CASE REPORT

Giant Cell Tumor of the Talus: A Case Report and Review of Literature

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Abstract

Aim and objective: This study aims to describe a rare case of giant cell tumor (GCT) of the talus.

Background: Giant cell tumors rarely present around bones of the foot and involvement of the talus is infrequent. In comparison to long bones, diagnosis and management of talus GCT is challenging and is sparsely reported in the literature.

Case description: We report a case of GCT arising from the talus in a 19-year-old boy, presenting as non-specific foot pain for the past 2 months. The diagnosis was established by open biopsy and treated with curettage, bone grafting, and subtalar joint arthrodesis. At 6 months of follow-up, the patient had painless arthrodesis of subtalar joint with functional ankle joint and no sign of recurrence at last clinicoradiological examination.

Conclusion: Diagnosis and management of GCT talus is challenging and can be treated with extended curettage with subtalar arthrodesis.

Clinical significance: Presentation of GCT talus may be missed at early stages. A high index of suspicion can help in diagnosis and appropriate management.

Keywords: Case reports, Giant cell tumor talus, Giant cell tumor, Management, Outcome, Review of literature.

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Background

A giant cell tumor (GCT) is described as a neoplasm of undifferentiated stromal cells with abundant mesenchymal multinucleated giant cells. Typically, the tumor involves the epiphyseal region of long bones with the most common site around the knee.[1] Foot involvement is rare and GCT arising from talus is rarer.[2] So far only 19 studies have reported cases of GCT of the talus. As a result of their rarity and unfamiliar presentation, diagnosis is usually missed or delayed. In previously reported cases, management ranged from intralesional curettage to total talectomy with stabilization of the subtalar joint.[3] We report a case of GCT arising from the talus, presenting as non-specific foot pain, in a 19-year-old boy, treated with curettage, bone grafting, and subtalar joint arthrodesis.

Case Description

A 19-year-old boy presented to the outpatient department of our institution with chief complaints of pain in the right foot for 2 months, swelling around the right ankle for 1 month, and difficulty in bearing weight on the affected side for 3 weeks. The pain was insidious in onset with increasing intensity over time while swelling was slowly progressive. There was no history of trauma, fever, loss of appetite, loss of weight, pain at neither other parts of the body nor any history of similar complaints in the past.

The general physical and systemic examinations were within normal limits. On local examination, the attitude of the limb was neutral. There was a 6 × 3 cm swelling over a dorsolateral aspect of the right foot and the anterior aspect of the ankle joint. There were no visible veins, sinus, or discharge from the swelling. The local temperature was raised slightly and the swelling was tender. All movements at the ankle and subtalar joint were painfully restricted. Routine blood investigations were within normal limits including erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP). Anteroposterior and lateral radiographs of the ankle showed a geographic osteolytic lesion in the body and neck of the talus with a narrow zone of transition and no cortical break (Fig. 1). Non-contrast computed tomography (NCCT) of the affected part was corroborative of X-ray findings. MRI was performed to delineate soft tissue extent, the lesion measured approximately 35 × 20 × 22 mm involving lateral part of the neck and lateral process of talus without soft tissue involvement (Fig. 2).

An open biopsy was performed for diagnosis confirmation. The histopathological examination was suggestive of GCT. As the lesion was localized to the talus, thorough curettage and bone grafting were planned. A standard anterolateral incision was made and the lateral flap raised followed by fibular osteotomy to expose the talus. A cortical window was made on the lateral surface to enter the lesion. Extended curettage was performed; tumor material was reddish-brown in color with a soft consistency. Curettage with a burr, cauterization with absolute alcohol was done to clear tumor tissues. The resultant cavity was filled with autologous cancellous bone graft harvested from the ipsilateral iliac crest mixed with synthetic bone substitute. Although the tumor had not eroded into the subtalar joint, the inferior talus cartilage was showing degradation. Hence, it was decided to fuse the subtalar joint (Fig. 3).

After preparation of joint surfaces, subtalar arthrodesis was performed using cancellous screws. Fibular fixation was done

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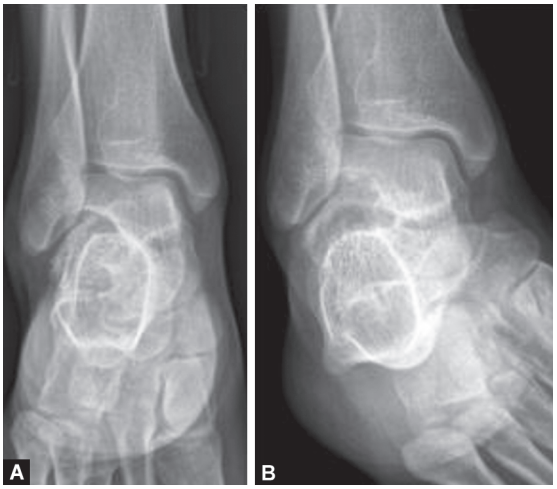
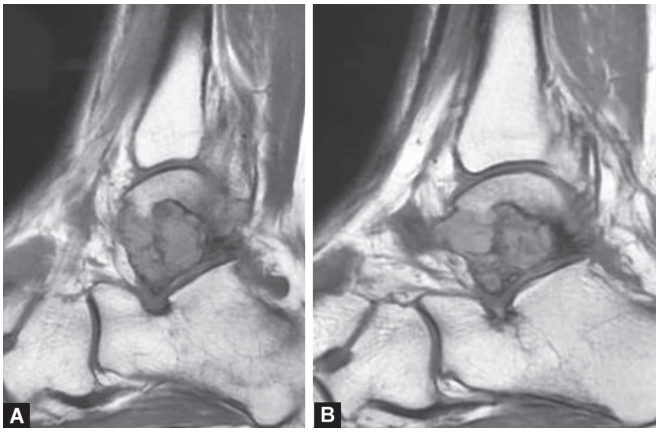


Figs 1A and B: AP and oblique X-ray showing lytic lesion of talus



Figs 2A and B: MRI of ankle showing extensive lysis of talus

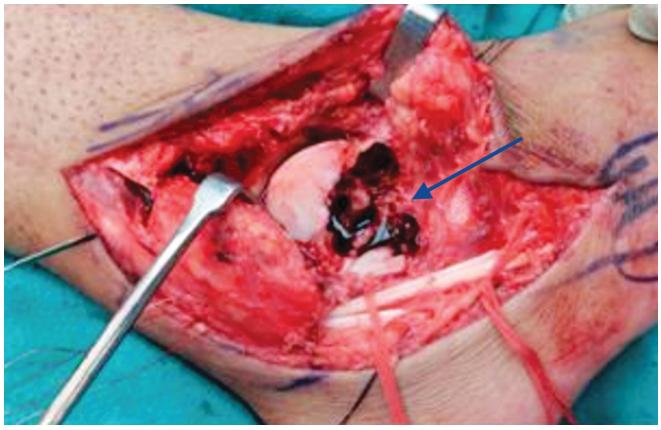
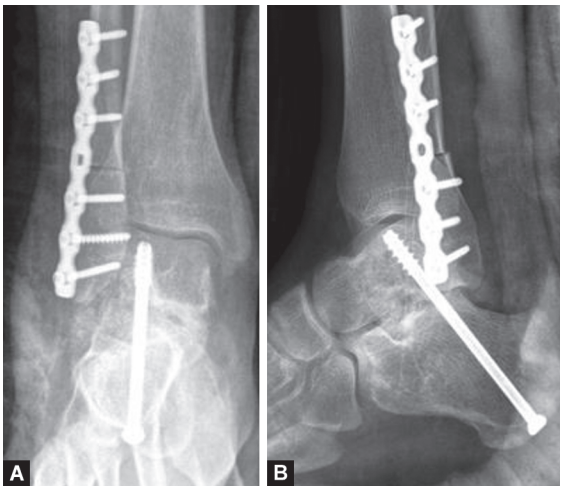


Fig. 3: Intraoperative photograph showing involved part of talus



Figs 4A and B: Postoperative X-rays showing subtalar arthrodesis

with a semi-tubular plate (Fig. 4). Hemostasis was maintained throughout the procedure and the wound was closed in layers over a negative suction drain. Below knee plaster of Paris (POP) slab applied. The intraoperative and immediate postoperative period was uneventful and non-weight bearing crutch walking started on the second day. The patient was followed up every 15 days initially for the first 2 months, then monthly for 6 months. Sutures were removed at 3 weeks. At the last follow-up visit, there were no complications, the patient was able to walk with partial weight-bearing with a fair range of motion at the ankle joint and minimal pain. Radiological examinations showed healing with no sign of recurrence.

Discussion

Femur, tibia, and distal radius are typical locations for the occurrence of GCT while foot bones, hand, and spinal involvement are rare.[4] Giant cell tumor foot are typically solitary lesions, but 1–2% may be multicentric. Minhas et al.,[5] in their study at the tertiary care center, found 240 cases of GCT but only 5% related to foot bones and of them, only 0.4% of cases involving talus. Similarly, Goldenberg et al.,[6] in their series of 218 cases of GCT, found only one case involving the talus; as also by Sung et al.,[7] one talus GCT case in their series of 208 cases. To date, 25 cases of talus GCT have been reported in the literature by various studies (Table 1).

The usual clinical picture of the talus GCT is that of insidious onset pain, which in many cases may be mismanaged as an ankle sprain. A history of preceding trivial trauma may be present. Other features are non-specific. Radiologically, the tumor appears as an eccentric lytic lesion with cortical thinning and expansion. Reactive new bone formation is absent. The tumor may erode the cortex and invade the subtalar joint or may cause a pathological fracture.[24] Intralesional curettage and bone grafting have been reported by several authors with satisfactory results.[25] However, curettage alone has a high rate of recurrence, and adjuvants like methyl methacrylate (bone cement), cryotherapy, and phenol have been suggested.

In cases where there is substantial involvement of the talus, partial or complete talectomy can be contemplated. Arthrodesis may or may not be performed, depending on the involvement of the surrounding joints.[26] Modality of treatment has changed over the past from amputation to reconstruction. Among the published literature for talus GCT, the management has varied from resection, excision intralesional or wide margin, curettage to amputation.[13,24] Result-wise, the outcomes of talectomy and ankle arthrodesis were satisfactory with no recurrence on follow-up. Total talus replacement for GCT has not been reported so far.

Due to the non-specific nature of the symptoms, our patient was diagnosed late as he was receiving treatment for non-specific ankle pain elsewhere. This underlines the importance of investigations

Table 1: Review of literature

S. no.	*Author(s)*	*Cases of GCT talus*	*Age (years)*	*Sex*	*Management*	*Outcome*	*Follow-up*
1	Malawer and Vance (1981)[8]	2	25, 22	Male, male	Extended curettage	No recurrence	60 months
2	Mechlin et al. (1984)[9]	2	18, 19	Female, female	Intralesional curettage	No recurrence	9 months
3	Wold and Swee (1984)[10]	1	18	Male	Extended curettage	No recurrence	12 months
4	Makashir et al. (1991)[11]	1	18	Male	Intralesional curettage	No recurrence	36 months
5	Dhillon et al. (1994)[12]	2	23, 18	Male, male	Extended curettage	No recurrence	18 months
6	Biscaglia et al. (2000)[4]	3	21, 17, 19	Male, male, female	Curettage	No recurrence	18 months
7	Bapat et al. (2000)[13]	1	18	Female	Extended curettage	No recurrence	6 months
8	Schoenfeld et al. (2007)[14]	1	17	Male	Curettage	No recurrence	9 months
9	Sharma et al. (2009)[3]	1	19	Male	Total talectomy and ankle arthrodesis	No recurrence	18 months
10	Mondal et al. (2009)[15]	1	18	Male	Curettage	No recurrence	9 months
11	Bhattacharyya et al. (2010)[16]	1	22	Male	Extended curettage	No recurrence	24 months
12	Mohan et al. (2011)[17]	1	23	Male	Talectomy and subtalar arthrodesis	No recurrence	9 months
13	Young et al. (2012)[18]	1	22	Male	Below knee amputation	NA	NA
14	Song et al. (2014)[19]	1	30	Female	Curettage	No recurrence	12 months
15	Minhas et al. (2015)[5]	1	19	Male	Extended curettage	No recurrence	18 months
16	Bell et.al (2018)[20]	1	19	Male	Extended curettage	No recurrence	6 months
17	Islam et al. (2019)[21]	1	25	Male	Intralesional curettage	No recurrence	24 months
18	Galvan et al. (2020)[22]	1	43	Female	Intralesional curettage	No recurrence	12 months
19	Rahul and Sinha (2020)[23]	1	22	Male	Intralesional curettage	No recurrence	24 months

and radiology. Given the good outcomes in published literature, we decided to go with extended curettage with an autologous bone graft from the ipsilateral iliac crest followed by subtalar arthrodesis as the talus articular cartilage was not of good quality due to the underlying tumor, although there was no breach. On the last follow-up, the patient was able to walk partial weight-bearing, without any surgical site complication, and had a good range of movement at the ankle joint with no sign of recurrence on radiographs and CT scan.

Conclusion

The primary GCT arising out talus is a rare disease and can masquerade clinically as an ankle sprain initially. Early diagnosis and management is key to the successful and complete removal of the tumor. Extended curettage and grafting are still considered as the best treatment modality for GCT given the least recurrence rates. Special attention should be given to the articulate cartilage intraoperatively, and arthrodesis should be done if any doubt regarding the involvement.

Clinical Significance

Presentation of GCT talus may be missed at early stages. A high index of suspicion can help in diagnosis and appropriate management.

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