

**PERIPHERAL OSTEOMA OF THE HARD PALATE: A RARE CASE REPORT
 AND LITERATURE REVIEW**

**Sert Damakta Periferik Osteoma: Ender Görülen Bir Olgu Bidirisi
 ve Kaynak Derlemesi**

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ABSTRACT

Osteomas are benign osteogenic tumors that are composed of cancellous or compact bone. They are usually sessile tumours composed of dense sclerotic, well-formed bone projecting out from the cortical surface. They commonly occur in skull and facial bones, however, excluding the maxillary sinuses, the maxilla is a rare site for osteomas. Therefore, we aim to present a case of maxillary osteoma in a 19-year-old male patient and to provide an overview of current literature on the diagnostic options and treatment modalities. To best of our knowledge, in the English literature, this is the seventh reported case of maxillary osteoma which is located on the hard palate.

Keywords: Hard palate; Maxilla; Osteoma

ÖZ

Osteomlar süngerimsi veya kompakt kemikten oluşan iyi huylu osteojenik tümörlerdir. Bunlar; genellikle kemiklerin kortikal yüzeyinden dışarıya doğru çıkıntılar oluşturan, belirgin şekilli, yoğun sklerotik doku içeren sesil tümörlerdir. Kafa ve yüz kemiklerinde daha sık görülürler. Ancak, osteomalar maksiller sinüs haricinde maksillada ender olarak izlenebilirler. Bu olgu sunumunun amacı; 19 yaşındaki bir erkek hastada saptanan maksiller osteoma olgusunu incelemek ve tanı seçenekleri ile tedavi yöntemleri hakkındaki güncel kaynakları değerlendirmektir. Bildiğimiz kadarı ile, bu İngilizce kaynaklarda bildirilen sert damakta konumlanmış 7. olgudur.

Anahtar kelimeler: Sert damak; Maksilla; Osteoma

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Introduction

Osteomas are benign, slow-growing tumors of the cancellous or compact bone, which may be classified as solitary or multiple. Multiple osteomas are mainly associated with Gardner's syndrome, while solitary lesions are usually independent of diseases or syndromes. Solitary osteomas can be classified as peripheral, central, or extra-skeletal depending on the origin (1). The central types arise from the endosteum, the peripheral types arise from the periosteum, and the extra-skeletal soft tissue types arise within a muscle (2). The most commonly affected sites are the mandible and paranasal sinuses in the maxillofacial region (3-5). Excluding the maxillary sinuses, maxillary osteomas located on the hard palate are very rare (6, 7). Their etiology is unknown. Traumatic, infectious or developmental causes have been speculated (8). As to the gender predilection, some investigators suggest that there is a higher incidence of osteomas in males, particularly between the second and fourth decades of life (8, 9). Osteomas may remain silent for years without any symptoms. They are often detected incidentally in a routine radiographic survey or when they cause facial asymmetry or functional impairment. Osteomas generate symptoms by compression, rather than by invasion or destruction (9, 10). Depending upon the location, they might cause headaches, facial pain or swelling, and/or limited mandibular movements (6, 10). Osteoma treatment is based on complete surgical removal, and there are no reports of malignant transformation (11, 12). Its recurrence is rare, with only one case described in the literature (11, 13).

Apart from osteomas related to Gardner's syndrome, few cases involving the maxilla have been reported (9, 10). The aim of this paper is to present an unusual case of maxillary osteoma and to provide an overview of the current knowledge concerning the pathogenesis, differential diagnosis and treatment of these lesions.

Case Report

A 19-year-old male patient was referred to our clinic for evaluation and treatment of the swelling on his palate. The patient complained of a slow growing, painless mass in the right palate which was present for two months. The patient's history revealed no trauma or relevant diseases. He was asymptomatic except for mild discomfort and difficulty in mastication.

Oral examination revealed a firm, painless, well-circumscribed exophytic mass, 15 mm × 11 mm in size on the right posterior palatal area in relation to teeth numbered 13,14, and 15 according to the FDI classification. The pulp vitality test confirmed that all teeth were vital. Clinically, the teeth demonstrated no mobility. The lesion was non-tender, non-compressible, non-fluctuant and non-pulsative on palpation and it was covered by normal oral mucosa.

The intra-oral periapical (Figure 1) and panoramic radiographs revealed a well-demarcated radioopaque mass superimposing the roots of teeth numbered 13, 14, and 15, which is clearly demarcated from the surrounding healthy bone. Based on the clinical and radiological findings, a provisional differential diagnosis of cementoblastoma, osteoma or maxillary torus was made.



Figure 1. The appearance of the lesion in the periapical radiograph.

Under local anesthesia, mucoperiosteal flap was reflected on the palatal surface starting from the left central incisor and reaching to the left first molar teeth. Surgical removal was performed (Figures 2 and 3). Curettage and debridment were done. After hemostasis, the area was irrigated with saline solution and flap was primarily sutured back in position (Figure 4). Postoperative instructions were given, with emphasis to the maintenance of appropriate oral hygiene, ingestion of cold and soft meals. Also, he was advised to refrain from having physical exercise for 48 hours. Routine post-operative medication was prescribed to prevent pain and infection. Postoperative healing was uneventful with no sensory deficit and the involved teeth were not devitalized. The specimen

was processed for histopathological examination. Histopathologically, the lesion was diagnosed as peripheral (mature) osteoma (Figure 5). The patient recovered without any complications.

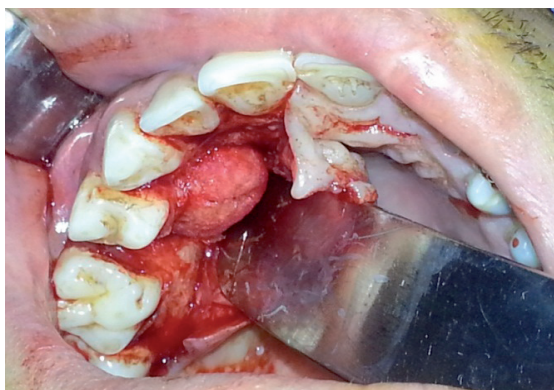


Figure 2. Intraoperative view of the lesion.



Figure 3. Macroscopic view of the surgical specimen.



Figure 4. Postoperative view.

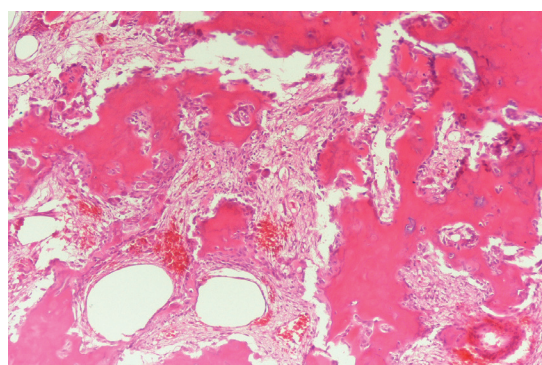


Figure 5. Histopathological view.

Discussion

There are two different types of peripheral osteoma which is a tumor of mature bone. The compact or “ivory” osteoma usually has a sessile base, normal-appearing dense bone with minimal marrow spaces and occasional Haversian canals. The cancellous osteoma is usually a pedunculated, mushroom-like mass and resembles to the cancellous bone. It is characterized by bony trabeculae and fibrofatty marrow with osteoblasts. The surface can be irregular or smooth, with cortical bone at the margin. The sizes of both compact and cancellous osteomas range from several millimeters to several centimeters; however, part of the lesion may be in the bone, masking its true size (2, 14). The case presented here was diagnosed as a compact osteoma. Clinically, osteomas appear as circumscribed, rounded and protuberant (15). They may remain silent for years without any symptoms and they are often diagnosed only when they are large enough or incidentally during radiological investigations (4). In most cases, osteomas have a very slow growth rate and they cause swelling and asymmetry (16).

The peripheral osteomas' pathogenesis is controversial (10, 13, 15). Some investigators classify this lesion as a hamartoma while others consider it as a true neoplasia (11). Reactional mechanism, trauma or infections are also proposed as possible causes (17). According to Thoma and Goldman (18), growth is associated with trauma, not with the inflammation, and starts spontaneously. Schneider *et al.* (19) reported a positive history of prior trauma in six cases. Osteomas are usually located in muscle insertions, suggesting that the muscle pull acts on the development of the lesion. Minor traumas which are not even remembered by the patients may cause a subperiosteal hematoma that, associated

with the muscle pull, starts the lesion (11, 12, 17). Varboncoeur *et al.* (20) considered osteomas to arise either from embryological cartilaginous rests or from persistent embryological periosteum. According to the developmental or embryological theory, the osteomas would then originate from the suture between bones with different embryological derivation (membranous/enchondral). But this seems unlikely, because in most cases they develop in adults and not during childhood or adolescence (10). This patient was in adolescence and did not have a trauma history.

According to Cutilli *et al.* (17) there is no predilection for gender (21). However, Bosshardt *et al.* (13), Kaplan *et al.* (12) and Sayan *et al.* (2) reported that males are affected more frequently than females, at a 2:1 ratio. Schneider *et al.* (19) and Sah *et al.* (16) reported a 3:1 female predilection, and Horikawa *et al.* (11) found a slightly higher prevalence among women, with a 1.5:1 ratio. In our case, the patient was male. Bodner *et al.* (21), Longo *et al.* (22), Sugiyama *et al.* (23), Horikawa *et al.* (11) and Sayan *et al.* (2) reported that osteomas have no age predilection. However, according to Longo *et al.* (22), there is a higher frequency of occurrence between the third and fifth decades of life. Kashima *et al.* (15) reported that osteomas are more often seen in the sixth decade of life. Sah *et al.* (16) reported that the mean age range of the patients is 29.4 and 40.5 years. Our patient was 19 years old. Peripheral osteomas of the craniofacial region occur more often in the paranasal sinuses. The external auditory canal, the orbit, the temporal bone and the pterygoid processes are less commonly affected (2, 11, 21). The mandible is more commonly involved than the maxilla, and the mandibular body and angle are the most commonly affected. It is a rare entity in the maxilla when the maxillary sinuses are excluded (15, 21, 23). Conventional radiographic images are generally adequate in diagnosing an osteoma (10, 22). Radiographically, the presence of an oval, radiopaque, well-circumscribed mass attached by a broad base or pedicle to the affected cortical bone is a hallmark of peripheral osteomas (2, 6, 15). In addition to panoramic radiograph, Water's view or tomograms usually helps to reveal the lesion. The computerized tomography (CT) scan can show more details concerning the relationship between the osteoma and the adjacent structures and location with a better solution, especially with 3D reconstruction (9, 10, 15, 22). In the present case, periapical and panoramic radiographs revealed the bony mass.

The differential diagnosis includes exocytosis and several other pathologic processes, including inflammatory and neoplastic lesions. Exocytosis can be distinguished from the osteoma by its tendency to stop growing after puberty (6). Periosteal osteoblastoma, osteoid osteoma, and paraosteal osteosarcoma usually grow rapidly and they cause painful swellings (2, 16). Patients presenting with osteomas should be investigated for Gardner's syndrome. These patients may also present with symptoms of rectal bleeding, diarrhoea, and abdominal pain (2). The triad of colorectal polyposis, skeletal abnormalities, such as peripheral and endosteal osteomas, and multiple impacted or supernumerary teeth is consistent with this syndrome (1, 2). Osteomas generally develop before the formation of colorectal polyposis. Therefore, the syndrome might be early recognized and this might be a life-saving event (1). In our case, the lesion was solitary and there were neither intestinal problems nor impacted or supernumerary teeth. Without Gardner Syndrome, having solitary osteoma of the maxillofacial region is rare (10, 11, 15, 21). Apart from osteomas located in the maxillary sinuses, there are 19 cases of maxillary osteomas previously described in the English language literature (Table 1) (1, 2, 4, 6-8, 10, 12, 16, 24-28). Among these, there are 10 male and 9 female patients. Their ages ranged from 16 to 76 years, with a mean age of 45.07 years however, the complete clinical data of five cases are lacking. These osteomas usually occur in the peripheral type, but two of them were central variants. Maxillary osteoma is most commonly observed in the alveolar ridge. Eleven of these cases were found in the alveolar ridge, two of them in the alveolar process, and six of them in the hard palate. Our patient is the 20th reported case of maxillary osteoma in the jaws and the 7th case found in the hard palate.

Asymptomatic osteomas of the jaws are mostly managed conservatively, but in symptomatic cases, the treatment of peripheral osteoma is based on complete surgical removal (10, 11). Recurrence is rare with only two case reported in the literature. Bosshardt *et al.* (13) described the first case of recurrence nine years after surgical treatment and Horikawa *et al.* (11) reported the second case of recurrence two years after surgical excision. No malignant transformation has been reported in the English literature (10, 11). Therefore, the treatment of asymptomatic lesions is controversial (11). There was no sign of recurrence in our patient during the follow-up period.

Table 1. Summary of clinical data of maxillary osteomas previously published in the English language literature (NA; Not Available).

Case	Authors	Year	Age	Gender	Histology	Site	Region
1	Seward (28).	1965	50	Female	Compact	Peripheral	Alveolar ridge
2	Sayan et al.(2).	2002	NA	Female	NA	Peripheral	Alveolar ridge
3	Sayan et al.(2).	2002	NA	Female	NA	Peripheral	Alveolar ridge
4	Sayan et al.(2).	2002	NA	Male	NA	Peripheral	Alveolar ridge
5	Sayan et al.(2).	2002	NA	Female	NA	Peripheral	Hard Palate
6	Sayan et al.(2).	2002	NA	Male	NA	Peripheral	Hard Palate
7	Dalambiras et al.(11).	2005	16	Female	Compact	Peripheral	Alveolar ridge
8	Woldenberg et al.(25).	2005	76	Female	NA	Peripheral	Alveolar ridge
9	Durinheto et al.(7).	2007	40	Male	Cancellous	Peripheral	Alveolar ridge
10	Iatrou et al.(27).	2007	64	Male	Compact	Peripheral	Alveolar process
11	Kaplan et al.(13).	2008	46	Male	Compact	Central	Alveolar ridge
12	Chaundhry et al.(26).	2009	73	Male	Cancellous	Peripheral	Alveolar ridge
13	Wong and Peck (1).	2010	20	Male	Compact	Peripheral	Alveolar ridge
14	Sah et al.(17).	2011	50	Female	Cancellous	Peripheral	Alveolar process
15	Prabhujji et al.(8).	2011	45	Male	Compact	Peripheral	Hard Palate
16	Nah (3).	2011	69	Female	NA	Central	Hard Palate
17	França et al.(6).	2012	53	Male	Compact	Peripheral	Alveolar ridge
18	Viswanatha. (29).	2013	14	Female	NA	Peripheral	Hard Palate
19	Viswanatha (29).	2013	15	Male	NA	Peripheral	Hard Palate

Conclusion

Osteomas occur predominantly in the maxillofacial region, but maxillary osteomas are extremely rare. In patients with osteoma, Gardner's syndrome should be considered as the possible underlying cause. These lesions are slow growing, well-circumscribed and lobulated masses. Complete surgical removal is the ideal treatment choice and the recurrence is very rare.

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Conflict of interest

None declared

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