Postoperative Fracture Risk in Giant Cell Tumor: A Case Report and Review of Literature

Amit Kumar [1] , Kumar Keshav [1] , Siddhartha Singh [1] , Amarendra Singh [2]

1. Orthopaedics, Sanjay Gandhi Postgraduate Institute of Medical Sciences, Lucknow, IND 2. Trauma and Orthopaedics, Sanjay Gandhi Postgraduate Institute of Medical Sciences, Lucknow, IND

Corresponding author: Siddhartha Singh, sid.dt24@gmail.com

Abstract

Giant cell tumor (GCT) of the proximal femur poses various challenges in its management and recurrence. We present a rare case of GCT of proximal femur in which recurrence and coxa vara deformity were encountered after index surgery. Management of the recurrence was done with intramedullary fixation with extended curettage and bone grafting. Different aspects of management such as the role of defect size, adjuvants, bone cement/bone graft, implants, and bisphosphonates have been highlighted in this article.

Categories: Pathology, Oncology, Orthopedics
Keywords: defect size, bisphosphonates, proximal femur, recurrence, gct

Introduction

Giant cell tumors of the bone (GCTB) are intermediate-grade primary bone tumors [1]. They are mostly treated by intralesional curettage to maintain joint integrity and functional outcome. Post curettage, the defect that is created can be filled with bone cement, bone autograft/allograft, or bone graft substitutes. However, there is still an imminent risk (up to 14%) of fracture in the postoperative period [2-4].

Giant cell tumor (GCT) of the proximal femur (PF) accounts for around 6% of all GCTs [5]. The treatment of a GCT around the PF is unclear. In most cases, the management relies on the surgeon's experience and the patient's age [6]. Various treatment modalities have been used depending on the defect size created by the tumor. However, there is still a paucity of literature on the treatment of GCT in the PF.

In this case report, we describe a rare case of GCT of the PF where the patient developed post-surgery deformity and fracture and how we managed it with a review of the literature.

Case Presentation

A 17-year male patient presented to us with dull aching pain in the left hip for one year. X-ray showed a lytic lesion in the PF suggestive of a benign bone tumor (Figure 1).

FIGURE 1: Lytic lesion in the left proximal femur (first visit)

Core needle biopsy was suggestive of GCTB (Figure *2*).
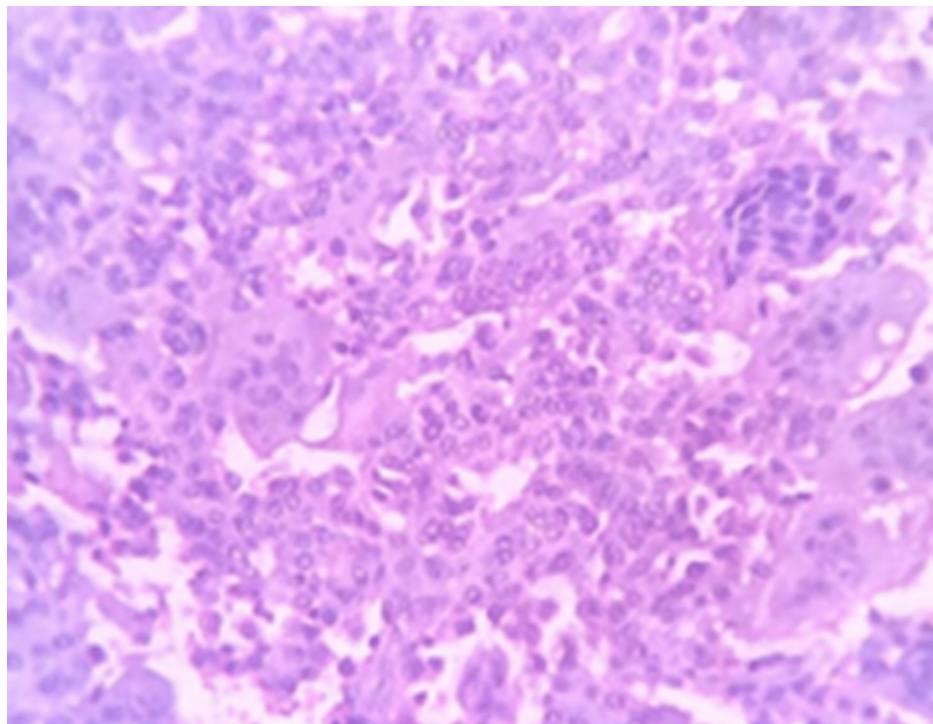

FIGURE 2: HPE suggestive of GCT

HPE: Histopathological examination; GCT: Giant cell tumor.

He underwent curettage, the use of phenol as an adjuvant, and vancomycin-coated antibiotic cement augmentation (Figure *3*).

FIGURE 3: Curettage and cement augmentation

The HPE report of the curetted material confirmed the lesion to be GCTB. Subsequently, he remained asymptomatic for two years. After two years, the patient started having a painful limp. An X-ray revealed a coxa vara deformity, which was then managed conservatively (Figure 4).
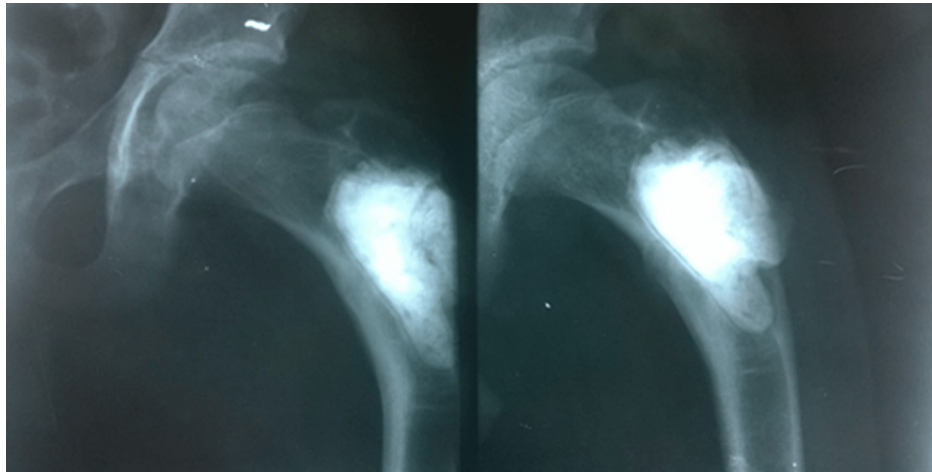

FIGURE 4: Development of coxa vara deformity after two years

He continued to walk with a short-limb gait, but after one month, he sustained a fatigue fracture in the subtrochanteric region (Figure 5).

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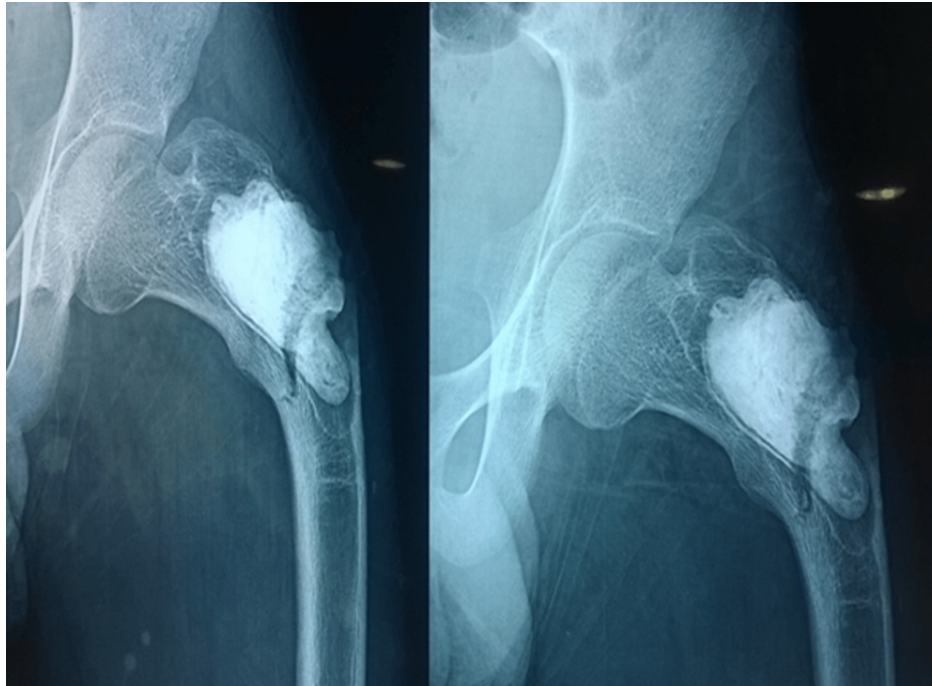
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FIGURE 5: Subsequent fatigue fracture in the subtrochanteric region

Following this, the patient was admitted and managed conservatively with skin traction under continuous monitoring for three months, which resulted in fracture union and improvement in the neck-shaft angle, after which the patient was gradually rehabilitated to full weight bearing. At the one-year follow-up subsequent to the initiation of conservative management of fracture, the X-ray showed complete union and normal neck-shaft angle (Figure 6).

FIGURE 6: One-year follow-up following conservative management of subtrochanteric fatigue fracture

However, after two years (four years post-index surgery), the patient redeveloped coxa vara deformity (Figure 7) with an evident subtrochanteric fracture three months later (Figure 8).
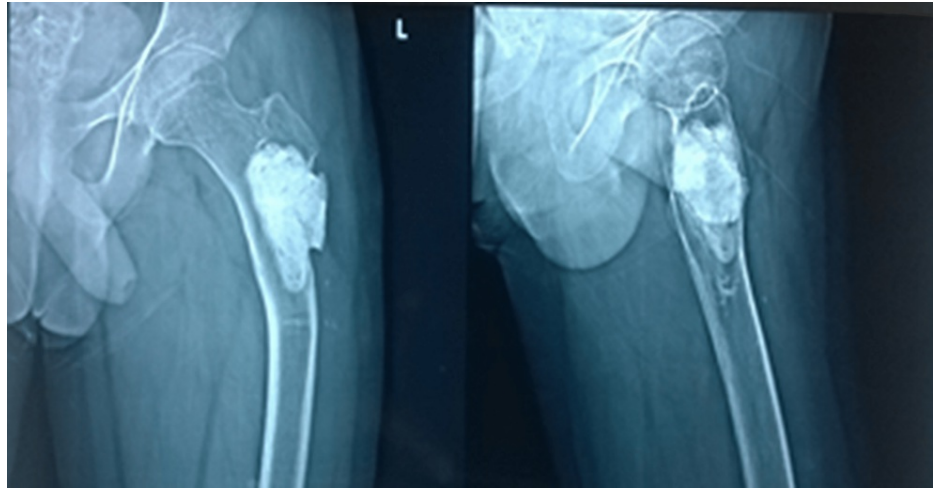
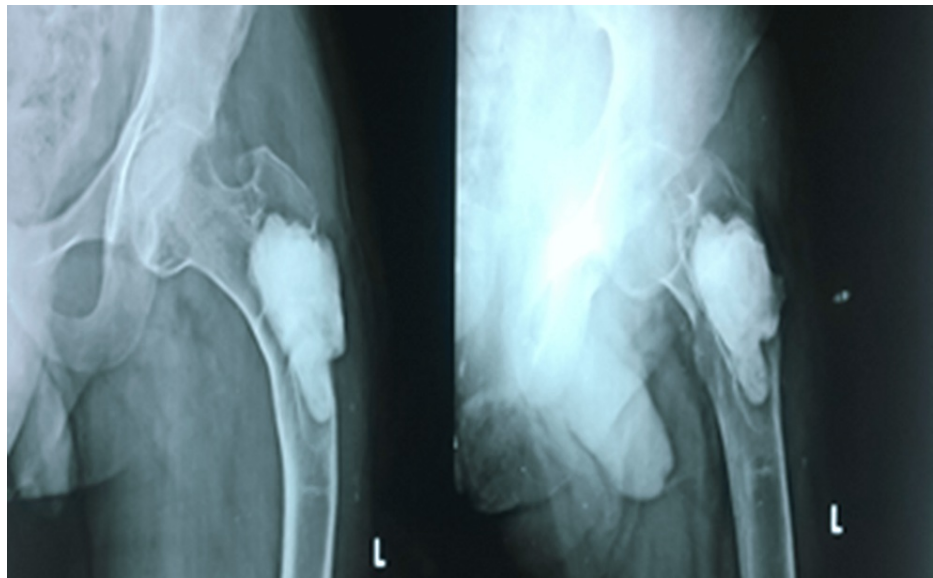**FIGURE 7: Redevelopment of coxa vara four years post index surgery**

FIGURE 8: Re-fracture in the subtrochanteric region

He underwent removal of bone cement, intralesional curettage, internal fixation with PFN, and morselized autofibular bone grafting (Figure 9).

FIGURE 9: Removal of bone cement, IC, and IF with PFN + morselized autofibular bone grafting

IC: Intralesional curettage; IF: Internal fixation; PFN: Proximal femoral nail.

Biopsy confirmed a multinucleated giant cell suggesting a recurrence of GCT in the PF. He was started with bisphosphonates (Tab Alendronate 70 mg weekly once) along with calcium and vitamin D3 supplements postoperatively for two years. The two-year follow-up showed fracture union without any signs of recurrence (Figure 10).

FIGURE 10: Two-year follow-up showing no signs of recurrence

Discussion

GCTB poses several challenges in its management. There are a number of complications that can be encountered while surgically managing a GCTB case. The most common complications are tumor recurrence, failed reconstruction, and postoperative fracture. The lack of conclusive assessment criteria for postoperative fracture risk and appropriate implants for augmentation/construct reinforcement are concerns that need to be addressed.

This article reviews the various factors responsible for postoperative fracture after curettage in

GCT. Marcove et al. stated that curettage combined with cementation bears an increased fracture risk because of the premature load bearing of the involved bone as well as inadequate cement strength in the bone defect after curettage [7].

Role of defect size

According to Jeys et al. [8], a tumor volume to distal femoral volume ratio of 54% or greater was linked to a higher risk of pathological fracture. Amanatullah et al. showed an increased risk of fractures with defect sizes that damaged more than 50% of the cortical width in their biomechanical investigation [9]. On the distal femur, finite element calculations revealed that the bone strength dropped when the defect represented 35% or more of the epiphyseal volume and was greater in the medial defect than in the lateral defect [10,11].

Hirn et al. found that certain lesions were never fully filled up with new bone, while others were. They concluded that a key element in the development of postoperative fracture is the size of the defect. They claimed that when a bone defect's volume was higher than 60 cm^3 and its maximal diameter was greater than 5 cm, the risk of fracture increased dramatically.

Role of adjuvants

Phenol has a lower fracture rate than liquid nitrogen because it penetrates the bone less extensively [12,13]. Infection, postoperative fracture or femoral condyle collapse, nonunion, nerve palsy, and PMMA leakage were the more common non-oncological problems found by van der Heijden et al. when liquid nitrogen was used by direct pour method [3]. The average depth of necrosis, according to Bombardier et al., was 2.54 mm for the liquid nitrogen spray and 0.3 mm for the phenol [14]. As a result, cryosurgery utilizing liquid nitrogen spray may maintain bone union ability. Cryosurgery using liquid nitrogen spray has been shown to lower fracture rates when compared to the direct pour technique (0% versus 17%) [15].

The majority of authors concur that high-speed burring of the cavity's borders is advantageous for sufficient exposure, but in some cases, especially when the bone is already thin, this may increase the risk of fracture [16].

Bone grafting versus bone cement

The defect once created after curettage of the tumorous area causes the host bone to weaken structurally. There are various advantages and disadvantages of using bone graft/cement found in the literature. However, there is no common consensus regarding the choice of material to be used for filling the bone defect created after curettage.

As cement has a higher Young's modulus than trabecular bone, reconstruction by bone cement leads to the transfer of load to the stiffer material causing resorption at the bone cement interface [17]. As a result, it weakens the nearby trabecular bone, which leads to fracture. This effect is more extensive near the joint where extensive curettage and cementation are done.

Benevenia et al. [2] reported that the cement with bone graft groups (5%) have a low risk of postoperative periarticular fracture compared to the cement group (23%). Wallace et al. discovered a statistically insignificant increase in postoperative fracture in patients who underwent bone graft reconstruction [18].

Role of implants in reconstruction

Construct reinforcement after curettage and cementation offers a mechanical advantage in reducing postoperative fracture. However, there is a paucity of literature that can guide in choosing implants. Large cavitary lesions are often supported by pins, screws, and locking plates as they provide immediate stability and structural support [19-23].

Various in vitro studies and finite element analyses on the bone model have supported the need for construct reinforcement with internal devices in larger cavitary lesions and concluded the superiority of nails/plates over pins and screws as they span the proximal and distal end of bone cement interface and their screws cross the interior wall of the interface [24,25].

There is a scarcity of literature on stabilizing a PF GCT with intramedullary fixation following adequate curettage. Even though four of the 10 patients in his group who received this type of treatment experienced local recurrences, Sakayama et al. advised against replacement if at all possible [26].

Role of bisphosphonates and denosumab

By making the bone lesions harden, the monoclonal antibody denosumab aids in halting the breakdown of the bone. It works by attaching to the nuclear factor-kappa ligand's active receptor. According to studies, tumor cells are concealed in osteosclerotic lesions, and keeping them untreated increases the risk of

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recurrence [27,28]. Every four weeks, 120 mg of denosumab can be injected subcutaneously, with an extra 120 mg loading dosage given on days 8 and 15 of the first cycle. It can be continued until either radiographic disease stability is achieved at six months [29] or there is nearly complete eradication of large cells on repeat biopsies following treatment (for all evaluable patients) [30]. Due to denosumab's tumoristatic properties, stromal cells in the sclerotic rim may take longer to reactivate, increasing the likelihood of recurrence [31].

On osteoclasts, which are precursors of GCTB, zoledronic acid has an apoptotic impact. A consensus has not been reached because there is conflicting evidence supporting the use of zoledronic acid in GCTB prevention. Supplementing with zoledronic acid has been linked to noticeably decreased rates of tumor recurrence [32]. Preoperatively, three doses of intravenous zoledronic acid (4 mg) were administered in GCTB at intervals of three weeks in a trial by Zile et al. Two weeks following the last infusion, the extended curettage was carried out. Recurrence only happened once among the 18 patients in the infusion group, while it happened four times among the 19 patients in the control group [33].

Bisphosphonates such as oral alendronate have been shown to have similar effectiveness in reducing the recurrence of GCTB [34]. Alendronate can be given orally to patients with GCTB who have undergone surgery in the dosage of 70 mg/week or 10 mg once every day [35]. It can be given for a period of two to three years. It has been shown to prevent recurrence along with fewer side effects when compared to zoledronic acid. Other bisphosphonates such as ibandronate have also been used and have been found effective in preventing the recurrence of GCTB [36].

There is a need for assessment criteria that can guide the surgical approach in GCT. Chinese doctors devised a grading system for surgical recommendations for treating GCTB that identified articular surface involvement, pathological fracture, and cortical bone degradation as risk factors [36].

In our study, we attempted to correct the coxa vara deformity after curettage and filled the cavity with a morselized fibular graft, and the construct was reinforced with an intramedullary device PFN. The follow-up of the patient has highlighted a promising result of the construct in GCT of the PF.

Conclusions

Removal of tumor mass weakens the parent bone. This requires augmentation either with autologous/allogenic bone graft, bone graft substitutes, cement, or a combination of the two with or without instrumentation. Concerns that need to be addressed include the lack of clear assessment criteria for postoperative fracture risk and suitable implants for augmentation/construct reinforcement. Based on our experience regarding this case and available literature, we recommend instrumentation to prevent the deformity and help early healing of bone, when the cavity size is large and high stress area are involved.

Additional Information

Disclosures

Human subjects: Consent was obtained or waived by all participants in this study. **Conflicts of interest:** In compliance with the ICMJE uniform disclosure form, all authors declare the following: **Payment/services info:** All authors have declared that no financial support was received from any organization for the submitted work. **Financial relationships:** All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. **Other relationships:** All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

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