Received: 3 December 2022 | Revised: 9 May 2023 | Accepted: 28 May 2023

DOI: 10.1002/ccr3.7652

CASE REPORT

Clinical Case Reports Open Access WILEY

Malignant, fungating giant-cell tumor of the tendon sheath (GCT-TS) of the foot: A case report

Hassan Elbahri[1] | **Hozifa Mohammed Ali Abd-Elmaged**[2] | **Sawsan A. M. Babiker**[3] | **Alaa Hatim Ameer Mohamed**[3] | **Mohammed Mubarak Mohammed Ahmed**[4] | **Mohamed Abdulkarim**[5]

[1]Department of Orthopedics, International University of Africa, Khartoum, Sudan

[2]Department of Orthopedics, Alzaiem Alazhari University, Khartoum, Sudan

[3]Department of Pathology, University of Khartoum, Khartoum, Sudan

[4]Ibrahim Malik Teaching Hospital, Khartoum, Sudan

[5]Department of Surgery, Alzaiem Alazhari University, Khartoum, Sudan

Correspondence
Hassan Elbahri, Department of Orthopedics, International University of Africa, Khartoum, Sudan.
Email: hassan.elbahari.ortho@gmail.com

Key Clinical Message

Giant-cell tumors of the tendon sheath (GCT-TS) are relatively common benign tumors that arise in close proximity to joints and tendons. Malignant GCT-TS are extremely rare. Surgery with wide resection remains the cornerstone for treating malignant giant-cell tumors of the tendon sheath especially in large tumor cases.

KEYWORDS

dissection, Fungating, Giant cell tumor, tendon sheath, Tenosynovial

1 | INTRODUCTION

Giant-cell tumors of the tendon sheath (GCT-TS) (also called tendosynovial giant cell tumors) are rare disease entity that can face orthopedic surgeons. They are tumors, of possible benign or malignant nature, affecting soft tissues and can arise from the tendon sheaths or the synovium of the joints. No more than 8% of all soft tissue tumors, whether they are benign or malignant, are found in the foot.[1]

These tumors usually affect middle-aged persons (30–50 years) with female prediction. Although many cases have been reported to occur in pediatric population of different ages and sex. They have an incidence of 1/50,000.[2–8]

Malignant GCT-TS is a rare disease entity. In this article, we report a large fungating malignant GCT-TS in a 35-year-old female patient which is not a common entity and less is known about it apart from the surgical approach to the disease management.

2 | PATIENT INFORMATION

The patient is a 35-year-old married female patient, a mother of seven children the youngest of whom is 14 months old. She has no known chronic illnesses including diabetes mellitus and hypertension. She has no history of previous surgery. She does not smoke nor drink alcohol. She is not known to have any allergies. She has no

family history of similar presentation nor taking chronic medications.

3 | CLINICAL FINDINGS

Generally, at presentation, she was unwell, cachexic and pale. She presented with a mass in the dorsum of her left foot. Local examination revealed a fungating lesion that measures approximately 20 × 25 cm, discharging a bloody purulent material with no grains (increased by squeezing) with intact distal neurovascular examination. There was no proximal lymphadenopathy. Her vitals were within normal range.

4 | TIMELINE

The history started 12 years ago when the patient noticed a small mass on the dorsum of her left foot at the level of MTP joint at her 4th and 5th toes. The mass was slowly and gradually increasing in size over these years with no associated skin changes, numbness or tingling. It had no effect on toes range of movement (R.O.M).

During the last year, the lesion started to rapidly increase in size with ulceration of overlying skin and a bloody purulent discharge increased by squeezing the mass or walking for a long distance. All foot and ankle become swollen. She developed secondary infection for which she received antibiotics for 2 weeks without improvement.

In the last 4 months, her condition deteriorated and she developed prolonged fever with loss of appetite and weight loss. She was then referred to our orthopedic oncology department unit where systematic work up was done and final diagnosis was made.

5 | DIAGNOSTIC ASSESSMENT AND INTERPRETATION

In the orthopedics oncology department, laboratory workup showed low hemoglobin elevated levels of C-reactive protein. Her liver and renal function tests were normal. FNAC was done and showed malignant cells. An X-ray of the affected region revealed erosion and destruction of the forefoot bone. CT scan of the chest showed no metastatic lesions. Whole left foot and leg MRI showed soft tissue mass involving extensor and flexor compartments of the lateral column of forefoot, heterogeneous low T1 and high T2 with malignant behavior consistent with a malignant soft tissue tumor (Figures 1 and 2). Bone scan revealed no other lesions. FNAC is done and showed malignant cells.

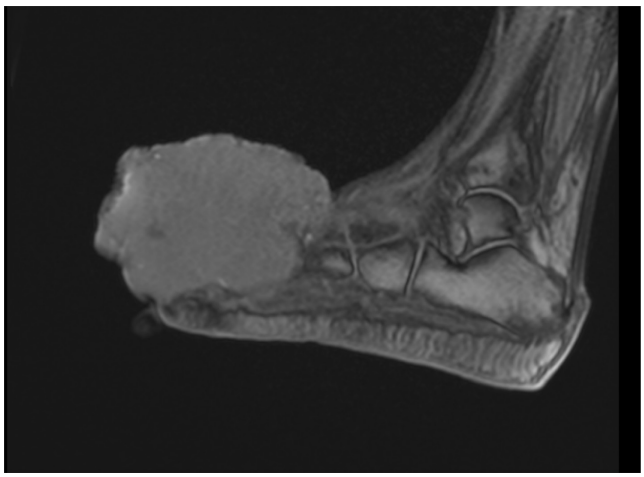

FIGURE 1 Sagittal view of T1 sequence MRI of the left foot showing a heterogenous mass at the distal tarsal, metatarsal and toes.

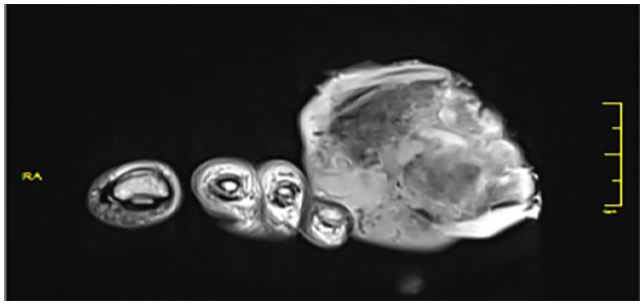

FIGURE 2 Axial view T1 MRI of the left foot showing the heterogenous mass at the distal tarsal, metatarsal and toes.

6 | INTERVENTION

The patient was planned for Chopart amputation after counseling and a fully-informed consent. Surgery was done under spinal anesthesia and a tourniquet with no exsanguination. Meticulous dissection and hemostasis with excellent bone handling to ensure complete tumor excision was performed. Tendo-Achilles Lengthening (TAL) was done with transfer at TA tendon to talar neck. Closure in layers with compressive dressing was ensured followed by insertion of A drain. Histopathology showed cellular neoplasm composed of sheets of polygonal mononuclear cells with oval nuclei admixed with many multinucleated giant cells of the osteoclast type. The neoplasm showed focal marked nuclear pleomorphism and increased mitotic activity with hemorrhage and necrosis (Figures 3 and 4). Bone invasion was identified with clear surgical cut edges. The malignant diagnosis of malignant and fungating GCT of tendon sheath was made.

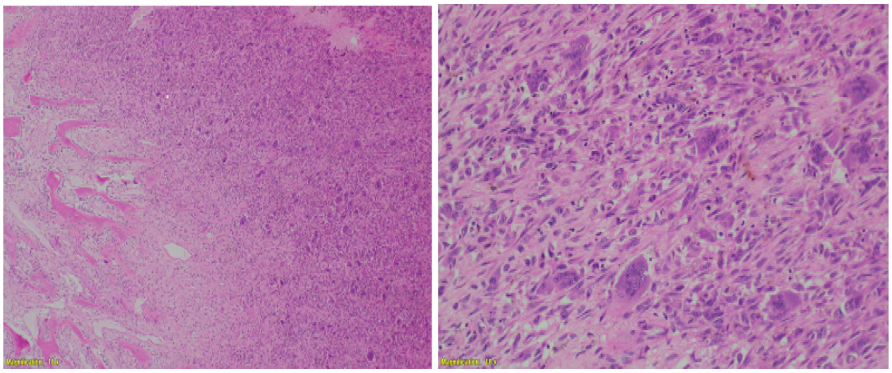

FIGURE 3 Left: low power view showing neoplasm invading bone. (H&E ×4). Right: Cellular neoplasm composed of large number nonneoplastic osteoclast-like giant cells, between which mononuclear neoplastic cells are embedded. (H&E ×10).

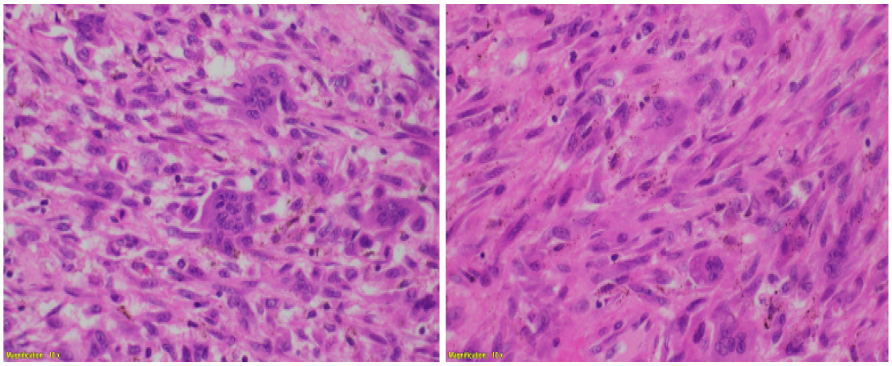

FIGURE 4 Highly cellular neoplasm with nuclear pleomorphism and brisk mitotic activity. (H&E ×40).

7 | FOLLOW-UP AND OUTCOME

Patient underwent an uneventful post-recovery course apart from a superficial wound infection treated by multi-dressing and antibiotics.

After 3 months of follow-up, she is well and the wound has totally healed with healthy stump. Her chest X-rays and CT imaging are clear. She is planned for a sophisticated forefoot prosthesis.

8 | DISCUSSION

Giant-cell tumors of the tendon sheath (GCT-TS) (also called tendosynovial giant cell tumors) are rare diseases with no clear etiology been identified yet; however, a strong correlation to trauma has been described in the literature. Additional etiologies have been suggested including inflammation, metabolic disorders, or infections.[2,9] It is still not known whether the tumors are lesions reactive to above mentioned potential causes or a true neoplasm.

Patients usually present with a slowly progressive and gradually increasing painless mass (it can extend to many years as in this case). The tumor usually measures less than 10 cm in length, but it can sometimes reach up to 15 cm in.[2] It usually causes pain, discomfort, decreased range of motion with involvement of adjacent structures or markedly swollen joints. Neurovascular complaints, though uncommon, may happen if there is an invasion or compression to adjacent structures. Some studies reported mild motor limitation of plantar flexion of the interphalangeal joint with sensory loss due to digital nerve compression,[6] in contrast to our case which showed no neurovascular involvement or decreased range of motion. Although the disease has benign microscopic features and nature; it is a locally and have a high rate of local recurrence (up to 44%). This fact has made the recurrence rate one of the possible factors affecting prognosis and outcome.[10]

Making the diagnosis solely on the basis of the clinical presentation is difficult since many other soft tissue tumors can show similar features. X-rays can show soft tissue lesions with possible bony involvement such as erosions, mainly in the cortices. While ultrasonographic scan is not usually done, it can show a hypoechoic mass that is solid and homogenous in nature. Magnetic resonance imaging (MRI scan) is done for more better lineation of the mass, and it usually helps distinguishing GCT-TS from other soft tissue tumors such as lipomas, malignant fibrous histiocytomas, or synovial cysts among others. These tumors are characterized by a low-signal intensity on both MRI T1 and T2-weighted images that bear resemblance to that intensity of skeletal muscles which can be ascribed to the presence of hemosiderin with its paramagnetic impact and the tumor's high content of collagen stroma. When

contrast is added, the tumors usually take the contrast material and appear with homogenous enhancement.[6,10–12] These findings are of a great importance as they aid in differentiating it from other soft tissue tumors like hemangiomas and neurofibromas and other nerve sheath tumors preoperatively.[6,10–12]

Malignant variant of the disease is not common, and benign variant is much more common. Yong-wei Pan et al. described malignant giant cell tumor of the tendon sheaths in the hand in their retrospective study conducted between January 1991 and December 2001 in 10 patients with histologically proven MGCTTS, and they have been treated by surgical option alone without chemo- or radiotherapy.[13]

Surgical management with removal of all the affected tissues is not only the usual treatment, but also is a crucial and most critical step in management as these tumors have high rate of recurrence which was reported to be up to 45%.[14] Poor surgical technique resulting in insufficient resection is thought to be associated with recurrence; therefore, a meticulous and thorough dissection with the use of magnification devices helps maximizing the benefit from the surgery.[15] While adjuvant treatment with radiotherapy has been suggested, it has not reported to be performed for the purpose of preventing recurrence of these tumors.[16]

What is unique about this case, is not only that the GCT is a rare disease entity but also the fungating nature of the tumor that is complicated by infections and erosion of the bone and the large size of the tumor can be attributed to the prolonged course of the illness. In addition, despite the prolonged course of the mass, it has not metastasized anywhere in the body.

This case has been reported in line with the SCARE criteria.[17]

AUTHOR CONTRIBUTIONS

Hassan Elbahri: Conceptualization; data curation; formal analysis; investigation; methodology; project administration; resources; supervision; validation; writing – review and editing. **Hozifa Mohammed Ali Abd-Elmaged:** Conceptualization; data curation; formal analysis; investigation; methodology; resources; software; validation; visualization; writing – original draft. **Sawsan A. M. Babiker:** Conceptualization; data curation; formal analysis; investigation; methodology; project administration; resources; validation; visualization; writing – original draft. **Alaa Hatim ameer Mohamed:** Conceptualization; data curation; formal analysis; investigation; methodology; project administration; resources; validation; visualization; writing – review and editing. **Mohammed Mubarak Mohammed Ahmed:** Conceptualization; data curation; formal analysis; investigation; methodology; project administration; resources; validation; visualization; writing – review and editing. **Mohamed Abdulkarim:** Conceptualization; data curation; formal analysis; investigation; methodology; project administration; resources; validation; visualization; writing – original draft; writing – review and editing.

FUNDING INFORMATION

The authors received no funding from any source and this work is completely a voluntary work.

CONFLICT OF INTEREST STATEMENT

The authors report no conflict of interest of any sort.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

No ethical approval was needed for this case.

CONSENT

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editor in chief of this journal on request.

ORCID

Hassan Elbahri https://orcid.org/0000-0002-3056-4419
Mohamed Abdulkarim https://orcid.org/0000-0001-7888-4158

REFERENCES

1. Kennedy JG, Ross KA, Smyth NA, Hogan MV, Murawski CD. Primary tumors of the foot and ankle. *Foot Ankle Spec*. 2016;9(1):58-68. doi:10.1177/1938640015620634
2. Awad P. Atypical Tenosynovial Giant cell tumor of the foot and ankle: a case report. *J Am Podiatr Med Assoc*. 2021;111(3):Article_19. doi:10.7547/18-038
3. Ueno T, Ansai S, Omi T, Kawana S. A child with giant cell tumor of tendon sheath. *Dermatol Online J*. 2011;17(5):9.
4. Ansari SM, Tambe SA, Nayak CS. Giant cell tumor of tendon sheath in children. *Indian J Dermatol Venereol Leprol*. 2019;85:496-498.
5. Asaad SK, Bapir R, Salh AM, et al. Giant cell tumor of the tendon sheath in a 5-year-old child; a case report. *Ann Med Surg (Lond)*. 2021;69:102599. doi:10.1016/j.amsu.2021.102599
6. Zheng S, Lee PY, Huang Y, Wang A, Li T. Giant cell tumor of tendon sheath and tendinopathy as early features of early onset sarcoidosis. *Front Pediatr*. 2019;7:480. doi:10.3389/fped.2019.00480
7. Lee DH, Lim SY. A rare giant cell tumor of the tendon sheath of the dorsal hand in a child. *Arch Plast Surg*. 2014;41(5):605-606. doi:10.5999/aps.2014.41.5.605
8. Gholve PA, Hosalkar HS, Kreiger PA, Dormans JP. Giant cell tumor of tendon sheath: largest single series in

children. *J Pediatr Orthop*. 2007;27(1):67-74. doi:10.1097/01.bpo.0000242380.95348.8b
9. Paez H, Vuletin JC, Soave RL, Sellitto RV. Pedal giant cell tumor of tendon sheath. *J Am Podiatr Med Assoc*. 1999;89(7):368-372. doi:10.7547/87507315-89-7-368
10. Vasconez HC, Nisanci M, Lee EY. Giant cell tumour of the flexor tendon sheath of the foot. *J Plast Reconstr Aesthet Surg*. 2008;61(7):815-818. doi:10.1016/j.bjps.2006.09.006
11. Levi M, Crafton J. Rare Giant cell tumor of the distal flexor Digitorum longus tendon sheath and early diagnosis with use of magnetic resonance imaging. *J Am Podiatr Med Assoc*. 2017;107(4):333-336. doi:10.7547/16-163
12. Jelinek JS, Kransdorf MJ, Shmookler BM, Aboulafia AA, Malawer MM. Giant cell tumor of the tendon sheath: MR findings in nine cases. *AJR Am J Roentgenol*. 1994;162(4):919-922. doi:10.2214/ajr.162.4.8141018
13. Pan YW, Huang XY, You JF, Tian GL, Li C. Malignant giant cell tumor of the tendon sheaths in the hand. *Zhonghua Wai Ke Za Zhi*. 2008;46(21):1645-1648.
14. Kotwal PP, Gupta V, Malhotra R. Giant-cell tumour of the tendon sheath. Is radiotherapy indicated to prevent recurrence after surgery? *J Bone Joint Surg Br*. 2000;82(4):571-573.
15. Fotiadis E, Papadopoulos A, Svarnas T, Akritopoulos P, Sachinis NP, Chalidis BE. Giant cell tumour of tendon sheath of the digits. A systematic review. *Hand*. 2011;6(3):244-249.
16. Goni V, Gopinathan NR, Radotra BD, Viswanathan VK, Logithasan RK, Balaji S. Giant cell tumour of peroneus brevis tendon sheath--a case report and review of literature. *BMJ Case Rep*. 2012;2012:bcr0120125703. doi:10.1136/bcr.01.2012.5703
17. Agha RA, Franchi T, Sohrabi C, Mathew G, for the SCARE Group. The SCARE 2020 guideline: updating consensus surgical CAse REport (SCARE) guidelines. *Int J Surg*. 2020;84:226-230.

How to cite this article: Elbahri H, Abd-Elmaged HMA, Babiker SAM, Mohamed AHA, Ahmed MMM, Abdulkarim M. Malignant, fungating giant-cell tumor of the tendon sheath (GCT-TS) of the foot: A case report. *Clin Case Rep*. 2023;11:e7652. doi:10.1002/ccr3.7652