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A Rare Case of Giant Cell tumor of Bone in Distal Tibia

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Learning Point of the Article:

Giant Cell Tumor of bone should be considered as a differential diagnosis for any lytic lesion, even at uncommon sites and in treatment, along with adjuvants, bisphosphonates also can be used in decreasing the chances recurrence.

Abstract

Introduction: Giant cell tumors of bone commonly seen in the distal femur, proximal tibia and distal radius and are seen in the age group of 20–40 years with female preponderance. Being close to the joint, the management is formidable, and there are no clinical, radiological or histological parameters to predict chances of recurrence or metastasis. Here, we present a rare case of giant cell tumor of the distal tibia treated successfully with no recurrence at the end of 4 years.

Case Report: A 33-year-old female presented with complaints of pain, swelling and decreased range of motion (ROM) at the left ankle joint for 6 weeks. On examination, we observed swelling and tenderness on left ankle joint with decreased ROM. X-rays and computed tomography revealed a well-defined expansile osteolytic lesion in the distal epiphysio-metaphyseal region of the left tibia without breach in the tibial cortex or ankle joint. Surgery by extensive curettage, adjuvants, cementation and prophylactic fixation was performed. Oral alendronate was started and continued for 24 months after surgery.

Conclusion: The patient at 4-year follow-up is doing well, walking without pain, able to squat, having full ROM of the ankle joint with no signs of recurrence.

Keywords: Giant cell tumor of distal tibia, curettage, cementation and fixation, bisphosphonates.

Introduction

Giant cell tumor of bones (GCTB) is a primary benign but locally aggressive bone tumor, most commonly occurring in long bones of young adults with a female preponderance affecting people aged 20–40 years [1, 2]. It accounts for approximately 5% of all primary bone tumors [3]. It is characterized by the presence of large multinucleated osteoclast-like giant cells, facilitating bone destruction [4]. The commonly affected site is around the knee joint, followed by the distal radius [3]. We are here reporting a rare case of GCTB in the lower end of the tibia in a 33-year-old female. The treatment in GCTB is surgical removal, and the surgical options vary from intralesional curettage (IC) to en bloc resection [5]. Preferably, the IC is combined with local adjuvants, including alcohol, phenol, liquid nitrogen, argon beam coagulation, cryo-ablation, hydrogen peroxide and mechanical high-speed burr [2,4,6]. The cavity after IC is usually filled with autologous or allogenous bone graft or preferably with polymethylmethacrylate cement (PMMA), as PMMA has many advantages over bone graft [6,7]. Bisphosphonates with anti-osteoclastic activity have been investigated and found to be

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Figure 1: Initial X-ray showing bone lesion.

efficient in controlling disease progression and reducing recurrence [4,8]. The drug is used in pre-operative and post-operative periods for a long duration.

Case Report

A 33-year-old female presented with pain and swelling at left ankle from last 6 weeks after a twisting injury. She first visited another doctor 2 weeks back, but pain persisted. On our evaluation, there was swelling at the anterolateral corner of the left ankle with mild tenderness and painfully decreased range of motion at the left ankle. Ankle radiograph demonstrated a well-defined, radiolucent lesion in the distal tibia antero-laterally extending to subchondral bone. The tibial cortex appeared intact, and the ankle joint was congruent and apparently not breached (Figs. 1 and 2).

Computed tomography (CT) left ankle revealed an expansile, eccentric osteolytic lesion in the distal tibia with cortical thinning but continuity still maintained and similarly intact tibio-talar joint surface (Fig. 3 and 4). GCTB was the most probable provisional diagnosis. The patient was planned for incisional biopsy/frozen section and treatment accordingly as soon as possible. We applied a back slab and started oral Alendronate 70 mg/week.

She got herself admitted after 4 weeks for the planned surgery. Required blood investigations, chest X-ray and chest CT were done, which were clear.

Figure 2: Initial X-ray showing bone lesion.

Under spinal anesthesia and the patient in supine position, tourniquet was applied, an anterolateral approach to sufficiently expose the tumor site was used. The lesion was curetted a bit, and sampled tissue sent for frozen section, which confirmed it as consistent with GCTB. On the confirmation of being benign, we moved as per the treatment plan, which was extensive IC, use

Figure 3: Computed tomography of bone lesion in distal tibia.

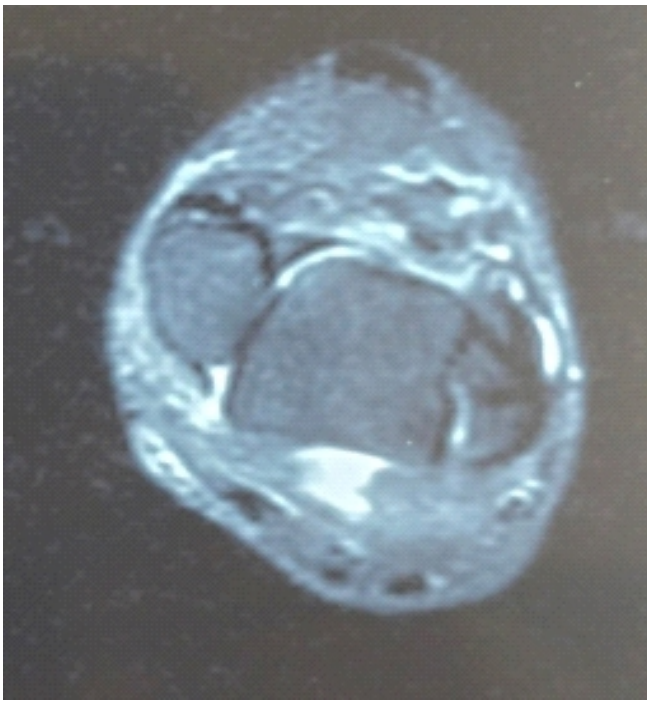

Figure 4: Computed tomography of bone lesion in distal tibia.

of hydrogen peroxide, high-speed burr, cementation and prophylactic fixation with plate.

The distal tibia was considerably exposed with mobilization of the tendon and neurovascular bundle. A cortical window of good size was made, and extensive curettage beyond the tumor margin into the normal bone was done, carefully preserving the ankle joint line. As an adjuvant, a hydrogen peroxide wash and high-speed burr were used. From a medial approach, a plate with screws was used for prophylactic fixation of the distal tibia and the screws in the distal tibia used as rebar to maintain the cement plug. Under fluoroscopic guidance, the big cavity formed after complete resection of the tumor was packed with bone cement. Closure was done, and pressure dressings were completed. Tourniquet was deflated.

Post-operative care and follow-up

In the immediate post-operative time, limb elevation was done, and the patient was encouraged for active ROM exercises of the ankle after 24 h of surgery. Strict non-weight-bearing for 6 weeks after surgery with physiotherapy. Then, partial weight-bearing with crutches was allowed and full weight-bearing at 12 weeks. Alendronate was advised for 24 months, which she continued. She was back to her office job in 14 weeks after surgery. She gained full ROM of the ankle, pain-free walking and able to squat. She was followed for 4 years, and she is doing well. Repeat X-ray at intervals reveals no abnormality or recurrence (Fig. 5 showing radiograph at 6 months and Fig. 6 at 4 years).

Figure 5: Post-operative X-ray 6 months after surgery.

Discussion

GCTB accounts for around 5% of all primary bone tumors [3]. They are expansile, osteolytic, benign but locally aggressive neoplasm but rarely can metastasize (3%) [1,4]. These are commonly seen around the knee and distal radius. Only rarely they can be seen in other sites, such as the distal tibia, as in this case presented here. Involvement of the foot and ankle is rare and comprises <4% of all GCTB [9]. The typical appearance of GCTB is best demonstrated on conventional radiographs, which show a lytic lesion with a geographic type of bone

Figure 6: Post-operative X-ray 4 years after surgery.

destruction, a well-defined but non-sclerotic margin, eccentric in location, that extends to the subchondral bone, and occurs in patients with closed physis [10]. The abundance of multinucleated osteoclast-like giant cells is a histological hallmark of the tumor which mediates bone destruction [4,5,11]. The tumor tissue contains spindle-shaped stromal cells, which are the primary neoplastic cells [1,4,12]. Their biologic behavior and typically juxta-articular location require specific surgical management with adjuvants. Extensive curettage is the surgical treatment of choice, especially if the cortex is intact and the joint line not penetrated [2,10,13]. Literature suggests that IC strikes the best balance between controlling disease and preserving function in most of the cases [14]. Chances of recurrence and loss of support of the articular surface have continued to plague the treatment of GCTB. The recurrence rate is nearly 50% with conventional curettage and bone grafting [6]. Surgeons face difficulty in differentiating the radiographic changes seen with the bone graft incorporation and recurrence of the tumor [9]. PMMA is an ideal filler of the cavity created as the cement provides early weight-bearing, early rehabilitation, no donor site morbidity, ease of detection of recurrence, and has a tumoricidal effect by its exothermic reaction, monomer diffusion, and hypoxemia [9,13]. Bone cement has been proposed to reduce local recurrence, resulting in disease-free survival rates as high as 85% [2]. It was documented that polymethylmethacrylate enhances the margin of tumor cavity by 1.5–2 mm in cancellous bone and 0.5 mm in cortical bone [6,12]. Local adjuvant therapy with cytotoxic agents like hydrogen peroxide and the use of high-speed burr have been used intra-lesionally to improve local margins and reduce recurrence [6,10]. In one study, it was recorded that use of cementation, prophylactic fixation, argon laser, and hydrogen peroxide perhaps contributed to no recurrence after 3.5 years [11]. Several other authors have since documented recurrence rates between 8% and 15% using these methods [6,12]. It was seen that the use of prophylactic fixation to stabilize the distal tibia cortex during surgical treatment of GCTB of the distal tibia contributes to the prevention of pathological fracture [11].

Bisphosphonate with anti-osteoclastic effect was introduced with good efficacy [2]. This drug promotes apoptosis of the stromal cells in GCTB [8, 15]. It was found to reduce recurrence after surgery. Balke et al reported that patient treated with oral alendronate (70 mg/week) for 24–32 months show that most tumors did not increase in size, and control of recurrence and pain was achieved, and did not report any untoward effect [8].

Conclusion

We must think of the rare occurrence of GCTB in unusual sites, as it is scarcely seen in the distal tibia and demands careful management to have a disease-free and quality life. In the 4 years of follow-up in our case, we observed that extensive IC, adjuvants, cementation and prophylactic fixation supports joint preserving surgery and prevent pathological fracture. Use of bisphosphonates can halt progression of disease and prevent recurrence, and this case, as well few other studies, highlight the need to establish the efficacy of bisphosphonate and inclusion in standardised protocols for the treatment of GCTB.

Clinical Message

Giant cell tumors deserve consideration as a differential diagnosis of any lytic bone lesions in young adults. Although they are common at the distal femur, proximal tibia, and distal radius but can also occur at a rare location like the distal tibia, as seen in our patient. Extensive intralesional curettage, hydrogen peroxide, high-speed burr, cementation, and alendronate give excellent results and reduce the chances of recurrence, and prophylactic fixation prevents pathological fracture.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given the consent for his/ her images and other clinical information to be reported in the journal. The patient understands that his/ her names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflict of interest: Nil **Source of support:** None

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