CASE REPORT

The Enigma Unveiled: Expansile Compound-complex Odontoma in the Anterior Maxilla of a Teenager

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Abstract

Aim and objective: The objective of this case report is to offer insight into an expansive compound-complex odontoma located in the anterior maxilla of a 15-year-old male. The focus is placed on the importance of early detection and the progressive comprehension of odontomas.

Background: Odontomas are common odontogenic lesions that are frequently discovered during examinations for delayed tooth eruption. There are two distinct classifications for odontomas—compound odontomas and complex odontomas. With its own each set of characteristics. A timely diagnosis is critical for avoiding complications.

Case description: A male individual aged 15 years exhibited an expansive compound-complex odontoma located in the anterior maxilla. The clinical examination showed delayed tooth eruption and asymptomatic swelling. The radiographic images showed a radiopaque mass with tooth-like structures and radiolucent borders affecting the surrounding dentition. A surgical excision procedure was conducted, followed by a subsequent histopathological examination confirming the diagnosis of compound-complex odontoma. The patient continued orthodontic treatment after a 1-year follow-up without recurrence.

Clinical significance: This case emphasizes the importance of regular dental exams in detecting odontomas early. This observation also highlights the growing understanding of odontomas as hamartomatous odontogenic malformations and the challenges of diagnosing them clinically. Additional molecular investigations are required to facilitate the classification and elucidation of genetic factors.

Keywords: Case report, Complex odontoma, Compound odontoma, Impacted tooth, Maxilla, Odontoma, Odontogenic tumors.

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Introduction

Odontomas, the most common intraosseous developmental odontogenic lesions, constitute a significant aspect of oral pathology due to their diverse clinical and histopathological presentations.[1] Initially diagnosed as neoplasms, odontomas were soon recognized as hamartomas—malformations that result from the development of odontogenic epithelium and ectomesenchyme. These hamartomas produce enamel, dentin, and cementum.[2] The World Health Organization (WHO) has established a classification for odontomas comprising two variants: compound and complex.[3] This odontogenic anomaly, which includes compound and complex odontomas, can be challenging to diagnose. Compound odontomas exhibit organized tooth-like structures, primarily in the anterior maxilla, while complex odontomas are characterized by haphazard conglomerates of enamel, dentin, cementum, and pulpal tissue, primarily in the posterior mandible.[4]

Various factors can influence the development of odontomas, including trauma during primary dentition, infection, the persistence of periodontal Malassez remnants, hyperactivity of odontoblasts, as well as hereditary anomalies and genetic mutations.[1,5,6] While the majority of odontomas are asymptomatic, some patients may experience symptoms, such as unerupted or impacted teeth, retained deciduous teeth, swelling, or signs of infection.[2] The occurrence of compound-complex odontoma, a distinct subtype characterized by both compound and complex radiological and histological features, is rare in scientific literature.[6,7] Although odontomas are easily diagnosed radiographically, their size, clinical behavior, and histological differences require careful examination.[5,7]

This case report offers an analysis of the clinical, radiological, and histopathological characteristics of a 15-year-old male patient who presented with an extensive compound-complex odontoma in the anterior maxilla. It underscores the importance of routine dental checkups, the evolving classification of odontomas as hamartomatous odontogenic malformations, and the challenges in diagnosing them. The multidisciplinary diagnosis, treatment, and favorable patient outcome make this case clinically significant, further highlighting the diagnostic challenges associated with this unique odontoma subtype.

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Case Description

A 15-year-old male patient presented at the dental clinics of the Department of Oral and Maxillofacial Surgery, College of Dentistry, Jazan University, with the chief complaint of edema in the anterior maxillary region and the absence of the maxillary left central

incisor. The patient's medical and family history did not reveal any significant medical issues or relevant information, and there was no documentation of any traumatic events or infections in the orofacial region.

Clinical examination revealed that the maxillary left central incisor exhibited delayed eruption, characterized by an asymptomatic, expansive swelling extending from the anterior maxillary ridge to the vestibular area, covered by normal oral mucosa. A provisional diagnosis of an odontoma or a dentigerous cyst was made due to the possible impaction of the maxillary left central incisor. A panoramic radiograph exhibited a substantial radiopaque mass in the midline of the maxilla, surrounded by a radiolucent rim. The mass exhibited structures resembling teeth. The maxillary left central incisor was found to be horizontally impacted, and the maxillary right central incisor and both lateral incisors were deviated distally due to the expansile behavior of the lesion (Fig. 1). A cone-beam computed tomography (CBCT) scan revealed a dense mass involving the midline maxilla, resulting in expansion and slight thinning of the facial bone (Fig. 2).

A differential diagnosis considered odontogenic ghost cell tumor and ameloblastic fibro-odontoma, but the clinical and radiographic presentation ultimately led to the diagnosis of an odontoma.

A multidisciplinary treatment plan was devised involving oral surgery, oral pathology, orthodontic treatment, and the potential placement of a single implant. A comprehensive surgical procedure was conducted, including the complete removal of the lesion and subsequent curettage. The excised specimen was fixed and subsequently submitted for histopathological analysis. To ensure adequate bone healing and support for the soft tissue profile, 2cc allograft particulate-derived matrix (CenoBone®) and a graft collagen matrix membrane (CenoMembrane®) was placed (Fig. 3). A postoperative panoramic radiograph confirmed the complete excision of the odontoma (Fig. 4).

Histopathological examination revealed sections of fish-scale-like enamel matrix, tubular dentin, and primitive dental papilla. Additionally, cementum-like material was observed, and some sections revealed a haphazard mixture of dense dentinoid-cemento-osseous material. No ghost cells or cyst lining were observed. Based on clinical, radiographic, and histopathological characteristics, the diagnosis of a compound-complex odontoma of the anterior maxilla was made. A visual representation is provided for reference and analysis, denoted as shown (Fig. 5).

The radiograph obtained after the surgical procedure confirmed the successful removal of the odontoma, and the subsequent wound healing was deemed satisfactory, with no observed complications. After a 1-year follow-up, no significant recurrence was observed, allowing for the continuation of orthodontic treatment.

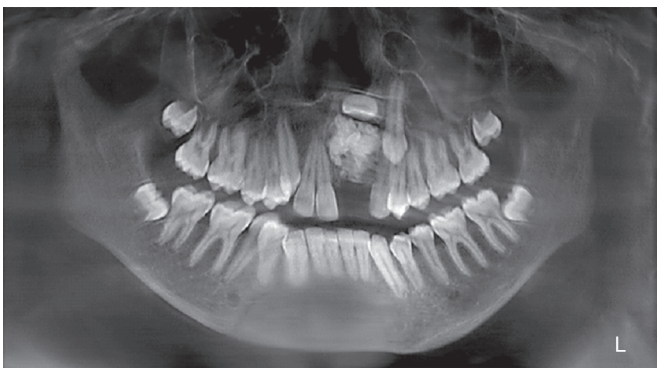


Fig. 1: Panoramic radiograph demonstrating large radiopaque mass with numerous tooth-like structures surrounded by a thin radiolucent rim and associated with impacted left maxillary central incisor

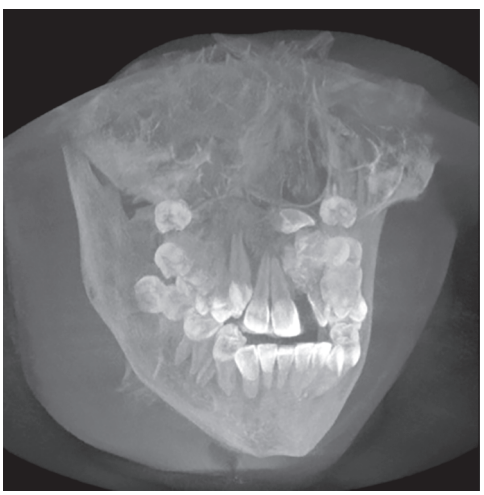


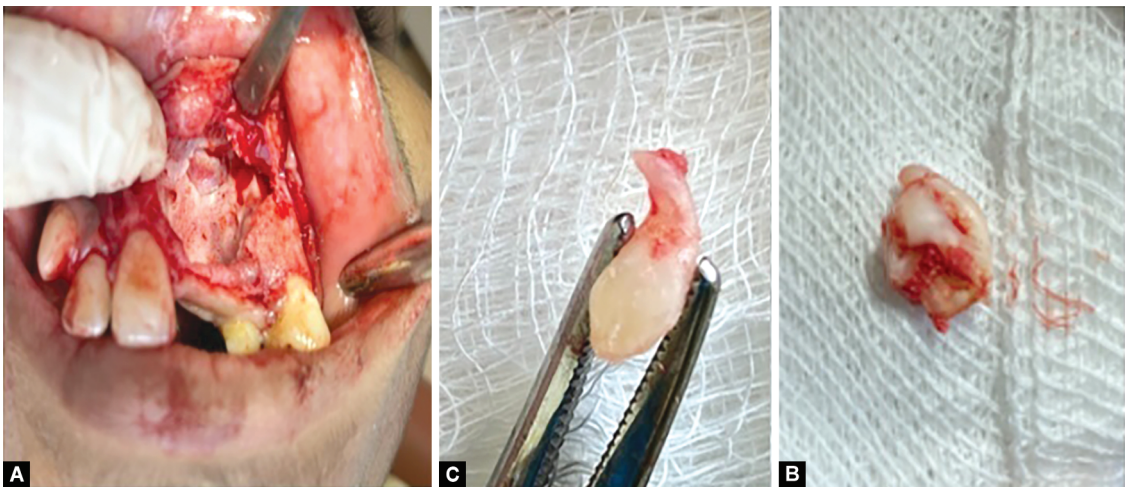
Fig. 2: Axial section of CBCT

Discussion

A substantial body of literature affirms that odontomas are the most common oral odontogenic tumors.[4,5,8] Previously classified as benign odontogenic neoplasms, they are now recognized as hamartomatous odontogenic malformations. The WHO conducted a comprehensive review in 2022, particularly focusing on odontogenic hamartomatous lesions, specifically ameloblastic fibro-odontoma (AFO) and ameloblastic fibrodentinoma (AFD). In both the 2017 and 2022 classifications, AFO and AFD were omitted due to their presumed association with developing odontomas. Odontomas are genetically characterized by the absence of BRAF p. V600E mutations, commonly observed in ameloblastic fibro-odontomas (AFO) and AFD, similar to AF.[9] The ongoing debate regarding the classification of AFO and AFD as odontomas necessitates further investigation through molecular and genetic analyses.

Benign tumors of this nature are characterized by their asymptomatic nature and typically do not manifest noticeable symptoms. However, they have the potential to disrupt the normal process of tooth eruption, leading to delayed emergence of primary or permanent dentition or the persistence of primary dentition.[10–12] Odontomas are typically detected in the majority of patients through a routine radiographic examination.[13] However, the current case report presents a particularly expansive and rare clinical scenario, detailing a unique odontoma subtype exhibiting radiographic and histological characteristics of both compound and complex odontomas concurrently. The author conducted a review of bibliographic data on the PubMed platform, identifying only six studies documenting compound-complex odontomas. These included five case reports and one retrospective analysis.[4,6,7,14,15]

Compound odontomas commonly favor the anterior region of the jaw, particularly the maxilla, while complex odontomas are predominantly observed in the posterior alveolus.[16] The current case identifies the occurrence of the compound-complex type in the anterior maxilla, while a recently published case report documented this distinctive subtype in the mandibular posterior



Figs 3A to C: (A) Surgical excision and curettage of the lesion; (B and C) Gross examination of a compound odontoma reveals multiple small malformed tooth-like structures

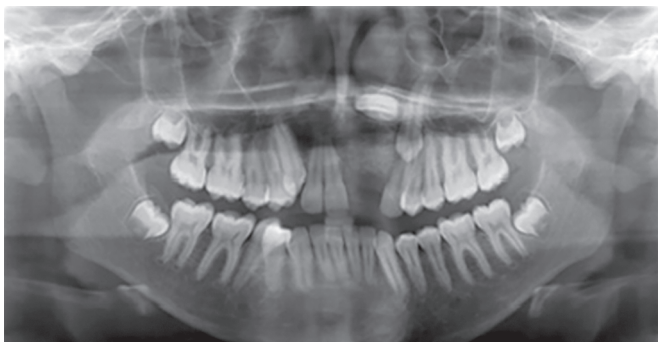


Fig. 4: Postoperative panoramic radiograph showed complete excision of the odontoma

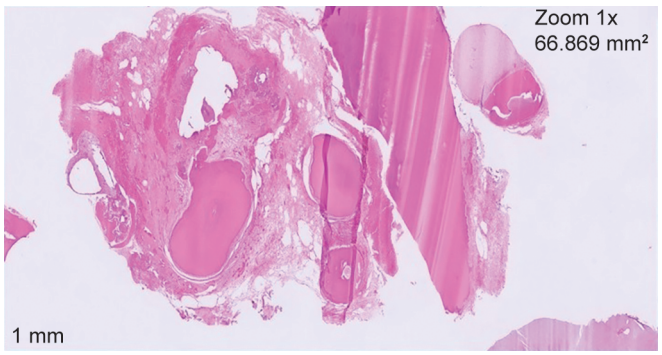


Fig. 5: The microscopic examination revealed sections of fish-scale like enamel matrix, tubular dentin, and primitive dental papilla. Cementum-like material was seen as well. Few sections reveal haphazard mixture of dentinoid-cemento-osseous material

region, highlighting the potential for these lesions to develop in different regions of the oral cavity.[6,14] The age of the patient in the current case report, 15 years, aligns with the usual age range for odontoma occurrence in adolescents, as supported by existing literature.[1,10,17,18] In contrast, the case presented by Khalifa et al.[15] pertains to a 24-year-old male patient, emphasizing the possibility of odontomas manifesting in individuals beyond adolescence.

On radiographs, odontomas typically exhibit a radiolucent halo surrounding a radiopacity encircled by a thin sclerotic line. Compound odontomas display a diffuse radiopaque appearance, composed of numerous radiopacities indicative of denticles. Complex odontomas exhibit a diffuse radiographic appearance with a single irregular mass or a collection of disorganized masses. In the present case, radiographic features show both tooth-like and dense radiopaque structures. It is crucial to consider other differential diagnoses, such as ghost cell tumor and AFO, odontoameloblastoma, especially when expansile behavior and deviation of surrounding dentition are evident, as in our current case, consistent with those typically associated with odontomas.[2,14,15,19] This underscores the significance of implementing routine radiographic screening for odontomas.

Histopathology images revealed a matrix of enamel similar to fish scales, dentin tubules, primitive dental papilla, and a cementum-like substance. Some sections showed a haphazard mixture of dense dentinoid-cemento-osseous material. Complex odontomas may contain ghost cells.[19] While this case's histopathology indicates a compound-complex odontoma, a recent publication highlighted a case of a compound-complex odontoma with an exceptionally high number of denticles (526), demonstrating the wide spectrum of histological variations within compound-complex odontomas.[6]

The initial diagnosis was odontoma, but other potential diagnoses considered included odontogenic ghost cell tumor and AFO. This cautious approach is driven by the need to distinguish odontomas from other dental pathologies that share similar clinical and radiological characteristics.[20] The standard treatment typically involves the surgical removal of the lesion, along with curettage, as was performed in the current case. Enucleation is straightforward due to the encapsulated nature of the tutors.[8] While the likelihood of recurrence is low, a thorough observation over time, particularly in young children, is imperative.[21] Early detection and intervention enable a more conservative approach during the surgical procedure, minimizing the risk of lesion degeneration and preserving the vitality and positioning of the neighboring tooth. Ultimately, this contributes to a favorable prognosis. The present case involved the implementation of a multidisciplinary treatment plan encompassing oral surgery, oral pathology, orthodontic intervention, and the consideration of a solitary dental implant. To promote bone healing and provide support to

soft tissues, a combination of surgical excision, curettage, and the utilization of graft materials was employed. The patient underwent uninterrupted orthodontic treatment following a 1-year follow-up period with no signs of recurrence.

Conclusion

Complex and compound odontomas are two distinct types of odontogenic tumors classified by the WHO. Complex odontomas are characterized by the disorganized aggregation of mineralized dental tissues, including enamel, dentin, cementum, and dental pulp, in an anarchic manner. In contrast, compound odontomas feature agglomerations of diminutive rudimentary structures resembling teeth. The co-occurrence of characteristics from both types in a single odontoma, as observed in the current case, is a rare phenomenon. This case report presents an uncommon combination of features found in complex and compound odontomas in a 15-year-old male patient.

The successful implementation of a multidisciplinary treatment approach resulted in a 1-year follow-up period without any recurrence of the condition. This particular case serves to emphasize the significance of regular dental examinations, enhances our understanding of odontomas as hamartomatous odontogenic malformations, and highlights the complexities involved in diagnosing such lesions. Furthermore, it underscores the paramount importance of routine dental check-ups, where early detection can significantly impact the prognosis and overall treatment outcome. In addition, the author suggests that additional molecular investigations are necessary to facilitate the classification and elucidation of genetic factors.

Clinical Significance

The clinical significance of this case underscores the criticality of routine dental examinations in the timely identification of odontomas. This observation further emphasizes the increasing recognition of odontomas as hamartomatous odontogenic malformations and the difficulties associated with their clinical diagnosis. The present case serves to enhance the existing body of knowledge regarding compound-complex odontomas, thereby prompting additional investigations within the field of oral pathology.

Author's Contribution

Yaser A Alhazmi has written the manuscript, participated in the design, the acquisition of data as well as the revision of the manuscript.

Data Availability Statement

The datasets generated during the current study are not publicly available but are available from the corresponding author on reasonable request.

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