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Unusual case of massive, erupted complex odontome associated with congenitally missing permanent molars

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ABSTRACT

The term 'Odontoma' has been used as a descriptor for any tumor of odontogenic origin. However, odontoma has been known as mixed odontogenic tumor composed of epithelial and ectomesenchymal components. Odontomas are non-aggressive, hamartomatous developmental malformation rather than a true neoplasm and usually they are asymptomatic and most common in mandible in posterior region. Here, we report a case of unusually large, erupted complex odontoma associated with pain and swelling, present at a rare site of maxillary posterior region demonstrating missing permanent 2nd and 3rd molars.

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1. Introduction

The World Health Organization (WHO) defined a complex odontoma as a malformation in which all the dental tissues are represented in which individual tissues being mainly well formed but occurring in a more or less disorderly pattern.[1] Odontoma is considered as hamartoma/developmental anomaly rather than a true neoplasm.[2] The term 'Odontoma' was coined by Paul Broca in 1867.[2,3]

Odontoma is the most common odontogenic tumor frequency of which varies from 5 to 30%.[1] They are usually associated with unerupted, impacted or retained deciduous teeth.[3,4] WHO (2005) defines odontoma being of two types: complex and compound.[4] 83.9% of cases occur before the age of 30 with peak incidence in second decade of life. The male: female ratio varies between 1.5:1 and 1.6:1.[5] Complex odontomas have predilection for posterior jaws (59%), anterior maxilla (34%) and lastly premolar area (7%). The size of the complex odontoma varies from 3 to 4 cm.[1] They are usually

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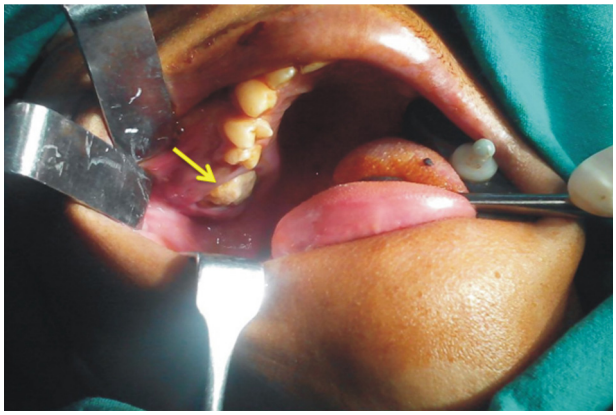


Fig. 1 – Large brownish tooth like conglomerate present in right maxillary posterior teeth.

small; however occasionally grow bigger resulting in bony expansion.[2]

The purpose of the article is to add to the report of extremely rare case of large, massive erupted odontoma present unilaterally in posterior right maxilla which is associated with congenitally missing permanent maxillary 2nd and 3rd molars.

2. Case report

A 16-year-old male patient reported with pain, swelling and tooth like mass in upper right posterior region since 10 days. Pain was spontaneous in onset, dull, intermittent, non-radiating and aggravated by chewing, lasting for few minutes. Patient's past medical and dental history was non-significant.

Intra-oral examination revealed an irregular, tooth like mass associated with right maxillary posterior region. In the region of maxillary molars, we could reveal an irregular shaped brown mass approximately of dimension 3.8 cm × 3.5 cm just distal to 2nd premolar (Fig. 1). On palpation, the mass was bony hard, with slight expansion of buccal and lingual cortical plates. Panoramic radiograph showed irregular but well defined radiopaque mass surrounded by radiolucent capsule except toward the occlusal surface where it was exposed to oral cavity, in right 1st maxillary molar region. Radiographic examination fails to reveal presence of tooth bud for permanent maxillary right second and third molars. The radio-opaque mass was highly radiodense, large and irregular. OPG also revealed impacted maxillary first permanent molar, distally rotated right maxillary second premolar with root being mesially inclined (Fig. 2).

Clinical and radiographic examination led to the diagnosis of complex odontoma. Clinical differential diagnosis included cementoblasoma, osteoblastoma, central ossifying fibroma, ameloblastic fibro-odontoma, etc.

Routine hematological investigations were performed and found to be within normal limits. Lesion was completely excised under GA. Macroscopically; the specimen was 3.8 cm × 3.5 cm in dimension, lobular, irregular, yellowish white, hard showing conglomeratic solid lesion (Fig. 3). The specimen did not bear resemblance to tooth structure and hard mass was covered with soft, yellowish white and thin fibrous capsule. Cut surface showed striated appearance (Fig. 4). Radiograph of surgical specimen revealed highly radiodense, large and irregular mass with radiating radiopaque stria at periphery (Fig. 5). Decalcified and H&E stained section revealed a thin fibrovascular capsule surrounding the admixed tissue was evident. It also showed irregular and disordered mixture of enamel matrix, dentin, small spaces with pulp tissue and cementum like structures. Empty clefts due to decalcification resulted into loss of mature enamel (Fig. 6). Higher magnification revealed that the major bulk was composed of dentinal tubules. Ground section also revealed irregularly arranged enamel, dentin and cellular cementum (Fig. 7).

All these features led to the confirmatory diagnosis of complex composite odontome.

3. Discussion

The term 'Odontoma' by definition alone means any tumor of odontogenic origin. Odontoma is considered to be the hamartomas of aborted tooth development and seems to result from budding of extra-odontogenic epithelial cells from

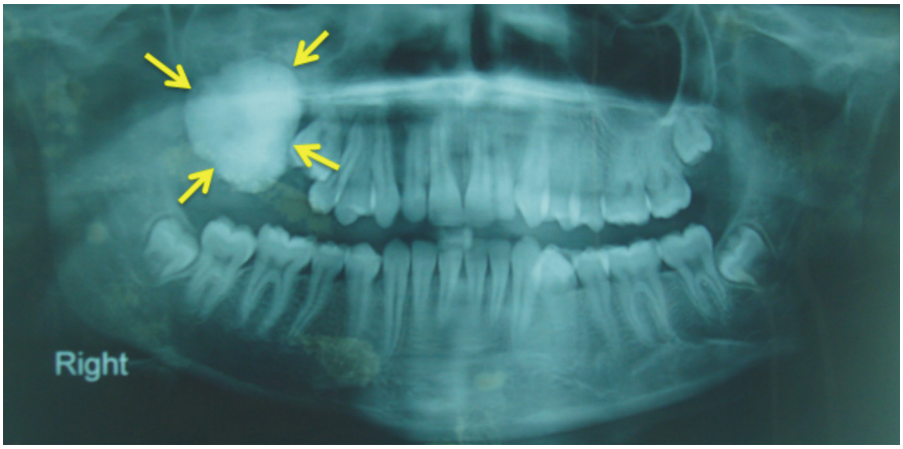


Fig. 2 – Panoramic radiograph showing massive radio-opaque mass present in right maxillary posterior region.

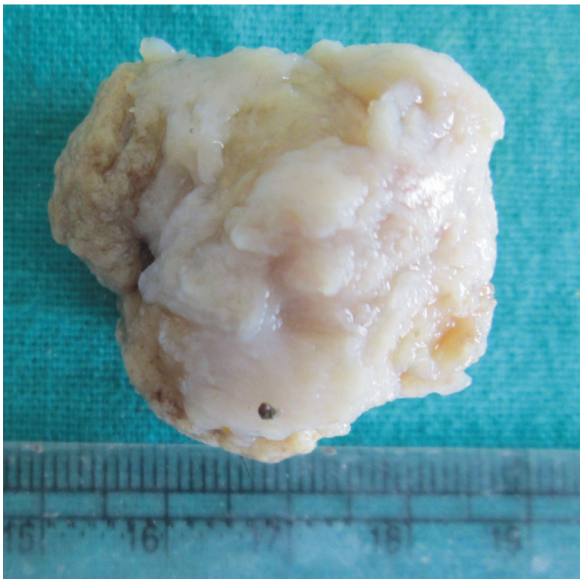


Fig. 3 – Excised specimen of odontoma.

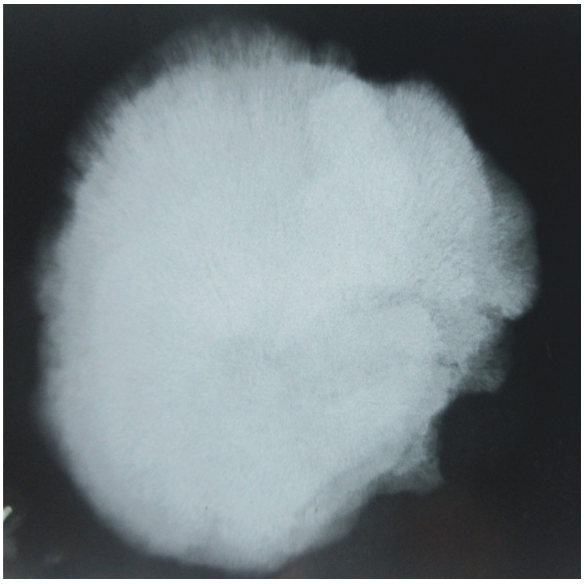


Fig. 5 – Radiograph of excised specimen demonstrating highly radio dense mass.

the dental lamina resulting from the growth of completely differentiated epithelial and mesenchymal cells that give rise to ameloblast and odontoblast. They accounts for 22% of the odontogenic tumors,[4,6] 9–37% odontomas are complex.[7] Odontomas have a slight predilection for occurrence in males (59%) as compared to females (41%). Philipsen et al. suggested that 83.9% of the cases occur before the age of 30 with peak incidence in second decade of life.[5,8] Present case shows similar age of occurrence in a male patient.

The majority of complex odontomas are found in the posterior mandible, followed by the anterior maxilla, as reported by Philpsen et al. (1997).[8] Clinical symptoms are uncommon however affected patients may show failure of eruption of tooth or teeth. Additionally large odontomas are associated with bony expansion and spontaneous eruption of it leads to pain and inflammation of adjacent soft tissues. Quite commonly, complex odontome occur on right side associated with pain and swelling.[9] Our case too showed similar clinical presentation.

Our case of complex odontome presented a rare feature such that it was associated with congenitally missing permanent molars present at a rare site of posterior maxilla. To our knowledge, congenitally missing deciduous lateral incisors of mandible followed by deciduous lateral incisors of maxilla associated with odontome have been commonly reported by Yanagide and Mori (1990).[8] However congenitally missing molars associated with odontoma is extremely rare

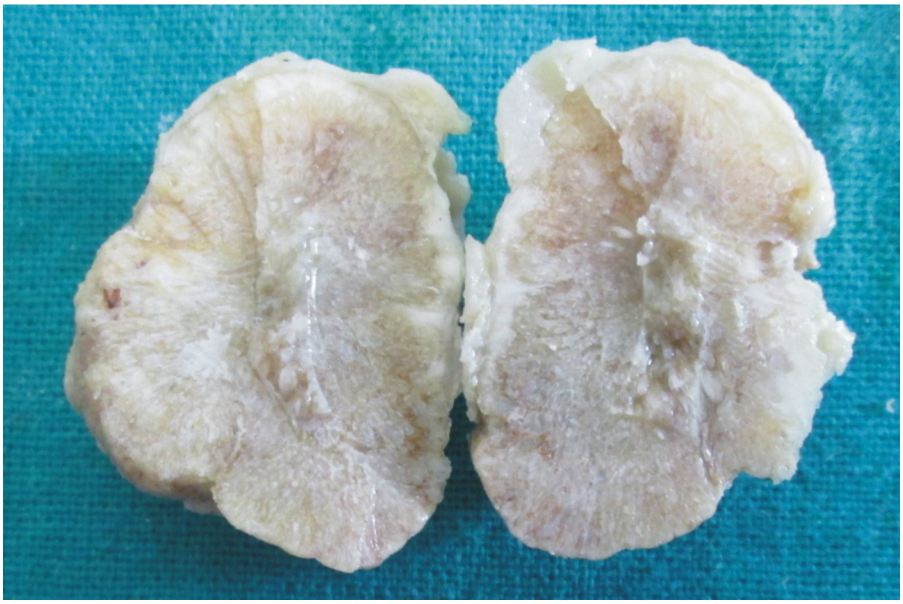


Fig. 4 – Cut surface of excised specimen.

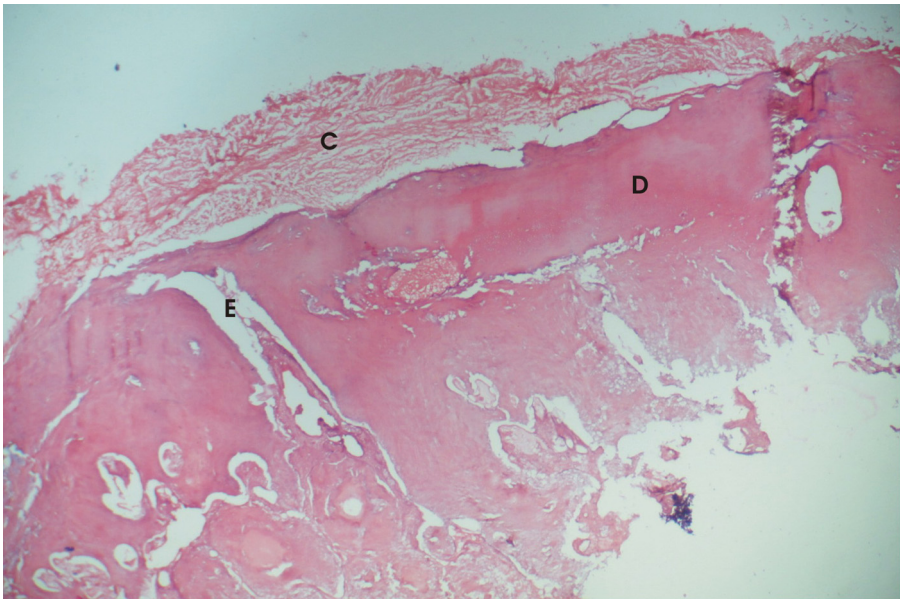


Fig. 6 – Decalcified and H&E stained section demonstrating fibrovascular connective tissue capsule (C) and irregularly arranged enamel space (E) and dentin (D) (40×).

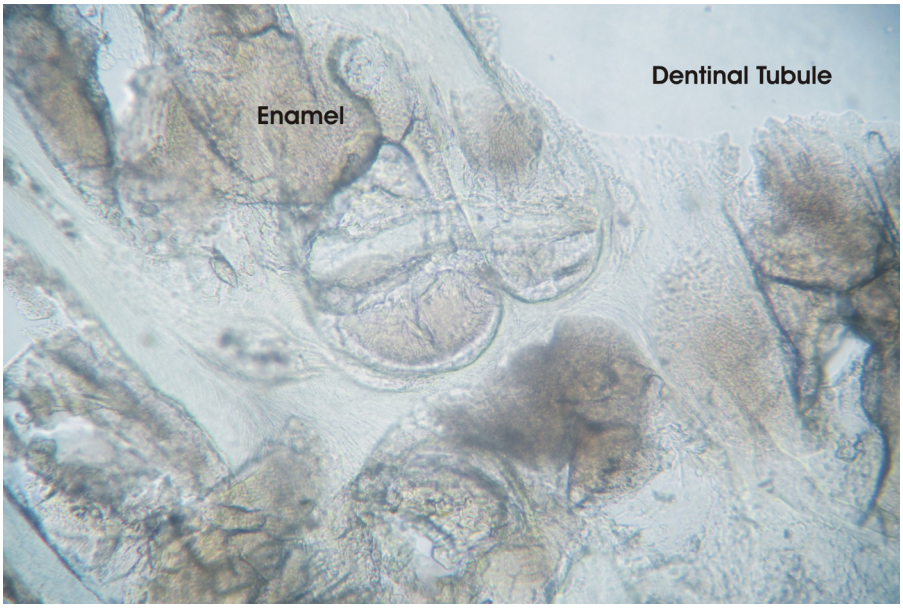


Fig. 7 – Ground section showing enamel and dentin (40×).

which was found in our case. Only 7.2% cases of odontoma are associated with agenesis of permanent teeth.[6]

The first case of an erupted odontoma was described in 1980 by Rumel et al.[5,9,10] To the best of our knowledge since then very few cases have been reported. A literature review by Litonjua et al. (2003) recorded only 14 cases in English literature from 1980 to 2003.[10]

Rarely odontoma may form peripheral or soft tissue lesion in which they arise outside alveolar bone and may erupt.[5] Such a rare erupted odontome was present in our case. Potential age in case of erupted odontoma has been reported to be 25–35 years[9] which was matching in our case too.

The etiology of odontoma has been attributed to local trauma, infection, family history and genetic mutation and hereditary anomalies like Gardner's syndrome, Hermann's syndrome,[1,2] odontoblastic hyperactivity of mature ameloblasts and cell rest of serre.[7,11]

The mechanism of eruption appears to be different from tooth eruption because of lack of periodontal ligament and root in odontoma. Therefore the force required to move the odontome is not linked to contractility of the fibroblasts as in the case of teeth. Although there is no root formation in odontoma, its increasing size may lead to sequestration of the overlying bone and hence occlusal movement or eruption.[9] An

increase in size of the odontoma over time produces a force sufficient to cause bone resorption.[9,11] Another reason for odontoma eruption could be bony remodeling of the jaws. However for this to occur dental follicle is required which indirectly provides both conductance and chemoattraction for osteoclasts necessary for tooth eruption. The reduced enamel epithelium initiates a cascade of intercellular signals by expressing epidermal growth factor β (EGF β) and transforming growth factor. These factors, in turn stimulate the follicular cell to produce colony stimulating factor (CSF) which recruit osteoclasts to the follicle. The reduced enamel also secretes proteases which assist in breakdown of follicle to produce a path of least resistance. The epithelial signaling could the remarkable consistency of eruption times as it is likely that the enamel epithelium is programmed as part of its functional life cycle.[12]

Radiographic appearance depends upon stage of mineralization. The first stage is characterized by radiolucency due to lack of calcification (Weiches odontome-soft odontome).[1] Partial calcification is observed in intermediate stage while in the third stage the lesion usually appears as a dense, radiopaque mass surrounded by thin radiolucent area corresponding to connective tissue capsule.[13] Our case presented radiographic appearance similar to third stage.

Thoma and Goldman classified odontoma into:

Germinated composite odontoma – Two or more, more or less developed teeth fused together.
Compound composite odontoma – Made up of more or less rudimentary teeth.
Complex composite odontoma – Calcified structures which bear no great resemblance to the normal anatomical arrangement of dental tissue.
Dilated odontoma – Crown or root part of tooth shows marked enlargement.
Cystic odontoma – Normally encapsulated by fibrous connective tissue in a cystic wall of cyst.[6]

Further WHO (2005)[4] divided odontoma into complex, compound and ameloblastic fibro-odontoma.

Complex odontoma is described as a calcified dental tissue simply arranged in an irregular mass bearing no morphologic similarity to rudimentary teeth where as compound odontome is composed of all odontogenic tissue in an orderly pattern which result in many teeth like structures but without morphologic resemblance to normal teeth. Ameloblastic fibro-odontoma consists of varying amounts of calcified dental tissues and dental papilla like tissue. It is an immature precursor of complex odontoma.[12]

Furthermore odontoma may also be classified as:

Intraosseous – Occur inside the bone and may erupt into the oral cavity (erupted odontome).
Extraosseous/peripheral odontome – Occurring in the soft tissue covering the tooth bearing portion of the jaws and having a tendency to exfoliate.[12]

Based on radiographic and clinical appearance, we classify present case under intraosseous complex composite odontoma. Histologically the odontoma is not a diagnostic dilemma. It is composed of irregularly arranged dentin, cementum, pulpal tissue and enamel. However, mature enamel is lost during the decalcification process. A thin, fibrous capsule can surround the lesion. Histopathologically our case presented in a similar manner reflecting high degree of differentiation of dental tissue and late stage of morphodifferntiation and maturation of odontogenesis.[5] With the haphazard conglomerates of mature hard dental tissue, we confirmed the diagnosis of complex odontoma in our case.

Differential diagnosis included hypercementosis which is usually attached to a part of the root and separated from peripheral bone by radiolucent periodontal ligament space which surrounds entire root. Condensing osteitis is ruled out as it occurs at the periapex of non-vital tooth and does not have radiolucent rim. Periapical osteosclerosis is ruled out as it is quite regular in shape without radiolucent border.[13] Eruption mesenchymal hamartoma which is almost similar to odontoma except it is not well separated from surrounding connective tissue and does not contain enamaloid.[14] Odontogenic calcified masses are relatively small, opaque, white in color with smooth texture. Histopathology revealed that they content osteodentine, cementum and pulp like component but on epithelial cells or enamaloid. Further mesenchymal multinucleated giant cells and dysplastic dental matrix were observed in connective stroma.[14] Also case need to be differentiated from supplemental and supernumerary teeth. Supplemental tooth is the one that resembles normal tooth both morphologically and histologically and is located adjacent to normal tooth where as supernumerary tooth is a tooth like structure of variable size and shape however represents the normal organization of the tooth structure such as enamel, dentin, pulp and cementum including periodontal ligament.[12]

In our case, histopathological as well as ground section examination confirmed the presence of enamel and the diagnosis.

Present case was treated by conservative surgical enucleation. In our case using an intraoral approach, a gingival mucoperiocteal flap was reflected and an occlusal–buccal window created to expose the mass. As it was surrounded by connective tissue capsule complete removal of the lesion was possible. Patient was advised for regular follow up to 3 months to assess healing and recurrence. Impacted maxillary first permanent first molar was left for observation so that it will erupt by itself. Regular follow up for up to 1 year was advised. If the tooth does not erupt then orthodontic treatment will be planned.

4. Conclusion

Despite the benign nature of odontoma pain, inflammation and infection may be associated with it. Odontoma, very rarely erupt into oral cavity and can be associated with congenitally missing impacted or supernumerary teeth. Although clinical, radiographic and histopathological examination usually leads to good diagnosis, it is the moral duty of surgeon to opt for conservative surgical excision of odontoma and follow-up regularly.

Conflicts of interest

The authors have none to declare.

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