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(CASE REPORT)

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Massive allograft in salvage surgery for giant cell tumor of the proximal humerus

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Abstract

Introduction: The giant cell tumors are characterized by being locally aggressive and are associated with a wide biological spectrum. Proximal humerus tumors represent between 4% and 10% of all primary bone tumors.

There are various surgical limb salvage treatment strategies for proximal humeral giant cell tumor such as shoulder arthroplasties, arthrodesis, and massive allografts.

In this article, we report a 20-year-old young adult with a giant cell tumor of the right proximal humerus who was managed using a limb salvage technique with the use of a massive allograft.

Case presentation: A 20-year-old man presented with right shoulder pain associated with a sensation of a mass. He was diagnosed with a giant cell tumor. One year later, he developed a pathological fracture, and limb salvage surgery was performed, which included resection of the tumor lesion in the proximal humerus and reconstruction with a massive osteochondral allograft.

Conclusion: Giant cell tumors of the proximal humerus, although rare, represent a significant diagnostic and therapeutic challenge due to their anatomical location and proximity to crucial vascular and nerve structures. Salvage limb surgery using massive allografts proves to be a viable option for preserving the functionality.

Keywords: Giant Cell Tumor; Proximal Humerus; Massive Allograft; Salvage Surgery

1. Introduction

Neoplasms of the upper limb encompass a broad spectrum and are generally divided into benign and malignant types, which vary depending on the location in the upper extremity, the type of tissue involved, and the age at presentation. Malignant bone and soft tissue neoplasms in the upper limb are relatively rare; however, proximal humerus osteosarcoma, scapular chondrosarcoma, rhabdomyosarcoma, and synovial sarcoma are among the most prevalent. (1)

On the other hand, benign tumors of the upper limb are more common than malignant ones. These include ganglion cysts, lipomas, giant cell tumors of the tendon sheath, and in bone tissue, osteochondromas, simple and aneurysmal bone cysts, osteoid osteomas, enchondromas, and giant cell tumors. (2,3)

Bone tumors can be classified histologically as bone-forming, cartilage-forming, connective tissue, or vascular. They are also categorized by their biological behavior, such as locally aggressive and rarely metastatic (4), as in the case of giant

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cell tumors, which are locally aggressive and associated with a wide biological spectrum—from latent benign to highly recurrent malignant (15–25%) and occasionally metastatic. (5,6)

Giant cell tumor is generally benign and is histologically characterized by multinucleated giant cells against a background of mononuclear stromal cells. The latter are the main tumor cells, as they express receptor activator of nuclear factor kappa-B ligand (RANKL), leading to the recruitment of monocytes to the tumor site. RANK receptors on monocyte surfaces bind to RANKL, inducing differentiation into multinucleated giant cells. These giant cells have osteoclastic properties, mediating bone resorption, and thus the tumor appears as a purely lytic lesion on radiographs. These lesions typically have well-defined, non-sclerotic margins and are eccentrically located near the articular surface, especially in patients with closed growth plates. (6,9)

Giant cell tumors represent between 4% and 10% of all primary bone tumors and 15% to 20% of benign bone tumors. They primarily affect young adults in their third and fourth decades of life, with a female-to-male ratio of approximately 1:1.13 and 1:1.5 a higher incidence in Asian populations compared to Western ones. (5)

These tumors typically arise in the epiphyseal of long or flat bones such as the pelvis or sacrum. About 44% are located around the knee joint (distal femur or proximal tibia), 13% in the hands and feet, and 10% in the distal radius. Although benign, they may metastasize in approximately 1% to 5% of cases, with the lungs being the most common site. (5,7)

The proximal humerus is a rare site of involvement, accounting for only 4.1% to 8.3% of cases. In this location, giant cell tumors are asymptomatic in early stages, but moderate to large osteolytic lesions reduce bone strength, leading to pathological fractures and pain, severely affecting patients' quality of life. Due to its rarity, treatment principles and surgical efficacy for giant cell tumor in the proximal humerus remain unclear. (8)

The first systemic drugs studied in the multidisciplinary treatment of giant cell tumors were bisphosphonates. Zoledronic acid showed promise by inducing apoptosis of neoplastic stromal cells and promoting osteogenic differentiation. However, the introduction of denosumab—a fully human synthetic IgG monoclonal antibody that binds RANKL with high affinity and prevents its interaction with RANK receptors—has proven to be highly effective, especially in advanced cases. (9,10)

Several limb-salvage surgical strategies exist for proximal humerus giant cell tumors. These are limited by the region's complex anatomy and the proximity to major blood vessels and nerves, requiring precise delineation of the resection zone and preserved structures. (11) Available strategies include hemiarthroplasties, shoulder arthroplasties with reverse or anatomical prostheses augmented with allograft or vascularized fibula autograft, megaprostheses, arthrodesis (12–18), and massive allografts (22–25). Amputation may still be necessary as a radical option depending on malignancy transformation, functional impairment, and neurovascular involvement.

This article presents a case of a 20-year-old male with a giant cell tumor in the right proximal humerus managed using a limb salvage technique with a massive allograft.

2. Case presentation

A 20-year-old male with no past medical history presented with right shoulder pain, a palpable mass, and limited range of motion. An initial shoulder radiograph showed an expansile osteolytic lesion with poorly defined cortices in the proximal humerus (Fig. 1). MRI revealed a T2 hyperintense mass (Fig. 2). A biopsy was performed, reporting a mesenchymal neoplasm predominantly composed of giant cells. A CT scan of the chest, abdomen, and pelvis ruled out metastatic disease.

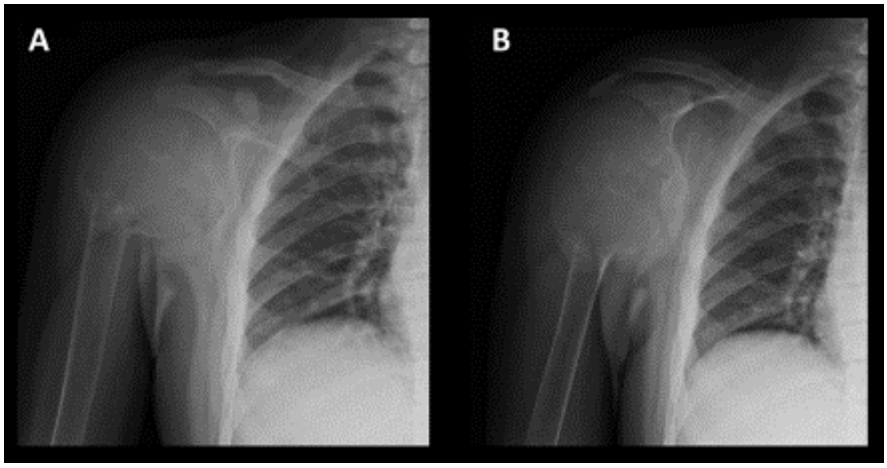

Figure 1 A–B Anteroposterior radiograph (Fig. 1-A) and internal rotation view (Fig. 1-B) of the right shoulder showing a radiolucent lesion with a non-sclerotic margin and eccentric cortical erosion of the proximal humerus, extending from the humeral head to the proximal third of the diaphysis, with involvement of surrounding soft tissues.

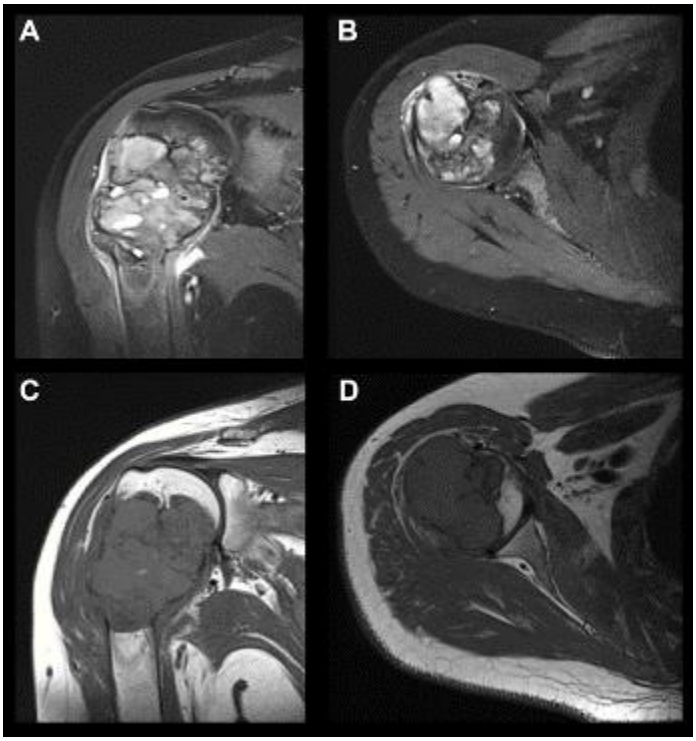

Figure 2 A–D. MRI images of the tumor showing heterogeneous high signal intensity on T2-weighted coronal (Fig. 2-A) and axial (Fig. 2-B) views, and low signal intensity on T1-weighted coronal (Fig. 2-C) and axial (Fig. 2-D) views.

One year later, the patient experienced sudden shoulder pain during work activity and presented with limited motion. A new shoulder X-ray showed a pathological fracture at the site of the lesion.

Limb salvage surgery was performed, involving resection of the tumor in the proximal right humerus and reconstruction with a 14 cm osteochondral massive allograft.

Under general anesthesia, with the patient in a supine position, an anterolateral approach to the right shoulder was used. Planes were dissected, and medial and lateral flaps were developed. The long head of the biceps tendon was tenotomized proximally. Dissection extended 14 cm distally, and osteotomy was performed with an oscillating saw. The conjoint tendon, rotator cuff, and joint capsule were identified and dissected. Vascular structures in the axillary fossa

and terminal branches of the brachial plexus and humeral artery were freed. Glenohumeral disarticulation was performed and sent for pathology.

A proximal humeral allograft from a bone bank was used to reconstruct the defect. The graft was reshaped with a Midas drill and fixed with a long locked proximal humerus plate and screws. Anchors were used for reattachment of the joint capsule and rotator cuff, along with tenodesis of the long head of the biceps. No injury to adjacent structures was noted. The wound was closed in layers, and the limb was immobilized with a sling. Postoperative X-ray showed proper positioning of the stabilized allograft (Fig. 3).

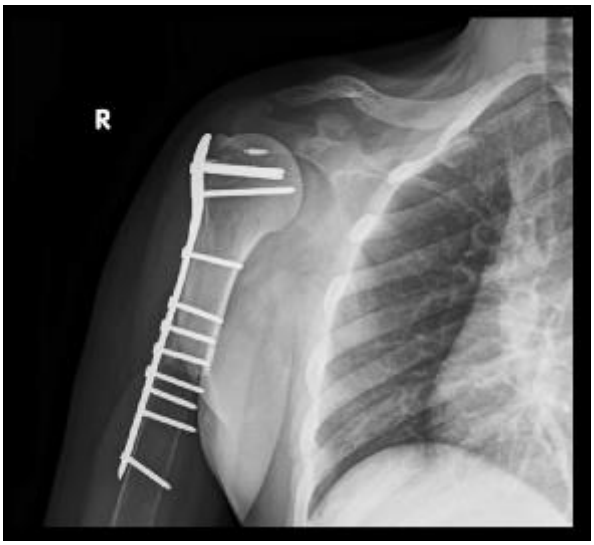

Figure 3 Postoperative radiograph showing a proximal humerus allograft fixed with a proximal humerus locking plate and an anchoring system for reinsertion of the joint capsule and rotator cuff.

3. Discussion

Proximal humerus giant cell tumors are rare, comprising 4.1% to 8.3% of reported cases. Their anatomical location poses unique diagnostic and surgical challenges due to proximity to critical neurovascular structures. The patient in this case matched the demographic pattern described in the literature, typically affecting young adults in their 20s or 30s. (8)

Biologically, the case underscores the local aggressiveness of giant cell tumors and their potential to cause pathological fractures, significantly impacting quality of life. Histological findings of multinucleated giant cells and mononuclear stromal cells reaffirm the importance of a multidisciplinary approach that includes surgery and targeted therapies. (9)

A key point is the role of denosumab in systemic management, which has revolutionized treatment by inhibiting RANKL-mediated osteoclastic activity, reducing tumor size, and facilitating more conservative resection. (9,10,20)

Surgical limb salvage with massive allograft, as performed here, remains a valid strategy, particularly when more advanced options like reverse prostheses are unsuitable. Allografts maintain anatomical and functional structure, especially in young patients. (11,19)

Allografts avoid complications associated with prosthetic wear and improve long-term viability compared to hemiarthroplasties or megaprostheses. (22–25) However, such techniques demand meticulous planning due to the region's anatomical complexity.

Complete resection with clear margins and massive allograft use reduces recurrence, as shown by Wen-zhe Bai et al., with recurrence rates of 7–11% at 1-, 2-, and 8-years post-op in patients undergoing curettage and segmental resection. (8)

Complications of allografts include infection, non-union, graft resorption, and peri-implant fractures. Additionally, rotator cuff detachment and reattachment lead to longer immobilization and limited function, particularly in abduction and external rotation.

Shoulder arthrodesis may be considered for young, active patients with extensive resections. Although it ensures stability, it sacrifices shoulder movement and strength. (12)

This case highlights the viability of massive allografts in limb salvage surgery for proximal humerus giant cell tumors, particularly in young, active individuals. Despite low recurrence rates, the approach requires high surgical expertise and thorough preoperative planning due to the complexity of the region's neurovascular structures.

4. Conclusion

Although rare, giant cell tumors of the proximal humerus pose significant diagnostic and therapeutic challenges due to their anatomical location and proximity to vital neurovascular structures. The presented case aligns with literature trends and emphasizes the importance of a multidisciplinary approach, including surgery and pharmacological treatment with agents like denosumab.

Limb salvage surgery using massive allografts proves to be a viable option for preserving limb function in young patients with high activity demands. While this technique offers advantages such as anatomical preservation and reduced prosthetic wear, it also carries inherent risks like infection and complex rehabilitation. Surgical planning must be precise given the region's anatomy.

In summary, massive allografts in salvage surgery for proximal humerus giant cell tumors show promising outcomes in preserving function and minimizing recurrence, though a highly specialized approach is essential for long-term success.

Compliance with ethical standards

Disclosure of conflict of interest

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Statement of informed consent

The patient signed an informed consent form authorizing the use of his clinical data for the preparation and publication of this case report.

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