like endodontic surgery or extraction in the case of type III DI. Periapical surgery is indicated either when conventional non-surgical root canal treatment has failed because of the anatomic variations of the canals [21], or when the apexification is unsuccessful in cases of immature DI with nonvital pulp [11]. Extraction is required in extremely rare cases of complex root anatomy combined with a morphology incompatible with the long-term preservation of the tooth [3]. In our case, the impaired anatomy of the tooth 42 (oval and oversized shape, incompatible with function and esthetics), led to the decision of extraction. Multidisciplinary management (radiologist, oral surgeon, pediatric dentist and orthodontist) is necessary for the diagnosis and treatment of such rare cases.

Conclusion

This case report highlights the usefulness of the three-dimensional imaging by cone beam computed tomography that provided relevant information to investigate the complex anatomy of the lesion and thus determine the diagnosis and the appropriate treatment.

Conflicts of interests: The authors declare no conflicts of interest.

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