



Compound-Complex Odontoma in a Pediatric Patient-Diagnosis and Treatment: A Case Report

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ARTICLE INFO

Article Type: Case Report

Received: 13 October 2025

Revised: 15 January 2026

Accepted: 8 March 2026

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ABSTRACT

Odontomas are benign odontogenic tumors classified as either complex or compound, depending on the organization of dental tissue. Complex odontomas are typically located in the posterior mandible and anterior maxilla, whereas compound odontomas are more commonly found in the anterior maxilla. It is rare for both types to be present simultaneously. These tumors usually manifest during the first two decades of life and are evenly distributed between genders. Often asymptomatic, they are frequently discovered during routine dental check-ups, especially when over-retained primary teeth or delayed eruption of permanent teeth are noted. Early diagnosis is crucial, as odontomas can interfere with the normal formation and eruption of teeth. Given these lesions' benign nature and characteristic appearance, clinical and radiographic evaluations are generally sufficient for initial diagnosis and treatment. However, histopathological analysis and follow-up are advisable to confirm the diagnosis and ensure appropriate management, particularly for younger patients. This study presents an unusual case of complex and compound odontomas in an 8-year-old boy. The odontoma was located in the anterior mandible, an atypical site. Clinical examination revealed slight intraoral swelling in the anterior mandible, an over-retained right primary mandibular central incisor, and an unerupted right permanent mandibular central incisor. Surgical enucleation of the lesion was performed, and histological examination confirmed the diagnosis. Subsequent clinical and radiological assessments did not indicate any recurrence.

Keywords: Compound odontoma; Complex odontoma; Odontogenic tumor; Impacted tooth; Delayed tooth eruption.

Please cite this Article as:

Shariati M, Nikparto N, Bahrami R. Compound-Complex Odontoma in a Pediatric Patient-Diagnosis and Treatment: A Case Report. J Craniomaxillofac Res 2026; 13(2): 238-245. DOI: [10.18502/jcr.v13i2.22552](https://doi.org/10.18502/jcr.v13i2.22552)



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Introduction

Odontomas are the most common benign tumors of odontogenic origin. They are classified as hamartomas due to their epithelial and ectomesenchymal composition [1]; these tumors consist of morphologically normal cells but demonstrate defective structural organization [2,3]. According to the World Health Organization (WHO), odontomas are divided into two main types: compound and complex [4]. A complex odontoma is characterized by a malformation in which all dental tissues are present, but their arrangement tends to be more disorderly. Although the individual tissues are generally well-formed, they occur randomly within the lesion. On the other hand, a compound odontoma represents a malformation in which all dental tissues are orderly and arranged in a more structured pattern than complex odontomas, forming multiple tooth-like structures, called “odontoids,” within the lesion [3–5].

Complex odontomas are usually found in the posterior mandible and anterior maxilla, while compound odontomas are more commonly seen in the anterior maxilla [6]. The exact cause of these lesions remains unclear. Several possible causes have been suggested, including trauma during the baby teeth period, remaining Malassez from the periodontal ligament, inflammatory processes, odontoblastic hyperactivity, and hereditary anomalies such as Gardner’s syndrome [7,8]. Odontomas can occur at any age, but they are most prevalent during the first two decades of life, with the average age being 14.8 years. There is no significant difference in occurrence between males (59%) and females (41%) [9–12].

Patients with odontomas often do not show symptoms. However, they are sometimes associated with problems in tooth formation and eruption, such as malformation, aplasia, malposition, and retained [5]. Early diagnosis of the lesion is essential to prevent these complications. Diagnosis relies on clinical signs and radiographic findings. Clinical signs may include intra- and extraoral swelling, pain (especially in erupted odontoma), over-retained primary teeth, permanent teeth’ failure to erupt, cortical bone expansion, and tooth displacement [5]. In a panoramic view, an odontoma typically appears as a well-defined radiopaque area within the bone, with a density higher than that of bone and equal to or greater than that of a tooth. It contains areas of varying density surrounded by a radiolucent halo, usually enclosed by a thin sclerotic line, representing the connective tissue capsule of

a normal tooth follicle [13]. While odontomas rarely display both radiological and histological features of compound and complex types simultaneously, when they do, this type is referred to as a compound-complex odontoma in the literature. Treatment consists of complete tumor removal [14]. Because odontomas are encapsulated in a fibrous capsule, they can be carefully separated from the surrounding bone during surgery [8]. The prognosis for patients with odontomas is generally good [14], and recurrence is rare [15]. In this study, we report a rare case of an unerupted compound-complex odontoma in the anterior mandible of an 8-year-old boy with no medical history. The nature of the lesion (compound-complex), the uncommon location of the odontoma, and its impact on neighboring teeth (over-retained right primary mandibular central incisor and an unerupted right permanent mandibular central incisor) make this case particularly noteworthy.

Case Report

An 8-year-old boy with no previous medical history was referred to the Department of Pediatric Dentistry at the Iran University of Medical Sciences, School of Dentistry, for a routine dental check-up. His chief complaints included an abscess on the anterior mandibular without any pain, and an unerupted right permanent mandibular central incisor. There were no significant details in the patient’s family history. Clinical intra-oral examination revealed decayed teeth in his mixed dentition. Also, slight swelling, with no pus discharge, on the labial side of the alveolar gingiva of the right anterior mandibular region, over-retained right primary mandibular central incisor, and unerupted right permanent mandibular central incisor, were observed in clinical examination (Figure 1).

The consistency was hard to firm, and the overlying mucosa was normal without any ulceration. An intraoral panoramic X-ray was recommended, which revealed multiple radio-opaque, tooth-like structures in a narrow radiolucent zone adjacent to an impacted right permanent mandibular central incisor (Figure 2). Cone beam computed tomography (CBCT) scans were suggested for a more accurate diagnosis and comprehensive evaluation (Figure 3). Upon examination, an assemblage of tooth-like structures was identified near the root apex of the right primary mandibular central incisor, located immediately posterior to the buccal plate and buccal to the impacted right central incisor, which caused anterior mandibular bulging. Additionally, the CBCT scans showed that the impacted right permanent mandibular central incisor had a standard

shape and size. Based on these findings, an odontoma was provisionally diagnosed. The surgeon (N.N.) recommended proceeding with surgical excision under infiltration anesthesia. Additionally, the orthodontist (R.B.) suggested extracting the over-retained right primary mandibular central incisor along with enucleation of the odontoma. This approach was based on a CBCT analysis indicating that removing the odontoma and the primary tooth could create space for the impacted tooth to erupt.

The pressure exerted by the lip and tongue post-extraction may help the impacted right permanent mandibular central incisor to erupt closer to its correct position. Based on WHO recommendations and previous studies [3,16–18], it is recommended to wait 5 months for the impacted tooth to erupt. Suppose it does not, consider surgical exposure with or without orthodontic traction. The pediatric dentist (M.Sh.) confirmed their suggestion: three-quarters of the impacted tooth's root had formed and was developing typically; there-

fore, surgery did not affect its development. Therefore, the surgical enucleation of the odontoma commenced with the extraction of the over-retained primary central incisor. To access the lesion, a quadrangular flap was designed with a sulcular incision and two releasing incisions. Following subperiosteal dissection and careful removal of the buccal cortical bone, a surgical window was created (Figure 4). The lesion was then enucleated using a dental extraction elevator, exposing the impacted central incisor. After thorough irrigation and meticulous removal of the odontoma capsule, the surgical site was closed using 4-0 Vicryl sutures. The excised specimen was fixed and subsequently sent for histopathological examination. Based on the histopathological and clinical findings (Figure 5), a diagnosis of compound-complex odontoma was confirmed. Following a five-month follow-up period, the lesion exhibited no signs of recurrence, and the impacted right permanent mandibular central incisor also successfully erupted.

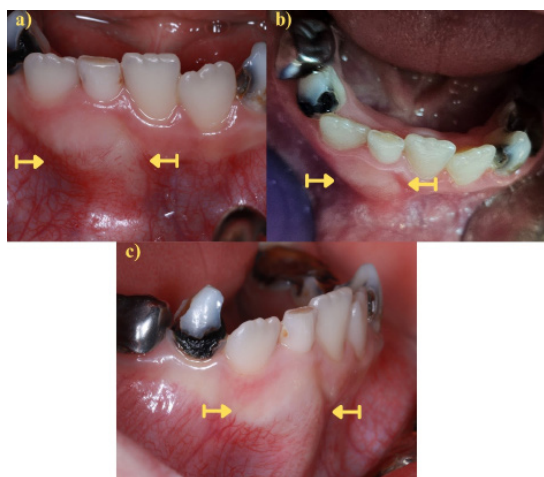


Figure 1. The intra-oral photograph showed slight swelling on the labial side of the alveolar gingiva of the right anterior mandibular region, the over-retained right primary mandibular central incisor, and the unerupted right permanent mandibular central incisor. a) Frontal view, b) Occlusal view, c) Lateral view.



Figure 2. The pre-operative panoramic image displays multiple tooth-like structures and impacted the right permanent mandibular central incisor.

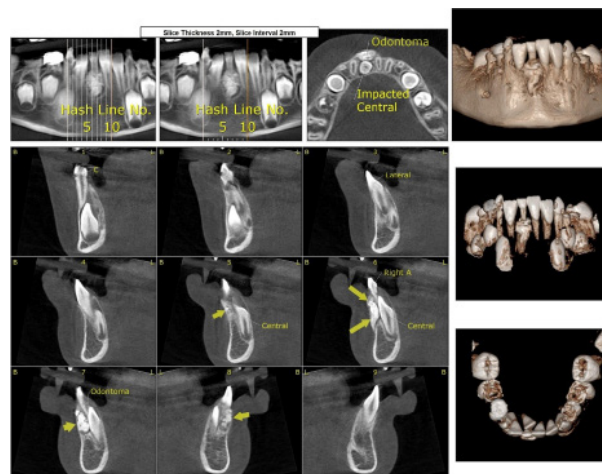


Figure 3. Pre-operative cone beam computed tomography views.

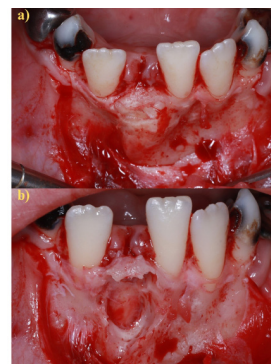


Figure 4. Surgical management of odontoma associated with an unerupted tooth. a) The quadrangular flap reflection and bone removal, along with the extraction of the over-retained primary central incisor; b) The odontoma was carefully enucleated and separated from the surrounding bone.

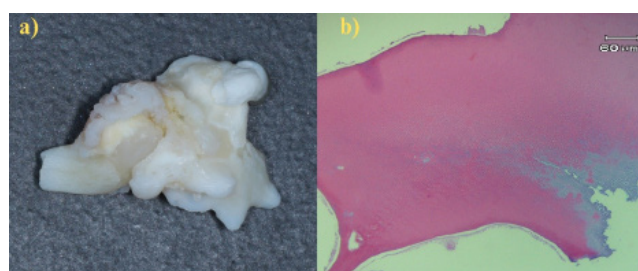


Figure 5. Gross (a) and histopathological (b) evaluation.

Table 1. Review of the literature related to complex–compound odontoma.

Authors	Gender/ Age	Location	Clinical findings	Radiology findings	Histology findings	Treatment	Follow-up
Our case	Male/ 8-year-old	Anterior mandible	Intraoral slight swelling Painless No pus Over-retained primary tooth (81) Unerupted per- manent tooth (41)	Panoramic: Multiple ra- dio-opaque tooth-like structures surrounded by a narrow radiolucent zone. Impaction of 41. CBCT: A well-defined solitary mass with numerous tooth-like structures (near the root apex of 81, posterior to the buccal plate, and buccal to 41). Normal size and shape of 41.	Histological examination confirmed that the lesion was a compound-com- plex odontoma.	Surgical enucleation (Quadrangular flap) under infiltra- tion anesthesia with the extraction of 81.	6 months No recurrence Spontaneously eruption of 41
Khalifa et al. (2022) [19]	Male/ 24-year-old	Posterior mandible	Pain	Panoramic: A large radiopaque lesion surround- ed by a narrow radiolucent zone (an assemblage of multiple odontoids and an anarchic radiopaque). CBCT: An assemblage of teeth-like structures.	Histological examination confirmed that the lesion was a compound-com- plex odontoma.	Surgical enucleation with the extraction of 48.	6 months No recurrence
Marimuthu et al. (2022) [20]	Male/ 7-year-old	Posterior mandible	Diffused and firm swelling Painless no signs of paresthesia No changes in overlying skin Unerupted permanent tooth 46	Panoramic: Conglomerate mass of mixed radio density	Histological examination confirmed that the lesion was a complex- compound odontoma.	1: Aspiration was done and was negative 2: Incisional biopsy of the palpable mass was performed and a provisional diag- nosis of ameloblas- tic fibro-odontoma was arrived at 3: Surgical excision (Mucoperiosteal flap) under local anesthesia with bone removing. 526 denticles were removed.	No recurrence

Authors	Gender/ Age	Location	Clinical findings	Radiology findings	Histology findings	Treatment	Follow-up
Ahuja et al. (2020) [21]	Female/ 13-year-old	Posterior mandibular (second premolar)	No swelling (no facial asym- metry) Painless No pus Unerupted per- manent tooth	Panoramic: Multiple radi- opaque miniatures tooth-like structures located in the superior and distal aspect of impact- ed 35	Histological examination re- vealed two types of odontomas.	Surgical enucleation (Mucoperiosteal flap) under local anesthesia with bone removing.	6 months No recurrence Spontaneously eruption of 35
Girish et al. (2016) [22]	Male/ 16-year-old	Anterior mandible	No swelling (no facial asym- metry) Painless No pus Unerupted per- manent tooth (31, 32)	Panoramic: Ill-defined mixed radio-opaque and radiolucent lesion in the region of 31, 32 of approximately 1 cm × 1.5 cm a thin radiolucent band. (compound odontoma was determined)	Histological examination confirmed that the lesion was a compound-com- plex odontoma.	Surgical excision (Mucoperiosteal flap) under local anesthesia with bone removing.	No recurrence
Abdul et al. (2014) [8]	Female/ 13-year-old	Posterior mandible	No swelling (no facial asym- metry) Painless No pus Over-retained primary tooth (85)	Panoramic: There were multiple small, radio-opaque structures at the apex of the distal root of 85, with a thin radiolucent zone measuring approximately 1.5 × 1.0 cm.	Histological examination confirmed that the lesion was a compound-com- plex odontoma (The connective tissue capsule is similar to that of dental follicle).	Surgical excision with curettage (Mucoperiosteal flap) under local anesthesia with bone removing and primary tooth extraction.	6 months No recurrence
Khanum et al. (2014) [23]	Male/ 13-year-old	Anterior maxilla	Ill-defined swelling No pus Blood or watery discharge Over-retained primary tooth (23,24,25) Unerupted per- manent tooth (43)	Panoramic: A well- defined mixed radi- opaque-radiolucent lesion with thin radiolucent rim extended superiorly up to the floor of the maxillary sinus and the nasal cavity (measuring about 3 × 3 cm)	Histological examination confirmed that the lesion was a compound-com- plex odontoma.	Surgical excision under local anes- thesia.	No recurrence

Abbreviations: CBCT, Cone beam computed tomography; N/A, Not applicable.

Discussion

As shown in Table 1, a limited number of cases of compound-complex odontoma in the anterior mandible have been reported [8,19–23]. Within these cases, four were male and two were female, aged 7 to 16; however, the study by Khalifa et al. included a patient aged 24 years [19]. These lesions are usually asymptomatic and are often found during routine examinations. However, in some cases, patients may report symptoms such as pain [19]. Intraoral assessments, which include counting teeth, checking for over-retained primary teeth, identifying unerupted permanent teeth, and observing any swelling, can prompt further investigation through radiography. Similar to our case, the common symptom observed in the reviewed studies was impact-

ed permanent teeth or over-retained primary teeth. A previous study has established a correlation between delayed tooth eruption and the presence of odontomas; these lesions can obstruct the natural pathway of tooth eruption [24]. During normal tooth development, the completed crown of the anterior permanent teeth is located lingual to the apical third of the root of the primary teeth. The eruptive movement of the permanent tooth occurs in a labial and incisal direction. This movement initially causes resorption of the root of the primary teeth on the lingual side of the apical third, followed by resorption on the labial side of the apical third—subsequently, the permanent tooth positions itself beneath the root of the primary tooth. Over time, resorption progresses horizontally in an incisal direction until the primary tooth is shed and the permanent

tooth erupts in the oral cavity. So, the lack of eruption of permanent teeth can prevent the physiologic root resorption of primary teeth, leading to over-retained primary teeth [25]. In our case (Figure 3, Slice 6), we observed that the unerupted right permanent mandibular central incisor only caused resorption on the lingual side of the root of the primary tooth. The report did not mention the significant resorption on the labial side, which resulted in the delayed shedding of the primary right mandibular central incisor. Based on the patient's report, poor oral hygiene, and decayed teeth, the initial diagnosis was a dental abscess. However, clinical findings including the hard consistency, fixed relative to the mandible, lack of pus discharge, absence of a fistula and the normal overlying mucosa, ruled out this possibility.

Upon reviewing both panoramic and intraoral findings, we proposed several alternative diagnoses. These included odontogenic hamartomas and tumors such as periapical cemento-osseous dysplasia, compound and complex odontoma. Periapical cemento-osseous dysplasia typically presents with specific characteristics, such as being more common in females than males, predominantly affecting the mandible over the maxilla, occurring more frequently in the anterior region than the posterior, typically manifesting in patients over 30 years old, and progressing through stages where the lesion changes in appearance from radiolucent to mixed and finally to completely radiopaque [26]. Additionally, we considered compound odontoma as a potential diagnosis. This condition is characterized by a radiopaque mass of multiple small calcified structures resembling normal teeth, surrounded by a narrow radiolucent rim [27].

Compound odontoma is more prevalent in children and adolescents, with a higher occurrence in the maxilla compared to the mandible and a tendency to manifest in the anterior region over the posterior region. We also suggested complex odontoma as a possible diagnosis. This type of odontoma is characterized by an amorphous and disorganized calcified mass that does not resemble normal teeth, encircled by a narrow radiolucent rim [27]. Complex odontomas are more common in females than males, more frequently found in the mandible than the maxilla, and tend to occur in patients under 30 [27]. Further examination using CBCT (Figure 3, Slice 6) revealed a collection of compound-complex odontoma, along with the impaction of the right permanent mandibular central incisor [28]. When dealing with odontomas, it's important to have a comprehensive understanding and accurate as-

essment of X-ray records for a precise diagnosis. Although X-rays can provide valuable insight, it is often necessary to conduct histological analysis to confirm the clinical and radiographic diagnosis [16,29,30]. The preferred treatment for compound-complex odontomas usually involves a conservative surgical approach to preserve dental structures and promote optimal tissue recovery. This method has high success rates with minimal recurrence, allowing for bone regeneration after the treatment. Literature suggests that when managing impacted teeth, the focus should be on facilitating spontaneous eruption whenever possible. In our case, the impacted permanent tooth successfully erupted within five months, possibly due to opening the eruption pathway after removing the interferences and acceleration of the remodeling of alveolar bone, which plays a crucial role in tooth movement, during the healing process by stimulating the maturation and proliferation of the main bone cells (osteoclasts and osteoblasts) [31]. Clinicians can effectively address odontomas and impacted teeth by combining careful analysis of X-ray findings with histological confirmation and appropriate treatment strategies, leading to positive patient outcomes.

Conclusion

Odontomas can significantly affect tooth formation and eruption, potentially leading to issues such as devitalization, malformation, aplasia, malposition, and embedding. In the present case, a compound-complex odontoma was found in the anterior mandible, associated with the over-retained right primary mandibular central incisor and the unerupted right permanent mandibular central incisor. This case underscores the importance of early diagnosis in preventing the adverse effects of odontomas. Therefore, it's crucial to use radiography as a regular tool in dental clinical examinations, especially during routine dental check-ups for children. By using radiographic imaging during dental assessments, practitioners can detect odontomas early, allowing for timely intervention and management to reduce potential complications and ensure optimal oral health outcomes.

Conflict of Interest

There is no conflict of interest to declare.

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