Giant cell tumor of distal radius: En bloc resection with allograft reconstruction: A case report

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Key Clinical Message

Giant cell tumor of bone (GCT) is a rare neoplasm which often presents as a lytic lesion in the epiphyseal region of long bones and which are usually accompanied by pain, swelling, and restricted movement.

Abstract

Giant cell tumor of bone (GCT) is a rare neoplasm that affects individuals in their third and fourth decades of life. Clinically, it often presents as a lytic lesion in the epiphyseal region of bones, notably the distal femur and proximal tibia. Radiologically, GCT appears as a distinct lytic lesion in the epiphyseal region. Histopathologically, GCTs are composed of mononuclear cells, macrophages, and multinuclear giant cells, indicative of osteoclastogenic stromal tumors. A 37-year-old man presented with left wrist pain, swelling, and restricted movement persisting for a year, worsening over the last 7 months. Radiographic assessments revealed a distal radius bone mass involving the radiocarpal joint. Biopsy confirmed a GCT with extension into peripheral muscle. PET/CT scan showed localized pathology without metastasis. Histopathologically, GCT exhibited multinucleated giant cells, spindle cells, and aneurysmal bone cyst-like regions with coagulation necrosis. Surgical resection involved en-bloc removal and reconstruction with a non-vascularized radius bone graft. Postoperatively, the patient showed no complications at the one-year follow-up, suggesting successful intervention.

KEYWORDS

allograft, distal radius, en bloc resection, giant cell tumor

1 **INTRODUCTION**

A giant cell tumor (GCT) is a benign neoplasm characterized by progressive and destructive features. Typically manifesting in individuals during their third or fourth decade of life, GCT exhibits a higher prevalence in women than in men. Although categorized as a benign bone tumor, GCT displays local aggressiveness and a propensity for recurrence.¹ Accounting for approximately 6% of all bone tumors, 4% of primary bone tumors, and 20% of benign bone tumors, GCTs are relatively uncommon. The distal femur, proximal tibia, and distal radius are the most frequently affected long bones, with a particular predilection for the distal end of the radius, making it the third most common location for GCT

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after the distal femur and proximal tibia. Despite their benign classification, GCT can exhibit local aggressiveness, causing bone erosion, pain, and potentially leading to fractures.²

GCT, also known as osteoclastoma, is a rare yet locally aggressive primary bone tumor characterized by multinucleated giant cells dispersed throughout the tumor, along with mononuclear stromal cells producing osteoid matrix. Histologically, GCT exhibits areas of hemorrhage and necrosis indicative of its aggressive nature, and in some cases, increased mitotic activity.³ The stromal cells often generate fibrous bands and septae, contributing to tumor compartmentalization. Histological grading based on features like mitotic activity guides prognosis, with higher grades associated with increased recurrence rates. Immunohistochemistry, utilizing markers such as CD68, CD163, and RANKL, aids in distinguishing GCT from other bone tumors. Understanding these histopathological features is crucial for accurate diagnosis and management decisions, which often involve a multidisciplinary approach tailored to the tumor's behavior and grade.⁴

GCTs pose a challenge in terms of management, especially when located in the distal radius. En bloc resection, a surgical technique involving the removal of the tumor along with a margin of surrounding normal tissue, is often employed in cases of large, aggressive, or challenging-toreach tumors. This method aims to reduce the risk of local recurrence.⁵ However, the optimal approach for treating GCTs of the distal radius remains controversial, with enbloc excision showing decreased recurrence but posing challenges for wrist reconstruction due to the complex functional requirements of the wrist.⁶ The multidisciplinary management of GCTs involves various treatment options such as curettage, cryotherapy, radiation therapy, monoclonal antibody therapy (denosumab), and surgery. Recurrence rates after surgical treatment may vary based on factors like tumor location, extent of surgical resection, and patient age. The distal ulna and radius have been reported as especially susceptible to GCT recurrence after resection.7

This study aims to report a case of GCT of the distal radius treated with en-bloc resection, providing insights into an effective strategy against GCT and offering valuable guidance for similar cases.

2 | CASE REPORT

2.1 | Patient information

A 37-year-old man presented with complaints of joint pain, swelling, and restricted movement in his left wrist. These symptoms had persisted for a year before his hospital visit and had exacerbated over the last 7 months. The patient characterized the pain as a stabbing sensation, intensifying during wrist flexion and subsiding with wrist extension. After conducting a physical examination of the distal radius area, a bony mass was palpated, which, in addition to acute pain, led to restricted movement during radiocarpal joint extension.

2.2 | Diagnostics assessment

Radiographic assessments, including standard anteroposterior and lateral views of the left wrist, revealed the presence of a bone mass in the distal radius with evident involvement of the radiocarpal joint (Figure 1A-E). Subsequent biopsy conducted on the left metaphysis of the distal ulna under general anesthesia yielded brown spongy material. Histologically, the specimen exhibited a distinctive composition characterized by a significant presence of multinucleated giant cells and spindle cells within a densely collagenous background. These histological findings were indicative of a diagnosis consistent with GCT, with extension into the peripheral striated muscle. Pathologists recommended a comprehensive correlation with the clinical and radiological findings to enhance diagnostic accuracy (Figure 2A,B). In the PET/CT scan, there was no evidence of tumor metastasis observed, providing valuable insights into the localized nature of the pathology (Figure 3A-C).

2.3 | Histopathological evaluation

The histopathological analysis of the tumor mass revealed a hypocellular capsule containing scattered fibroblasts (spindle or stellate cells) in a mucoid background. Sections exhibited bony trabeculae engaged by a neoplastic lesion consisting of cells with round to oval pleomorphic nuclei, with numerous evenly distributed osteoclastic giant cells interspersed among them. The mitotic activity was low (3-4/10 hpf), and signs of osteoid bone formation were evident. Additionally, histopathological sections revealed secondary aneurysmal bone cyst-like regions accompanied by mononuclear cells and multinucleated giant cells, as well as evidence of coagulation necrosis. Furthermore, fibrohistiocytic proliferation, hemorrhage, necrosis, and the formation of aneurysmal bone cysts were noted within the bone trabeculae. Notably, the neoplastic cells extended into striated muscles, and the lesion contained thin collagen fibers along with small cells featuring hyperchromatic nuclei and scanty cytoplasm scattered throughout. Multinucleated giant cells, characterized by multiple

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FIGURE 1 Preoperative anteroposterior and lateral x-ray radiographs (A–C) and T1 (D) and T2 (E) -weighted MRI of the left radiocarpal joint and distal end of radius.

nuclei within a single cell, were prevalent and distributed throughout the tumor tissue. The stromal cells displayed a spindle-shaped morphology with variable appearances, ranging from a more uniform to an irregular and plump structure. The tumor exhibited heightened vascularity, accompanied by observable hemorrhage and cyst formation containing hemosiderin-laden macrophages, contributing to its distinct brownish appearance. Additionally, a peripheral rim of osteoclast-like cells was evident at the interface between normal and tumor bone tissue (Figure 2A,B).

2.4 | Surgical technique

The patient was positioned supine in the operating suite and placed under endotracheal anesthesia after proper identification. Following standard protocol, a tourniquet was applied to the arm post-administration of preoperative antibiotics. Exsanguination of the upper limb was achieved using an Esmarch bandage, and a pre-placed, well-padded pneumatic tourniquet was then inflated to 250 mmHg. The total tourniquet time was less than 1 h. Subsequently, the left arm was positioned on a wellpadded surgical table. A dorsal incision was made at the wrist, extending both proximally and distally through the tumor zone. En bloc removal of the distal 7 cm of the radius tumor bone, including periosteum and soft tissue in the region of recent biopsy or cortical breakthrough, was performed. The allograft was tailored to size, and the reconstruction involved en-bloc surgical excision. A non-vascularized radius bone graft, affixed with a locking compression plate (LCP), was inserted into the defect, replacing it with a suitable allograft. The tourniquet was then deflated, and hemostasis of small bleeding points was achieved using monopolar electro

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surgery. The skin wound was meticulously closed in layers with buried deep dermal 2–0 Vicryl and a running over-and-over technique using 3–0 nylon. A bulky dressing was applied and reinforced with a short-arm splint to maintain the wrist and forearm in a neutral position. The patient demonstrated good tolerance to the procedure, awakening in the operating room, and was subsequently transported to the recovery room in a stable condition (Figure 4A-E).

2.5 | Post-operation and follow-up

During the 1-year follow-up, the surgical site was initially monitored for signs of infection, hematoma, infection, bleeding, or nerve damage. Early rehabilitation exercises were initiated to prevent stiffness and enhance wrist and forearm function. Range of motion, grip strength, and any functional limitations were assessed during follow-up visits every 3 months. Regular imaging, including x-rays, was conducted every 3 months to assess bone healing, graft integration, and the absence of tumor recurrence (Figure 5A–E).

3 | DISCUSSION

GCTs are locally aggressive primary bone tumors characterized by the presence of multinucleated giant cells and mononuclear stromal cells. While the exact etiology remains unclear, several factors have been implicated, including genetic mutations (such as those in the **FIGURE 2** Histopathology of the tumor resected at the first surgery showing polynuclear giant cell surrounded by mononuclear stromal cells [H&E, (A, B)×100, scale bar=100 μ m and (C, D)×400, scale bar=25 μ m].



FIGURE 3 PET (A) and CT (B, C) scan was showed there was no evidence of tumor metastasis.

FIGURE 4 Intraoperative image showing the GCT in the radius. The image shows the treatment through En bloc removal of the distal 7 cm of the radius tumor bone and reconstruction with allograft (A–E) affixed with a locking compression plate (LCP).



H3F3A gene), trauma, and hormonal influences.⁸ GCTs most commonly affect the epiphyses of long bones, particularly around the knee joint, but they can occur in any bone. The proximal tibia is the most frequent site for GCT. Histopathologically, GCTs exhibit multinucleated giant cells interspersed within a background of mononuclear stromal cells. These stromal cells produce osteoid, resulting in a characteristic appearance on histological examination. Additionally, features such as hemorrhage, necrosis, fibrous bands, and aneurysmal bone cyst-like areas may be observed. Diagnosis of GCTs involves a combination of clinical, radiological, and histopathological findings. Clinically, patients may present with pain, swelling, and limited range of motion. Radiologically, GCTs typically appear as lytic lesions with well-defined margins and cortical thinning. Definitive diagnosis relies on histopathological examination of biopsy specimens, which reveal the characteristic features mentioned earlier.^{3,4}

Treatment of bone GCTs depends on various factors, including tumor location, size, grade, and patient factors. The mainstay of treatment is surgical resection, which aims to achieve complete tumor excision while preserving maximal function and minimizing recurrence. Curettage with or without adjuvant therapy (such as bone cement, phenol, cryotherapy, or adjuvant medications like denosumab) is commonly employed. Recurrence rates of up to 50% have been observed in GCTs within a 3-year period following curettage.9 Approximately one fifth of GCT cases show recurrence. In cases where complete resection is not feasible or in recurrent or metastatic disease, other modalities such as radiotherapy or systemic therapies may be considered. Despite its generally benign behavior, bone GCTs has a significant propensity for local recurrence, particularly in cases of incomplete excision or aggressive variants. Long-term follow-up is therefore crucial to monitor for recurrence and to assess treatment efficacy and functional outcomes.¹⁰



FIGURE 5 Immediate postoperative surgical area (A) and postoperative lateral (B) and anteroposterior (C) x-ray radiographs. Functional outcome at the end of 1 year showing (A) a complete palmar flexion (D) and a complete dorsiflexion (E).

4 | CONCLUSION

GCT of bone presents a complex interplay of clinical, radiological, histopathological, and therapeutic aspects. A multidisciplinary approach involving orthopedic surgeons, radiologists, pathologists, and oncologists is essential for accurate diagnosis, appropriate treatment selection, and optimal patient outcomes.

AUTHOR CONTRIBUTIONS

Parviz Ahangar: Conceptualization; supervision. **Alireza Rahimnia:** Supervision; visualization; writing – review and editing. **Mohsen Akbaribazm:** Conceptualization; writing – original draft; writing – review and editing. **Abbas Khalilpour:** Supervision; writing – review and editing. **Mohsen Rahimi:** Investigation; supervision; validation; writing – review and editing. **Hosein Pirmohamadi:** Conceptualization; supervision; writing – review and editing.

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The authors declare that there is no conflict of interest.

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All data associated with the article is available if required.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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