Recurrent giant cell tumour of distal Tibia: Case report and review of the literature

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Introduction
First described by Jaffe et al in 1940, Giant Cell Tumours [GCT] constitute 20% of benign bone tumours [1]. Less than 4% of these involve the foot and ankle region, but the exact prevalence in the distal Tibia is not known. Age group most commonly affected is from 20-40 years with a slight female preponderance. Typically, it is described as an expansile lytic lesion in the epiphysio-metaphyseal region in a skeletally mature bone. Clinically, patient presents as a dull aching or a vague pain around the affected joint and sometimes trauma brings notice to the existence of this lesion. Swelling and joint stiffness can also be the presenting complaints. Pathological fractures are seen in 12% of patients at the time of presentation [2]. We have briefly reviewed various treatment modalities for the management of this tumour in distal end Tibia.

Case Report
A 53 year old street vendor came to outpatients department with history of pain and swelling around the left ankle. He related pain to a trivial trauma suffered 2 months back. Pain was dull aching and aggravated with prolonged standing and walking.

BACKGROUND:
Giant Cell Tumour [GCT] is a familiar benign but locally aggressive bone tumour especially around the knee joint. Less than 4% of these tumours are known to affect the ankle joints. But, its biological behavior at this rare location is quite unpredictable. Moreover, restoring the ankle joint functionality following tumour resection is a challenging task.

CASE REPORT:
We report a case of Giant Cell Tumour of distal end of left Tibia in a 53 year old male patient. Initially the condition was treated by curettage and bone grafting. But, due to recurrence of the condition within 6 months, he was treated with extended curettage and bone cementation. At One year follow up there is no recurrence and reasonably good function around the ankle joint is maintained.

CONCLUSIONS:
Primary GCTs have been traditionally treated with curettage of the lesion followed by bone grafts/bone cement. Recurrent cases often require aggressive management. In our case, despite recurrence, extended curettage of the lesion was done and the defect was packed with bone cement. This added adjuvant treatment offered good stability and allowed early mobilization of the ankle joint. This case substantiates the use of bone cement in the treatment of recurrent GCT of distal Tibia whenever the articular integrity is intact with reasonably good functional outcomes. However, a periodic follow-up is still recommended to watch-out for late recurrences.

KEY WORDS: Giant cell tumour
Bone cement
Distal tibia
Recurrence
Curettage

There was no history of constitutional symptoms. On examination, diffuse swelling at the anteromedial border of the left ankle with normal ankle movements and intact neurovascular status was noted. X-rays showed expansile lytic lesion in the distal end of tibia (Figure 1). Plain and contrast MRI revealed a well-defined lytic lesion measuring 7x3.4x4.5cms having narrow zone of transition in the distal tibial epi metaphyseal region. Areas of hemorrhage

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ABSTRACT

Background: Giant Cell Tumour [GCT] is a familiar benign but locally aggressive bone tumour especially around the knee joint. Less than 4% of these tumours are known to affect the ankle joints. But, its biological behavior at this rare location is quite unpredictable. Moreover, restoring the ankle joint functionality following tumour resection is a challenging task.

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and necrosis within the lesion were also noted, but there was no evidence of periosteal reaction, sclerosis or matrix calcification. Cortical thinning with minimal soft tissue extension along antero-inferior aspect was also observed. These features were suggestive of GCT of distal tibia. Through an antero-lateral incision entire lesion was curetted out. The defect was filled with cortico-cancellous bone graft harvested from ipsilateral iliac crest (Figure 2). Specimen sent for histopathological evaluation confirmed as GCT (Netherlands grade II). Post-operative days were uneventful.

Unfortunately, patient returned back within 6 months with complaints of dull aching pain around the same ankle. Repeat x-rays and MRI confirmed the recurrence of the lesion (Figure 3). As the articular surface still appeared intact, we planned to use bone cement as fillers. Lesion was approached through the previous surgical incision. Extended curettage was done till only a rim of subchondral bone was left intact (Figure 4). Thorough debridement and pulsatile lavage followed by high speed burring was also employed this time. The defect was filled with bone cement spacer. Wound was closed over a suction drain. Repeat histopathological evaluation reported it as GCT of same grade (Netherlands grade II). Post-operative, the patient was given below knee slab support for 6 weeks and non-weight bearing with the help of crutches was encouraged. After 6 weeks, slab was discarded and active ankle mobilization was initiated. Weight bearing was allowed as tolerated only at 3rd month follow-up. He was followed up periodically at 3 weeks, 6 weeks, 3 months, 6 months and 1 year (Figure 5). His Revised Musculoskeletal Tumor Society Rating at last follow up was 27/30 (90%).

**Figure 1.** X-rays antero-posterior and lateral view of ankle at 1st presentation.

**Figure 2.** X-rays following autologous bone grafting

**Figure 3.** Recurrence within 6 months (articular surface intact).

**Figure 4.** Intra-operative. Extended curettage of the cavity.

**Figure 5.** Last follow-up at one year after recurrence. Bone cement filling up the cavity.
Discussion
The changing trends in the management of bone tumours are from limb sacrifice to limb salvage and currently preservation of limb functionality. Amputation surgeries have become only of historical significance. Small cavitary lesions have been traditionally treated by intralesional curettage with adjuvant chemical cauterization using phenol, hydrogen peroxide, and zinc chloride. Saleh.A et al in their retrospective study of 31 cases of distal tibia GCTs have suggested that extended curettage of the lesion alone is sufficient in local control of the disease, even in recurrent conditions [3]. Cribb et al have described a case of GCT of distal end of Tibia managed with curettage followed by high speed burring and the ankle was stabilized using an Ilizarov frame. Despite a prolonged course of rehabilitation, excellent functional recovery was noted [4].

A large bone defect needs to be filled with either autografts / allografts. Blackley et al reported an overall 12% recurrence in the treatment of GCTs of long bones with curettage and bone grafting. Of 59 cases studied, 3 were in distal tibia [5]. However, there are concerns regarding donor site morbidity (autograft), risk of disease transmission (allograft), and difficulty in visualizing (in x-rays) recurrence with grafts occupying the cavity.

Alternative to bone grafts, bone cement is quite popular as void fillers following tumour resection. According to Thomas JL et al use of polymethylmethacrylate in large osseous defects in foot and ankle following tumour resection provides stability and allows early weight bearing [6]. The rationale behind the use of bone cement is that the exothermic reaction during polymerization of cement aids in killing residual tumour cells without damaging the native host tissue. Similar to our case, Monish Bami et al have reported a case with good results with the use of bone cement, albeit in primary GCT of distal tibia [7]. Pan et al described a case with defect of anterior and posterior cortex in distal tibia due to recurrence. A double layer of polypropylene mesh was used for the containment of cement and the lesion healed uneventfully [8].

Due to proximity of these lesions around the joints, it becomes a daunting task to address both the tumour lesion as well as joint functionality. As these tumours predominantly arise in the epiphyseal-metaphyseal region, there may be breach in the adjacent articular surface by the primary tumour as a part of the disease progression or due to a pathological fracture. Moreover, with each recurrence these tumours tend to get aggressive and invariably affect the adjacent joint. Attempts at aggressive tumour resection may control the condition but, at the expense of joint mobility. Such aggressive lesions can be treated with extended tumour resection followed by ankle arthrodesis or recently, endoprosthetic replacement.

Ankle Arthrodesis following tumour excision can be achieved by vascularized autografts, non-vascularized autografts, allografts, or pasteurized autografts. Despite good functional outcomes, limitations for this procedure include stiff ankle, a long period of recovery, infection, and non-union. Saglik et al have described the use of Fibular autografts to achieve arthrodesis in 2 patients with aggressive GCT of distal Tibia [9]. Economopoulos et al achieved arthrodesis using a custom made porous tantalum spacer after tumour resection in distal Tibia and suggested that Tantalum to be a feasible structural substrate and a suitable substitute to bone grafts [10].

Similar to mega-prosthesis in the treatment of aggressive GCTs around the knee, custom–made ankle endoprosthesis have been introduced. V A Singh et al treated 4 cases (1 primary and 3 recurrent) by endoprosthetic reconstruction. One developed deep infection and one had talar collapse [11]. In another series by Shekkeris et al, 2 out of 6 cases developed deep infection [12]. Although ankle endoprostheses looks a viable option, concerns regarding long term outcomes, deep infection, implant loosening and cost of implant need to be addressed.

Conclusion
GCTs affecting the distal tibia are rare to encounter. Bony defects can be filled with autografts. However, when there is recurrence, it is still amenable to do extended curettage and use bone cement as void fillers to achieve local control of the disease. This modality of treatment offers good stability and early ankle mobilization is possible. Nevertheless, a periodic follow-up is still warranted to watch out for late re-recurrences.

Conflict of Interest
We declare that we have no conflict of interest.
References


