

Mandibular Osteosarcoma with a Preliminary Diagnosis of Ossifying Fibroma: Case Report

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ABSTRACT

Osteosarcoma is the most common primary malignant bone tumor, and most often occurs in the metaphysis of long bones of the extremities. Osteosarcomas seen in the craniofacial bones account for only 5-13% of all cases, and generally appear in the mandible and maxilla. Ossifying fibroma is a benign, non-odontogenic, expansile but commonly slow-growing lesion of the mandible. Patients with mandibular osteosarcoma usually apply with a painful swelling in the jaw over the previous 2-3 months. The swelling could easily be misdiagnosed as a benign lesion. Computerized tomography (CT) scan is the most helpful imaging method for such cases, providing an excellent view of the lesion and its correlation with bony structures. Thus, CT scan must be performed immediately in all suspected cases for early diagnosis. This article presents the case of a 44-years-old female patient with mandibular mass, which was considered a malignant tumor but was misdiagnosed as ossifying fibroma according to the pathological analysis of the incisional biopsy material.

Key words: Mandibular neoplasms, Craniofacial osteosarcoma, Ossifying fibroma

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ÖZET

Ossifiye Fibrom İnsizyonel Biyopsi Tanılı Mandibuler Osteosarkoma: Olgu Sunumu

Osteosarkom, en sık görülen primer malign kemik tümörüdür. En çok ekstremitelerin uzun kemiklerinin metafizinde meydana gelir. Kraniofasial kemiklerden kaynaklanan osteosarkom ise tüm olguların ancak %5-13'üdür ve yerleştiği kemikler genellikle mandibula ve maksilladır. Ossifiye fibrom ise mandibulanın benign, non-odontojenik, ekspansil ancak sıklıkla yavaş büyüyen bir lezyonudur. Mandibuler osteosarkomlu hastalar, genellikle 2-3 aylık çenede ağrılı şişlik öyküsüyle başvurur. Bu şişlik, kolaylıkla benign bir lezyon olarak yorumlanabilir. Bu tür olgularda; kitleyi ve kemikle ilişkisini oldukça iyi gösteren bilgisayarlı tomografi en yararlı görüntüleme yöntemidir. Erken tanı için, şüphelenilen olgularda mutlaka gecikmeden bilgisayarlı tomografi çekilmelidir. Bu makale, radyolojik tetkikleri sonucu malign olduğu düşünülen ancak yapılan insizyonel biyopsinin patolojik incelenmesi, ossifiye fibrom olarak raporlanan mandibuler kitleli 44 yaşındaki kadın hasta sunulmaktadır.

Anahtar kelimeler: Mandibuler kitleler, Kraniofasial osteosarkom, Ossifiye fibrom

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INTRODUCTION

Osteosarcoma (OS) is a malignant mesenchymal tumor, in which there is production of osteoid matrix and bony tissue by sarcomatous connective tissue cells. It is the most common primary malignant bone tumor. These tumors are typically seen in the long bones of the extremities, with only 5% of OSs located in the jaw. OSs are usually observed in the 3rd and 4th decades. The male/female ratio is 2/1^[1]. The precursor factors are exposure to radiation, trauma and bone pathologies such as fibrous dysplasia and Paget's disease.

The tumors of the mandible are divided as odontogenic and non-odontogenic. The primary malignant non-odontogenic tumors of the mandible are rare and aggressive in nature, with poor prognosis. Among those are OSs, chondrosarcomas, fibrosarcomas, Ewing's sarcoma, Burkitt lymphoma, plasma cell neoplasms, and malignant lymphomas. The clinical symptoms of the mandibular OSs are rapid swelling and pain in the affected site. Paresthesia and mobility of the teeth at the affected site are among the other clinical symptoms. Sixty percent of mandibular OSs are observed in the body of the mandible.

On the other hand, ossifying fibroma is among the benign non-odontogenic fibroosseous lesions of the mandible. They originate from undifferentiated cells of the periodontal ligament. Histopathologically, they are composed of fibrous tissue and of varying amounts of calcified material. They are slow-growing and expansile lesions. They are frequently asymptomatic, and if left untreated, may cause facial asymmetry.

In this case report, we aimed to present the findings, clinical findings and treatment of our mandibular OS patient, who was initially diagnosed as ossifying fibroma.

CASE REPORT

A 44-year-old female patient was admitted to the Clinic of Otorhinolaryngology (ORL) with complaints of swelling and pain on the right mandible for the past month. On the physical examination, a stiff mass was found, located in the posterior one-third of the right mandibular body, at the level of the 3rd molar tooth, showing expansion extending to the zygomatic arch superiorly and fixed to the mandible (Figure 1). Maxillofacial computerized tomography (CT) and facial magnetic resonance imaging (MRI) revealed a wide destructive mass lesion, characterized by a soft tissue component, 4.5 x 4.5 x 7 cm in size, destructing and expanding the right mandibular body, extending

from the masseter muscle laterally, temporal muscle medially, up to the lateral and posterolateral border of the right maxillary sinus, and causing thinning of the lateral wall of the maxillary sinus (Figures 2, 3). The pathology of the incisional biopsy taken from the mass lesion was reported as fibroosseous lesion, and in the pathology report, it was noticed that it was most likely an ossifying fibroma. The patient was referred to us from the ORL clinic, and subsequent surgical treatment was undertaken by our clinic. During the surgical intervention, the mass lesion was excised together with the 3rd molar tooth (Figure 4). Additionally, the superior half of the bony tissue of the posterior one-half segment of the right mandibular body, anterior half of the bony portion of the inferior two-thirds of the ramus and the whole coronoid were excised (Figures 5, 6). Mandibulectomy was not done. Based on the pathology report that defined the lesion as benign according to the incisional biopsy, and considering the fact that it has low rates of postoperative morbidity, the mass lesion was just resected from its boundaries. Subsequent intermaxillary fixation was done. After the pathological examination, the mass was reported as OS. Observing tumor tissue as close

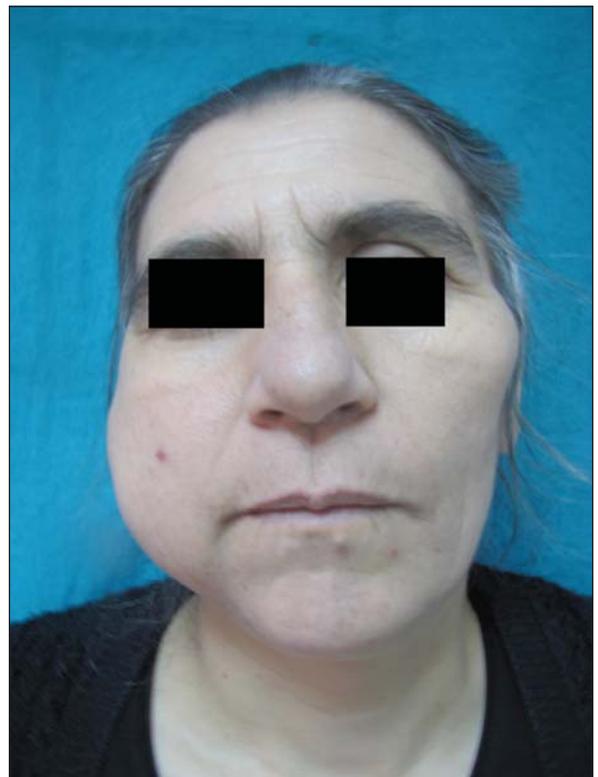


Figure 1. Pre-operative appearance of the mandibular osteosarcoma.

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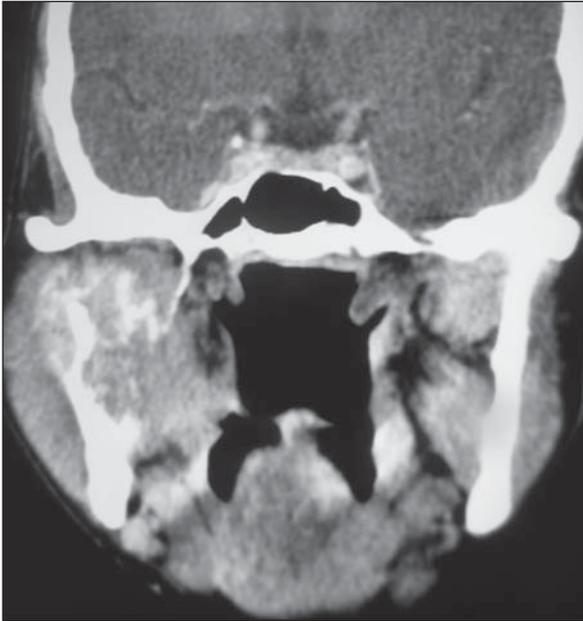


Figure 2. CT imaging of the mass.

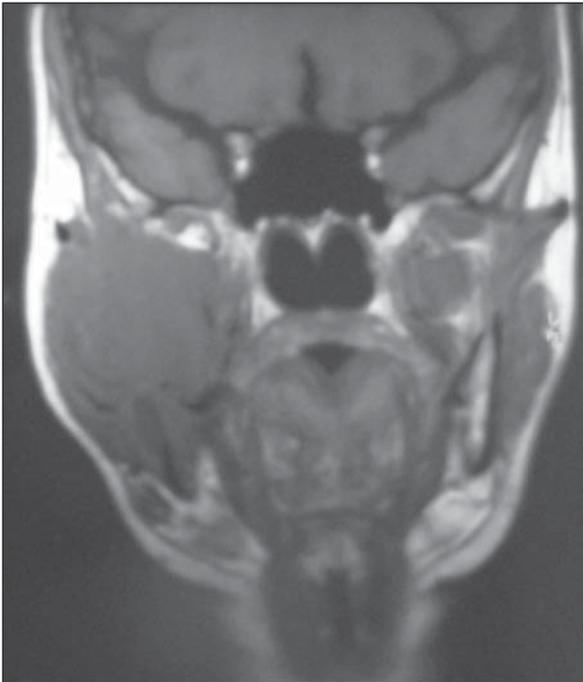
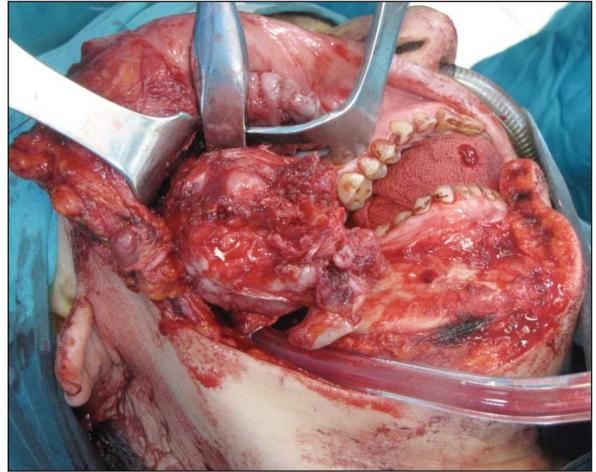


Figure 3. MRI of the mass.

to the anterior and superior surgical margins of the mass as 0.1 cm, partly adjacent to the posterior, lateral and medial surgical margins, and totally adjacent to the inferior surgical margin, the patient was presented to the Tumor Council, which included the



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Figure 4. Intraoperative appearance of the mandibular osteosarcoma.



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Figure 5. Intraoperative view of the mandible after the first bone resection.

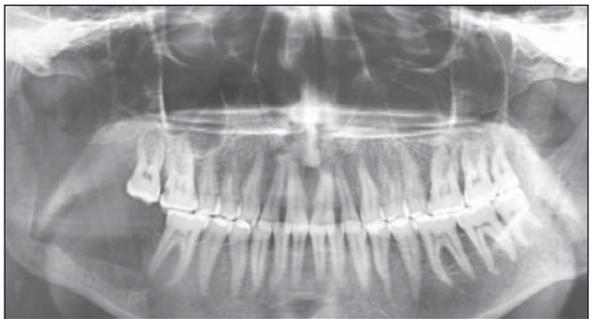


Figure 6. Postoperative radiography of the mandible after the first bone resection.

Plastic Surgery, ORL, Oncology, and Pathology Clinics. It was decided to proceed with systemic chemotherapy after complementary surgical intervention. The patient was prepared for hemimandibulectomy.



Figure 7. The radiography of the mandible after hemimandibulectomy.

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Figure 8. Image of the patient 2 weeks after surgery.

tomy. Meanwhile, the necessary imaging was done to rule out possible remote metastases, but nothing was found in favor of metastases.

The mandible was excised in total from the right subcondylar region up to the right parasymphysis. As reoperation will be required in case of tumor positivity in the surgical margins, bone reconstruction was not planned after hemimandibulectomy in the early stages. As a space occupier, a reconstruction plate was placed between the subcondylar region and parasymphysis (Figures 7,8). No additional surgical resection was performed, since the surgical margins were free of tumor, even though the pathological examination of the mandibulectomy material implicated tumor tissue. The patient was referred to the Medical Oncology Clinic for further chemotherapy. After completion of oncologic treatment, bone reconstruction was planned.

DISCUSSION

OS has a high potential for malignancy. OSs originating from the maxilla and mandible in comparison to those that originate from long bones are mostly observed in later adulthood (3rd and 4th decades) and rarely metastasize. Although maxillofacial OSs do not show the same aggressive course as in long-extremity OSs, early diagnosis is very important for better aesthetic results^[2]. It is stated that the most important negative prognostic factor for OSs is insufficient radical surgical intervention^[3]. Local recurrence is the most important cause of mortality. It is reported that five-year survival for mandibular OSs is 37% and for maxillary OSs is 27%^[4]. Radiological findings differ, depending on the calcified component of the lesion. The tumor can be seen as totally radiolucent or radiopaque, as well as with mixed pattern. One of the radiological clues of early-stage OS is symmetrical widening in the periodontal membrane space of affected teeth. A “sunburst” pattern, which is the radiographic appearance of soft tissue components as a result of ossification, is classical for OSs, but it is neither specific nor sensitive.

In our case, planning the surgical treatment of the patient in accordance with the diagnosis, based on incisional biopsy, led to the need for complementary surgical intervention. From this experience, we learned that, in the case of mass lesions originating from maxillofacial bony structures, it is not always optimal to rely on the pathological diagnosis made by incisional biopsy, and interventional treatment should be designed taking into consideration the clinical

course of the mass lesion and radiological examinations. In expansile mass lesions of the mandible, in order to increase the probability of precise pathological diagnosis of incisional biopsies, it is very important to obtain the biopsy material from the deepest portions of the mass, and it should be as abundant as possible^[5]. Superficial biopsy materials may be reported as benign lesions, as they may not carry malignant features. This, in turn, can misguide the surgical team and may lead to inevitable delay in radical surgical treatment of the patient. Ossifying fibroma, which was the initial diagnosis made by incisional biopsy in our case, is one of these benign lesions. Unfortunately, since the biopsy was not made by our clinic, the precise information regarding the depth and size of the biopsy sample was lacking.

In conclusion, based on our experience and the literature review, we advise our colleagues that in such cases initially suspected as malignant lesions based on the clinical history and radiological imaging but reported as benign lesions based on incisional biopsy, the surgical treatment algorithm should be planned based on the pathological diagnosis made by frozen section examination of the sample obtained by intra-operative total excision of the mass lesion.

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