Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology 31 (2019) 254–257

Contents lists available at ScienceDirect

Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology

journal homepage: www.elsevier.com/locate/jomsmp

Case Report

Compound odontoma as secondary manifestation following impaction of supernumerary tooth in a single region: A case report

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ARTICLE INFO

Keywords:
Odontogenic tumors
Compound odontoma
Supernumerary teeth
Tooth retention

ABSTRACT

Compound odontoma, a benign epithelial odontogenic tumor, manifests disordered clinical and pathological features due to the formation of a variant tooth-like structure; moreover, it is frequently associated with disrupted permanent tooth eruption, which is a characteristic of supernumerary teeth. Here, we present a rare case of compound odontoma in a 7-year-old male with a unique and an informative clinical course. The patient presented compound odontoma and supernumerary tooth both in the same perifollicular region of the left permanent central incisor of the maxilla; however, each lesion was radiographically detected at different times. Interestingly, an odontoma-like entity appearing as a dense radiopaque lesion was recognized only 1 year and 6 months after extracting the primary incisor and impacted supernumerary teeth, respectively. A secondary lesion subsequently migrated to the impacted crown of the permanent incisor. The surgical specimen revealed partial involvement with a developing odontoma.

1. Introduction

Odontomas, classified as benign mixed epithelial and mesenchymal odontogenic tumours by the World Health Organization [1], are most commonly found in the oral cavity. In most cases, these tumors are asymptomatic and small and are therefore usually accidentally discovered during routine radiographic examinations or during the disturbed eruption of permanent teeth [2]. Unlike other benign intraosseous tumors, odontomas are considered as hamartomas or malformations and their growth typically stops upon the completion of mineralization, as observed with supernumerary teeth [2]. The anterior section of the maxilla is the preferred site of occurrence of ondontomas [3] similar to that observed for supernumerary teeth [4,5].

Odontomas can manifest as either compound or complex types. Compound odontoma (CpOD), formed by tooth-like structures, is favorably associated with tooth retention and tends to erupt earlier than complex odontomas [6]. Therefore, although CpOD and supernumerary teeth are defined as distinct entities, they have the same pathologic origin, developmental process, and clinical characteristics [7].

Herein, we report a rare case of CpOD, which was incidentally diagnosed as a secondary manifestation of an impacted supernumerary tooth from the same site but of a different origin. It was associated with the retention and migration of a permanent tooth in the anterior section of the maxilla. We present the case's unique clinical course, including a review of the literature on this condition.

2. Case report

A 7-year-old male was referred to our institution with a chief complaint of delayed eruption of the left permanent central incisor of the maxilla. The patient had no significant medical or family history or dental/maxillofacial trauma or infectious diseases history. Intraoral examination showed few symptoms around the left anterior region of the maxilla and alveolus, except a persistent primary central incisor tooth. Radiographic examination revealed a supernumerary tooth adjacent to the embedded tooth germ of the left permanent central incisor (Fig. 1Aa). Additionally, coronal and sagittal computed tomography scans confirmed the exact location of the supernumerary tooth, which seemed to interfere with the eruption of the successor tooth.

Based on the radiographic findings, extraction the supernumerary tooth along with the persistent primary incisor tooth was planned. Postoperative healing was uneventful; however, the permanent central

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https://doi.org/10.1016/j.ajoms.2019.02.006
Received 7 January 2019; Received in revised form 5 February 2019; Accepted 22 February 2019
Available online 08 March 2019

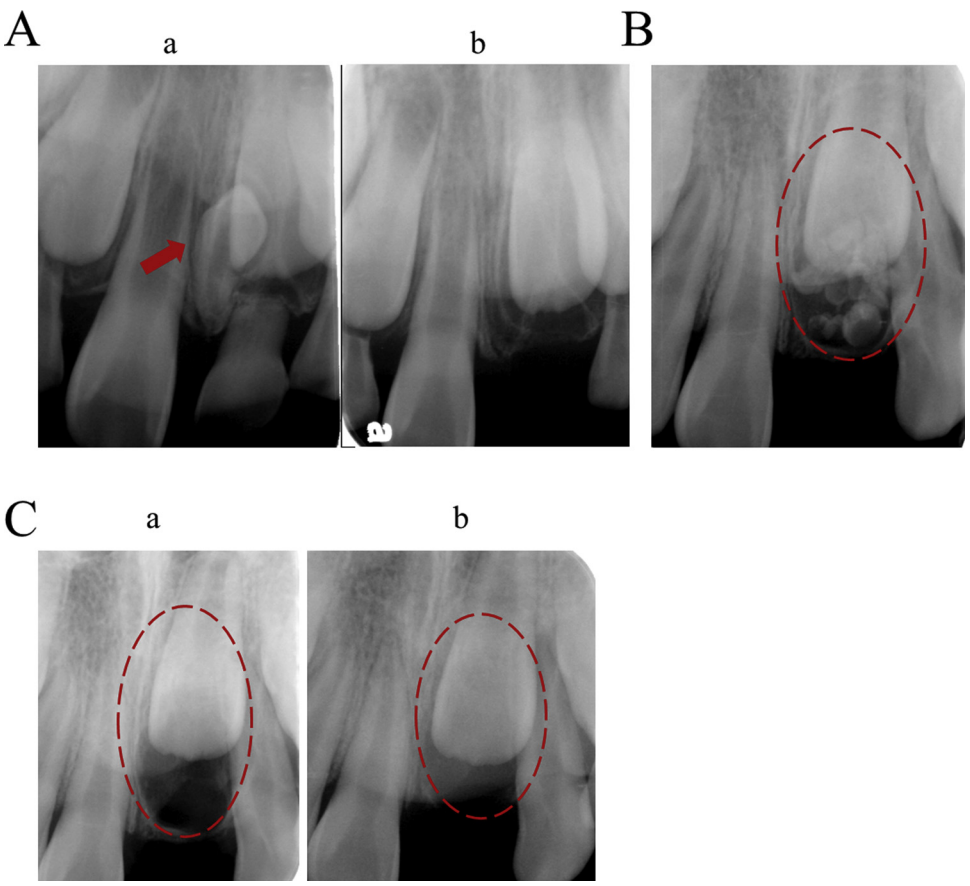

Fig. 1. Intraoral radiographic views taken during the course of treatment.
A: Dental radiographic images taken before (Aa) and 6 M after (Ab) the extraction of supernumerary tooth (arrow). B: Follow-up radiograph obtained 18-months after the initial surgery revealed an odontoma-like radiopaque lesion. C: Postsurgical radiographic images taken 40-days (Ca) and 6-months (Cb) after tumor resection. The ellipses in B and C represent follicular region of the left permanent central incisor tooth. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

incisor did not erupt until 6-months after the surgery (Fig. 1Ab). Follow-up radiographs obtained after 1 year without routine visits to our department. This revealed an odontoma-like radiopaque lesion that was located in the follicular region of the permanent central incisor. Relative to its presurgical location, the crown of the successor tooth had migrated in the apical direction (Fig. 1B). Thus, the patient was provisionally diagnosed with an odontogenic benign tumor; the intraosseous lesion was resected under general anesthesia (Fig. 1Ca).

The postoperative process was uneventful and without any evidence of recurrence. A follow-up radiograph obtained 6-months after the surgery showed poor eruptive movement of the impacted permanent incisor (Fig. 1Cb). Additional orthodontic traction was applied for further treatment.

The extracted lesion was capsulated and was found to have 11 malformed tooth-like structures (Fig. 2A). Microscopic examination of the surgical specimen revealed layered hard tissue comprising an enamel matrix, mineralized dentin, and pulp (Fig. 2Ba). Immature structures that resembled the tooth germs from the periphery were noted; these findings were compatible with the diagnosis of developing odontoma (Fig. 2Bb). Based upon these histopathological findings, the patient was definitively diagnosed with CpOD.

3. Discussion

Odontomas are considered as hamartomas or malformations that resemble tumor-like lesions rather than true neoplasms. Although their etiology is variable, inflammation, injury (including trauma), and genetic issues are potential causes of odontomas; however, its pathogenesis is not completely understood [8–10].

A previous study reported that traumatic injury to the primary dentition potentially affects the development of the permanent successor, resulting in an odontoma-like malformation [11,12]. Previous reports highlighted odontoma as a developmental anomaly that exhibits a close relationship between primary teeth apices and the germs of the permanent successors, which is the reason for the developmental disturbance in the permanent dentition caused by dental trauma to the primary teeth [11,12]. Although dental injuries are capable of disturbing the normal development of the permanent teeth, odontoma-like malformations or odontomas arising during infancy are extremely rare [11,13,14]. Considering odontogenesis, the calcification of permanent central incisors of the maxilla begins at 3–4 months and is completed by 4–5 years of age. The severity of the morphological disturbance depends on the age and stage of dental morphogenesis that was exposed to injury [11,12,15]. Accordingly, many cases of odontomas are not associated with impacted permanent teeth and are instead accidently diagnosed until a disturbance in the eruption of successor teeth becomes apparent during the second decade of life [16].

In the present case, preliminary examination revealed no trauma or inflammatory symptoms in the anterior region of the maxilla. Subsequent radiographic investigation revealed that the supernumerary tooth embedded above the left permanent central incisor resulted in the persistence of primary incisor tooth and delayed the eruption of the permanent tooth. Although there were no postsurgical complications, the permanent incisor tooth remained embedded and CpOD clearly appeared as a radiopaque lesion within the intrafollicular region. This was accompanied by the migration of the crown of the permanent central incisor in the apical direction during 12–30 months after removing both the persistent primary tooth and the impacted supernumerary tooth.

Pathologically, both the embryonic residual epithelium such as the rests of Malassez, rests of dental lamina, rests of Serres, and pericoronal follicular tissue related to mature unerupted or impacted teeth have been proposed as the potential sources of central epithelial odontogenic tumors [3,17,18]. CpOD more frequently interferes with the eruption of

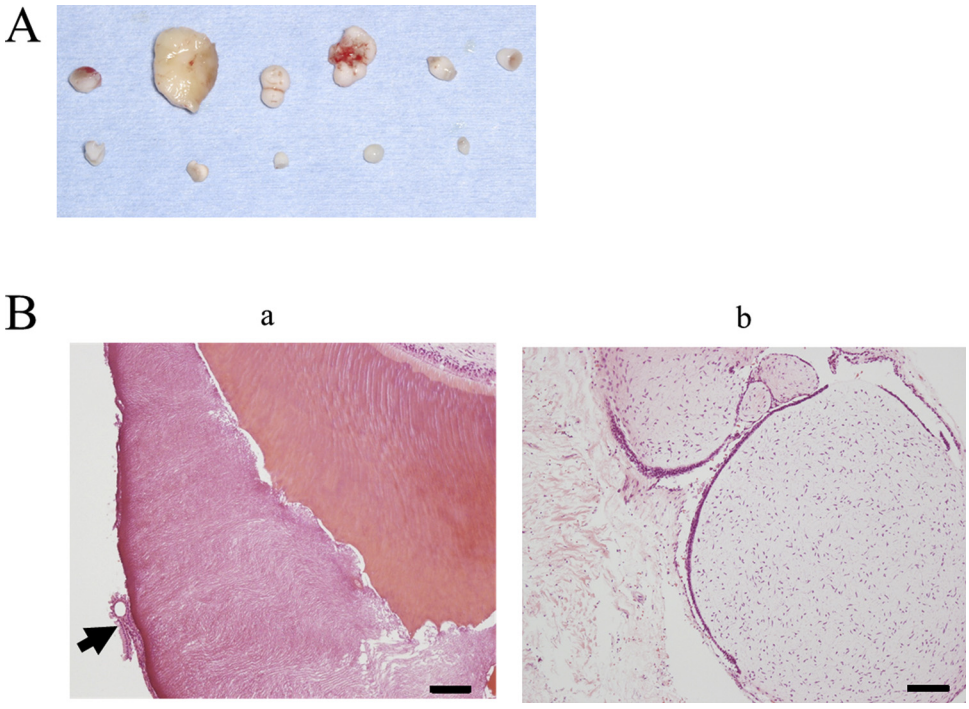

Fig. 2. Macroscopic and microscopic evaluation of the surgical specimen.
A: Examining the surgical specimen revealed the presence of various malformed tooth-like entities. B: Histopathological examination (Hematoxylin and Eosin staining) with higher magnification showed layers of enamel matrix, dentin, and pulp tissue. Black arrow shows the follicular epithelium (Ba). An immature structure equivalent to the developing odontoma was partially observed in the periphery (Bb). Bars, 100 μm.

permanent teeth and is generally diagnosed at an earlier age than complex odontoma [7] as observed in supernumerary teeth [19]. Previous studies have supported the hypothesis that CpOD should be considered a malformation, potentially formed as a result of locally-conditioned hyperactivity of the dental lamina as observed with supernumerary teeth [7,20]. As for the extra-osseous counterpart of odontomas, peripheral odontoma, is extremely rare and considered to be originated from the remains of odontogenic epithelium embedded in the oral soft tissue [14]. Although the morphological features of peripheral odontomas are dependent on the developmental stage of each embedded tooth germ at discovery, most of cases reported were found at earlier aged period of infancy than CpODs because they are not intraosseously existed [14]. Considering the clinical course of the patient in the present case and the aforementioned assumption, two distinct origins of CpOD were hypothesized: as a duplicated tooth bud or intrafollicular-conditioned hyperactivity related to the permanent central incisor. During the early disease stage, a supplementary tooth is formed at the same time or just before the formation of the crown of the permanent central incisor. This may be followed by the occurrence of a primitive or precursor lesion, eventually forming a mature CpOD after the extraction of primitive and impacted supernumerary teeth.

To prevent later complications such as disrupted eruption of the permanent teeth, as observed in the present case, early diagnosis is necessary. A previous study revealed the evidence of incipient or precursor lesions related to intraosseous epithelial odontogenic tumors such as ameloblastomas, calcifying epithelial odontogenic tumors, and calcifying cystic odontogenic tumors [3]. Developing odontomas are uncommon and occur in the peripheral soft tissue [21,22]. Although detecting an incipient lesion before mineralization may be difficult, as in our case, periodical radiographic observations should be emphasized, especially in cases with supernumerary teeth and disrupted eruption of the permanent teeth in the anterior maxillary region. This report serves as a guideline considering the possibility of coexistence or subsequent development of odontoma-like lesions.

Conflict of interest

None.

Patient consent

Written consent was obtained from the patient's parents.

Funding support

The authors declare that they did not receive any support in the form of grants.

Acknowledgements

None.

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