

Multiple Complex Odontomas in Maxilla and Mandible: A Rare Case Report

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DOI: [10.36348/sjpm.2022.v07i11.001](https://doi.org/10.36348/sjpm.2022.v07i11.001)

| Received: 23.09.2022 | Accepted: 30.10.2022 | Published: 03.11.2022

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Abstract

Odontomas are among the most common type of benign odontogenic tumors comprising 22% of all odontogenic tumors of the jaws. Rather than true neoplasm, odontomas are considered to be hamartomas. Usually odontomas are found to be asymptomatic, although a few incidences of swelling, delayed eruption of permanent tooth and in severe cases, infection or lymphadenopathy has been reported. Multiple complex odontomas pose a therapeutic challenge in some cases especially in the mandible due to proximity of vital anatomic structures. They have also been associated with complications like cystic transformation. Here we are presenting a case of multiple complex odontomas in the maxilla and mandible in a 32-year-old male patient.

Keywords: Multiple complex odontomas, benign odontogenic tumors, hamartomas.

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INTRODUCTION

Odontomas are among the most common type of benign odontogenic tumors. Rather than true neoplasm, odontomas are considered to be developmental anomalies (hamartomas) [1]. Paul Broca in the year 1867 coined the term odontoma. He defined odontomas as ‘tumors formed by the overgrowth of transitory or complete dental tissues [2]. Odontomas are mixed odontogenic tumors since they are composed of both epithelial and mesenchymal dental hard tissues [3, 4]. They are predominantly composed of enamel and dentin along with variable amount of cementum, and pulp [1].

The WHO, in 2005, categorized odontomas based on the radiographic features into 2 types: 1) complex odontoma which are calcified irregular mass not resembling teeth and 2) compound odontoma which are calcified mass bearing resemblance to the teeth [5]. Complex odontomas are less common when compared to the compound variety in the ratio of 1:2 [6]. The occurrence of odontomas have been attributed to

etiologic factors such as local trauma and infection. Genetic factors are also another one of the suggested causes [7].

The odontomas are usually found have a higher rate of incidence in the areas of the upper incisors and canines, followed by the anterior and posterior mandibular regions. Complex odontomas are found more commonly in the area of the second and third lower molars (34%) [8-10]. This case report describes a rare case of multiple complex odontomas involving both jaws in a 32-year-old male. Informed written consent was obtained from the patient regarding the use any of accompanying images and publication of this report.

CASE REPORT

A 32-year-old apparently healthy male patient was referred to Department of Oral Pathology and Microbiology with a complaint of mild pain on the right side of mandible. The patient's medical, family and dental history were non-contributory. The patient was

found to be healthy on general physical examination. No evident swelling was noted on extraoral examination. Intraoral examination revealed supraerupted 18, missing 48 and a whitish mass present in 48 region (Figure 1). On palpation the mass was found to be tender and hard in consistency.



Fig 1: Intraoral photograph showing a whitish mass present in 48 region

Panoramic radiograph showed well defined radiopaque lesion involving crown portion of 48 and extending from root towards ramus of mandible approximately 3 to 3.5 cm below the right sigmoid notch. Radiopacity was also noted in relation to mesial root of supraerupted 18.



Fig 2: Panoramic view showed well defined radiopaque lesion involving crown portion of 48 extending from root towards ramus of mandible and mesial root of supraerupted 18

Based on all these clinical and radiological examination findings, a provisional diagnosis of complex odontoma was made. The patient was convinced for surgery after being made aware of the existing pathology. Haematological investigations were done and values were found to be within normal range. Written informed consent from the patient was obtained prior to the surgical procedure. Surgical excision of the mass was performed under local anaesthesia along with removal of supraerupted 18 and impacted 48.

The gross specimen consisted of eight hard tissue bits of which two were retrieved from 18 region and six were from 48 region. The specimen included three fractured tooth segments of 48, one large (2 x 1.4 x 0.8cm), three medium (1 x 1 x 0.5cm) and one small (0.4 x 0.3 x 0.2 cm) (Figure 3) irregular masses of calcified hard tissue bits. They were sent for histopathological evaluation after decalcification.

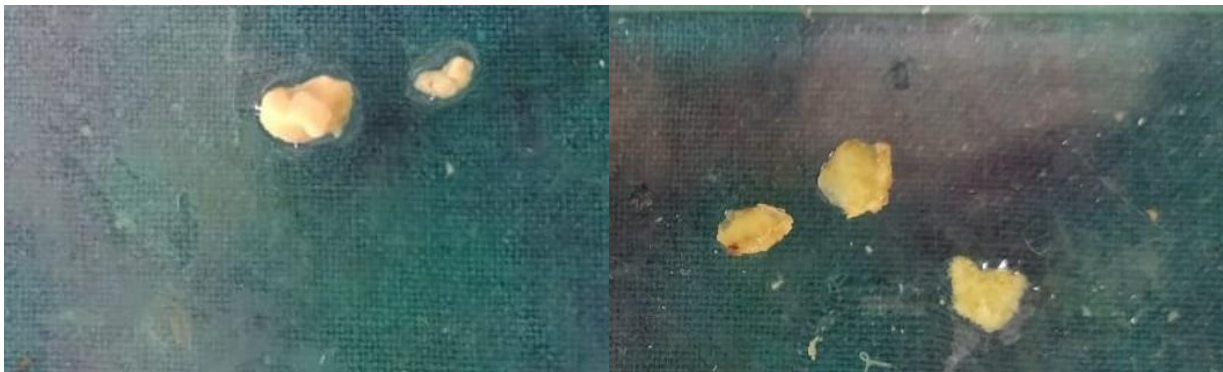


Fig 3 (a and b): Gross specimen showing five irregular masses of calcified hard tissue bits

Histopathological examination of H and E stained decalcified sections revealed haphazardly arranged dentine globules. Loose connective tissue resembling pulp was noted in close association with the dentinal tissue. Bony trabeculae with lacunae

containing osteocytes and intervening fibrofatty connective tissue were noted in some bits. The histopathological features were suggestive of “Complex Odontoma”.

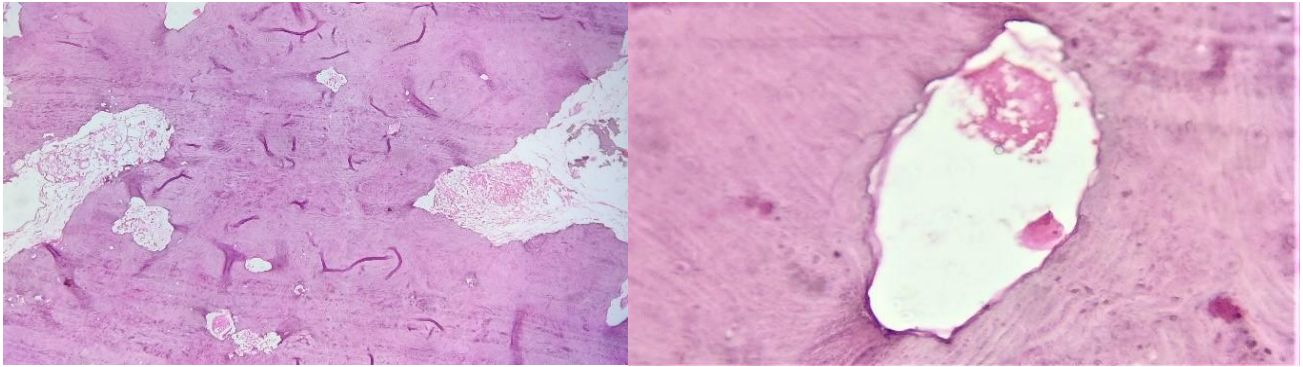


Fig 4 (a and b): H & E stained sections showing haphazardly arranged dentine globules with loose connective tissue resembling pulp was noted in close association with the dentinal tissue in 10x(a) and 40x(b)

DISCUSSION

Odontomas comprises 22 % of all odontogenic tumors of the jaws [11]. Odontomas are slow growing, expansile lesion commonly occurring in the second and third decades of life. Even though odontomas are among the most common odontogenic tumors, the occurrence of multiple odontomas are rare, usually being associated with some systemic syndromes such as familial adenomatous polyposis [9, 12]. There has been only 15 reported cases of multiple odontomas, out of which only nine were complex odontomas [13]. Usually odontomas are found to be asymptomatic, although a few incidences of swelling, delayed eruption of permanent tooth and in severe cases, infection or lymphadenopathy has been reported [14]. In most of the cases odontomas are detected incidentally on routine radiographic examinations [15]. The complex form of odontomas are less common than the compound type, known to occur mostly in the posterior mandibular region. The male/female ratio in complex odontomas has found to vary from 1:0.8 to 1.6:1. Complex odontomas were reported to measure around 1-2 cm in diameter according to majority of the literature [7]. In the present case they measured around the same size.

The complex odontomas are more frequently associated with unerupted teeth [16]. They rarely erupt into the oral cavity but when this occurs, it is different from a normal tooth eruption. This is due to lack of the periodontal ligament. Increase in size can lead to the sequestration of the overlying bone, causing pressure and possible movement in the occlusal direction, leading to eruption. In this case retention of the right third molar was noted [17].

The exact etiology of odontomas remains unknown. A history of trauma to the deciduous dentition, as well as inflammatory and infectious processes have been associated with occurrence of some odontomas. They have also been attributed to hereditary anomalies like Gardner's syndrome and Hermann's syndrome, alterations of the genetic components responsible for controlling tooth development, or odontoblastic hyperactivity [18].

Torreti *et al.*, suggested the role of mature ameloblast in the etiology of odontomas. According to him these specialized cells have potential of giving rise to tumors with wide variation in their appearance and content [19]. According to Fijerskov *et al.*, cell rest of serres of the retained tooth with some epithelial islands have the capability of undergoing proliferation giving rise to odontomas [20].

Another aspect of the etiology of odontomas is the extraneous odontogenic epithelial cells.[13] The division of the tooth buds occur into several parts, they may develop individually to form numerous, closely positioned malformed teeth or tooth-like structures. When the buds develop without such uncommon division and consists of haphazard collection of dental tissues, they give rise to complex odontomas [21]. Recently, a possible genetic etiology was suggested in a study by Ziebart T *et al.*, for multiple odontomas. According to his study a gain of function may be conferred to FGF3 and FGF4 genes on the partial duplication of chromosome 11 q13.3 [22]. In this case there was no relevant family history, history of trauma nor was the patient associated with any syndromes.

The differences between the complex and compound type of odontomas cannot be ascertained only by visual examination of the lesions, as the odontomas usually lie in the bone and do not usually exhibit any outward signs, such as expansion of the bone. Even in the rare instances of odontomas erupting into the oral cavity, differentiating between them visually is difficult as the surface appearances of both types are similar [23]. Radiographic examination has been deemed as an effective method of discriminating between the two types clinically. Well-organized malformed teeth or tooth-like structure are noted in case of compound odontome. Complex odontoma presents as a well-demarcated radiopaque mass consisting of less well-organized dental tissue, occasionally surrounded by a narrow, radiolucent zone. Another useful procedure in the effort to establish a definite diagnosis is microradiography. It enables the recognition of histologic structures based on their differences in

radiopacities and/or radiolucencies [24]. Micro-CT has also been widely employed in many academic fields as a relatively new method of high-resolution three-dimensional imaging [25].

The indicated treatment for the odontoma is surgical excision. The fibrous capsule surrounding the odontomas facilitates easy dissection of the lesion away from the surrounding bone. Curettage of the region is done to prevent the possibility of cystic degeneration [26].

CONCLUSION

Multiple complex odontomas pose a therapeutic challenge in some cases especially in the mandible due to proximity of vital anatomic structures. They have also been associated with complications like cystic transformation. The present case shows five complex odontomas involving both maxilla and mandible. Surgical intervention was done and the odontomas along with 18 and impacted 48 were extracted. They have a favorable prognosis with rare chances of recurrence.

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