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Case Report

Orthopaedics Section

Aggressive Giant Cell Tumour of Talus with Pulmonary Metastasis-A Rare Presentation

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ABSTRACT

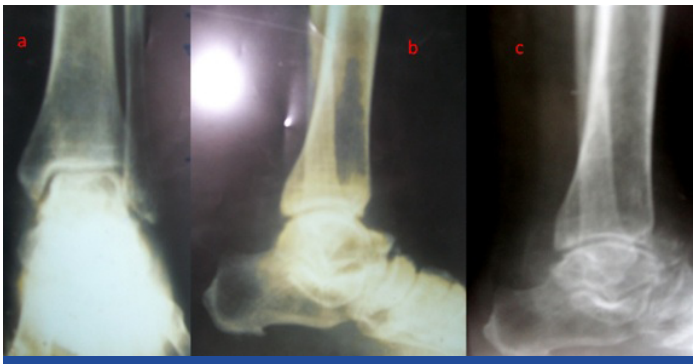
Giant cell tumour (GCT) is a primary benign neoplasm of bone. It is classically described as a locally invasive tumour that occurs close to the joint of a mature bone. It accounts for 5% of all skeletal tumours. It usually originates from long bones. Giant Cell Tumour of the small bones of the hand and foot are relatively uncommon. Giant Cell Tumour of talus is a rare occurrence. We report a rare presentation of giant cell tumour of the talus in a 62-year-old farmer treated by talectomy and tibiocalcaneal fusion, who later presented with features suggestive of recurrence and secondaries in chest within six months following surgical resection of the primary. Below knee amputation was performed to manage the recurrence. At two years follow-up the patient showed no further progression of pulmonary metastasis or local recurrence.

Keywords: Giant cell tumour of talus, Pulmonary metastasis, Talectomy, Tibiocalcaneal fusion

CASE REPORT

A 62-year-old farmer presented with complaint of pain around right ankle for about one month duration. He complained of dull aching, continuous pain persisting for almost a period of one month which interfered with his daily activities. The occupational work load, lack of accompanying complaints and radiological findings [Table/Fig-1a,b] of narrowed joint space of the involved joint all pointed out to one possible diagnosis of arthritis of the ankle joint. Below knee POP slab and analgesic medication were the treatment modalities offered to the patient. However, surprisingly he returned two weeks later with increased pain and significant limitation of daily activities than ever before. When the previously applied slab was removed, a swelling below the ankle joint involving the hindfoot was noticed. The swelling was lying anterior to the Achilles tendon and was tender, firm in consistency and yielding to touch. Repeat radiography revealed an expansile lytic lesion involving the talus [Table/Fig-1c] with no periosteal reaction, matrix calcification or soft tissue extension. The ankle joint was not infiltrated by the tumour. Further investigations revealed elevated serum acid phosphatase levels. Renal and hepatic profiles were all with normal limits. Blood glucose, CRP and serum calcium levels were also reported within the normal range. FNAC was performed, histopathology revealed a giant cell tumour. There was evident cortical breach on radiography itself which allowed us to refrain from the use of preoperative MRI or CT scan. General body survey and a chest radiograph were performed to rule out multicentric disease and pulmonary metastasis.

Talectomy with tibio calcaneal fusion was the initial surgical option that we chose based on patient's clinical and radiological features which correlated with a favourable outcome through such procedure. At the present time, recognized effective chemotherapeutic agents are also not available for the management of these tumours.



[Table/Fig-1a&b]: Radiograph at initial presentation
[Table/Fig-1c]: Radiograph taken after two weeks

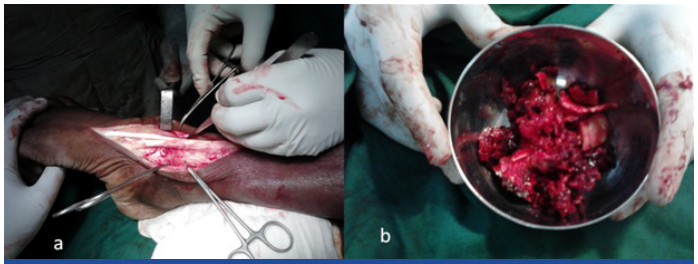
Procedure

The patient was operated under spinal anaesthesia in supine position with the involved limb under tourniquet control. Standard anterolateral incision was made to expose the talus and talectomy was performed [Table/Fig-2a]. Tibiocalcaneal fusion was performed with iliac crest and fibular strut graft [Table/Fig-2b]. Internal fixation using K wires was done to supplement the construct and above knee plaster cast was applied.

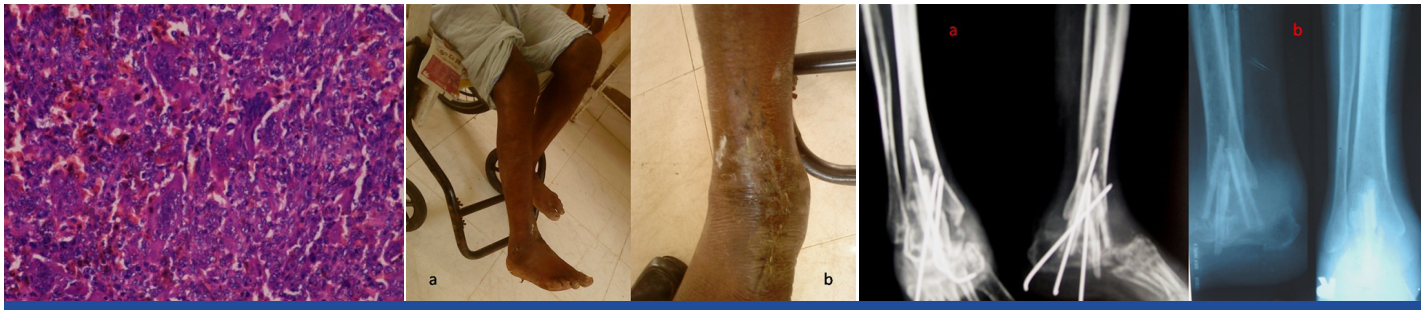
Postoperative period

Histopathological examination confirmed the diagnosis of giant cell tumour [Table/Fig-3]. The wound healed uneventfully [Table/Fig-4a&b]. The k wires were removed at two months follow up and the cast was reapplied [Table/Fig-5a,b]. The patient was kept on bisphosphonates in the postoperative period (Ibandronic acid 150 mg orally once a month) to prevent recurrence. Bisphosphonates act by targeting osteoclast-like giant cells inducing apoptosis and limiting tumour progression.

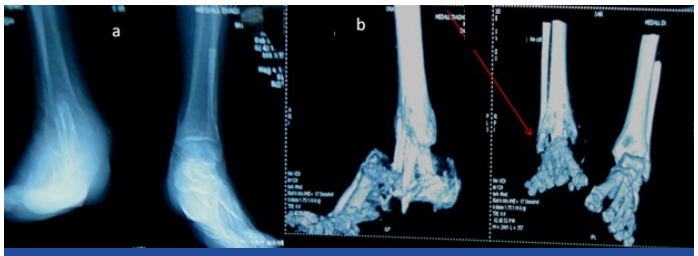
Six months following surgery the patient presented with increased pain and swelling of the right ankle. A CT scan performed at that stage showed recurrence [Table/Fig-6a&b] of the tumour, confirmed further, by a repeat biopsy. Chest CT scan of the patient taken at that stage revealed metastasis [Table/Fig-7]. In view of the aggressive recurrence and chest metastasis, below knee amputation was performed. The stump healed uneventfully. At two years follow-up there was no recurrence [Table/Fig-8a,b]. The chest metastasis did not progress.



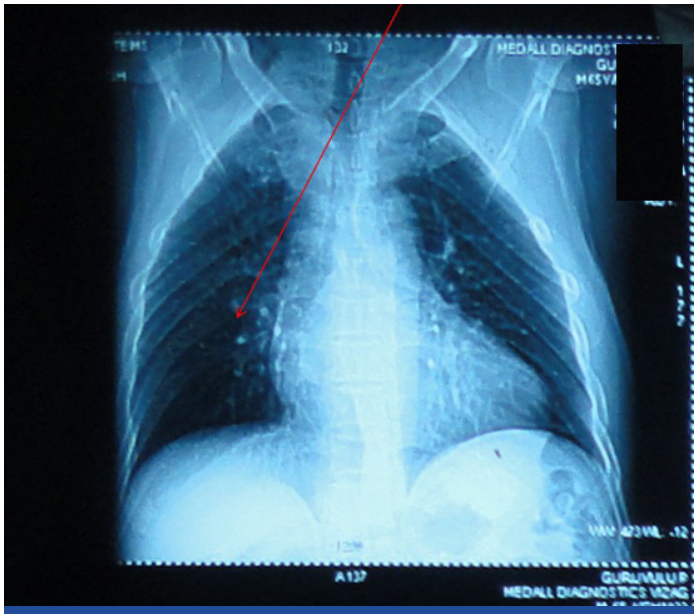
[Table/Fig-2a]: Intraoperative picture showing anterior approach to talus
[Table/Fig-2b]: Tumour mass following excision



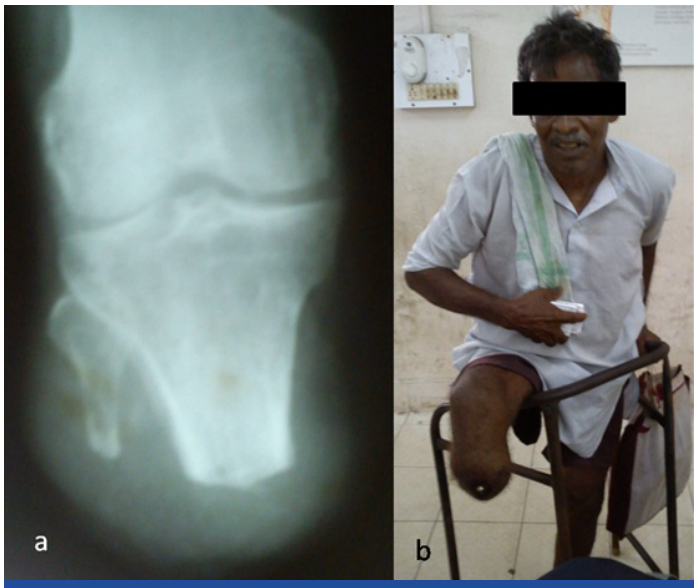
[Table/Fig-3]: Histo pathological slide showing features suggestive of giant cell tumour
[Table/Fig-4a&b]: Post Operative clinical picture of the patient and wound site
[Table/Fig-5a]: Immediate post op x ray showing internal fixation using k wires
[Table/Fig-5b]: X ray taken following removal of k wires (at 2 months follow-up)



[Table/Fig-6a]: X-ray at six months follow up
[Table/Fig-6b]: CT scan at six months follow up showing recurrence



[Table/Fig-7]: CT chest suggestive of metastasis



[Table/Fig-8]: X ray of stump at two years follow up
[Table/Fig-8b]: Clinical picture at two years follow up

DISCUSSION

Giant Cell Tumour has serious potential for local recurrence, malignant transformation and metastasis. It is usually seen in the third and fourth decade with a slight female preponderance. It is a relatively rare at the ankle but is known to behave unpredictably when situated at that location. Giant cell tumour of the talus is extremely rare and so far only 14 cases are reported in the literature [1].

Murari et al., found GCT to be the most common benign primary osseous neoplasms in the foot, comprising 19% of all lesions. GCT talus presents a difficult problem because the lesion is locally destructive, recurs even in the face of aggressive surgery and on occasion may even metasize. Goldenberg et al., in their series of 218 cases found only one case involving the talus [2]. Mirra et al., report an incidence of less than 2% in the foot and Huvos reports an incidence of 1.8% in the foot [3].

The clinical presentation of GCT is insidious onset of pain and swelling at the affected site. This non-specific symptom in many cases may be mismanaged as infection, arthritis or as chronic sprain [4]. GCT of foot usually occurs in a slightly younger age group [5] and is more aggressive than those of long bones.

The lesion in our patient involved the head and neck region of talus, was aggressive in nature which was evident by the rapid growth in a short term. Pulmonary metastasis developed within a short duration of six months following resection of the primary tumour. Although, recurrence of GCT is reported to be 10-30%, recurrence in talus is not been reported yet. The salvage options in recurrence of talus are limited.

Currently, the modalities of treatment for GCT include curettage and grafting, enbloc resection with grafting or arthrodesis, chemical cautery, talectomy and amputation. Talar lesions require partial or total talectomy with arthrodesis.

Arthrodesis is essential after resection of all tarsal bones except calcaneum [6]. Limb salvage and amputation is reserved for recurrences only [7]. Radiation therapy is considered for inoperable cases but it has no role in the treatment of completely resected tumours [8].

Recurrence of the tumour as a result of curettage has a high probability [9]. While the tumour is considered to be locally aggressive, occasional distal metastases are identified [10]. Fresh frozen osteochondral allograft reconstruction has also been described for an aggressive GCT of talus but there is paucity of literature on this particular modality of treatment [11]. In judiciously selected cases of giant cell tumour of the talus with the lesion well localized without a cortical break, properly performed extensive curettage and bone grafting can be a good option for complete removal of the tumour while preserving near normal structure and function of the ankle [12].

At about six months follow up following surgery our patient developed local recurrence and pulmonary metastasis even after complete talectomy and bone grafting. The only limited option left was amputation to prevent serious complications.

CONCLUSION

Giant cell tumours less commonly occur in small and flat bones and are described as being more aggressive in such less common sites. The diagnosis, management and prognosis (tendency for local recurrence and distant metastasis) of giant cell tumour of small bones of foot especially talus is still a matter of controversy. Hence, prompt aggressive management is essential for the tumour presenting in such sites. Talectomy with tibiocalcaneal fusion has a strong indication for aggressive giant cell tumours with extensive bony destruction, collapse and adjacent articular degeneration. In our case report even with an early diagnosis and aggressive treatment we could not prevent recurrence as well as chest metastasis. This case report also illustrates that metastasis in a giant cell tumour does not hold the same prognosis as metastasis in a malignant tumour. Due to the risk of local recurrence and pulmonary metastasis, the follow up of patients including radiological imaging of the involved area and the lungs is important.

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