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Case Report

Aggressive giant cell tumour of the talus: a rare case report and challenges faced during its management

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ABSTRACT

Giant cell tumour (GCT) of the bone is a locally aggressive benign tumour that commonly occurs in the epiphyseal ends of long bones. Its occurrence in the small bones of the foot like the talus is rare. In this case report we described a case of aggressive GCT of the talus (Campanacci grade 3) in a twenty-one years old male diagnosed by the presence of a well-defined osteolytic lesion seen involving the talar body with cortical breach and soft tissue extension confirmed to be a GCT by closed needle biopsy. The patient was managed surgically by excision of the tumour mass in-toto along with the talus followed by tibio-calcaneal fusion using free fibular strut graft and Ilizarov frame. At 3 months follow up no evidence of fusion was seen and therefore secondary augmentation with a cortico-cancellous graft from the iliac crest was done to aid fusion and Ilizarov fixator was removed 6 months after the surgery. The patient at one year follow up had shown no signs of recurrence of the tumour and has a pain free, stable fused tibiocalcaneal joint with a bipedal unassisted gait. Our case report found excision of talus combined with tibio-calcaneal fusion using free fibular strut graft along with cortico cancellous iliac bone graft to have a good outcome in terms of aiding tibio-calcaneal arthrodesis and thereby providing pain relief and providing a stable foot to the patient.

Keywords: GCT, Talus, Tibiocalcaneal fusion

INTRODUCTION

GCT of the bone is a locally aggressive benign tumour that accounts for approximately 5% of all bone tumours.[1,2] It was first described by Cooper in 1818.[3] The tumour is commonly seen in females in the 3rd to 4th decade of life, often arising from the epiphyseal ends of long bones.[2] Most common sites include the distal end of the femur, proximal end of the tibia and distal end of the radius.[4] The involvement of the small bones of the foot like the talus is rare and these tumours tend to behave more aggressively and are seen in younger age groups.[5,6] Radiologically GCT's are osteolytic in nature, eccentrically placed and aggressive tumours that may show thinned out cortices or cortical breach. Microscopically they are composed of multinucleated giant cells along with proliferating mononuclear spindle cells and mitotic figures. Owing to the rarity of this tumour, aggressive nature, preponderance to recur and lack of a well-defined treatment protocol we report a case of aggressive GCT of the talus in a twenty-one years old male managed by excision of the tumour, total talectomy and tibio-calcaneal fusion using free fibular strut graft and an Ilizarov frame, later augmented with cortico cancellous iliac crest graft to aid in tibio-calcaneal fusion and help provide a tumour free stable joint.

CASE REPORT

A twenty-one year old male student presented with complaints of pain over the left ankle region for the past five months associated with a progressive swelling over the left foot and ankle region for the past 2 months. The pain was insidious in onset, dull aching, diffuse type of

pain with no aggravating or relieving factors. There was no history of trauma and no history of fever episodes. Clinical examination of the foot and ankle revealed diffuse swelling over the dorsal aspect of the mid foot. Warmth and tenderness were noted over the same region. Ankle movements were restricted and painful with 0 degree of dorsiflexion and 10 degrees of plantar flexion and subtalar movements were terminally restricted. Sensations over the foot were intact. On plain radiograph of the ankle, a well-defined eccentrically placed osteolytic lesion was noted involving the talar body. Cortex appeared to be thinned out with evidence of cortical breach and soft tissue extension of the lesion. The lesion appeared to have a narrow zone of transition (Figure 1).

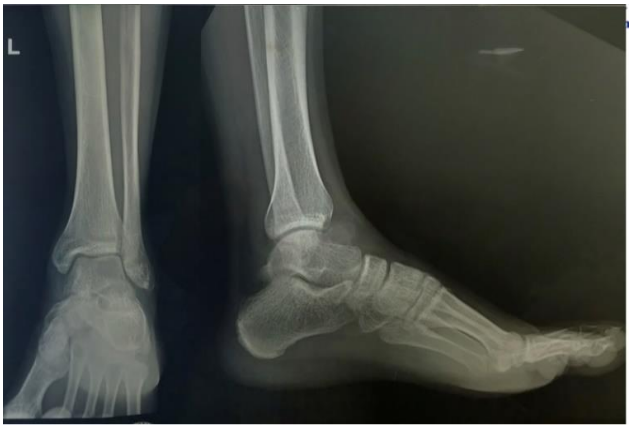

Figure 1: Plain radiograph of left ankle showing osteolytic lesion involving the talus with cortical breach and soft tissue extension.

MRI study confirmed the presence of a bony lesion involving the talar body extending into the soft tissue along with evidence of marrow edema, ankle and subtalar effusion (Figure 2 and 3). General body examination and chest radiograph were done to rule out metastasis and were found to be normal. Blood parameters including complete haemogram, erythrocyte sedimentation rate and C-reative protein were analyzed and found to be within the normal range. A closed needle biopsy done in an outside hospital confirmed the diagnosis of a GCT.

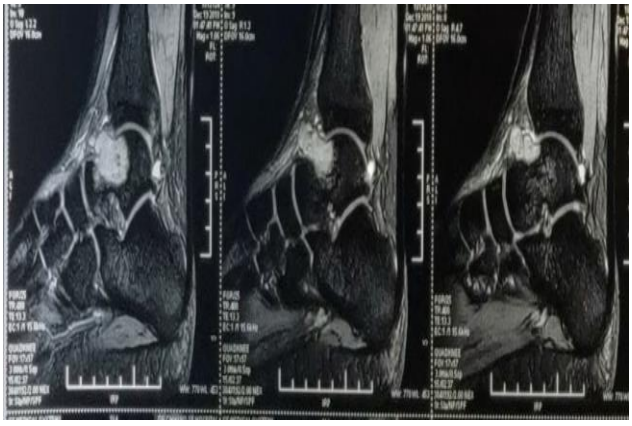

Figure 2: MRI sagittal section of the left ankle.

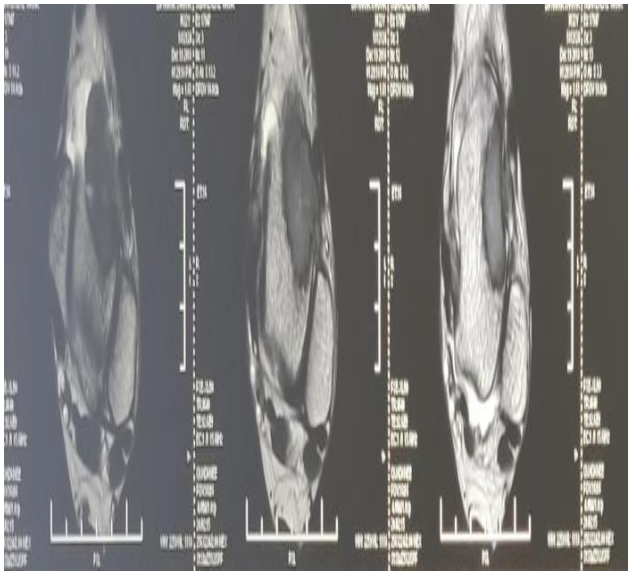

Figure 3: MRI axial section of the left ankle.

Based on the above findings the tumour was classified as a Campanacci grade 3 aggressive GCT of the talus with soft tissue extension. Owing to the aggressive nature of the tumour dilemma over the mode of management existed. After a detailed discussion on the treatment protocol the patient underwent excision of the tumour in toto along with total talectomy through an anterolateral approach that included the previous closed biopsy tract in its incision. The dorsalis pedis artery and superficial peroneal nerve were identified and isolated. The reddish-brown tumour mass was visualized and excised along with the removal of the entire talus and the entire specimen was sent for histopathological examination (Figure 4 and 5). HPE showed non neoplastic multinucleated osteoclast like giant cells with mono nuclear neoplastic cells arranged in sheets in between (Figure 6).

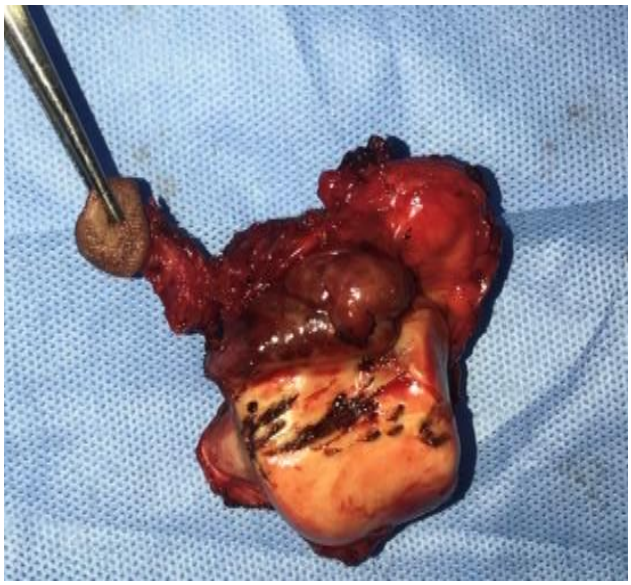

Figure 4: Tumour mass removed into along with skin tag from the biopsy site.

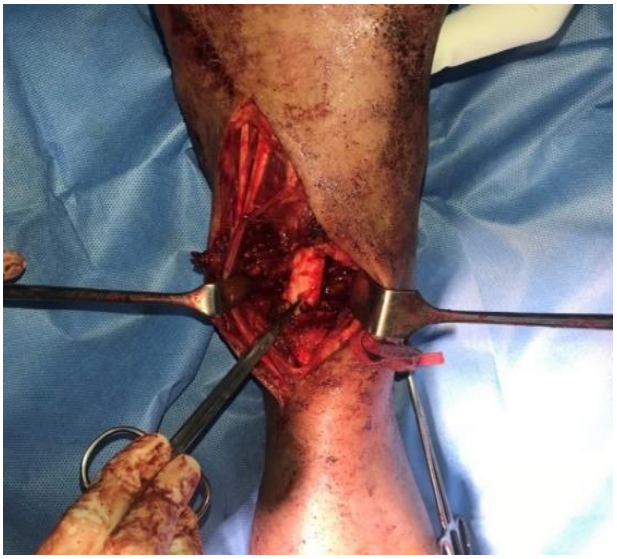

Figure 5: Placement of free fibular strut graft.

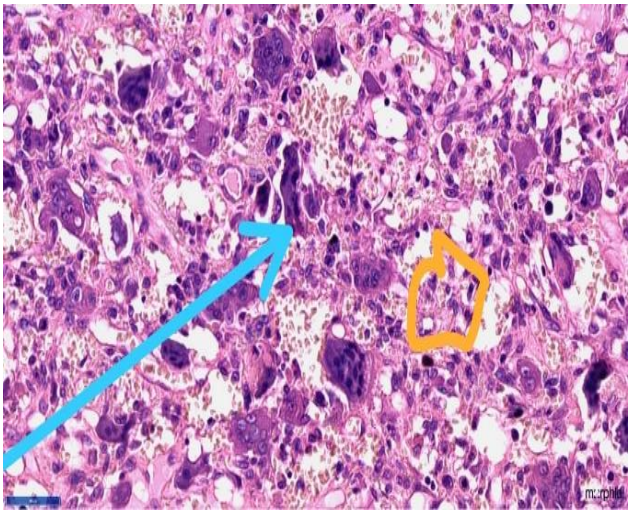

Figure 6: No neoplastic multinucleated giant cells. Orange: mononuclear tumour cells.

The defect created was filled using an autologous ipsilateral free fibular strut graft that was placed between the tibial end proximally and the calcaneal surface distally the articular cartilage of distal tibia and superior calcaneum was denuded and a shallow slot was created to hold the inserted fibula graft. Tibio-calcaneal fusion was attempted and the graft held in place by using an ilizarov fixator ensuring that the heel was in neutral position. The patient was then mobilized with strict non-weight bearing measures. At 3 months of follow up, the plain radiographs of the ankle showed delayed healing of the fibula graft to tibia and calcaneum (Figure 8). Osteo integration had failed to occur between the tibial and calcaneal ends of the fibular graft owing to its cortical nature. The patient was then planned for secondary augmentation of tibio-calcaneal arthrodesis using autologous ipsilateral cortico cancellous bone graft from the iliac crest. The graft was harvested and packed in and around the free fibular strut graft after making multiple drill holes in the exposed surface of distal tibia and superior calcaneum. The Ilizarov frame was maintained and the patient was advised to continue non-weight bearing mobilization. At 6 months of follow up the plain radiographs revealed signs of tibio calcaneal fusion, clinically no abnormal mobility was noted and the Ilizarov frame was removed and the limb immobilized using a below-knee cast (Figure 9). The cast immobilization was continued for two months following which sequential partial and then full weight-bearing mobilization was allowed. At 12 months of follow up plain radiographs of the foot and ankle revealed no radiological signs of recurrence and the fibula graft has united to the distal tibia and calcaneum (Figure 10). Apart from mild pain, the patient is otherwise satisfied with his treatment and is able to carry out his daily activities.

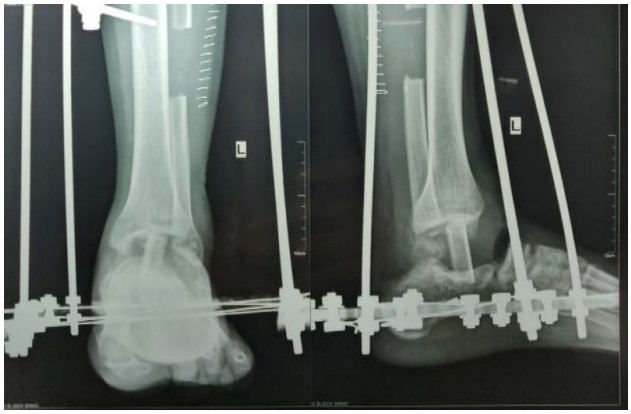

Figure 7: Immediate post-op radiograph showing complete resection of tumour along with total talectomy and tibio-calcaneal arthrodesis using free fibular strut graft and Ilizarov frame.

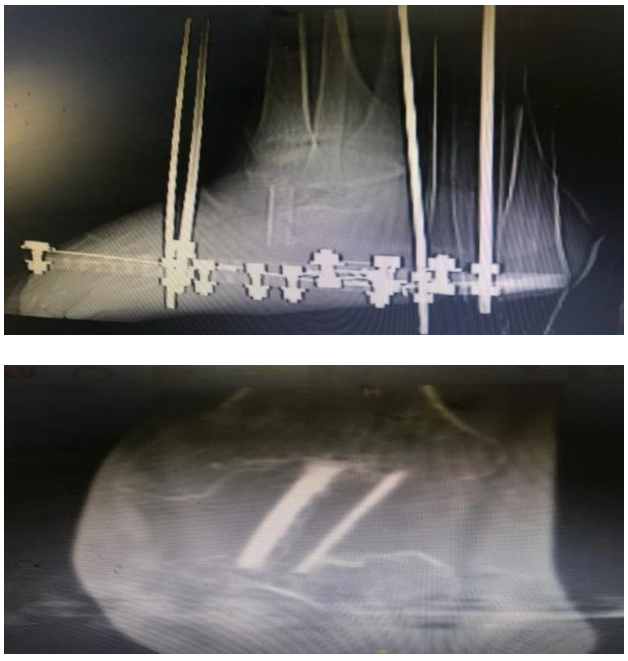

Figure 8: Three months post-op CT scan of the left ankle showing a fibular strut graft *in-situ* with no signs of fusion.

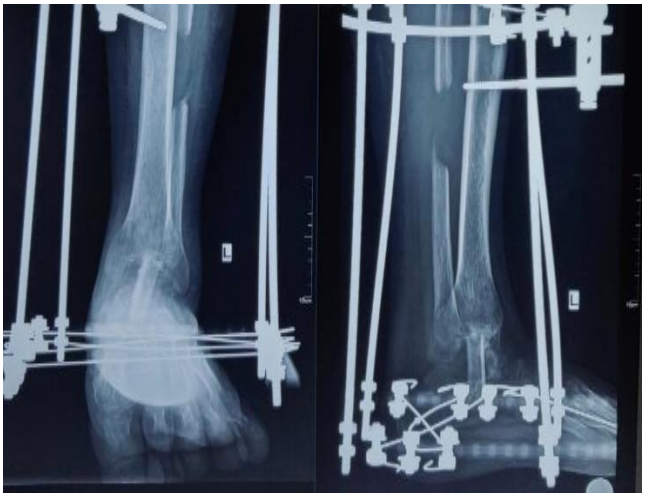

Figure 9: Six months post-op radiograph of left ankle showing fibular strut graft *in situ* with evidence of tibio-calcaneal fusion.

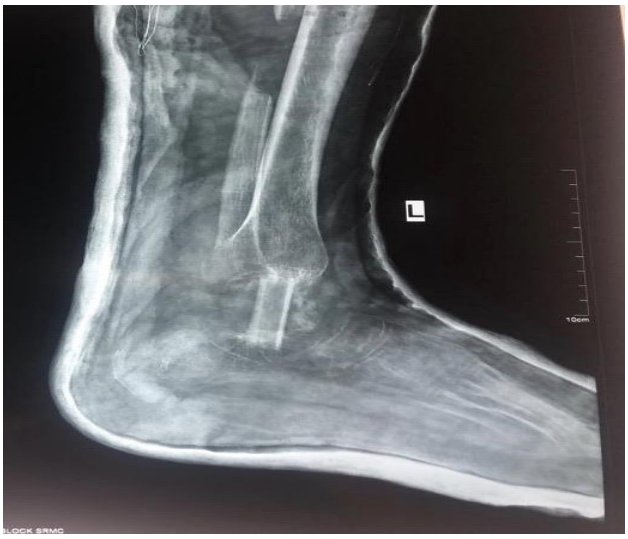

Figure 10: Post-op plain radiograph of the ankle following removal of the Ilizarov frame and application of a below-knee plaster of Paris cast.

DISCUSSION

The talus bone is a relatively uncommon site for GCT occurrence. Several studies like those by Dhillon et al, Goldenberg et al and Sung et al document the rarity of talus and other small bones of the foot as sites for GCTs and stress upon the need for anatomic and biomechanical considerations while planning its management.[6-8] These tumours tend to behave differently, are more multicentric and have a high propensity to recur.

Clinically these patients may present with dull aching diffuse pain around the ankle region occasionally associated with swelling, with or without any preceding history of trauma. Presentation is vague and may often be misdiagnosed as an ankle sprain during the early stages of the disease.

Radiologically these tumours present as eccentrically located osteolytic lesions with thinning of cortices and narrow zone of transition. Unusual sites of GCT may not always present with the classical radiological picture mentioned above and sites of bone destruction and soft tissue expansion may often be noted. In our case, an osteolytic lesion was noted within the talus with cortical break and soft tissue extension that aided in narrowing our diagnosis. Campanacci et al in their study graded a system to help classify these tumours to aid in their management.[9] In our case report, the tumour fell under grade 3 based on Campanacci's grading system denoting it to be an aggressive GCT and increasing its risk of recurrence.

Management of GCT aims at local control without sacrificing the joint as it is a benign locally aggressive lesion. This is usually achieved by extended intralesional curettage and reconstruction of the cavity using an autologous bone graft or bone substitute. But high rates of recurrence have been reported following these procedures.[10] Several other treatment options like fresh frozen osteochondral allograft, partial or total resection and arthrodesis have also been described.[11] As in our case, the lesion already had a soft tissue extension, therefore resection of tumour along with the complete resection of talus and tibio-calcaneal fusion was planned. Several studies have adopted the same line of management. Dhillon et al in their study treated two cases of aggressive talus GCT with tumour resection along with talectomy without fusion and noted instability as a complication. Sharma et al in their study reported a case of GCT talus with extensive talus destruction and thinned out cortices that they managed with tumour resection, total talectomy and tibio-calcaneal fusion using a Steinman pin and cortico cancellous iliac crest graft and reported good results.[12] In our patient, the initial tibio-calcaneal fusion was attempted using an ipsilateral free fibular strut graft and stabilised using an ilizarov frame. Owing to the failure of fusion the graft was augmented 3 months postoperatively using autologous cortico cancellous iliac crest graft that later yielded good fusion. Arthrodesis is essential for all tarsal bone resection except that of the calcaneum. Amputation is reserved as an option only for recurrences. Radiotherapy can be used in inoperable cases.[13,14]

Limitation of the study was that the patient was followed up to 1 year after the surgery, more years of follow up was needed to identify any recurrence in future and to access the functional outcome.

CONCLUSION

Owing to the paucity of such cases, the treatment protocol is still ill-defined. All GCT's of the foot should be considered locally aggressive and we advocate adequate surgical management of the same. As curettage has a high risk of recurrence, GCT's of the talus of grade 2 and 3 should be considered for tumour resection along with partial or total talectomy and fusion. Although amputation may prevent recurrence it should be saved for recurrent

cases only as the surgery is cosmetically deforming and has a low rate of acceptance from the patients.

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Ethical approval: Not required

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