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Case Report | Olgu Sunumu

A Rare Osteochondroma of the Maxilla: A Case Report and Literature Review

Nadir Görülen Maksiller Osteokondroma: Olgu Sunumu ve Literatür İncelemesi

ABSTRACT

Osteochondroma is a benign osteocartilaginous tumor that is rarely encountered in the maxillofacial region, with localization in the maxilla being particularly uncommon. We present the case of a 29-year-old asymptomatic male patient with an osteochondroma arising from the left maxillary tuberosity and extending inferiorly and posteriorly, resulting in morphological alterations along the anterior border of the left mandibular ramus. The distinctive feature of this case is its resemblance to Jacob's disease through a mechanism that is anatomically opposite to the classical presentation of the condition.

Conclusion: The case was comprehensively evaluated with particular emphasis on its radiological characteristics and imaging findings and was compared with similar cases reported in the literature.

Key Words: Benign Tumors, Osteochondroma, Maxillary Osteochondroma, CBCT.

ÖZ

Osteokondroma, maksillofasiyal bölgede nadir görülen benign osteokartilajinöz tümörlerden biridir ve maksillada lokalizasyonu oldukça nadirdir. Bu olgu sunumunda, sol maksiller tüber bölgesinden inferior ve posterior yönde uzanarak sol mandibular ramusun ön sınırında morfolojik değişikliklere neden olan osteokondroması bulunan, 29 yaşında asemptomatik bir erkek hasta değerlendirilmiştir. Sunulan vakanın özgün yönü, klasik Jacob hastalığında görülen anatomik ilişkinin tersine bir mekanizma ile Jacob hastalığını andıran bir tablo oluşturmasıdır.

Sonuç: Olgu, radyolojik bulguları ve görüntüleme özellikleri açısından ayrıntılı olarak analiz edilmiş ve literatürde bildirilen benzer vakalarla karşılaştırılmıştır.

Anahtar Kelimeler: Benign Tümör, Osteokondroma, Maksiller Osteokondroma, KIBT.

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INTRODUCTION

Osteochondroma (OC) or osseocartilaginous exostosis is an exophytic bone lesion covered with cartilage. Although it is one of the most common benign tumors of the skeletal system, its incidence in the maxillofacial region is less than 1% (1-3). OC generally affects young adults between the ages of 10-20, but most maxillofacial OC cases have been reported in female patients between 30 and 40 years of age (4,5). Osteochondromas can be found in the skull base, maxillary sinus, zygomatic arch, and mandible. In the mandible, they are most commonly seen on the condyle or coronoid processes, probably because the embryological development of the temporomandibular joint occurs via endochondral ossification (6-8). Although it is a matter of debate whether OC is a developmental or neoplastic formation, trauma, and inflammation are among the predisposing factors (5,6,9). Osteochondromas are usually asymptomatic, but when they reach considerable dimensions, they can cause aesthetic problems and pain depending on the area where they are located. In such cases, the treatment option is surgery (10-12). The recurrence rate for OC is low and although it is a benign tumor, it can rarely show malignant transformation (13,14). Panoramic radiography may be sufficient for the initial evaluation in cases of OC in the maxillofacial region, but for detailed evaluation, examination with Cone Beam Computed Tomography (CBCT) is more useful in terms of the size and extent of the lesion and its relationship with the surrounding tissues (15).

CASE REPORT

This case report aims to describe the clinical and radiographic features, differential diagnosis, and treatment of a rare case of maxillary osteochondroma.
A 29-year-old male patient presented to the Oral and Maxillofacial Radiology Clinic of Bolu Abant İzzet Baysal University for routine examination. Following clinical examination, a panoramic image was taken for radiological evaluation, and a radiopaque structure with a pedunculated appearance and bone density extending inferiorly and posteriorly from the left maxillary tuberosity, parallel to the anterior border of the left mandibular ramus was noted. This structure appeared to induce morphological remodeling in the anterior border of the left mandibular ramus (Figure 1).

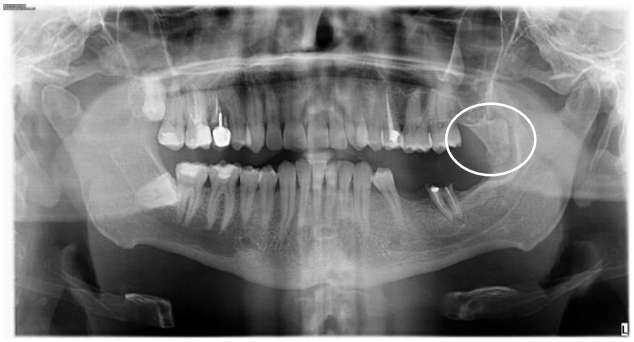

Fig. 1. Panoramic image of the OC in the left maxilla posterior region taken in our clinic during the diagnosis process.

Following identification of the lesion on the panoramic radiograph, the left maxillary tuber region and left dentoalveolar structures were re-examined intraorally. No expansion, morphological change, or pathological findings were found on the intraoral examination. It was determined that the lesion was present in the same location and a similar form on the panoramic radiograph taken 2 years before the current panoramic retrieved from the patient's previous radiographic records (Fig. 2).

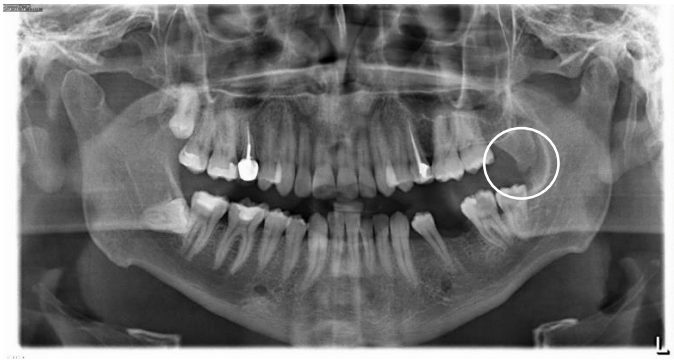

Fig. 2. Panoramic image of the OC in the left maxilla posterior region taken 2 years before the diagnosis process.

However, since the vertical angle and magnification ratio were different in the two panoramic views and standardization was not achieved, the dimensional change of the lesion could not be clearly interpreted. The patient underwent CBCT for detailed examination (Fig. 3-4). In the CBCT sections, an oval radiopaque lesion was observed, measuring approximately 19 × 17 × 13 mm, expanding from the left maxillary tuber region towards the buccal, lateral, and posterior directions, with regular and well-defined borders, surrounded by a thin sclerotic margin except at its junction with the maxillary bone. At this junction, the lesion and the bone trabeculae of the maxilla have fused and the border cannot be clearly observed.

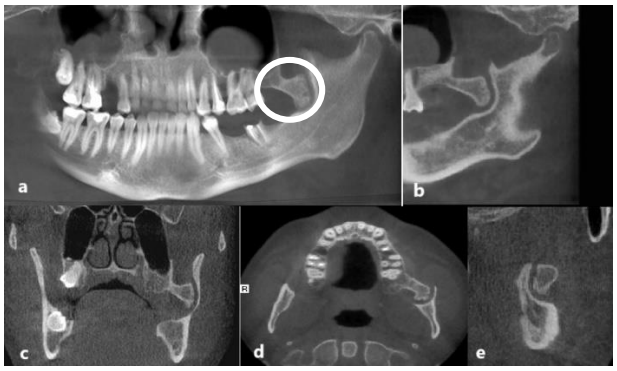

Fig. 3. CBCT images of the patient; (a) Crossectional panoramic view, (b) Reconstructed sagittal section view, (c) Coronal section view, (d) Axial section view, (e) Sagittal section view.

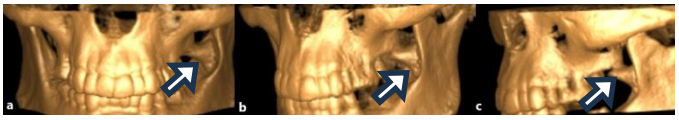

Fig. 4. 3D volumetric CBCT images of the patient (a) anterior, (b) oblique and (c) lateral view.

The lesion exhibited a homogeneous internal structure, and the internal structure of the lesion contains bone trabeculae. Mild expansion was observed in the maxilla, buccally and posteriorly, adjacent to the lesion. The lesion was located posteriorly, in close proximity to the ramus, causing a depression approximately 15 mm deep and 25 mm long on the anterior surface of the ramus under the coronoid process. This area on the ramus surface is surrounded by the cortical border. The patient reported no history of trauma and experienced no symptoms during functional activities such as mastication and swallowing. Based on the patient's anamnesis and examination data, a biopsy was requested with a preliminary diagnosis of osteoma. Exostosis, ossifying fibroma, chondroma, and osteochondroma were considered as differential diagnoses. The lesion was removed at the Department of Oral and Maxillofacial Surgery of the same university and the excised specimen was sent to the pathology department for histopathological examination. In histopathological examination, microscopic findings showed an exophytic lesion with a cap-forming cartilage layer and regenerated bone trabeculae under this area, and it was diagnosed as osteochondroma. In the patient's control examination performed 1 year later, no recurrence findings were found in the region examined with panoramic and CBCT. The patient was planned to be followed up at certain periods. (Fig. 5-6-7).

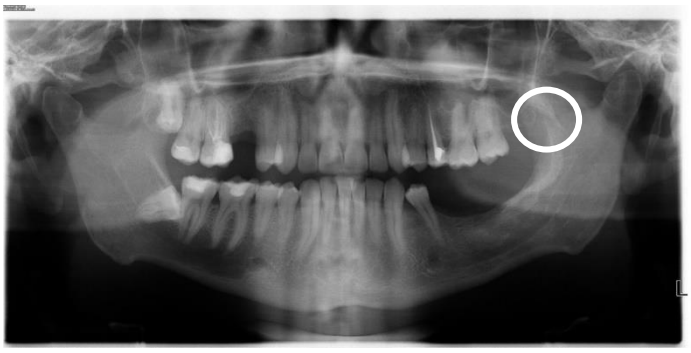

Fig. 5. Control panoramic image of the patient one year after the operation

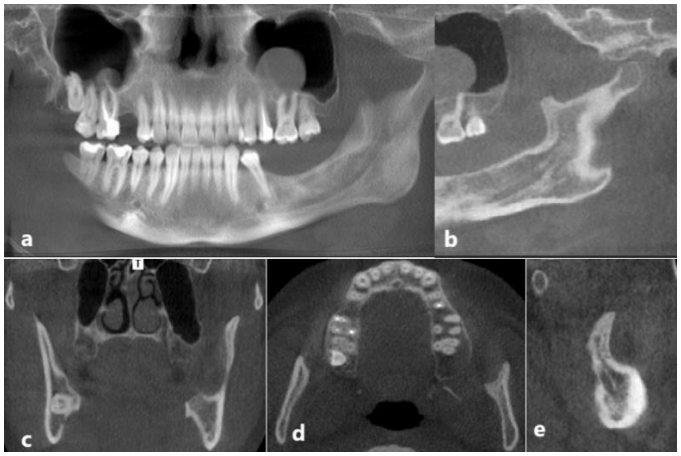

Fig. 6. CBCT images of the patient one year after the operation; (a) Crossectional panoramic, (b) Line section, (c) Coronal section view, (d) Axial section view, (e) Sagittal section view of the concavity in the anterior region of the ramus.

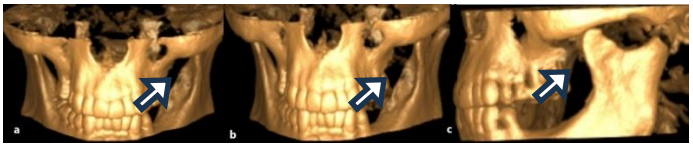

Fig. 7. 3D volumetric CBCT images of the patient one year after the operation (a) anterior, (b) oblique, and (c) lateral view.

DISCUSSION

Osteochondromas are characterized histopathologically by the presence of bone tissue covered with hyaline and fibrous cartilage. In the deep layer where cartilage and bone meet, the endochondral ossification process is observed (16). Histochemical and immunohistochemical analyses of the cartilage layer are an important method to identify the main components of the cartilage matrix. These analyses allow us to examine the components within the cartilage tissue in detail. Among the typical components of the cartilage matrix, cartilage proteoglycans, and type II collagen stand out (17).

The majority of maxillofacial osteochondromas reported to date have involved the mandible, particularly the condyle and coronoid process 5. This supports the theory that OC develops from abnormal foci of epiphyseal cartilage during the endochondral ossification process. However, it is observed that OC also occurs in tissues with intramembranous ossification 16. The underlying mechanism is thought to be the osteocartilaginous differentiation of ectopic mesenchymal cells (18, 19). Osteochondromas must be differentiated from pathologies such as exostosis, osteoma, ossifying fibroma, and osteosarcoma. Oral exostoses are defined as bony proliferations that occur in the cortical plates of both the maxilla and mandible in young individuals of unknown etiology. Lesions typically show slow but continuous growth. Unlike OC, exostoses are more common in the maxilla than the mandible (20). Osteomas are usually benign osteogenic lesions originating from cancellous or compact bone. Clinically, they appear well-defined, usually round and protruding; they are characterized by very slow and continuous growth (21). Osteomas can also be seen as multiple, and in multiple cases, patients should be evaluated for Gardner syndrome (22). Ossifying fibroma is a rare, benign, well-defined osteogenic neoplasm composed of fibrous tissue with variable amounts of calcified tissue, resembling bone, cementum, or a combination of both, and is derived from multipotential mesenchymal cells of the periodontal ligament (23-25). Ossifying fibroma has distinct borders and is separated from the surrounding bone tissue by these borders, and can cause resorption and displacement in neighboring teeth (23). It can be differentiated from OC by radiological features and histopathological examinations; the absence of cartilaginous cells is diagnostic in differentiating it from OC (26). Osteosarcoma is characterized by the production of osteoid by mesenchymal cells and is the most common malignant bone tumor, although jaw lesions are rare, accounting for approximately 4% of all osteosarcomas (27, 28). Although the age range for osteosarcomas seen in the jaws is wide, the incidence increases mostly in patients between the ages of 30-40 (29, 30). The less metastasis and better prognosis seen in osteosarcomas seen in the jaws make them different from osteosarcomas seen in long bones (27, 31). Radiologically, osteosarcomas show destructive growth with periosteal reaction and recurrence is common. Osteosarcomas are distinguished from osteochondromas by the production of malignant osteoid by neoplastic osteoblasts and by their radiological features (32, 33). Considering the similar features of the lesions included in the differential diagnosis, radiological and histopathological examination is required for the diagnosis of osteochondroma. Many cases of osteochondroma localized in the mandibular ramus, condyle, symphysis, coronoid and angulus regions have been presented in the literature. However, there were very few maxillary osteochondroma cases in the maxillary region. When the literature was reviewed, five cases of maxillary osteochondroma were reported between 1962 and 2012 (34-38). When we investigated more recent cases, in 2015, Jain et al. reported a case of osteochondroma located in the posterior right maxilla (39). In 2024, Klein et al. reported a case of osteochondroma in the anterior maxilla (40). It has been observed in the literature that the prevalence of OC in the maxilla is quite low, and it seems that this presented case of OC localized in the left maxillary tuberosity region can be added to the literature because of its unusual anatomical location. In the OC case reports presented in the literature, the lesion may be asymptomatic or symptomatic depending on its size, location, and relationship with adjacent anatomical structures. Mohanty et al. (16) presented a case report in 2016 of a well-defined hyperdense osteochondroma clinically located in the palatal region of the left maxilla posterior, observed as a hard and tender mass on palpation with a normal surface mucosa, originating from the left lateral pterygoid plate on CT and extending to the left retromolar trigone region, containing a medullary cavity measuring 10 mm × 15 mm. In our case, similar to this case, the cortex and medulla of the lesion showed continuity with the adjacent bone (left maxilla tuber region) and this continuity supported the diagnosis of osteochondroma. Our case was asymptomatic and in a similar location to the case reported by Mohanty et al. (16), but its size was larger and no findings were found in the intraoral examination. Our case was noticed incidentally and CBCT examinations showed that a fossa area developed in the ascending ramus adjacent to the osteochondroma, compatible with the lesion. Jacob's disease is a rare entity consisting of the formation of a pseudojoint between an abnormal coronoid process of the mandible and the inner surface of the zygomatic bone (41). Based on these findings, it is hypothesized that the lesion exhibited slow progressive growth, resulting in the development of a pseudoarticulation with the adjacent mandibular ramus and creating a radiological appearance that closely resembles Jacob’s disease, albeit in a reverse anatomical configuration (19, 42, 43). This osseous adaptation may have prevented the development of clinical manifestations such as intraoral swelling, trismus, or asymmetry in the tuber region. In this case report, an asymptomatic maxillary osteochondroma with no intraoral or extraoral clinical manifestations, and was detected incidentally during routine examination was presented. This case highlights the critical role of meticulous radiological assessment in the diagnosis of rare maxillofacial lesions. Exploring rare pathologies, such as benign maxillary

osteochondroma, is crucial as it enables dental professionals to identify diverse clinical presentations, enhance their diagnostic proficiency, and implement timely interventions, thereby reducing the risk of potential long-term complications.

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Data Availability

All the data are available upon reasonable request to the corresponding author.

Declarations

Declarations of interests

I have nothing to declare.

Ethical approval

All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration revised in 2024. IRB review was exempted because this paper is a case report. All procedures were performed in compliance with relevant laws and guidelines of Bolu Abant İzzet Baysal University.

Informed consent

Written informed consents were obtained from the patient for academic use of medical information and publication in journals.

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