CASE REPORT

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Reconstruction with Vascular Fibular Graft for a Patient with Recurrent Giant Cell Bone Tumor in Distal Ulna, a Rare Treatment Option

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ABSTRACT Giant cell bone tumors are benign lesions of the bone and they usually behave locally aggressive in the follow ups. The most common location of giant cell bone tumor is the metaphysis of distal femur, proximal tibia, distal radius, and proximal humerus. They rarely metastasize, and surgical methods are prominent in their treatment. Among the surgical treatments, curettage, cementation, and in some selected cases, reconstruction with vascular fibula graft can be applied. In addition to surgical treatments, denosumab treatments have also been put into practice. In this article we discuss a patient undergoing 3 surgeries due to giant bone cell tumor in right distal ulna in the forearm. We are going to discuss a patient that we performed a reconstruction operation with vascular fibula graft who had been operated on for giant cell bone tumor in his right ulna twice and received denosumab treatment and had good postoperative follow-up and functional results.

Keywords: Giant cell bone tumor; denosumab; locally aggressive; vascular fibular grafting

Giant cell tumor is a locally aggressive intermediate osteolytic tumor of bone. It accounts to 5% of all tumors and 20% of all benign bone tumors. Although rarely lethal, benign bone tumors may be associated with a substantial disturbance of the local bony architecture that can be particularly troublesome in peri-articular locations.[1] Its pathogenesis is characterized by a proliferation of mononuclear stromal cells and the presence of many multi- nucleated giant cells with homogenous distribution.[2] Over 80% of giant cell tumors present histone variants in the *H3F3A* gene.

Most giant cell tumors are benign tumors and occur near a joint in young adults. For this reason, many authors prefer intralesional surgical approaches that preserve bone anatomy over resection.[3] Around 5% present metastatic nodules whose most common site is lungs. The main treatment is surgery. Chemotherapy and radiotherapy options were applied in selected patients. En bloc resection is recommended in expendable bones and bones of the hand. In addition to these treatments, denosumab treatment has recently been applied to patients.[4,5]

But it is shown that local recurrence is a common problem in patients using denosumab.

Histological grading has no prognostic value.[6,7] Local recurrence depends on the stage treatment. Its course is unpredictable if untreated.

In this report we present a case with a reconstruction operation with vascular fibula graft on a patient who had been operated for giant cell bone tumor in his right ulna twice and received denosumab treatment and had good postoperative follow up and functional results.

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A 20 year old female patient applied to our hospital because of the pain in her right forearm. We learned that patient had been previously operated 2 times and

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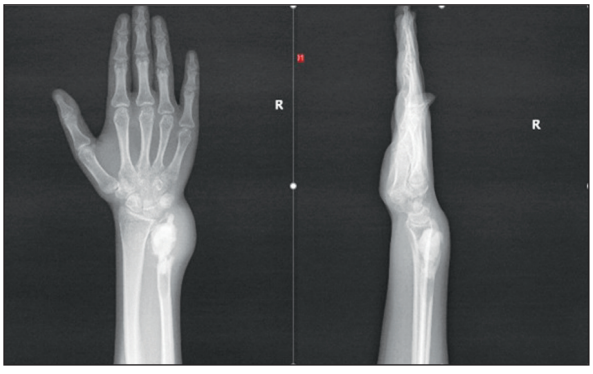

FIGURE 1: Direct X-ray of the patient with local recurrence of giant cell bone tumor in distal ulna, in the preoperative period.

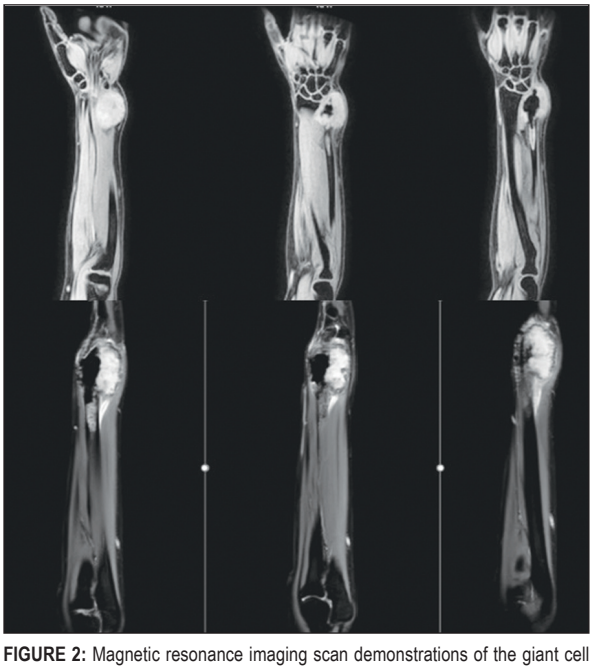

FIGURE 2: Magnetic resonance imaging scan demonstrations of the giant cell bone tumor in distal ulna after 2 surgeries and denosumab treatment. In the figures, we can see the giant cell bone tumor originating from distal ulna and spreading to the soft tissues around the wrist joint.

also taken denosumab treatment in a different hospital. At the state of admission, latest biopsy which was performed was reported as secondary proliferative changes due to denosumab use. Informed consent both from the patient and the doctors were taken before treatment.

After 2 surgeries and 6 cures of denosumab treatments, the patient applied to our hospital with pain in forearm. After making radiological assessments which were demonstrated above, patient was diagnosed with local recurrence of giant cell bone tumor in distal ulna (Figure 1, Figure 2). After the multidisciplinary sarcoma meeting which occurred in our hospital wide resection and reconstruction with bone graft decision was given. The patient underwent reconstruction with a vascular fibula graft surgery with the participation of the plastic and cardiovascular surgeons. Despite from the commonly used surgical techniques, this surgery was done with a different method. Mariappan et al. made a distal ulna reconstruction using proximal fibula and triangular fibrocartilage complex (TFCC) reconstruction using palmaris longus tendon following wide resection of giant cell tumor of distal ulna.[7] We performed a different technique because the patient didn't have palmaris longus muscle, with still using proximal fibula as a bone graft. Intraoperatively we noticed that the lesion was surrounding the extensor carpi ulnaris muscle 360 degrees and it was removed with the excision of the proximal and the distal ends of extensor carpi ulnaris tendon while preserving the ulnar nerve (Figure 3). Soleus fascia was than applied to the distal radius with one suture anchor. After this step, distal part of the vascular fibular autograft from left fibula diaphysis was reshaped as a new distal radioulnar joint with the help of high-speed burrs and was applied with 1 plate and 4 screws. The fibular vessels were moved through the subcutaneous tissue and was anastomosed with the ulnar vessels (Figure 4). After the anastomosis was performed, extensor carpi ulnaris muscles proximal end and extensor digiti minimi muscles distal end was sutured with the application of soleus fascia graft using 1 suture anchor to maintain distal radioulnar joint stability. As a result, fibulas distal end was connected with the extensor carpi ulnaris muscles distal end with a suture an-

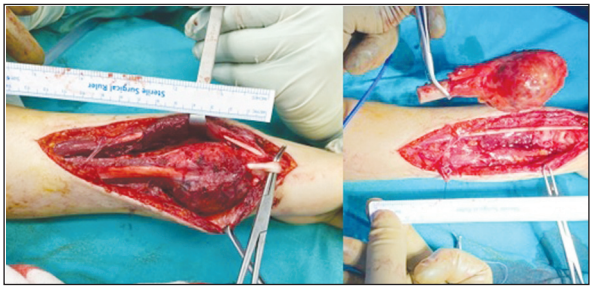

FIGURE 3: In the first image we can see that the lesion includes the extensor carpi ulnaris tendon. In the second image we can see the lesion after removal from the distal radioulnar joint including the extensor tendon ulnaris. Extensor carpi ulnaris lesion was excised because we saw the tumor surrounded the tendon 360°.

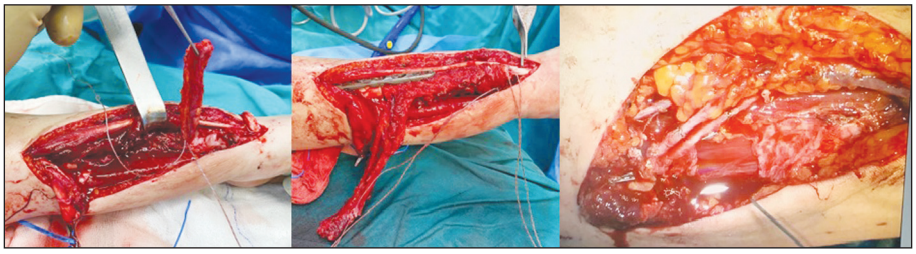

FIGURE 4: In the first image, we can see the application of the soleus fascia to suture with the extensor carpi ulnaris and extensor digiti minimi muscles. In the second image we can see the fixation with the plate and the soleal fascia. In the last image on the bottom we can see anastomosis of the fibular vessels with the ulnar artery and veins. The vessels were moved under the subcutaneous tissue.

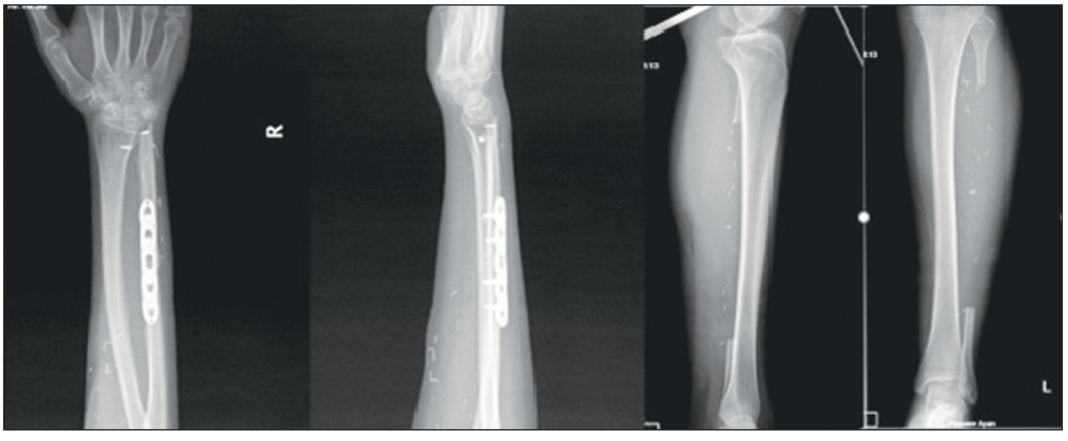

FIGURE 5: Direct X-rays of the patient who underwent a reconstruction by vascular fibula graft for cell bone tumor in the right ulna distal in the post-operative period.

chor and extensor digiti minimi muscle was sutured to extensor carpi ulnaris muscles proximal end. Unlike the previous surgery that we mentioned, we did not perform z-plasty, the patient didn't have palmaris longus muscle. The surgery was completed around 11 hours.

There was no discharge from the wound site and there were no other complications in the postoperative follow-up period of 12 months (Figure 5). The radial, ulnar and median nerves were intact, and their functions were preserved (Figure 6).

DISCUSSION

Giant cell bone tumors do not respond well to chemotherapy and radiotherapy and are therefore treated surgically. Prognosis is related with the distal organ metastasis. Most common metastasis site is lungs. Surgical treatment may vary depending on the localization of the lesion. Most common localization of giant cell bone tumors are knee joint in the lower extremity and distal radioulnar joint in upper extremity.[1,2]

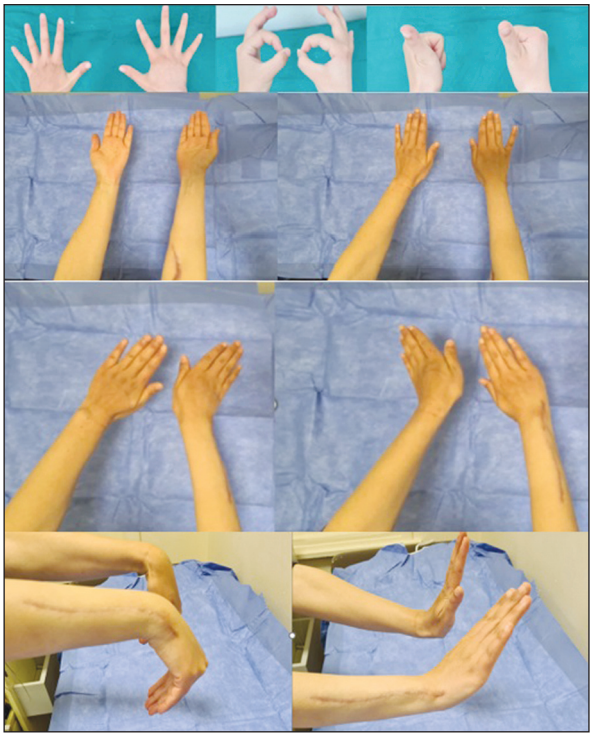

FIGURE 6: In the postoperative follow up, we can see the wrist range of motion is nearly complete and without pain. We observed that radial, ulnar and median nerve functioned properly. Patient was able to fully pronate and supinate the forearm. Dorsiflexion and palmar flexion of the hand is nearly same as the healthy hand.

Ulna is a less common site for this tumor. In the upper extremity, it usually occurs in distal radius and they are treated mostly with curettage and cementation or wide resection, and sometimes reconstruction techniques like vascular fibular grafting surgery. The latter appears to be a good option for recurrent lesions.[3]

Most commonly, the giant cell bone tumors are seen in younger patients between age 20-40. Because of this, some authors believe that vascular fibular grafting surgeries would give better results in general compared to elderly patients. It would be less common to do grafting surgeries for elder patients and curettage and cementation procedure is more often applied.

Some reviews indicate that application of denosumab treatment before and after surgeries may help recovery process. It is also very efficient to stop the cell growth.[4,5] However, it is more common for the patients with spine and pelvic lesions due to its difficulty to reach with surgeries and some reviews say that it induces cell proliferation which may cause malign transformation.[6] It may be better for the patient to directly undergo reconstruction with vascular fibular grafting without having denosumab application because in our case, the patient had secondary proliferative changes in the previous surgery sites due to drug application. During the surgery, there was no complication, but it would be easier for the surgeon to do the operation on a tissue without secondary proliferative changes.

Reconstruction of distal ulna is essential to restore normal function of the wrist following wide resection for tumors of distal ulna.[8] The distal end of the ulna is functionally important as it helps in pronation-supination of forearm and grip strength and in maintaining the relationship between the carpal bones and distal end of the radius through the ulnar collateral ligament and TFCC. The distal radioulnar joint instability appears to be the most common problem with these patients. Different types of surgery techniques were defined but most of them cause pain and instability in the long term follow ups.[9] Also, it is a known issue that these types of patients have the be operated with arthrodesis procedures. Because of this, we believe that the most important part of this reconstruction surgery is to obtain a stable distal radioulnar joint. Our patient had a stable joint since we reshaped the distal end properly with the help of high-speed burrs and other instruments.

We did not perform a z-plasty procedure unlike Mariappan et al.[7] Instead, we sutured the extensor carpi ulnaris and digiti minimi muscles with the application of soleus graft. We believe that it could give better results in functional status in the postoperative follow ups because it is known that z-plasty procedure in the distal ulnar regions may cause early arthrosis and instability.

Source of Finance

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: *Mahmut Kürşat Özşahin, Arın Celayir, Anıl Demiröz, Veli Muzaffer Murad Hız;* ***Design:*** *Mahmut Kürşat Özşahin, Arın Celayir;* ***Control/Supervision:*** *Mahmut Kürşat Özşahin, Arın Celayir, Anıl Demiröz;* ***Data Collection and/or Processing:*** *Mahmut Kürşat Özşahin, Arın Celayir, Anıl Demiröz, Veli Muzaffer Murad Hız;* ***Analysis and/or Interpretation:*** *Mahmut Kürşat Özşahin, Arın Celayir, Anıl Demiröz, Veli Muzaffer Murad Hız;* ***Literature Review:*** *Mahmut Kürşat Özşahin, Arın Celayir;* ***Writing the Article:*** *Arın Celayir;* ***Critical Review:*** *Mahmut Kürşat Özşahin, Arın Celayir, Anıl Demiröz;* ***References and Fundings:*** *Mahmut Kürşat Özşahin, Arın Celayir, Anıl Demiröz, Veli Muzaffer Murad Hız;* ***Materials:*** *Mahmut Kürşat Özşahin, Arın Celayir, Anıl Demiröz, Veli Muzaffer Murad Hız.*

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