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The Giant Cell Tumor of 1st Metatarsal in a Young Adult: A Rare Versatile Management with Fibula Cortical Graft

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Learning Point of the Article:

The treatment of Locally Malignant GCT of 1st Metatarsal should be meticulously planned, complete excision is required to prevent recurrence and reconstruction should achieve both mechanical stability and function.

Abstract

Introduction: Giant cell tumor is a benign aggressive tumor commonly affecting the 2nd decade. Most commonly seen in the ends of long bones like the distal femur, proximal tibia, distal radius, and proximal humerus, but it does occur in small bones like hands and feet in <2%.

Case Report: A young female adult of age 23 has been diagnosed with a giant cell tumor of her 1st metatarsal and underwent complete excision with reconstruction with non-vascularized autogenous cortical fibula strut graft using a reconstruction plate and screws and 1-year follow-up showed a good graft union and no signs of recurrence.

Conclusion: Local resection of the affected metatarsal combined with chemoablation reduces recurrence risk, while a fibula graft offers structural stability. In our case, there were no signs of recurrence, and the graft showed good incorporation.

Keywords: Giant cell tumors, young adult, female, metatarsal bones, neoplasms, fibula, follow-up studies.

Introduction

Giant cell tumor (GCT) is an osteolytic benign aggressive tumor that presents with bone destruction soft-tissue impingements and has malignant potential, and rarely metastasizes to pulmonary tissues (<2%) [1,2]. Sir Astley Cooper first described it in 1818. GCT most commonly occurs in young adults (20–30 years), approximately 4–9.5% of all primary osseous tumors and 18–23% of benign bone tumors [3,4]. It occurs most commonly at epiphyses-metaphysis of long bones 85–90% such as the distal femur, proximal tibia, distal radius, and proximal humerus, does occur in the spine, and pelvis 4–5% and rarely in small bones such as hands and feet 1–2% [4].

Radiologically, a large eccentric geographic osteolytic with no matrix mineralization and a thinned-out cortical lesion, Cam Panacci grade (1–3) [5] and LodWick type 1b/1c [5-8] commonly called a soap bubble appearance is seen. On histopathology, multiple giant cell lesions based on the aggressivity of the tumor presentation are seen [9].

GCT in small bones rarely occurs it needs to be managed meticulously and aggressively as it has a high malignant potential and recurrence with 0–65%. Hence, it has been classified as stage 3 of Enneking's benign bone tumors [5, 10]. Hence, these tumors should be managed with diagnostic fine needle aspiration cytology (FNAC) or TruCut biopsy and definite surgical intervention with extended curettage/high-speed burring, adjuvant Argon beam photocoagulation, intra-lesional hydrogen peroxide (H2O2) chemoablation plus auto/allogenous graft

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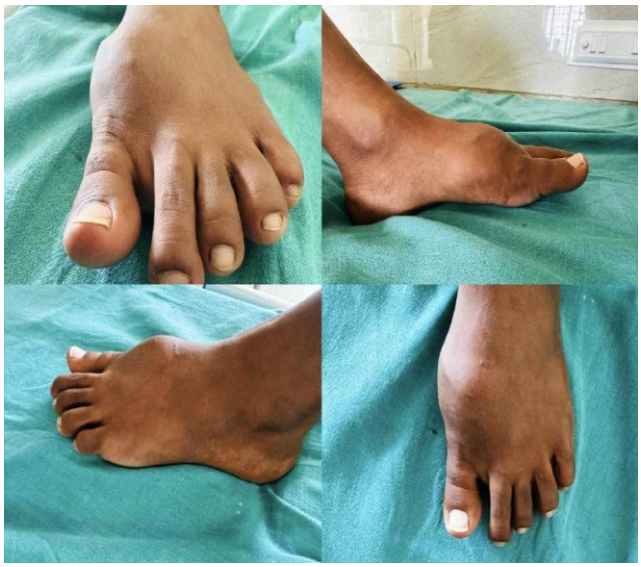

Figure 1: Pre-operative photographs show the swelling in the dorso-medial aspect of the left foot involving the 1st ray.

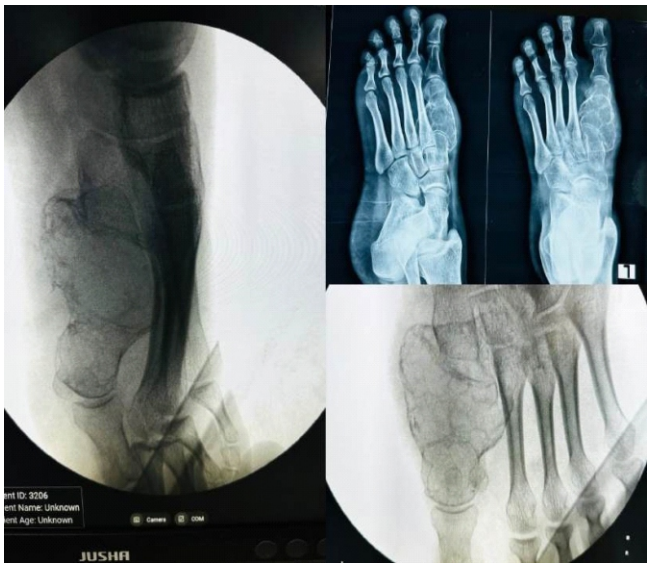

Figure 2: Pre-operative X-rays show large geographic expansile eccentric osteolytic with no matrix mineralization lesion of the entire 1st metatarsal of the left foot. The classical "Soap Bubble Appearance."

[11, 12].

We have encountered one such rare case presentation of GCT 1st metatarsal and managed with a unique reconstruction with autogenous fibular cortical strut graft.

Case Report

A 23-year-old female presented with complaints of swelling over the dorsum of her left foot for the duration of 2 years and pain in that foot for 1 year. The swelling was insidious in onset and progressive. Pain was mild, dull, aching, intermittent type, aggravated on activities of daily living such as squatting, standing, walking, climbing stairs, and running, and relieved on taking analgesics and rest. No history of swelling elsewhere, no constitutional symptoms, and no members of the family both paternal and maternal had similar complaints. On examination, there was a localized ovoid swelling 8 × 5 cm over the dorsum of the left foot, over 1st metatarsal area with well-defined margins, tender on deep palpation, firm in consistency, overlying skin was free, with shortening of 1st ray, with no sinus, discharge, and discoloration (Fig. 1).

Radiographs revealed a large geographic expansile eccentric

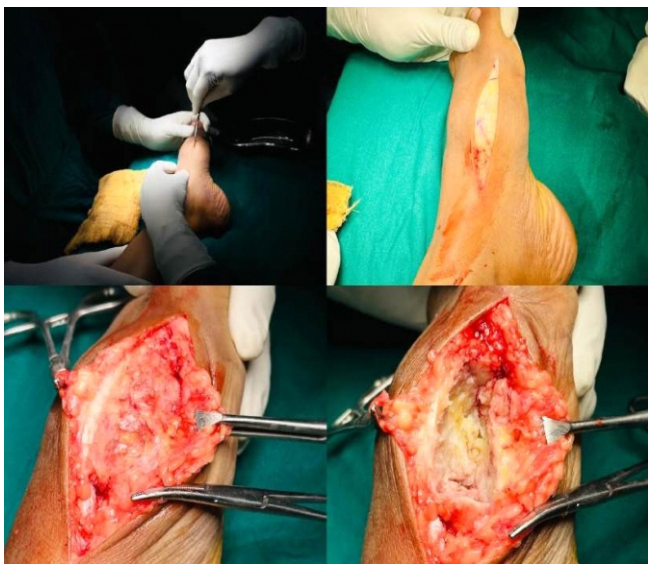

Figure 3: Intraoperative surgical images showing dorso-medial approach to 1st metatarsal and exposure of the bone tumor.

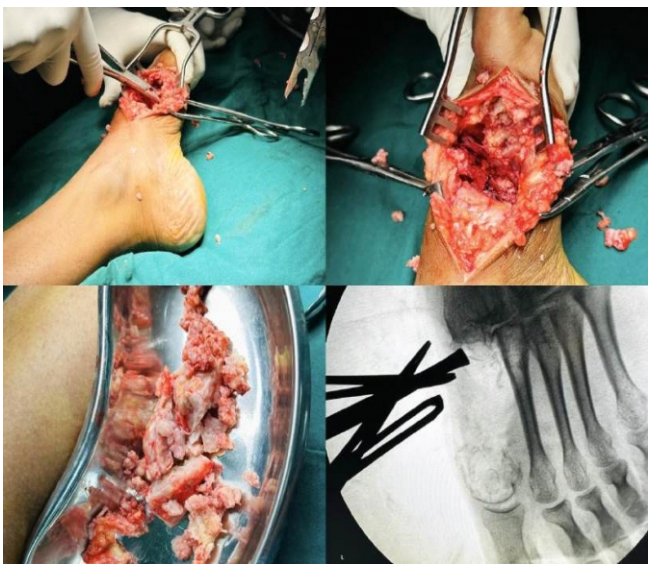

Figure 4: Intraoperative images showing the intralesional excision of the tumor with a radiograph depicting the complete excision.

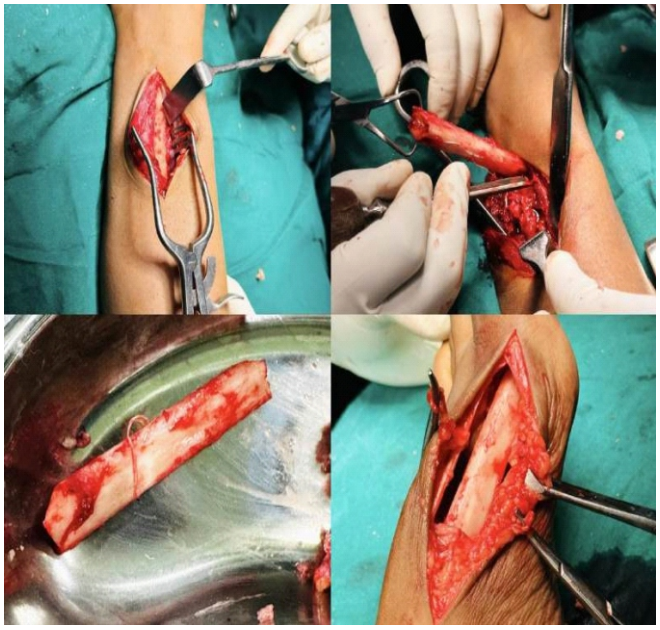

Figure 5: Intraoperative images showing the harvest of ipsilateral fibular avascular Strut Graft of 1 cm more than the measured length of 1st metatarsal in the contralateral normal foot.

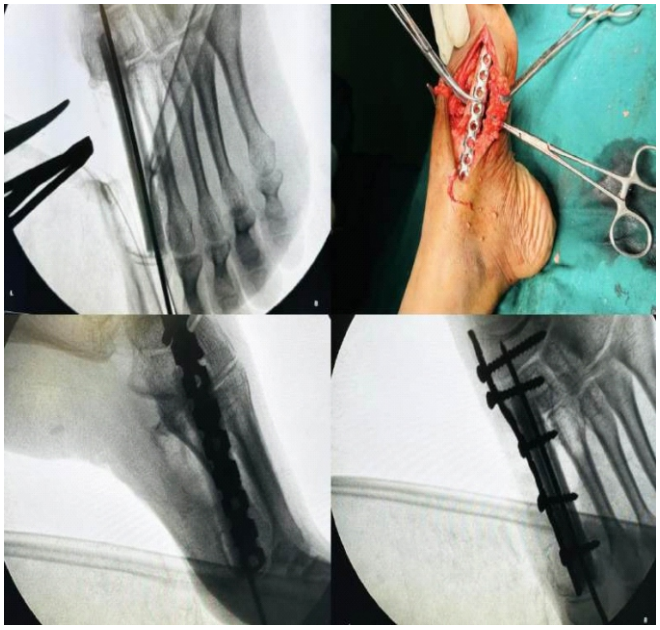

Figure 6: Intraoperative images showing the insertion of the fibular graft (gap filled) and stabilizing with K-wire and recon plate 10-holed and screws.

osteolytic lesion in the 1st metatarsal. The classical "soap bubble appearance" is seen. Stage 3 Enneking, Cam Panacci Grade 2 and LodWick 1c. A chest X-ray anteroposterior view showed normal and was negative for parenchymal lung lesions (Fig. 2). Before planning for a biopsy, an FNAC was sent and was diagnosed as a giant cell lesion.

Routine blood investigations include complete hemogram, C-reactive protein (CRP), erythrocyte sedimentation rate (ESR), liver and renal function tests were within normal limits and negative serology, with the patient and her attendant's written consent taken for proposed surgical management.

Surgical procedure

An incision was taken over the dorsomedial approach on the left foot, extending from the head of the 1st metatarsal to the navicular. The tumor site is exposed and the excision of the tumor with a distally preserved cuff of 1st metatarsal articular cartilage (Fig. 3).

Intra-lesional H2O2 chemoablation was done to remove all the nidus sites and the 1st metatarsal length measured from the contralateral foot and an ipsilateral fibular strut graft of 1 cm more than the measured length of the contralateral 1st metatarsal is taken and inserted into the troughs created in medial cuneiform and preserved 1st metatarsal distal articular

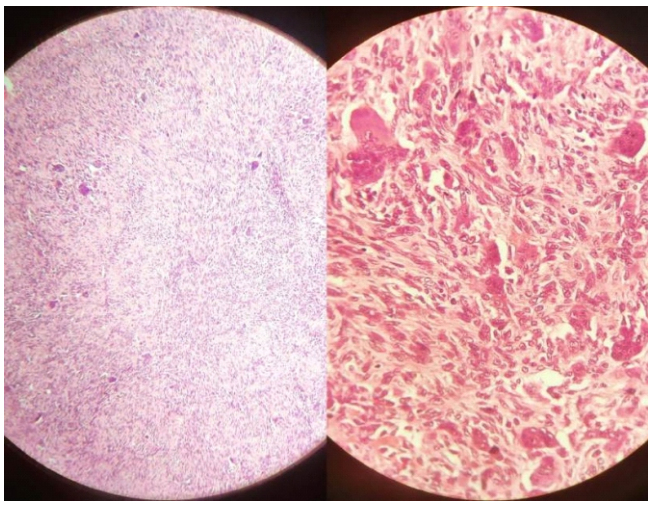

Figure 7: Histopathological images showing multiple giant cell lesions (×10 right image and ×40 left image).

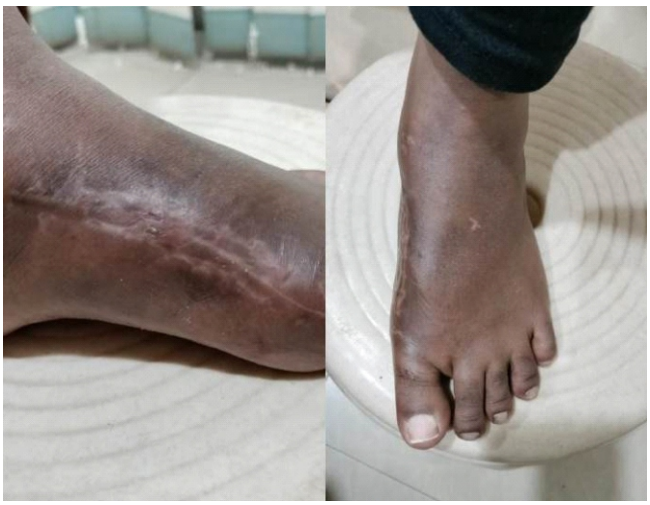

Figure 8: Post-operative images at 1 year showing healed surgical scar with primary intension with no sinus and discharge and no wound complications.

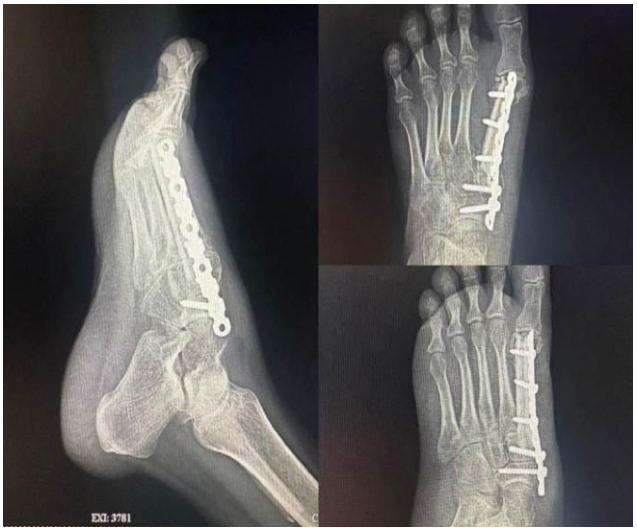

Figure 9: Post-operative radiographs taken after 1 year show no signs of recurrence with good graft incorporation.

cartilage and the specimen was sent for histopathological examination and inspected both proximally and distally (Fig. 4). 1st metatarsal length was measured from the contralateral foot, and an ipsilateral fibular avascular Strut graft of 1 cm more than the measured length of 1st metatarsal was taken and inserted into the troughs created in medial cuneiform and preserved (Fig. 5).

1st metatarsal normal articular cartilage and bone, fixed with intramedullary K-Wire and 10-hole re-construction plate with cortical screws from distal to proximal through normal viable 1st metatarsal bone and cartilage to medial cuneiform extended till navicular (Fig. 6).

Histopathological examination

The report showed to be a multiple multinucleated benign giant cell lesion on both 10 and ×40 (Fig. 7).

Follow-up protocol

Post-surgery, she was kept in below knee slab for 6 weeks and advised non-weight bearing. K-wire was removed after 6 weeks. She's advised to mobilize after 6 weeks with touch-toe (partially weight bearing) and complete weight bearing after 3 months. At 12 weeks, we noticed graft union radiologically. At 1 year follow-up, she's actively mobilizing and doing all the activities of daily living without hindrance with no signs of discharge/sinus (Fig. 8).

Radiographs after 1 year showed good graft integration with no signs of recurrence (Fig. 9).

Discussion

Giant cell tumor (GCT) are usually solitary lesions, but 1–2% can be synchronously or meta-synchronously multicentric [2, 13]. GCT in the small bones (i.e., metacarpals, metatarsals, phalanges) are very rare and are more malignant than other regions; hence, a thorough clinical history and examination of

S. No.	Publications	Place of study	Age (y)	Sex	Site	Management	Outcome
1	Bibbo (2010) [11]	Marshfield, Missouri, USA	Case 1: 13 Case 2: 15	M M	Right 1st metatarsal Left 5th Metatarsal	Extended Curettage + H_2O_2 Chemoablation + ICBG* Extended Curettage + H_2O_2 Chemoablation + ICBG*	No signs of recurrence after 2.5 y of follow-up
2	Yurdoglu et al. (2011) [12]	Turkey	CASE 1: 17 CASE 2: 33	F M	Right 3rd metatarsal right 2nd metatarsal	Complete extra-lesional Excision + Kwire	No signs of recurrence after 2 years of follow-up
3	Siddiqui et al. (2011) [15]	AMU, India	28	F	Right 1st metatarsal	Wide excision + Autogenous fibular cortical strut graft + ICBG* + K-wire	No signs of recurrence after 4 months of follow-up
4	Prashant et al. (2016) [22]	Uttar Pradesh, India	40	M	Left 1st Metatarsal	Complete excision + Fibular cortical strut graft + K-wire	No signs of recurrence after 9 months of follow-up
5	Chetia et al. (2017) [18]	Assam, India	26	M	Left 1st metatarsal	En mass resection + H_2O_2 Chemoablation + Fibula cortical Strut graft + K-wire	No signs of recurrence after 1 year of follow-up
6	Mahajan et al. (2021) [23]	Mumbai, Maharashtra, India	60	M	Right 3rd metatarsal	Complete excision of tumor with 3rd ray amputation	No signs of recurrence after 1.5 years of follow-up
7	Kamath et al. (2022) [16]	Mangalore, Karnataka, India	40	F	Right 1st metatarsal	En bloc Resection of tumor + Autogenous Fibular Cortical Strut Graft + K wire and SS wire as Double barrel Reconstruction	No signs of recurrence after 1 year of follow up
8	Florio et al. (2022) [24]	Italy	9	M	Right foot 4th Metatarsal	Wide local excision + Adjuvant phenol chemoablation + Fibular cortical strut graft + K wire	No signs of recurrence after 2 years of follow-up
9	Patel et al. (2023) [25]	Surat, India	Various age groups	-	16 cases with GCT of hand and foot	Among 16 cases, 2 were metatarsal GCT with the procedure including ray amputation and Wide excision + fibula bone graft	Follow-up were irreversibly lost (retrospective study)

GCT: Giant cell tumor, *ICBG: Iliac crest bone graft

Table 1: Over the past decade, published research articles on the management of GCT in small bones.

the hand and foot should be evaluated to rule out differential diagnoses such as an aneurysmal bone cyst, angiosarcoma, enchondromas, osteosarcoma, giant cell reparative granuloma, non-ossifying fibroma, brown tumor/hyperparathyroidism, metastatic tumor, and infective including acute/chronic osteomyelitis, and tubercular osteomyelitis [12, 14- 17].

Investigations include radiographs of affected extremities/regions, complete blood examination including CRP and ESR, and histopathological examinations (i.e., FNAC and Biopsy [incisional and excisional]) [11, 12, 15, 16, 18] to know the etiology and its pathogenesis.

GCT consists of 3 cell types: Neoplastic GCT stromal cells, represented as the proliferative population, mononuclear histiocytic cells recruited to the area, and multinucleated giant cells [9].

Genetic analysis of GCT of bone would reveal several cytogenetic abnormalities resulting from nonrandom translocations involving breakage and fusion of chromosome telomeres known as "telomeric associations" [14, 19].

The bone erosive tendencies of GCT of bone have been associated with the ability of tumor cells to express multiple cytokines. Stromal cells from excised GCT lesions in bone patients have demonstrated the production of vascular endothelial growth factor, a matrix metalloproteinase-9. Moreover, higher mRNA up-regulation (as detected by reverse transcriptase polymerase chain reaction) is evident in advanced-stage GCTs (stage II/III). This observation is also linked to increased bone destruction and the likelihood of local recurrence [20].

Aggressive treatment and meticulous excision are essential for managing GCT in the hand and foot [21]. Benign aggressive tumors like GCT in small bones demand careful management and thorough follow-up to monitor recurrence. Magnetic resonance imaging (MRI) of the foot can be utilized, if necessary, to detect soft tissue spillage, which could serve as a potential nidus for tumor recurrence.

Over the years, management of GCTs of the metatarsal (area of interest) and small bones (Table 1).

To the best of our knowledge and information available in the various search standard databases gathered the area of interest studies from the past 10 years [11, 12, 15-18, 22-25], we noticed a unique way of reconstruction could be done, and all the above-mentioned techniques and procedures showed less signs of recurrence with good stability and better functional outcomes.

Limitation of our study

It needs long-term follow-up because the recurrence of small bone GCT does occur at 2–3 years [14]. MRI was not taken hence missing out on the soft-tissue involvement for the nidus or site of GCT other than 1st metatarsal [11, 12, 20].

Conclusion

Benign aggressive bone tumors are managed with complete excision and reconstruction with autografts/allografts/bone cement for functional outcome and to prevent a recurrence. Our management showed the viability of the graft radiologically after 1 year with no recurrence and excellent functional foot outcome.

Clinical Message

Benign aggressive bone tumors include GCTs and aneurysmal bone cysts. GCT in small bones has high rates of malignant potential that should be addressed with thorough pre-operative evaluation and planned surgical excision with either chemoablation or radio-ablation to kill all the tumor cells in the surrounding tissues, and carefully monitor the patient with repeated interval follow-ups, also fibula graft reconstruction is encouraged to provide structural stability and cosmetically acceptable.

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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