



Case Report

The Unique Case of Rapid Developing Compound Odontomes Following Fibrous Tissue Removal Around Impacted Tooth

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Abstract

Odontoma is classified as an odontogenic tumor that develops slowly, asymptomatic, and usually found incidentally during the radiographic examination associated with the impeded eruption of teeth and displacement of adjacent teeth. This case report describes a unique encounter of rapid formation odontomas which formed and expand rapidly upon initial removal of fibrous tissue surrounding the impacted tooth and radiographically detected following the spontaneous eruption of the impacted tooth. Thus, odontomas which are known to be a slow-growing lesion may have a more rapid progression than previously thought.

Keywords: Compound odontoma, fibrous tissue, impacted tooth

Introduction

Odontoma is classified as an odontogenic tumor (hamartoma) which consists of enamel and dentin with multiple numbers of pulp and cementum when it is fully developed.[1] Based on the recent 2017 World Health Organization (WHO) classification of odontogenic tumors and cysts, odontomas can be classified into compound type and complex type.[2] Compound odontoma is identified by multiple, small tooth-like structures in a well-organized pattern. Meanwhile, complex odontoma is present as randomly arranged masses of enamel and dentin with no anatomic similarity to a tooth.[3] Odontomas are slow-growing, asymptomatic,

and usually found incidentally during the radiographic examination as odontomas are closely related to the late eruption of teeth and displacement of adjacent teeth.[4]

This case report illustrates a unique encounter due to the rapid formation of multiple odontomas and expansion of lesions after the removal of fibrous tissue surrounding the impacted tooth. Although calcifications were noted in the tooth germ lining, no presence of odontoma was detected. However, the removal of the fibrous tissue allowed spontaneous eruption of the impacted tooth while multiple odontomas were forming and maturing simultaneously. Therefore, a periodic review following surgical intervention for lesion removal is essential to monitor the condition of the area asso-

How to cite this article: Zainuren ZA, Abang Ibrahim DFA, Venkiteswaran A, Kamil WNWA, Reduwan NH. The Unique Case of Rapid Developing Compound Odontomes Following Fibrous Tissue Removal Around Impacted Tooth. J Pediatr Dent 2021;7(3):173-177

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ciated with the lesion. To our knowledge, this is the first case report of a rapidly developing compound odontomas following fibrous tissue removal around impacted tooth reported in the literature.

Case Presentation

A fit and healthy 9-year-old girl was referred to the clinic for unerupted tooth 21 (Fig. 1). According to the patient and her mother, the contralateral tooth (tooth 11) had erupted by the age of 8.5 while the upper left deciduous central incisor (tooth 61) remained firm in place. The patient had no previous history of trauma involving the 21 region. Intraorally, a bulge was palpable on the labial aspect of the 21 region.

Pre-operative intraoral periapical (IOPA) image (Fig. 2) revealed the presence of an unerupted upper left permanent central incisor. The coronal and root development of tooth 21 appear approximately similar to tooth 11 with no indication of root dilaceration. There was no significant presence of supernumerary or odontoma was noted. However, multiple ill-defined radiopaque speckles were detected overlying the cingulum area of tooth 21 (red arrows).

Extraction of the primary tooth 61 was performed. Upon review within the next 3 months, it was noted that there were no signs of the eruption of tooth 21. It was then decided that a surgical exposure of the tooth under local anesthesia would facilitate the tooth's eruption. A flap was raised, and a thick layer of fibrous tissue was found to be covering the crown of tooth 21. The specimen was dispatched for histopathological examination (HPE). The microscopic observation showed fragments of myxoid connective tissue lined by non-keratinized stratified squamous epithelium. In addition, scattered strands of odontogenic epithelial islands were seen in the stroma. Based on the clinical, radiological, and microscopic examination, the diagnosis established was dental follicle at 21 region (Fig. 3).

Due to the COVID-19 pandemic, we were only able to see the patient after a year. Upon one year's review, tooth 21 erupted and was well-aligned clinically (Fig. 4). The patient was thrilled with her appearance due to the erupted tooth 21. A post-operative IOPA of tooth 21 was obtained showing the presence of multiple well-demarcated tooth-like structures, besieged by a well-defined radiolucent rim. The radiopaque lesions appear located below the cervical segment of tooth 21 extending to the apical region. The radiographic impression is suggestive of a compound odontoma (Fig. 5).



Figure 1. The intraoral view showing unerupted 21



Figure 2. A pre-operative intraoral periapical image showing presence of multiple ill-defined radiopaque speckles overlying the cingulum area of tooth 21 (red arrows)

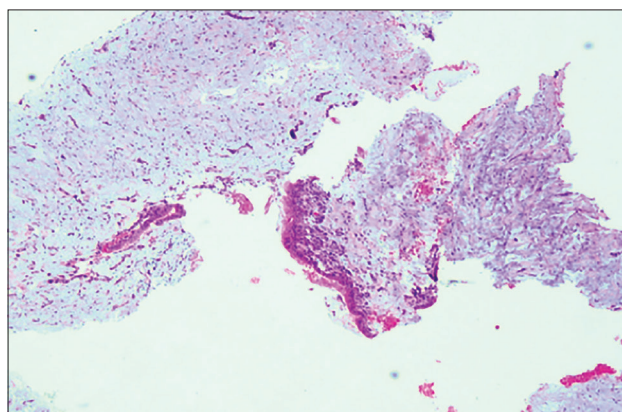


Figure 3. Photomicrograph showing myxoid connective tissue lined by non-keratinized stratified squamous epithelium and presence of odontogenic epithelial rest in the stroma (H&E staining, 20x)



Figure 4. The intraoral clinical pictures of erupted 21



Figure 6. Surgical removal of the lesion done under local anesthesia

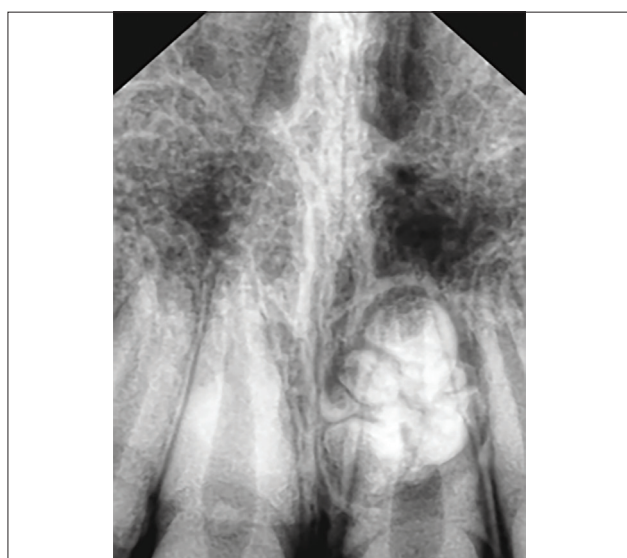


Figure 5. A post-operative intra oral peri-apical radiograph of tooth 21 was obtained showing presence of multiple well-demarcated tooth-like structures, besieged by a well-defined radiolucent rim suggestive of a compound odontoma

Cone-beam computed tomography (CBCT) images were obtained, which revealed a hyperdense mass with well-defined corticated borders external to the radiolucent margin. Internally, there are multiple radiopaque structures with variable odontoid-like opacity, resembling multiple malformed teeth were present. The mass was in contact with the palatal surface of tooth 21 and extended mesially invading the incisive foramen. Tooth 21 appeared labially displaced, but no sign of root resorption was detected. The radiographic impression of this lesion was compound odontoma.

Surgical removal of the lesion was performed under local anesthesia (Fig. 6) simultaneously with nitrous oxide sedation. Multiple tooth-like structures and the epithelial lining surrounded the lesion was removed during the procedure and sent for HPE. The micro-

scopic observation revealed myxoid connective tissue lined partially by a bilayer cuboidal epithelial cells resembling reduced enamel epithelium. Scattered strands of odontogenic epithelial rest were also evident in the stroma. Meanwhile, the decalcified section showed organize mass composed of enamel dentin and pulp tissue resembling the structure of a tooth. Based on the clinical, radiological, and microscopic examination, a diagnosis of compound odontomas surrounded by a dental follicle was made. The patient is still under periodic follow-up and at the latest visit, the surgical site is healing well with no signs and symptoms noted.

Discussion

Compound odontomas are an odontogenic tumor that emerges from both odontogenic epithelium and mesenchyme.[5] It is distinguished by the existence of dental tissues which include enamel, dentin, cementum, and pulpal tissues in a well-organized pattern but the size and shape are altered, lead to the formation of multiple small tooth-like structures called denticles.[3] Compound odontomas can be positioned in any place in the dental arches, but most frequently are found in the anterior maxilla region especially in upper incisors and canine areas.[6,7] These asymptomatic lesions are usually characterized by a slow and painless growth.[7]

The present case reported no obvious sign of calcified tissue obstruction associated with impacted tooth 21 except for a layer of thick dental follicle. The removal of the thick fibrous tissue around the impacted tooth 21 region was performed under local anesthesia. Unfortunately, we were only able to review this patient after a year due to the COVID-19 pandemic. During the one-year review, tooth 21 has erupted to the desired position and well-aligned. However, the post-operative radiographic examination revealed the

presence of multiple radiopaque lesions with well-defined corticated borders which were suspected as odontomas. This is considered an unusual finding as the impacted tooth 21 erupted uneventfully despite the rapid formation and maturation of multiple odontomas in its close proximity.

The etiology of odontoma remains obscure. However, it is believed to be related to a number of factors such as the history of trauma to the deciduous dentition, inflammatory and infectious process, or even alterations of the genetic components responsible for controlling dental development.[8,9] As the compound odontomas are asymptomatic, they are usually discovered as an incidental finding in children or adolescence as most of the cases emerge before the age of 20.[6] They are frequently associated with disturbance in tooth eruption, which results in impacted permanent teeth and sometimes it may appear as an expansion of bone, pain, and displacement of the adjacent tooth.[9,10] However, in this case report, tooth 21 managed to erupt even though multiple compound odontomas were forming in close relation to its root.

There are three clinical features of odontomas that have been acknowledged in the literature which include intra-osseous, extra-osseous, and erupted.[11,12] Intra-osseous odontomas are frequently reported in the anterior zone of the maxilla (67%) followed by the anterior and posterior zone of the mandible (33%).[12] Eruption of odontoma is rare but mostly observed with complex odontomas and in between age 12 to 59 years old.[13] The slow-growing odontomas are hardly ever exceeded the size of a tooth, but when the odontomas become bigger which are occasionally about up to 6 mm and more in diameter, these lesions can result in cortical bone expansion.[1] Bony expansion is seldomly associated with compound odontomas but frequently related to complex odontomas.[14] In severe cases, growth pressure from the odontomas may cause pain, infection, devitalization of adjacent teeth, and resorption of the tooth and the surrounding bone.[15,16]

As odontomas are asymptomatic, most of them are diagnosed incidentally during routine radiographic examination.[14] They are frequently located between the roots of teeth and appear either as an irregular mass of calcified material surrounded by a narrow radiolucent band with a smooth peripheral surface or as multiple tooth-like structures with the same peripheral outline.[17] For compound odontomas, radiographic examination frequently appears as comparative orderly malformed teeth or tooth-like structures surrounded by narrow radiolucent zone, meanwhile, complex onto-

mas often exhibit calcified mass with the radiodensity of tooth components, which is also besieged by a narrow radiolucent rim.[14] In the case of a developing odontoma, it can be noticed by routine radiographic examination but may cause difficulty in recognition due to lack of calcification.[17] This can be observed in this case report as only a single speckle of radiopacity was noted from the initial radiographic examination (Fig. 2) which is quite ambiguous to determine the diagnosis that resulted in impacted tooth 21 in the first place.

Histological features of compound odontoma show a number of structures resembling small, single-rooted teeth, contained in a loose fibrous matrix[1] and microscopically the compound odontomas' denticles consist of a central core, similar to pulp tissue, surrounded by primary dentin and covered with partially demineralized enamel and primary cementum.[18] Meanwhile, complex odontomas are majority comprised of mature tubular dentin. This dentin encircles clefts or hollow circular structures that contained the mature enamel that was removed during the decalcified process. The spaces may consist of little amounts of enamel matrix or immature enamel.[1] The finding of this case report was consistent with compound odontomas as the specimens consist of myxomatous and loose fibrous tissue partially lined by bilayer cuboidal epithelial cells that resemble reduced enamel epithelium. Scattered strands of odontogenic and calcified material were also observed.

Early recognition of odontomas as a diagnosis in the deciduous dentition is important to prevent the complications such as the failure of eruption of permanent teeth, cystic changes, and bone destruction.[15] Early diagnosis also ensures inexpensive treatment and a better prognosis.[19] In regards to this case report, the early diagnosis was not possible as the initial finding from the first surgical procedure did not reveal the presence of developing odontomas. Fortunately, as we called the patient for review, the impacted tooth 21 had already erupted at the desired position and well-aligned. To our surprise, the evident presence of multiple odontomas was observed from the recall radiographic investigation. Therefore, recall appointment plays an important role in this case as we were able to identify the abnormalities that emerged after the impacted tooth already erupted.

Even though odontomas have quite limited growth potential, they should not be left behind due to the presence of various tooth formulations that have the ability to emerge to cystic changes, disrupt the eruption of permanent teeth, and cause significant bone loss.[20] Due to the low recurrence rate, the treatment of choice will be surgical removal of the lesion. Odontomas are quite

easy to enucleate and as they are commonly separated by a septum of bone, the injury to adjacent teeth and surrounding structures during the procedure are quite rare.[21] However, particular attention should be taken into consideration to ensure the lesion was completely removed in order to prevent any recurrence in the future.[14] As reflected in this case report, even though the initial presentation of the lesion did not portray the presence of odontomas, we have to consider removing the entire dental follicle associated with the impacted tooth instead of just making a window by removing the thick fibrous tissues as this procedure might be one of the factors that triggered the rapid growth of the developing odontomas within a year. Besides, a periodic review following surgical intervention for lesion removal is essential to monitor the condition of the area associated with the lesion.

Conclusion

Therefore, periodic follow-up and radiographic investigations are vital even though the impacted tooth has already erupted to the desired position in order to detect any other emerging lesion. Besides, to allow spontaneous eruption of the impacted tooth, we must consider removing the entire dental follicle rather than just making a window by removing the thick fibrous tissues. Odontomas which are usually slow-growing and closely related to impacted teeth can sometimes develop rapidly and simultaneously while the tooth was erupting normally.

Financial Disclosure: Nil.

Conflict of Interest: None declared.

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