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# Giant cell tumour of the extensor tendon sheath of the ankle: A rare case report with review of literature

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**Abstract**
Giant cell tumour of the tendon sheath is also termed as tenosynovial giant cell tumour. It is relatively a rare tumour comprising only about 1.6% of all the soft tissue tumours. Giant cell tumour of the tendon sheath of the foot and ankle is much less reported as compared to giant cell tumour of the tendon sheath of the hand and knee. However giant cell tumor of the tendon sheath should be considered as a differential diagnosis of soft tissue tumours of the foot and ankle. We here present a rare case of giant cell tumour of the extensor tendon sheath of the ankle in a young female which presented with chief complaint of a painless swelling of size about 3.5×3.5cm over her right ankle from one year. The diagnosis was made initially by using ultrasonography which was supported by MRI and confirmed after the histopathological examination of the excisional biopsy. The giant cell tumour was found arising from the tendon sheath of extensor tendons - tibialis anterior and extensor hallucis longus. In our case the patient showed complete resolution of the symptom with no sign of recurrence at final follow-up after 1 year.

**Keywords:** Giant cell tumour of the tendon sheath, tenosynovial giant cell tumour, extensor tendon, ankle, xanthoma

## Introduction
Giant cell tumor of the tendon sheath is the second most common tumour of the hand after ganglion. It is a slowly growing and painless soft tissue tumour that develops over a period of months to year. Giant cell tumor of the tendon sheath is a benign, solitary, proliferating tumour that develops in the tendon sheath of small joints of the hand and foot [1]. Though it can occur at any age giant cell tumor of the tendon sheath most frequently affects individuals between the age of 30 to 50 years [2, 3, 4, 5]. There is no racial discrimination and the female to male gender ratio is 3:2 [6]. Giant cell tumor of the tendon sheath is significantly less common in the ankle and foot, where only 3 to 5% of giant cell tumour of the tendon sheath cases has been documented and it is estimated that 0.8 to 1.0% of foot and ankle masses are caused by giant cell tumour of the tendon sheath [7].

## Case report
We here present a case report of a 38 years old female presented with chief complaint of a painless swelling over the anterior aspect of her right ankle. The swelling had increased progressively in size from last one year without any constitutional symptoms. Her general examination was unremarkable and she was in a good health.
On local examination there was a non-tender, firm to hard, solitary swelling over the anterior aspect of the right ankle measuring about 3.5×3.5cm in size with restricted mobility from side to side direction without any effect on joint mobility; and the distal neurovascular examination was within normal limits.
Plain X-ray of the ankle revealed no bony abnormality but only a soft tissue shadow. Ultrasonography of the swelling showed a hard soft tissue density mass lesion at the ankle. MRI of the ankle revealed a moderate size multilobulated soft tissue lesion in the subcutaneous plane at the anterior aspect of the right ankle just superficial to the extensor tendons. The lesion was hypointense on T1WI and isointense to slightly hyperintense on T2WI. The lesion was closely abutting and partially encasing the tibialis anterior tendon without invasion of the body of the tendon.

The lesion was also focally abutting extensor hallucis longus tendon and extensor retinaculum with no evidence of deeper invasion. Medially the lesion was focally abutting great saphenous vein. No bony or ankle joint involvement was noted. No encasement of tibial artery was noted.
Intra-operatively a glistening yellowish white multilobulated mass with bony hard consistency and a smooth, regular surface was found arising from the sheath of extensor tendons (tibialis anterior and extensor hallucis longus). The mass was completely excised without any injury to the extensor tendon and sent for histopathological examination which revealed a giant cell tumour of the tendon sheath. The histopathology comprised of multinucleated osteoclast type giant cells, polyhedral histiocytes, fibrotic materials and xanthomatous changes with hemosiderin laden macrophages. The post-operative period was uneventful and the patient did very well. The ankle was mobilized from second postoperative day and she had no problem with weight bearing. On follow up there was no evidence of recurrence or functional debility after one year.

**Discussion**

About 1.6% of all soft tissue tumours are giant cell tumours of the tendon sheath, which are characterized by benign, sharply confined peritendinous fibrous masses in the synovial or tendinous spaces [8, 9]. Jaffe was first to defined the giant cell tumour of the tendon sheath as a tenosynovitis and a non-neoplastic swelling in 1941 [10]. According to Fotidas et al. the giant cell tumour of the tendon sheath affects more often females, with a female to male ratio 3:2 and the mean age of presentation ranged from 30 to 50 years. The vast majority of cases presents with a painless soft tissue swelling. Sensory disturbances noted in about 4.57% [11, 12, 13, 14, 15]
The hand and less frequently the foot and ankle regions are affected by giant cell tumours of the tendon sheath. After ganglion cyst, giant cell tumour of the tendon sheath is the second most common hand tumour [11, 16]. The most common site is the index finger. Other common sites are thumb, middle finger, ring finger, and little finger.
Only 3 to 5% of giant cell tumour of the tendon sheath cases have been described in the ankle and foot, making it substantially less common there [1]. Giant cell tumour of the tendon sheath is among the most prevalent soft tissue tumour of the foot [17]. The most common site is the great toe. It frequently involved the peroneus brevis tendon [9, 18] flexor hallucis longus tendon [18, 19] extensor hallucis brevis tendon [18], extensor digitorum longus tendon [20], Achille's tendon [21] and tibialis posterior tendon [8]. In this case report there was involvement of the tibialis anterior and extensor hallucis longus tendons with focal abutment of extensor retinaculum.
It is uncertain what causes the giant cell tumour of tendon sheath. Only 5% of the patients had a definite history of soft tissue trauma at the time of first presentation [11, 12, 13, 14, 15, 16, 22, 23, 24, 25]. Jaffe *et al.* had described a reactive or regenerative hyperplasia coupled with an inflammatory process as the most widely accepted theory [10, 25]. Byers classified giant cell tumours of the tendon sheath into two types - localized nodular type (common in the hand) and diffuse type (common in joints). Al-Qattan proposed a new classification of giant cell tumours of the tendon sheath
- Type I present as a single tumour which is round or multilobulated, and Type II present as a two or more distinct tumours which are unjoined [14, 22].
The giant cell tumour of the tendon sheath histopathologically comprised of multinucleated osteoclast type giant cells, polyhedral histiocytes, fibrotic materials and xanthomatous changes with hemosiderin laden macrophages [15, 26, 27].
Ultrasonography can detect whether a tumor is solid or cystic; and any accompanying satellite lesions can also be detected. Ultrasonography also describes the relationship of the lesion to the adjacent structures. Ultrasonography enables the determination of the degree of contact with the underlying tendon and the proportion of circumferential involvement [28]. MRI proved to be the most useful preoperative investigation as giant cell tumour of the tendon sheath has a characteristic appearance which allows planned local excision to be carried out.
The preferred course of treatment for giant cell tumour of the tendon sheath is the local excision by marginal resection with or without radiation [11, 12, 13, 14, 15, 16, 22, 23, 24]. The complete excision of the giant cell tumour of the tendon sheath is very difficult due to its close relationship with the tendon sheath or the synovium of the joint.
The incidence of local recurrence is high. Moore et al. Reported recurrence rate of 9% in 115 cases and Wright et al. reported recurrence rate of 44% in 69 cases [29, 30]. Despite being benign, local recurrence following excision has been reported in about 45% of the cases or more [12]. In more recent studies there is recurrence rate of about 14.7%. Various factors have been described including pressure erosion on X-ray, interphalangeal joint location, presence of degenerative joint disease and incomplete excision of the tumour. Reilly *et al*. and Grover et al. noticed that bone erosion for recurrence. However, Kitagawa did not support this theory, he advocated the bone involvement was due to simple erosion, caused by the pressure effect of the tumour, and was not a true invasion [14]. Lowyck did not find significant correlation of recurrence with pressure erosions, or degenerative joint disease, neither with the location at the distal interphalangeal joint [31]. Reilly et al. observed that recurrence of giant cell tumour was much higher at the interphalangeal (IP) joint of the thumb and distal interphalangeal joints of the fingers [32]. Williams et al. reported that the high risk group was defined as tumour involvement of the extensor tendon, flexor tendon or joint capsule [24]. As compared to Al-Qattan Type-I tumours, Type-II tumours have been linked to a higher recurrence rate due to missed satellite lesion and subsequent inadequate excision [22, 33, 34, 35]
The tumour can be excised completely without any difficulty from a nodular variety since the margin is clear, but it is more challenging to remove a diffuse type of tumour because of its infiltrating nature [15, 22].
The use of FNAC for pre-operative diagnosis aids in pre-operative planning to avoid recurrence. Ng V Y *et al*. proposed the use of FNAC as a primary diagnostic aid and helps in preoperative planning to prevent recurrence [36, 37].
The use of a tourniquet, microscopic excision using an operating microscope or magnifying loupe [22] and meticulous exploration for satellite lesions are necessary for the definitive removal of the tumour [14]. To reduce the chances of recurrence it is essential to surgically remove all tumour remnants [1, 8, 18].
When the excision of the tumour disrupts the continuity of the tendon, then tendon repair may be necessary. Bony

debridement and curettage are required to treat the involvement of the bone and bony erosions when they are present. In a small proportion of the cases if the skin is involved, then an elliptical excision and primary closure is done; and if the skin defect is large then a skin graft become necessary [38]. In our case the tendons were intact after excision of giant cell tumour of tendon sheath; and there was no bony involvement.

In cases where total excision may not be feasible then radiotherapy may play a part in the treatment of giant cell tumour of tendon sheath. Kotwal *et al.* advised postoperative irradiation of 20 Gy in divided daily doses of 2 Gy in cases of incomplete excision, presence of mitotic figures and bony involvement [12, 14].

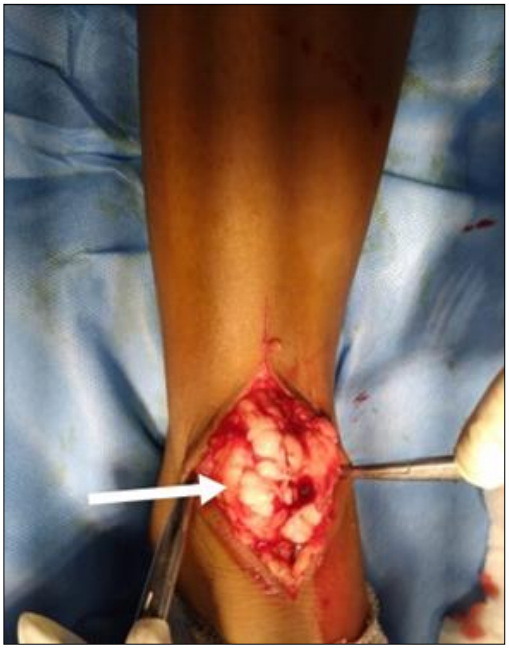

Fig 1.

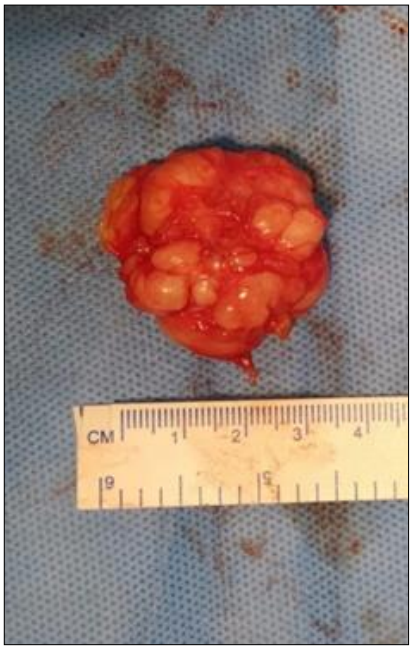


Fig 2.

**Fig 1 and 2:** Showing yellowish white multilobulated mass measuring about 3.5x3.5cm arising from the extensor tendon sheaths of tibialis anterior and extensor hallucis longus.

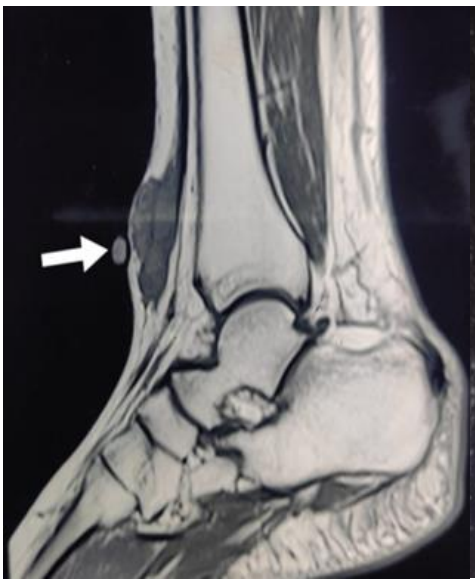

Fig 3

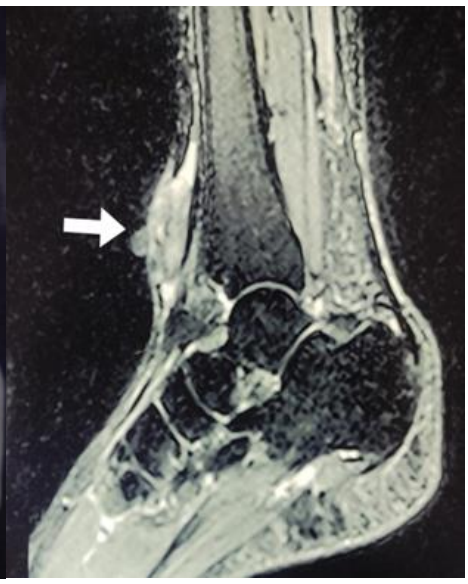

Fig 4

**Fig 3 and 4:** T1 and T2 sagittal sections showing a multilobulated lesion isointense to soft tissue seen along the anterior aspect of the ankle in the subcutaneous fat plane involving the tibialis anterior tendon.

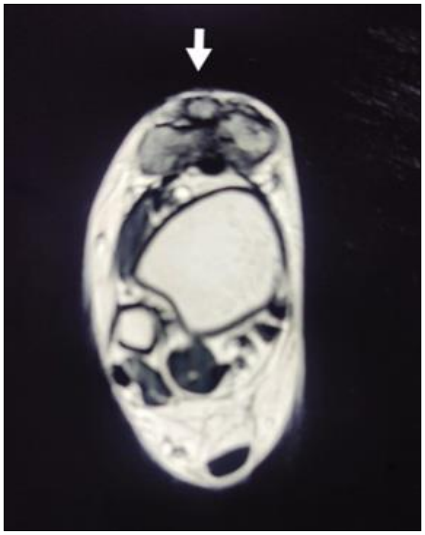

**Fig 5:** The T1 axial contrast lesion is showing moderate contrast enhancement.

## Conclusion

Giant cell tumour of tendon sheath is a rare condition in the foot and ankle. It usually presents as a painless mass in the foot and ankle which may cause compressive symptoms. Ultrasonography can detect whether a tumour is solid or cystic; and any accompanying satellite lesions can also be detected. Ultrasonography also describes the relationship of the lesion to the adjacent structures. Ultrasonography enables the determination of the degree of contact with the underlying tendon and the proportion of circumferential involvement. Imaging is essential in the work up and MRI is radiological investigation of choice. FNAC aids in pre-operative diagnosis and pre-operative planning to avoid recurrence. The optimal treatment strategy for giant cell tumour of tendon sheath in the foot and ankle is controversial due to the scarcity of the cases. Most widely accepted treatment for giant cell tumours of the tendon sheath is the excisional biopsy with or without radiation.

## Disclosures

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**Research ethics and patient consent:** Written consent for publication of patient details were obtained from parents.

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