

## COMPOUND ODONTOMA OF MAXILLA IN A 13 YEAR OLD PEDIATRIC PATIENT

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### ABSTRACT

Odontomas are odontogenic benign tumors composed of dental tissue. Most of these lesions are asymptomatic and are often detected on routine radiographs. Morphologically odontomas can be classified as complex, when present as irregular masses containing different types of dental tissues, or as compound if there is superficial anatomic similarity to even rudimentary teeth – the denticles. The pathological conditions in which odontogenic differentiation is fully expressed are the odontomas. We present a case of compound odontome in anterior maxilla in a 13 year old male child with retained maxillary deciduous right lateral incisor.

### INTRODUCTION

The term “odontoma” was introduced by Broca in 1863 to comprise all benign odontogenic tumors. At present, odontomas are classified by the World Health Organization as mixed benign odontogenic tumors because of their origin from epithelial and mesenchymal cells, exhibiting different structures of dental tissue (enamel, dentin, cementum and pulp).

Because of their slow progression, some investigators do not believe in the tumor characteristics of odontomas since they follow mechanisms similar to those observed during odontogenesis, i.e., their growth ceases once the process of calcification is completed. Odontomas are considered to be developmental anomalies resulting from the growth of completely differentiated epithelial and mesenchymal cells that give rise to ameloblasts and odontoblasts. These tumors are basically formed of enamel and dentin but they can also have variables amounts of cement and pulp tissue. During the development of the tumor, enamel and dentin can be deposited in such a way that the resulting structures show an anatomic similarity to

normal teeth, in which case the lesion is classified as a compound odontoma. However, when the dental tissues form a simple irregular mass occurring in a disorderly pattern, it is described as a complex odontoma [1].

Compound odontomas appear more frequently than complex odontomas. These odontogenic tumors can be found anywhere in the dental arches. In general they are asymptomatic, have slow growth, and seldom exceed the size of a tooth, but when large can cause expansion of the cortical bone. Odontomas are considered to be the most common odontogenic tumors of the jaw bones and may appear as numerous miniature or rudimentary teeth, called compound odontomas, or as an amorphous mass of mineralized tissue (complex odontomas). A compound odontoma still has the three separate dental tissues (enamel, dentin and cementum), but may present a lobulated appearance where there is no definitive demarcation of separate tissues between the individual "toothlets" (or denticles). It usually appears in the anterior maxilla. The complex type is unrecognizable as dental



tissues, usually presenting as a radiopaque area with varying densities. It usually appears in the posterior maxilla or in the mandible [2]. In addition to the above forms, the dilated odontoma is an infrequent developmental alteration that appears in any area of the dental arches and can affect deciduous, permanent and supernumerary teeth. Dens invaginatus is a developmental anomaly resulting from invagination of a portion of crown forming within the enamel organ during odontogenesis. The most extreme form of dens invaginatus is known as dilated odontoma. Odontomas are considered as hamartomas/developmental anomaly rather than a true neoplasm. A malformation in which all dental tissues are formed, but occurring in less orderly pattern is complex odontoma. Howards lists odontoma as fourth category of supernumerary teeth [3].

### **Etiology**

The etiology of odontomas is unknown, but it could be due to trauma during primary dentition, as well as to inflammatory and infectious processes, hereditary anomalies (Gardner's syndrome, Hermann's syndrome), odontoblastic hyperactivity, or alteration of the genetic components responsible for controlling dental development. Odontoblastic hyperactivity and alterations in genetic component is responsible for controlling dental development. Persistence of a portion of lamina may be an important factor in the etiology of a compound odontoma [4].

### **Clinical features**

Odontomas may be found at any age however, most are detected in the first two decades of life. There is no gender predilection and most lesions are detected on routine radiographs. Considerable controversy exists over gender distribution. While some studies consider odontomas to be more common in females than in males, others consider these lesions to be similarly distributed between both genders [5]. On the contrary, Iatrous et al. and Yadav et al. [6] found a male predilection. Clinical symptoms are uncommon however, an affected patient may present when a permanent tooth or multiple teeth that fail to erupt. Rarely, an odontoma may erupt into the oral cavity. Occurrence ratio of compound to complex odontomas is 2:1.8 [7].

Odontomas are generally small; however, they may occasionally grow large resulting in bony expansion. They can measure anywhere from a few millimetres to many centimetres in their greatest dimension. The largest found in a human as reported in literature weighed 0.3 kg. They are mainly intraosseous lesions although location in gingival soft tissue has been reported. There are very few reports of odontomas associated with primary teeth in the literature. In general, odontomas occur more often in the permanent dentition. The lesions tend to be located between the roots of erupted teeth or between the deciduous and permanent teeth. Anterior maxilla, followed

by anterior mandible and postero-inferior regions are the most common locations [8].

### **Epidemiology**

Odontomas are thought to be the second most frequent type of odontogenic tumor worldwide (after ameloblastoma), accounting for about 20% of all cases within this relatively uncommon tumor category which shows large geographic variations in incidence. Epidemiologically, odontomas are the most frequent odontogenic tumours, and according to different sources in the literature, it accounts for 22–67% of all maxillary tumours. Males and females are approximately equally affected. An increased prevalence of these tumours can be found in children and adolescents [9].

### **Histological features**

Histologically, odontomas are composed of different dental tissues, including enamel, dentin, cementum and, in some cases, pulp tissue. These dental tissues may appear normal but they seem to have a deficient structural arrangement. Mature enamel is lost during the decalcification process and will not be seen on conventional haematoxylin and eosin stained slides. The compound odontoma recapitulates the organization of a normal tooth, while the complex odontoma appears as a disorganized mass of hard odontogenic tissues. Loose, myxoid connective tissue with odontogenic epithelial rests may be seen in close association with the lesion, and most often represents normal dental follicular tissue [10].

### **Case Report**

A 13-year-old male child reported to Outpatient department, Department of Oral Medicine, Diagnosis & Radiology, Gwalior, Madhya Pradesh, with chief complaint of decayed teeth in upper front jaw region since 3 months. Medical history was non contributory and there were no hereditary disease in antecedents. Extra oral examination was unremarkable. Intra orally, right permanent maxillary lateral incisor was clinically missing and retained maxillary deciduous lateral incisor was present. Right permanent central maxillary incisor was mesially rotated. Intraoral periapical radiograph of retained maxillary deciduous lateral incisor was taken which revealed decayed tooth with small radio opaque structure just above it (Fig 1). An occlusal radiograph showed the presence of a lesion formed by many radiopaque small tooth-like structures constituting in right half of dorsum of hard palate and maxillary permanent right lateral incisor was missing (Fig 2). Considering the clinical and radiologic presentations, provisional diagnosis of compound odontoma was determined.

### **DISCUSSION**

In 1946, Thoma and Goldman formulated a classification for odontomas as germinated composite, compound composite, complex composite, dilated and



cystic odontomes. Eruption of odontomas is infrequent in the literature. Odontomas are benign tumors containing various tissue components of the teeth. Pain and swelling are the most common symptoms when odontomas erupt, followed by malocclusion. Recurrent infection following eruption into the oral cavity has been reported but the patient was asymptomatic & odontome did not erupt in oral cavity in our case. Forty-nine percent of odontoma diagnosed are the result of delayed eruption, 28% the retention of primary teeth, 20% are incidentally found on radiographs, and 3% the swelling of the jaws [10]. Our case is associated with overly retained deciduous teeth. Discovery often occurs due to radiographic investigation for the cause of a non-erupted permanent or retained primary tooth. An impacted tooth is present in more than half of the cases. In the 26 cases of odontomas analyzed by Iatrous et al. only 7% had the impaction of permanent teeth associated [11]. In our case the lesion was found due to a delay on the exfoliation of maxillary right deciduous lateral incisor and permanent tooth bud was congenitally missing. When an occlusal radiograph was taken an irregular radiopaque image with variations in contour and size, composed of multiple radiopacities corresponding to the so-called denticles, could be seen. Compound odontomas are more common in occurrence than complex odontomas. They are usually asymptomatic, and they may be detected by chance on a routine radiograph (panoramic and/or intra-oral X-rays), or when they are large enough to cause a swelling of the jaw. Clinical signs suggestive of an odontoma include a retained deciduous tooth or an impacted tooth. Odontoma is a generally asymptomatic, slowly progressing tumor that may pass unnoticed. The presence of odontomas may cause a series of disorders and sequelae in the patient, such as problems related to their interference with the process of tooth eruption, ectopic eruption, displacement and malformation of adjacent teeth, diastema, anodontia, and growth pressure exerted by the odontoma that may cause pain, devitalization, and tooth and bone resorption [12]. Some studies have reported a correlation between patient age and the type of odontoma involved – compound lesions being apparently more frequent in younger patients, which is in agreement with our case. Most authors agree that these lesions effectively appear more often in the maxilla. In our case the odontoma was also located in the upper jaw, what is in accordance with the previous studies. The reported tendency of odontomas to arise in the region of the incisors and canines is also confirmed in our case. Interestingly, both types of odontoma occurred more frequently on the right side of the jaw than on the left. In our case odontome was also present in right half of maxilla [13].

Ameloblastic odontoma and ameloblastic fibroodontoma bear great resemblance to the common odontoma, particularly on a radiograph, and thus it is suggested that all odontomas should be sent to an oral pathologist for microscopic examination and definitive diagnosis. In our case no syndromes were evident and no

episode of previous trauma was reported by the patient and family. In children, the impacted permanent teeth, depending on the age and on the tooth development, may be left to erupt spontaneously, or they may be guided to occlusion via orthodontic traction. In any case, follow-up is essential following odontoma excision. Besides, proper patient care should include careful clinical and radiographical follow-up. The ameloblastic fibro-odontoma is defined as a tumor with the general features of an ameloblastic fibroma but that also contains enamel and dentin. It is usually encountered in children with an average age of 10 years. It has been suggested that ameloblastic fibro-odontomas should not be considered as true neoplastic odontogenic lesions, but rather as a stage preceding the complex odontoma which would be the final stage in this line of development of hamartomatous lesions [14]. Other authors consider that, despite the fact that some lesions diagnosed as ameloblastic fibro-odontomas can be developing odontomas, all cases of ameloblastic fibro-odontomas should not be considered as hamartomatous in nature since there are rare cases of ameloblastic fibro-odontomas showing true neoplastic behaviour. The majority of the ameloblastic fibro-odontomas are found in the posterior region of the mandible. These lesions seem to be exclusively central or intraosseous tumours. They are also characterized by being painless and slow-growing. Radio graphically, the tumor shows a well-defined unilocular or, rarely, multilocular radiolucent defect that contains a variable amount of calcified material with the radio density of dental hard tissues. The calcified material within the lesion may appear as multiple, small radiopacities or as a solid conglomerate mass. It can be differentiated from the odontoameloblastoma by the fact that it is well circumscribed and usually separates easily from its bony bed. Whereas radio graphically, odontoma presents as a well-defined radiopacity situated in bone but with a density that is greater than bone and equal to or greater than that of a tooth. It contains foci of variable density. It is present with a radiolucent halo, typically surrounded by a thin sclerotic line, surrounding the radiopacity [15]. The treatment of choice of ameloblastic fibro-odontomas is conservative surgical enucleation and prognosis is excellent. However, development of an ameloblastic fibro sarcoma after curettage of an ameloblastic fibro-odontoma has been reported. The ameloblastic fibro sarcoma is considered to be the malignant counterpart of the ameloblastic fibroma and often represents a recurrence of a tumor previously diagnosed as an ameloblastic fibroma or an ameloblastic fibro-odontoma. It is characterized by a malignant transformation of the ectomesenchymal component of the tumor and not the odontogenic epithelium. The average age at time of diagnosis for the ameloblastic fibro sarcoma is 27.5 years, as opposed to 14.8 years for the ameloblastic fibroma and 9 years for the ameloblastic fibro-odontoma. This age difference supports a step-wise progression of a benign to a malignant tumor [16]. Radiographically, the



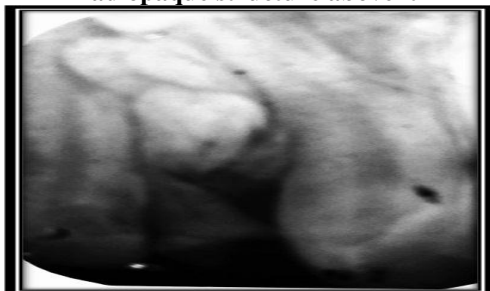
ameloblastic fibro sarcoma shows an ill-defined destructive radiolucent lesion that suggests a malignant process. Pain and swelling associated with rapid clinical growth are common complaints. Patients diagnosed with this condition should be treated by radical surgical excision, since the tumor is locally aggressive and infiltrates adjacent bone and soft tissues. Long-term prognosis is uncertain because of the few reported cases with adequate follow-up. The odontoameloblastoma is an extremely rare odontogenic tumor that contains an ameloblastomatous component together with odontoma-like elements. This lesion appears to occur more often in the mandible of young patients. Radio graphically, this tumor shows a radiolucent, destructive process that contains calcified structures. These have the radio density of tooth structure and may resemble miniature teeth or occur as larger masses of calcified material similar to a complex odontoma. Multiple recurrences of odontoameloblastomas have been reported after local curettage and it appears that this tumor has the same biologic potential as the ameloblastoma. So, it is considered wise to treat a patient with an odontoameloblastoma in the same manner as one with an ameloblastoma. However, because of the rarity of odontoameloblastomas, there are no valid data on the long term prognosis. When the odontomas are associated with unerupted teeth, orthodontic traction of the impacted tooth soon after removal of the lesion may be needed, especially if it is not diagnosed and treated early [17].

**Complex odontomas** generally reach diameters ranging from several millimetres to 3-4 cm. These odontomas preferentially involve the posterior portion of the jaw and might be associated with an unerupted or absent tooth in

the region. Caboc et al. [18] reported that odontomas in the maxillary sinus may cause pain, facial asymmetry and chronic congestion of the sinus. The radiopaque image of complex odontomas in the maxillary sinus can be characteristic of various bone lesions such as cementoblastomas, osteomas, ossifying fibromas, and osteoblastomas, as well as of other odontogenic tumors such as calcifying epithelial odontogenic tumors and ameloblastic fibro-odontomas. The presence of well-defined borders and a radiolucent halo around the radiopaque mass is a characteristic finding of odontoma. Complex odontomas can be associated with other more aggressive odontogenic lesions such as cysts and tumors. Several case series have documented that the majority of all odontomas were diagnosed in the first two decades of life. Although they may be discovered at any age, less than 10% are found in patients over 40 years old [19].

**Management:** Odontomas are common tumours that can be easily diagnosed and treated. However, these tumours should not be underestimated since they may show rare and aggressive features that can lead to serious disorders in the patient. Surgical removal is the treatment of choice. Care should be taken, however, not to harm adjacent teeth and permanent germens in children, while follow-up is essential for evaluation of further development of the permanent dentition at the removal location. Although the diagnosis of odontomas, in most cases, can be provisionally confirmed by radiographic examination, a histological study of the removed lesion must be done to confirm the diagnosis. Odontomas are treated by conservative surgical removal and there is little probability of recurrence [20].

**Figure 1. IOPA reveals grossly carious retained maxillary right deciduous lateral incisor with small radiopaque structure above it**



Courtesy: Department of Oral Medicine, Diagnosis & Radiology, Institute of Dental Education & Advance Studies, Gwalior, Madhya Pradesh

**Figure 2. Maxillary occlusal radiograph showing radiopaque tooth like denticle (compound odontome in right half of dorsum of hard palate & missing maxillary right permanent central incisor**



Courtesy: Department of Oral Medicine, Diagnosis & Radiology, Institute of Dental Education & Advance Studies, Gwalior, Madhya Pradesh

**CONCLUSION**

Odontomas are benign tumors frequently seen that may be symptomatic or sometimes produce no symptoms and are diagnosed as incidental findings on routine radiological studies. Routine radiographic examination is important for early detection of silent lesions such as

odontomas. They usually cause delayed eruption. If no signs or symptoms appear, and lesions go undetected, they can remain for many years without clinical manifestation. The recommended treatment is total surgical removal with proper histopathologic evaluation to confirm the diagnosis.



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**CONFLICT OF INTEREST:**

The authors declare that they have no conflict of interest.

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