Multi-centric Giant Cell Tumor of the Bone: A Rare Case Report

Kemiğin Multisentrik Dev Hücreli Tümörü: Nadir Görülen Bir Olgu Sunumu

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ABSTRACT

The giant cell tumor (GCT) of the bones is a benign but locally aggressive neoplasm that affects the epiphyseal-metaphyseal junction of long bones. It makes %5 of the bone tumors. Multi-centric giant cell tumor (GCT), consists %1 of the GCT of the bones (3, 4). The pathogenesis has not been identified yet because of the rarity of the multi-centric GCT. Paget disease, Brown tumor, and fibrous dysplasia should come up to mind at clinically and radiologically differential diagnosis. Definitive diagnosis is established by histopathological examination. The potential for the malignancy of giant cell tumors is yet to be clarified. Even the type of treatment is controversial the common surgical treatment is aggressive curettage with intra-operative grafting or cementing. The articles about multi-centric GCT are limited in the literature.

Keywords: Giant cell tumor, Curettage, Denosumab, Multicentric, Liquid nitrogen, Cementing

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ÖZET

Kemiğin dev hücreli tümörü (DHT) uzun kemiklerin epifizometafizyel bileşkesini etkileyen iyi huylu fakat lokal olarak agresif bir neoplazi türüdür ve tüm kemik türlerinin %5'ini oluşturmaktadır. Multisentrik DHT ise tüm kemik DHT'lerinin yalnızca %1'ini oluşturmaktadır. Nadir görülmesinden dolayı multisentrik DHT'nin patogenezi tam olarak aydınlatılamamıştır. Ayırıcı tanıda klinik ve radyolojik olarak Paget hastalığı, Brown tümörü ve fibröz displazi akla gelmelidir. Kesin tanı histopatolojik inceleme sonrası konulabilmektedir. DHT'lerin malignleşme potansiyeli henüz tam olarak belirlenememiştir. Tedavi tipinde görüş birliği sağlanamamış olsa da en sık uygulanan cerrahi tedavi türü agresif küretajı takiben intraoperatif graftleme ya da kemik çimentosu uygulamasıdır. Literatürde multisentrik DHT ile ilgili yayınlar oldukça sınırlıdır.

Anahtar Sözcükler: Dev hücreli tümör, Küretaj, Multisentrik, Çimentolama, Sıvı azot, Denosumab

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INTRODUCTION

The giant cell tumor (GCT) of the bones is a benign but locally aggressive neoplasm that affects the epiphyseal-metaphyseal junction of long bones. It most frequently occurs at 20-40 years old young adults. It rarely occurs before the maturation of epiphysis. GCT of the bones is slightly more common in females than the male gender. It consists the %5 of the bone tumors(1, 2).

Multi-centric giant cell tumor (GCT), consists %1 of the GCT of the bones(3, 4). The pathogenesis has not been identified yet because of the rarity of the multi-centric GCT. Paget disease, Brown tumor, and fibrous dysplasia should come up to mind at clinically and radiologically differential diagnosis. Definitive diagnosis is established by histopathological examination. Even the type of treatment is controversial the common surgical treatment is aggressive curettage with intraoperative grafting or cementing(5, 6). The articles about multi-centric GCT are limited in the literature. In this case report, we present a 27-year-old patient with a multi-centric GCT affecting the right knee.

CASE REPORT

A 27-year-old female patient was referred for pain and giving way feeling on the right knee. There was no history of trauma and family history. In physical examination even the range of motion was full it was painful and she had difficulty of weight-bearing. She had these complaints for three months and it was worsened over time. The x-ray (Figure 1) has shown cystic lesions expanding through osseous structures at the epiphyseal-metaphyseal junction of the right distal femur, the epiphyseal-metaphyseal junction of the proximal tibia and patella. The patient's laboratory findings were normal and no other lesions were found at thyroid ultrasonography and whole-body bone scintigraphy. Magnetic resonance imaging (MRI) (Figure 2) has shown lytic-destructive zones consisting of widespread cystic areas which cranial-caudal size reaching 7.5 centimeters (cm) at the epiphyseal-metaphyseal junction of the right distal femur. Another tumoral lesion has also been found at the epiphyseal-metaphyseal junction of the proximal tibia similar to the lesion defined in the femur. Also, another lesion has been found at the inferior-lateral section of the patella, characteristically expansive and consisted cystic component with septations.

An incisional biopsy was performed at the distal femur and proximal tibia. The result came as a proliferative lesion consists osteoclastic giant cells and a fibrohistiocytic component (Figure 3). Possible diagnosis was multi-centric GCT or hyperparathyroidism due to the multiple numbers of lesions. As a result of normal blood parathyroid levels and thyroid USG, with a pre-diagnosis of Multi-centric GCT, aggressive curettage with liquid nitrogen and cementing and internal fixation was performed (Figure 4).

No complication has occurred intraoperatively and postoperatively. On the postoperative first day after the drainage was removed, the patient was mobilized without weight-bearing. Rehabilitation was started. At the following third month; 8*6*5 cm size lesion completely invading the right acetabular roof with loss of cortical unity, ranging iliac fossa anteriorly and gluteal muscles, consisting cystic-necrotic zones and causing significant expansion of osseous structures were found (Figure 5). Denosumab was started after the result of the incisional biopsy came as GCT. After 1 year treatment, there was no growth in the lesions or no new lesion was occurred (Figure 6).

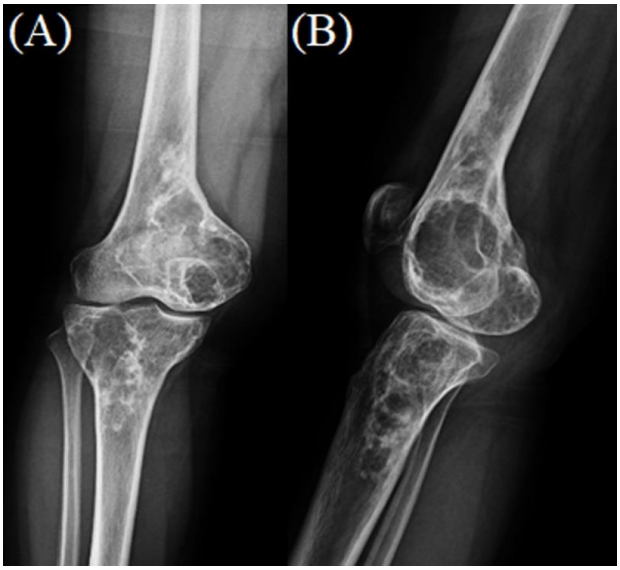

Figure 1: Cystic lesions resulting expansions of osseous structures at the epiphyseal-metaphyseal junction of the right distal femur, proximal tibia, and patella in anteroposterior (A) and lateral view (B)

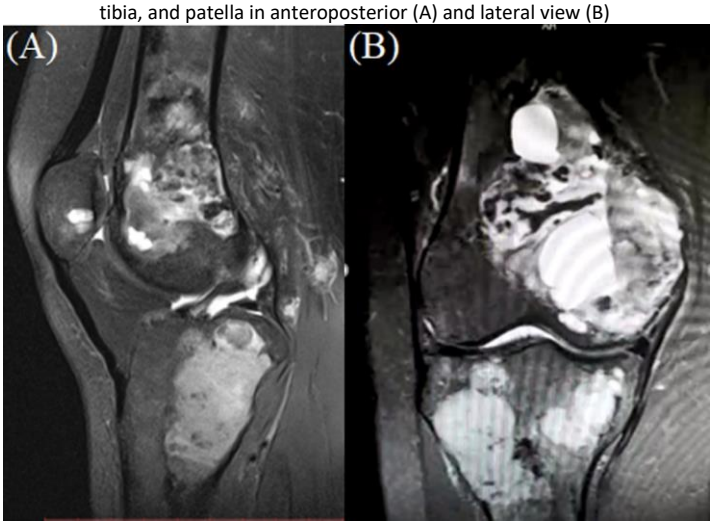

Figure 2: Lytic-destructive zones consisting of widespread cystic areas which craniocaudal size reaching 7.5 centimeters (cm) at the sagittal (B) coronal (A) plane of MRI scan

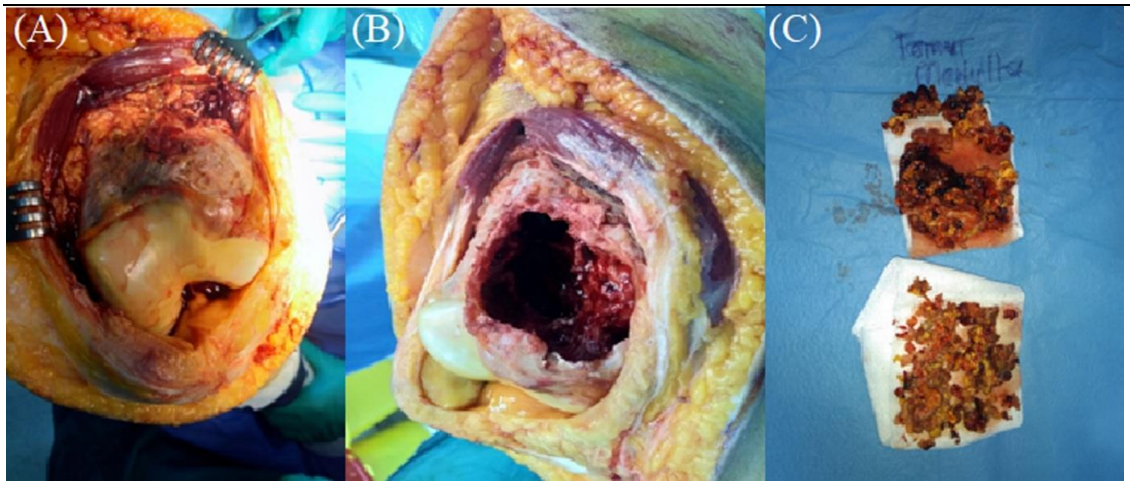

Figure 3: Intraoperative curettage and macroscopic image of the lesion

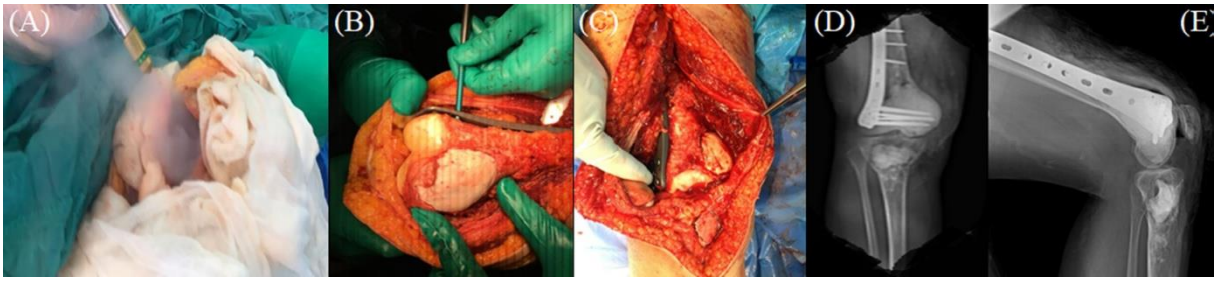

Figure 4: Intra-operative images of liquid nitrogen application (A), cementing (B), and internal fixation (C) with postoperative radiographs (D, E)

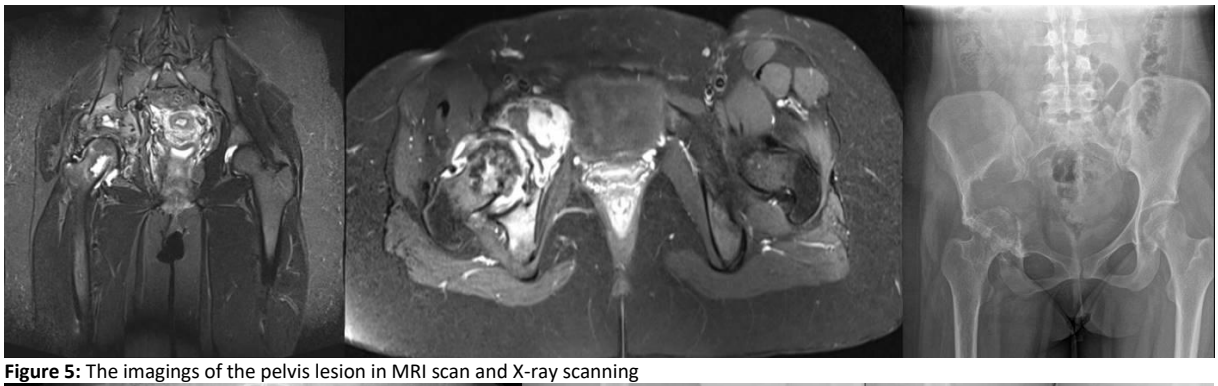

Figure 5: The imagings of the pelvis lesion in MRI scan and X-ray scanning

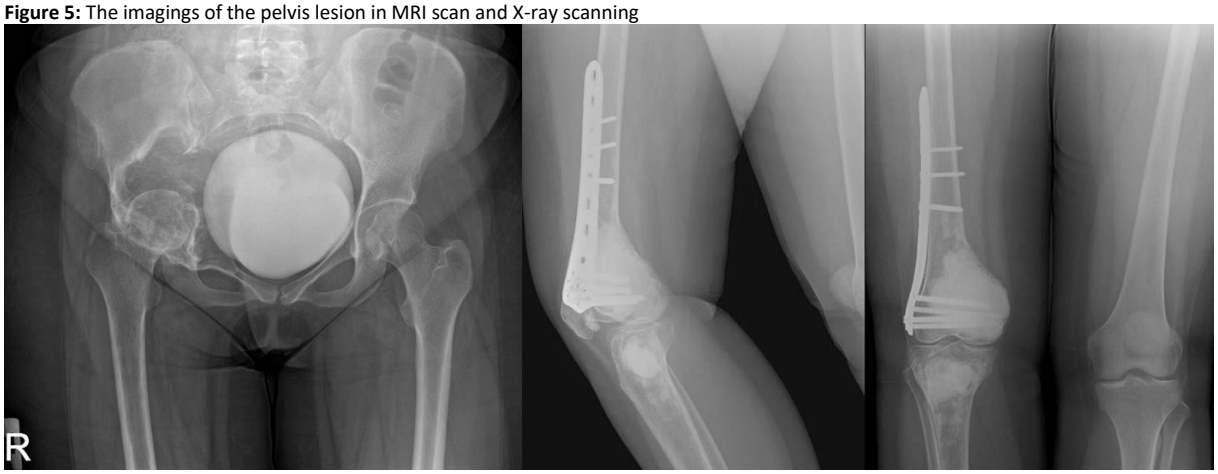

Figure 6: The imagings of both lesion after 1 year treatment of denosumab

DISCUSSION

Multi-centric GCT consists of % 1 of the total GCT of the bone (4). At differential diagnosis, hyperparathyroidism, Paget disease, and fibrous dysplasia should be considered. It is differentiated with normal blood calcium, phosphor, alkaline phosphatase, and parathyroid hormone level with normal thyroid USG and identification of osteoclastic giant cells in the pathological examination.GCT of the bones more commonly occurs at the ages between20-40 years old (7). Multi-centric GCTs can occur at slightly earlier ages, Dhillon et al. reported the mean age of Multi-centric GCT as 22,5 (9-62)(4). However even rare it can occur before the closing of the epiphyseal plate(8).In the literature, 82 patients' gender was reported as 44 female and 38 male(4). Our patient was a 27-year-old female.Multi-centric GCTs also most frequently occur in the knee area(4, 9). While the solitary GCT is more commonly located on epiphysis; multi-centric is more commonly located on the metaphysical segment(4, 10). In our patient GCT was placed on the knee and epiphyseal- metaphysical zone.Dhillon et al. has reported Multi-centric GCT has also another coexisting lesion(4). In our case, a new lesion was diagnosed in the right acetabulum at the third-month follow-up.The amounts of lesions of multi-centric DHTs are variable. In literature; 314 lesions have been reported at 83 cases. Most frequently 2 lesions (at 37 patients) have been reported(4). Our patient had 4 lesions.

The malignity rate of solitary GCT is below %1. The malignity rate of multi-centric GCT is reported as % 6,7 by Hoch et. al (3). However, all the other researchers have shown the malignity rate is not increased at Multi-centric GCT. However, all the other researchers have shown the malignity rate is not increased at Multi-centric GCT. Stratiland Stacy has asserted that Multi-centric GCT has no more potential of malignancy than solitary GCT(12). Our case has shown no potential for malignity.It is reported that the rate of pulmonary metastasis in solitaryGCT is between % 2-9 (8, 13). Dhillon et. al. have reportedthe rate of pulmonary metastasis of multi-centric GCT as % 5 with no increased risk of pulmonary metastasis(4).No pulmonary metastasis has occurred in our case.

In conclusion, Multi-centric GCT has the same pathologicalandclinicalfeatures with solitary GCT. Malignity potential andthe risk of metastasis is not higher than solitaryGCT. In case of the possibility of new lesion patients' follow ups should not be terminated and continue periodical examination.

Conflict of interest
No conflict of interest was declared by the authors.

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