

## \*UNILATERALLY ERUPTED BILATERAL SYMMETRICAL COMPOUND-ODONTOMAS AT THE POSTERIOR REGION OF HUMAN MAXILLAE: CASE REPORT

Maksiler Posterior Bölgede Unilateral Sürmüş Bilateral Simetrik Kompaund-odontomalar:  
Vaka Raporu

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### Abstract

Compound-odontoma, which is the subtype of odontoma, is composed of enamel, dentin, cementum, pulp tissues, and both ectodermal and mesenchymal cells exhibit complete differentiation resulting in the formation of small tooth-like structures. There have been many studies and reports of odontoma in the literature; however bilateral symmetrical occurrences of odontomas are limited with few reports in the literature. The etiology of bilateral symmetrical occurrences of odontomas is unknown. There is need more records to make accurate debates on its etiology. This case report, to our knowledge, constitutes the first describing a patient with bilateral symmetrical compound-odontomas in the posterior region of human maxillae, and discusses the clinical and histopathological features.

**Key words:** compound odontoma, developmental anomalies, odontogenic tumors, bilateral, symmetrical, maxillae

### Özet

Odontomanın bir alt türü olan compound odontoma, mine, dentin, sement, pulpa dokusundan oluşur ve hem ektodermal hem mezenşimal hücreler küçük diş benzer yapı oluşumu ile sonuçlanan tam diferansiyasyon gösterir. Literatürde pek çok odontoma çalışması ve raporları bulunmaktadır; ancak bilateral simetrik odontoma oluşumu literatürde bir kaç raporla sınırlıdır. Bilateral simetrik odontoma oluşumunun etiolojisi bilinmemektedir. Etiolojisiyle ilgili daha doğru tartışma yapmak için daha fazla rapora ihtiyaç bulunmaktadır.

Bu vaka raporu, bildiğimiz kadarıyla insan maksillasının posterior bölgesinde bilateral simetrik kompaund odontoması olan bir hastayı tanımlayan ilk rapordur ve klinik histopatolojik özelliklerini tartışmaktadır.

**Anahtar kelimeler:** Kompaund odontoma, gelişimsel anomaliler, ontojenik tümörler, bilateral, simetrik, maksilla

### Introduction

Odontomas are classified as the most common mixed odontogenic tumors (1,2). However, they have been also described as a

developmental anomaly in which all dental tissues are represented (3-7). Soames and Southam (6) indicated that the term odontoma should be used to delegate non-neoplastic developmental anomaly contain fully formed enamel and dentine. These calcified anomalies develop result from the growth of completely differentiated epithelial and mesenchymal cells that give rise to functional ameloblasts and odontoblasts. These cells in turn form variable amounts of enamel, dentine and pulpal tissues of the odontomas (8).

Odontomas, which are age and location related (9), are frequently diagnosed in the second decade of life (3-5,9-11) either during routine radiographic examinations or in association with impacted permanent teeth, with or without persistent primary teeth. Less frequently, it might be associated with asymptomatic swelling or other abnormalities

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(3,12,13). Odontomas rarely occur in association with primary teeth (1,10). Radiographically, they appear as radiopaque masses surrounded by radiolucent areas.

The cause of odontomas is unknown; however, inheritance or interference with genetic control of tooth development was attributed to possible causes (14). Trauma, infection or growth pressures which may disturb the biological mechanism that controls the tooth development are also regarded as causes leading to odontoma (14).

Two types of odontoma are recognized: complex- and compound-odontoma (4). In complex-odontoma, all individual dental tissues are mainly well-formed but represented in disorderly-pattern, whereas in compound-odontoma they are represented in orderly-pattern (3,4,9). The latter is a hamartomatous lesion which resembles miniature tooth structure (1,3,4,9). Its development passes through the same stages as developing teeth, and it is relatively small but rarely larger than a tooth (15). Therefore, radiographically appears as rudimentary miniature tooth structure and can be diagnosed from radiographs alone.

The frequency of occurrence of odontoma varies greatly in different populations. It is most common in the Caucasian (over 65% of all odontogenic tumors) (10), however, rare among Chinese (6-7%) (2) and Nigerians (2.5%) (16). There have been many studies and reports of odontoma in the literature; however, only few reported bilateral occurrences of odontomas in either human (17-20) or animal (21,22). Bilateral symmetrical occurrence of compound-odontomas is even rarer (22). We reviewed the literature and found only one report of bilateral compound-odontomas with unknown etiology in a dog mandible (22) and none in human. In the former, also limited clinical and histological information were provided. This case report, to our knowledge, constitutes the first describing a patient with unilaterally erupted bilateral symmetrical compound-odontomas in the posterior region of human maxillae. Since such presentation is unique and the etiology of bilateral symmetrical odontomas is unknown, we believe that the presentation of this case with detailed documentation is valuable to constitute data for its etiology.

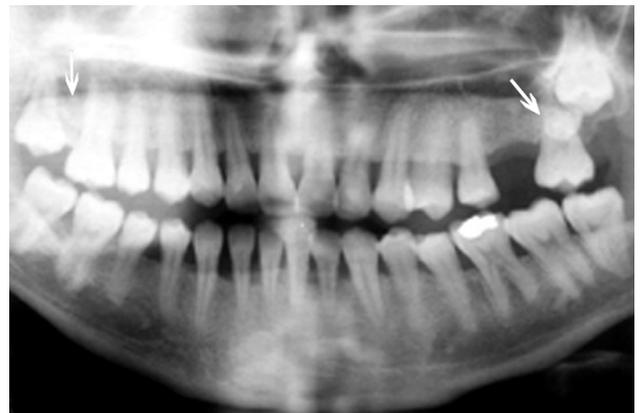
## Case Report

A 21-year-old female Turkish patient was referred to the Department of Oral and Maxillofacial Surgery for examination of a visible calcified oral mass at the right upper jaw (Fig. 1). Clinical examination revealed an erupted calcified oral mass on the mesiobuccal aspect of the maxillary right third molar tooth (M3) (Fig. 1).



**Figure 1** Intra-oral presentation of the erupted right compound odontoma at the mesiobuccal aspect of the third molar tooth

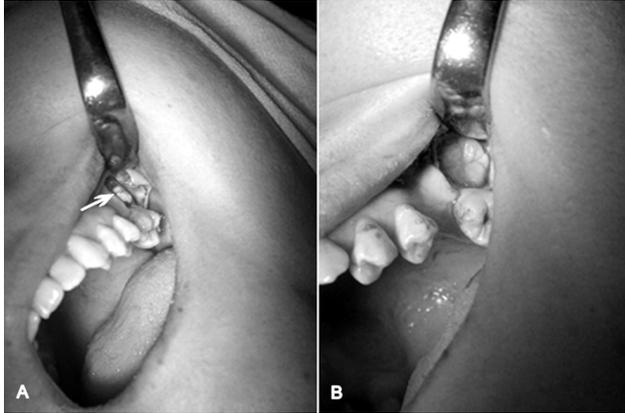
An orthopantomography (OPG) showed an impacted another radiopaque mass superimposed onto the root of the erupted left maxillary second molar tooth (M2) and also located at the occlusal aspect of the impacted left M3 (Fig. 2).



**Figure 2** An orthopantomography illustrates the right erupted (vertical arrow) and left impacted (oblique arrow) compound-odontomas. The left impacted compound odontoma is superimposed onto the root of the second molar

tooth and located at the occlusal aspect of the impacted third molar tooth.

Diagnosis of odontoma was made, initially, on the basis of OPG that showed separate small well-calcified masses with tooth-like appearance (denticles) with periodontal support. Among the asymptomatic odontomas, the right odontoma had erupted (Fig. 1) while the left odontoma was impacted (Fig. 2, 3a).



**Figure 3** Surgical removal of the left impacted compound odontoma (3a) and associated impacted third molar teeth (3b), which directed through the cheek.

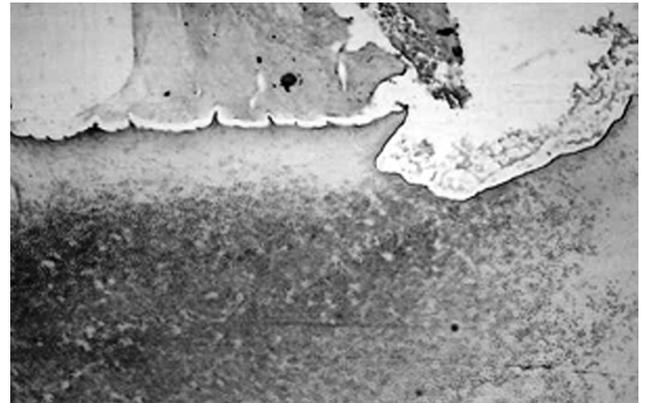
There was no family history of unerupted teeth, hypodontia or trauma to the orofacial region. Medical history was clear.

Dental history included extraction of the left maxillary first molar tooth (M1), restorative treatment of left mandibular M1 and recent periodontal treatment. Oral and radiographic examinations of the remaining teeth showed missing maxillary right M2 and mandibular right M3 and no other dental abnormalities. There was no palpable buccal or palatal swelling and the erupted mass was firm.

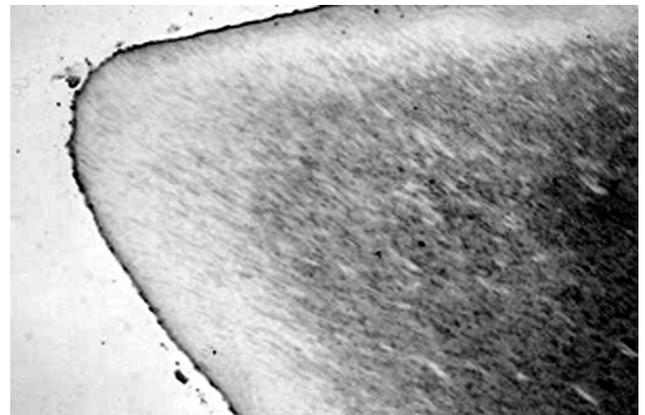
The treatment plan consisted of extraction of the right odontoma and surgical removal of the left odontoma (Fig. 3) with extensive curettage. Under local anesthesia, the right erupted odontoma was extracted and a labial mucoperiosteal flap was raised in the right maxillary molar region to remove left impacted odontoma and the associated M3. Bone was removed with a bur. Single compound-odontoma was removed. Although the root development of left M3 was incomplete (Fig. 2), it was removed since direction of eruption was through the cheek (Fig. 3b) and the patient did not accept any orthodontic treatment to correct

the direction. The surgical wound was closed primarily with sutures. The patient had uncomplicated healing period with only minimal edema for the first postoperative day. The sutures were removed a week later with satisfactory wound healing.

Macroscopically, both excised odontomas were solid and resembling miniature teeth which were shaped similar to a dental crown and root structures. The odontomas were examined by microscopy to determine whether the odontomas were complex or compound. Microscopically, tooth-like structure with rare irregular dentin tubules besides irregular spaces on the basophilic stained enamel surface in the right-odontoma (Fig. 4), and tooth-like tumor tissue consists of loose and seldom fibrous connective stroma in the left-odontoma (Fig. 5) were observed in light microscope.



**Figure 4** Light microscopic appearance of the right-odontoma shows tooth-like tissue with rare irregular dentin tubules besides irregular spaces on the basophilic stained enamel surface (Original magnification x 40; HE).



**Figure 5** Light microscopic appearance of the left-odontoma shows tooth-like tissue consists

of loose and seldom fibrous connective stroma (Original magnification x 40; HE).

The final diagnosis in each specimen was based on the 1992 histological criteria of the World Health Organization.<sup>4</sup> Microscopic examination of the excised tissues confirmed the clinical diagnosis and both odontomas were interpreted as compound-odontoma.

Patient had uncomplicated healing period. Clinical and radiographic recall examinations during last 4-years showed no recurrences to date.

## Discussion

Compound-odontoma occurs with equal frequency in both sexes either in the mandible or maxillae, and is usually asymptomatic (23). The incidence of compound-odontoma in the maxillae is 62% (24). It occurs most commonly in the incisor-canine region of the maxillae (1,10,11) and secondly in the anterior portion of the mandible (10,11). Although occurrence of compound-odontoma is not considered rare, its bilateral occurrence is indeed, rare. We reviewed the literature and found only six cases with bilateral odontoma in human (17-20) and animal (21,22). These extraordinary reports composed of composite odontoma (18), ameloblastic odontoma (19), complex-odontoma (21) and compound-odontoma (22).

The only case with bilateral compound-odontoma was in a six-month-old, male Black Russian terrier dog mandible and both completely erupted masses were located distolingual aspect of the M1 (22). However, limited clinical and histological information were provided with this report. We have reported this case, since, to our knowledge, the occurrence of bilateral symmetrical compound-odontomas in human posterior region of the maxillae have not previously been reported in the literature.

The etiology of bilateral symmetrical occurrences of odontomas is unknown. It is known that trauma to the permanent predecessor might result in alteration of the developing permanent tooth. However, bilateral symmetrical occurrences of trauma which lead to bilateral development of odontomas make it unlikely. Furthermore, the incidence of bilateral compound-odontomas is limited with six cases in the literature (17-22), and is disproportionate to the high prevalence of trauma. Traumatic

injuries can not, therefore, account for all cases of bilateral occurrences of odontomas. Secondly, bilateral symmetrical occurrences of idiopathic developmental disturbances, infections, coincidence or growth pressures which may disturb the biological mechanism that controls the tooth development at the same time also seem to be unlikely (14). Finally, inheritance or interference with genetic control of tooth development might be attributed to possible causes (14). However, there is need more reports to evaluate such hypothesis.

If the physical existence of the compound-odontoma influences adjacent tooth development lead to its absence is not precisely known. However, it is tempting to speculate that the right compound-odontoma appeared to replace the maxillary M2 in the arch. In other words the physical presence and/or development of the right compound-odontoma might have prevented development of right maxillary M2. On the other hand, the left compound-odontoma seemed to be an additional to the dentition. It is also possible to postulate that it has arisen from a supernumerary anlage. However, more data are required to justify these postulations.

Odontomas may cause various disturbances to tooth eruption; therefore, they are always included in the local etiologic factors of impaction or delayed eruption of teeth. While 41-47.6% of odontomas were found to be associated with unerupted teeth (9,11) up to 68% of impacted teeth were reported to be related with compound ones (3,5,11). Although impaction of a maxillary M3 is very common, a local presence of left compound-odontoma seemed to physically altered the direction of tooth eruption leading to its' impaction in the presented case. However, the maxillary right M3 was erupted. This may be due to missing right M2 tooth allowed sufficient place for the placement of right M3 and associated compound-odontoma.

Treatment advocated for odontoma is its surgical removal with or without associated impacted teeth (25). However, every effort should be made to preserve impacted permanent teeth. Therefore in cases of odontomas associated with impacted teeth, a combination of surgical removal of odontoma and orthodontic treatment of impacted teeth is also indicated (26). On the other hand, the teeth with incomplete root development may be

normally re-erupt spontaneously (27). Since odontomas are often well circumscribed and encapsulated, they can be removed easily.

Recurrence is rarely seen after the surgical removal of compound-odontomas (15). The presented case had a relatively small compound-odontomas and both were removed by means of conservative surgery. Associated impacted left M3 with incomplete root apex was also removed despite had potential to erupt, since direction of its eruption was through the cheek and the patient rejected further orthodontic treatment. The patient was followed up with clinical and radiographic examinations for 2.5-years after the removal of both odontomas, and surgical sites were characterized by the rapid post-surgical recovery without recurrences to date.

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