

CASE REPORT

Complex–compound Odontome with 526 Denticles: A Unique Case Report

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ABSTRACT

Aim: To report a unique case presentation of a complex–compound odontome with 526 denticles.

Background: Odontoma is a hamartoma of the jaws that has both epithelial and mesenchymal components differentiating to form enamel and dentin. It is of compound and complex types. Rarely, the features of both the types are present together in what is called the compound–complex type of odontoma.

Case description: The case report discussed here is that of a 7-year-old boy who presented with a compound–complex odontoma in the right posterior mandibular region.

Conclusion: Timely diagnosis and prompt surgical treatment aid in preventing complications and bony expansion. Thus, proper histopathological examination is essential for the confirmation of odontoma. Recurrence of odontoma is rare and usually has a favorable prognosis if diagnosed early.

Clinical significance: The odontome contained 526 denticles, the maximum reported in the literature so far, making this a case of extreme clinical significance.

Keywords: Complex, Compound, Denticles, Odontome.

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BACKGROUND

The term “odontoma” was first coined by Paul Broca in 1867. Initially, odontomas were classified as a true neoplasm of epithelial and mesenchymal origin¹ and the most common of all odontogenic tumors of the jaws. However, on further research, odontomas were proved to be hamartomatous malformations that occur due to defective structural organization of the epithelial and mesenchymal cell components exhibiting differentiation into functional ameloblasts and odontoblasts.² Based on the degree of differentiation, odontomas are classified as follows: (1) compound odontoma and (2) complex odontoma.^{1,3–5}

Compound odontoma appears radiographically as distinct, multiple, irregular, radiopaque, tooth-like denticles or odontoids in the center of a radiolucent lesion. Complex odontoma has enamel and dentin laid out in an irregular pattern, without any resemblance to a proper tooth structure, surrounded by a thin radiolucent line of demarcation.^{4–6} There exists a rare type of odontoma that exhibits features of both complex and compound types and is called the complex–compound odontoma.⁷ The incidence of compound odontoma ranges between 9 and 37% and complex odontoma ranges between 5 and 30%.⁸ The site of occurrence also varies between complex and compound odontoma. Compound odontoma has a predilection for the maxillary incisor-cuspid region. Complex odontoma has been more commonly reported in the mandibular molar-premolar region. Sex predilection of odontoma has been considered controversial and ruled out by many authors.⁶

Odontoma can be associated with syndromes like Gardner’s syndrome, Hermann’s syndrome, and basal cell nevoid syndrome.^{9–11} This proves the genetic predisposition associated with odontoma. An odontoma is frequently associated with unerupted teeth or in the place of a missing tooth. It is usually diagnosed in the second and third decades of life, and rarely during

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the first decade itself.¹² Interestingly, it has also been reported that both types of odontomas have a predilection for the right side of the jaw rather than the left.^{8,12} Odontoma is usually asymptomatic and is diagnosed incidentally during a radiographic examination. However, they may cause pain, swelling, and displacement of adjacent teeth in some patients.^{13,14} Occasionally, an odontoma may also erupt into the oral cavity.^{11,15} Radiographic examination alone cannot confirm an odontoma. Histopathological examination must be done to give a confirmatory diagnosis of odontoma. The conventional treatment of an odontoma involves complete removal along with any associated tissue.

In this case report, we discuss a rare case of a compound–complex odontoma in the right posterior mandibular region of

a 7-year-old boy. The presenting features, diagnosis, histopathology, and treatment method have been elaborated and compared with existing literature evidence.

CASE DESCRIPTION

A 7-year-old boy was referred to the department of oral and maxillofacial surgery with a complaint of swelling and a hard mass in the right posterior mandibular region for a duration of 6 months. The patient was asymptomatic. No previous dental treatments including extraction, medical history, or familial history were observed. Extraoral examination showed a diffused, firm, painless swelling in the right mandibular posterior region with no signs of paresthesia or changes in overlying skin. On intraoral examination, the swelling extended from the distal aspect of 84 to the retromolar trigone obliterating the buccal vestibule (Fig. 1A). The second deciduous and first permanent molar were missing. Considering the missing teeth and nature of the swelling, clinical examination differential diagnoses were dentigerous cyst, odontogenic keratocyst, or benign tumor. Radiographic investigation and orthopantomogram of jaws revealed a conglomerate mass of mixed radio density (Fig. 1B). Aspiration was done and was negative. Herein, the differential diagnoses were odontome, ameloblastic fibroodontome, or calcifying epithelial odontogenic tumor. An incisional biopsy of the palpable mass was performed and a provisional diagnosis of ameloblastic fibro-odontoma was arrived at.

Surgical excision of the tumor was performed under general anesthesia after informed consent was taken from the parents. A full-thickness mucoperiosteal flap was elevated, followed by removal of overlying bone and the excision of the mass *in toto*. The gross specimen was observed to have multiple tooth-like structures (Figs 2A and B). On subjecting to histopathological examination, the final diagnosis of a complex-compound odontoma was confirmed (Fig. 2C). A total of 526 denticles were removed from the excised tissue (Fig. 2D). The healing of the surgical site was uneventful and satisfactory (Fig. 3). There was no recurrence of the tumor during the postoperative follow-up period.

DISCUSSION

Odontoma is basically classified into compound and complex types. Compound odontoma is further classified into three

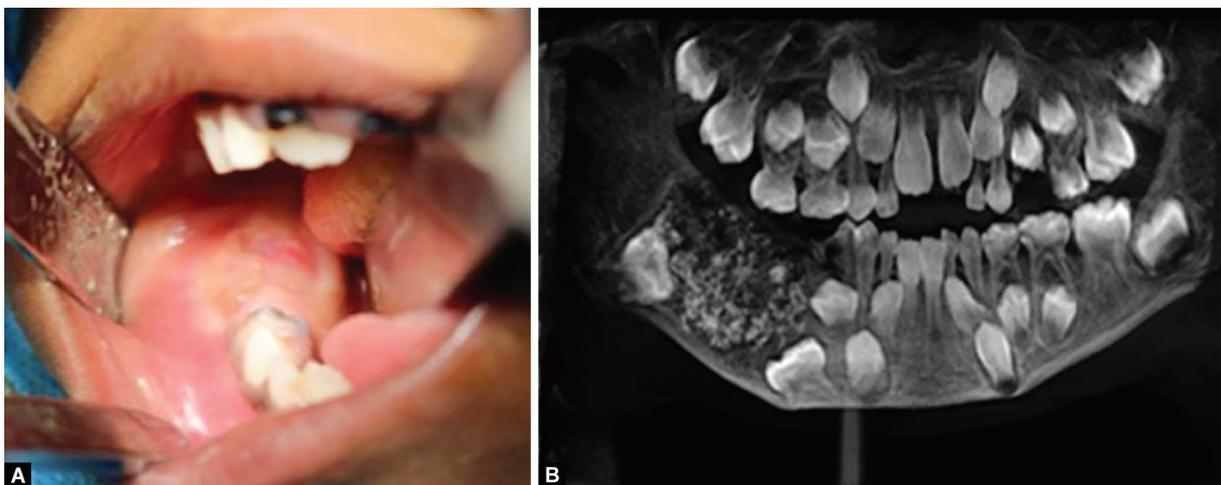
types: denticular type—has two or more separate denticles each representing a tooth; particulate type—has two or more separate masses of particles with tissues arranged irregularly; dentoparticulate type—denticles and particles present side by side. Compound-complex type is a rare entity, showing features of both types as in the current case scenario.

Compound odontomas generally have a predilection for the anterior portion of the jaw, particularly of the maxilla. Complex odontomas are mostly restricted to the posterior alveolus. In the current case, the compound-complex type was found to have occurred in the right posterior mandibular region. Odontomas have also been reported to occur in ectopic regions such as the maxillary antrum and the ramus of mandible.¹⁶ Odontomes usually occur as a mass consisting of denticles, with mostly a minimum of two or more denticles. Most studies have reported odontomes with an average of 8–10 denticles.² The highest numbers recorded in literature are the presence of 36 denticles in a massive compound odontoma of a 9-year-old child,¹² 62 denticles in the maxilla of a 19-year-old female patient,¹⁷ and 177 denticles in the mandible of a 17-year-old female patient.¹⁸ This case report is of high clinical importance as there were 526 denticles in a single odontoma of the complex-compound type in a pediatric patient. This number is incomparably one of the highest ever reported in literature evidence, making the case a rare entity and of extreme clinical significance. Further research must be initiated to understand the pathological process behind the variation in the number of denticles and the occurrence of denticles in such enormous quantities.

Clinical Significance

Why this paper is important to pediatric dentists:

- The incidence of odontome is common in younger age-group who are first seen by a pediatric dentist.
- The ability to diagnose and refer to a specialist maxillofacial surgeon appropriately with reassurance to patient is essential.
- As the primary caregiver to the pediatric patient, the pedodontist should actively participate in the treatment planning including the possibility to preserve any unerupted impacted teeth during the tumor excision.



Figs 1A and B: (A) Intraoral clinical appearance showing the diffuse swelling, missing teeth, and obliteration of buccal vestibule; (B) Cone-beam computed tomography revealing the lesion with mixed radiolucency and calcifications



Figs 2A to D: (A) Excised tumor; (B) Specimen grossing; (C) Histopathology showing mixed tooth-like structures formed from epithelial and mesenchymal components; (D) The isolated 526 denticles



Fig. 3: Postoperative cone-beam computed tomography showing complete removal of tumor

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