# A Pregnant Woman with Multi-Fragmented Giant Cell Tumor of Tendon Sheath

## *A Rare Anatomical Location*

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Giant cell tumor of the tendon sheath (GCTTS) in the foot is a rare pathology and is involved in the differential diagnosis of soft-tissue tumors of the foot and ankle. Although it can affect any age group, GCTTS mainly occurs at the 3rd and 5th decade and is more common in females. Histopathologic examination is a major definitive method for diagnosis, although physical examination and radiologic imaging are helpful in reaching a diagnosis preoperatively. Many treatment options exist but marginal excision is the most commonly used treatment. We describe the case of a 26-year-old pregnant woman with a multi-fragmented mass extending from the first web space to the plantar aspect of the metatarsophalangeal joint (MTP) of the left great toe associated with flexor hallucis longus tendon after trauma. She had pain that worsened with activity and wearing shoes. After pregnancy, a marginal excision with dorsal longitudinal incision in the first web space was performed under spinal anesthesia. The lesion was diagnosed as a localized type tenosynovial giant cell tumor. At the last follow-up appointment in the 23rd month, the patient was doing well and there was no recurrence of the lesion. GCSST should be considered in the differential diagnosis of plantar masses of foot. Although, GCTTS is frequently seen in females, it has not been previously reported in a pregnant woman with an extremely rare condition after trauma. (J Am Podiatr Med Assoc 110(3): 1-5, 2020)



Giant cell tumor of the tendon sheath (GCTTS) is a benign nodular tumor that has become increasingly frequent in the foot and ankle.[1-3] This tumor especially affects the small joints in the extremities and usually presents as a solitary soft-tissue mass; however, simultaneous multi-fragmented lesions in the foot have been reported as an uncommon presentation.[4,5] It can be seen at any age but mostly affects individuals in their third to fifth decades of life and is mostly seen in females.[6-8] GCTTS has not previously been reported in a pregnant woman. Etiology of GCTTS is not clear but there are many risk factors mentioned in the literature such as trauma, infection, vascular abnormality, lipid metabolism disorders, osteoclastic proliferation, and immune system disorders.[4] Most cases of GCTTS appear in the hand but any part of the extremity can be affected, such as the foot, knee, elbow, hip, and wrist.[4] We report the imaging and pathological findings of a very rare, localized, and multi-fragmented tenosynovial giant cell tumor in a 26-year-old pregnant woman and conduct a literature review of the imaging characteristics, unusual localization, and diagnosis.

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## Case Report

A 26-year-old female patient in the eight month of pregnancy was admitted to our outpatient clinics with a palpable mass located at the first web space of her left foot, which had been enlarging for the previous 6 months. She had a history of a foreign body penetrating the skin under the foot when walking with slippers in the second month of pregnancy has since had pain that worsened with activity and wearing shoes.

Clinical examination revealed a solid and partly mobile mass lesion on the first web space of her left foot approximately $1 \times 2$ cm in size. There was tenderness on the plantar aspect of her great toe and increased pain with active and passive motion of the first metatarsophalangeal (MTP) joint. She had a hypoesthesia at the great toe due to digital

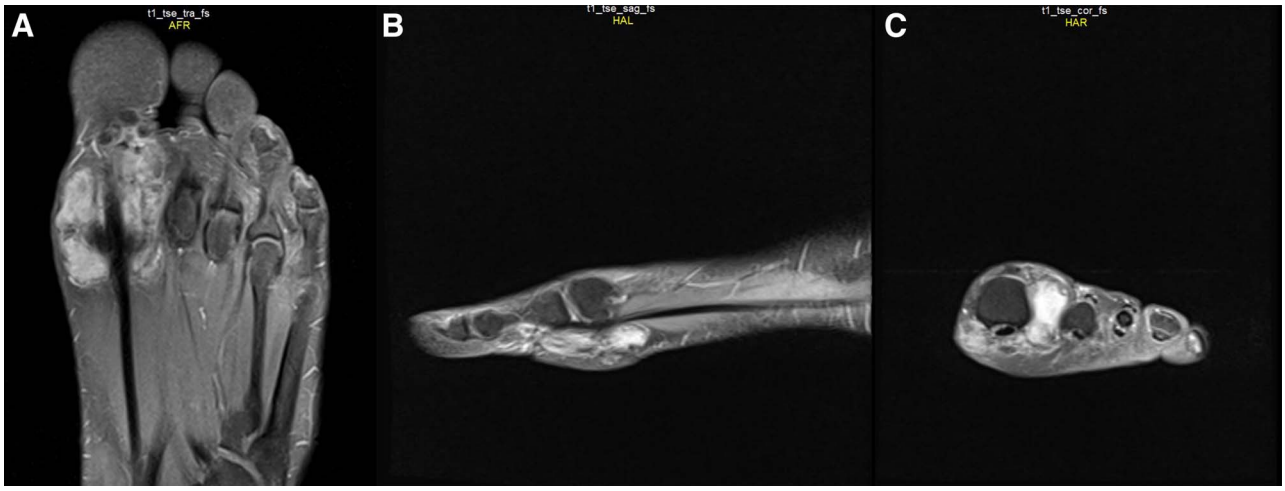


**Figure 1.** Coronal (A), sagittal (B), and transverse section (C) magnetic resonance imaging showed a solid-mass lesion, 48 × 38 × 17 mm in size, extending from the first web space to the plantar aspect of the metatarsophalangeal joint of the great toe.

nerve compression by the lesion. She had no medical and family history. There were no signs of inflammation (normal erythrocyte sedimentation rate, C-reactive protein, and total blood count). Ultrasonography showed a smooth, hypo-echoic solid lesion with internal vascularity.

The surgery was postponed to the postpartum period because of the risks of surgery during pregnancy. The magnetic resonance imaging after birth with gadolinium contrast administration showed a solid-mass lesion, $48 \times 38 \times 17$ mm in size, extending from the first web space to the plantar aspect of the MTP joint of left great toe. The lesion was hypo-intense on T1- and T2-weighted and diffuse homogenous contrast-enhanced images (Fig. 1).

The lesion was excised with marginal excision under spinal anesthesia after a longitudinal incision over the first web space of the left foot (Fig. 2). The lesion was gray-yellow in color, and there was no relationship between the mass and the adjacent bone. It mainly originated from the flexor hallucis longus tendon adjacent to the plantar aspect of the

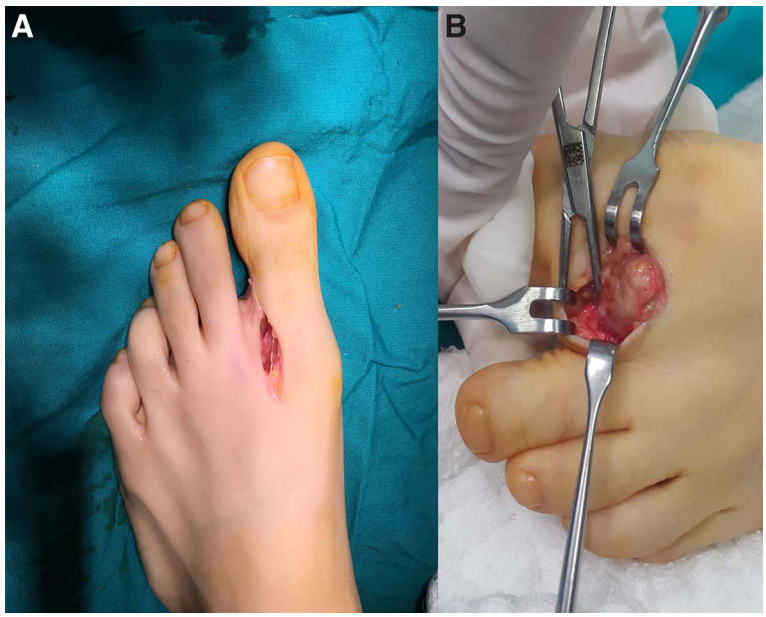


**Figure 2.** Intraoperatively, longitudinal incision over the first web space of the left foot (A) and a gray-yellow color lesion (B).

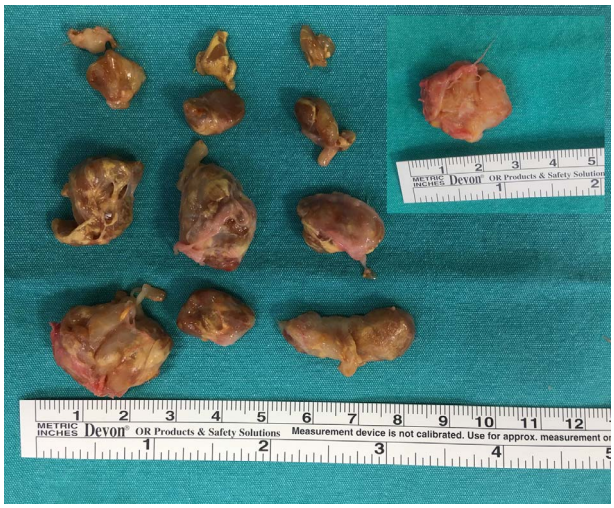


**Figure 3.** Macroscopically, the lesions were composed of multiple fragments ranging in size from 2.5 × 2 × 1 to 0.8 × 0.4 × 0.4 cm.

first MTP joint. Macroscopically, the lesion was composed of multiple fragments, ranging from 2.5 × 2x1 cm to 0.8 × 0.4x 0.4 cm in size (Fig. 3). Histopathologically the tumor consisted of broad, patternless sheets of histiocytes with bland nuclei and variable cytoplasm in a haphazardly arranged hyalinized stroma (Fig. 4A). Numerous osteoclast-like giant cells were also seen (Fig. 4B). There was no necrosis. The lesion was diagnosed as a localized type tenosynovial giant cell tumor. At the final postoperative visit (month 23) the patient was doing well and there was no recurrence of the lesion (Fig. 5). A written informed consent was obtained from the patient.

## Discussion

Although the GCTTS, also known as the pigmented-villonodular tumor of the tendon sheath, is a benign nodular tumor of the hand arising from the tendon sheaths,[5,8] the incidence of cases originating from the foot and ankle is increasing.[1,2] The localized GCTTS especially affects the small joints in the limb and usually presents as a solid-solitary soft-tissue mass, but simultaneous multi-fragmented lesions in the foot are uncommon.[4,5] When the tumor presents in the foot and ankle, it is primarily seen arising from second toe[2]; the dorsal aspect of the foot is more commonly affected than the plantar aspect.[9,10] The lesion in our patient was seen in an uncommon location, extending from the first web space to the plantar aspect of the MTP joint of left great toe.

Pain and neurological symptoms are not common in GCTTS of the hand[5,7,8] but they could be present when the foot is affected.[1] Zhang et al[2] retrospectively evaluated 20 patients with GCTTS and reported that three had discomfort on weightbearing because the mass was located on the plantar aspect; only one patient had neurologic symptoms.[2] In a study by Kant et al[6] of 26 patients with GCTTS, only 4 cases involved the lower extremity; three of those patients sensory impairment. In our work, the patient had hypoesthesia due to compression of the digital nerve by the mass.

The etiology of GCTTS is not clear but there are many risk factors such as infection, vascular abnormality, trauma, lipid metabolism disorders, osteoclastic proliferation, and immune system disorders.[4] Trauma is one of the rarest etiological factors causing GCTTS. In a study evaluating 17

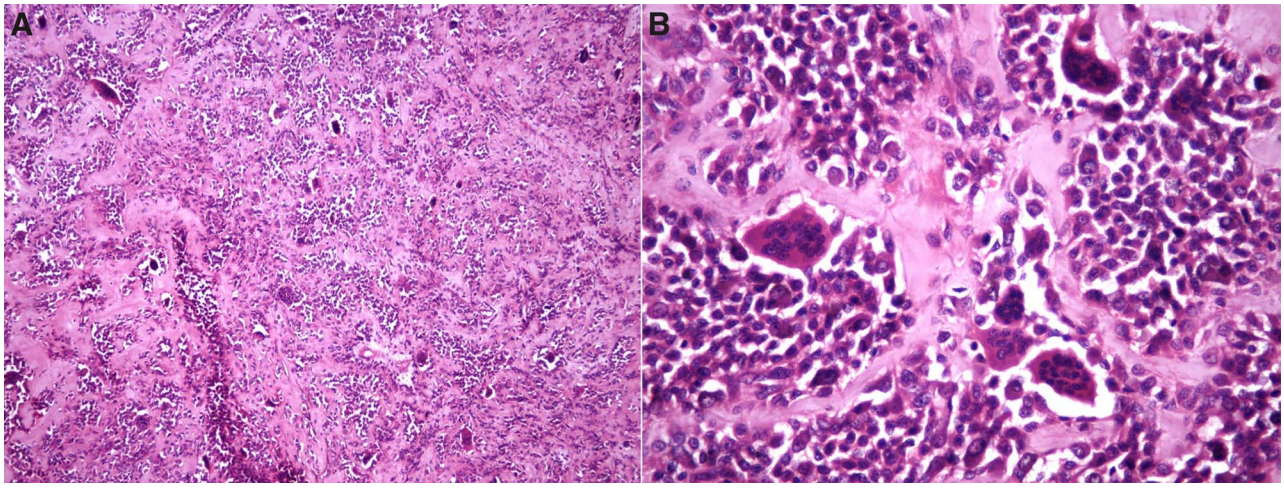


**Figure 4.** A, At low magnification, localized-type tenosynovial giant cell tumor (HEx40), B, Histiocytes and osteoclast like giant cells in a hyalinized stroma (H&E x200).

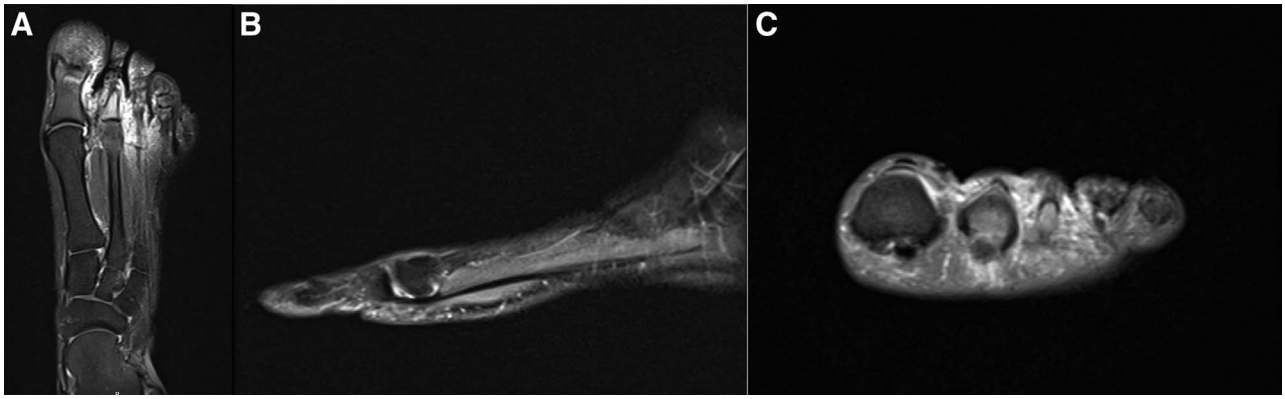


**Figure 5.** Coronal (A), sagittal (B), and transverse section (C) magnetic resonance imaging of the left foot at the 23-month postoperative follow-up. There was no recurrence of the lesion.

patients with GCTTS of the foot, only two patients had a history of trauma.[1] In the study by Zhang et al,[2] trauma was an etiological factor in two of the 20 patients having GCTTS. The patient in our study had a history of foreign body penetration under the foot while walking with slippers.

Histopathologic examination is a definitive method of diagnosis, although physical examination and radiologic imaging are helpful preoperatively. GCTTS is characterized by proliferating histiocytes and moderately cellular appearance. Cellular components are sheets of rounded or polygonal cells. In addition, hemosiderin (brown color) may be present but is typically less than what is seen in pigmented villonodular synovitis (PVNS) ; multinucleated giant cells are common.[4,6,7] Differential diagnosis includes ganglion cyst, PVNS, desmoid tumor, fibroma/fibrosarcoma, and glomangioma.[5,7,8] Pigmented villonodular synovitis is histologically identical to GCTTS but it most commonly involves larger joints; hemosiderin is more common in PVNS than in GCTTS. Giant cell tumor of the tendon sheath and ganglion cysts are virtually indistinguishable by clinical examination but are differentiated by ultrasound; GCTTS is solid, nodular, and has internal vascularity whereas ganglion cyst has a cystic component with well-defined borders and no internal vascularity.[5] Desmoid tumors occur in deeper limb tissues and have no obvious contact with the tendon sheath or joint capsule. The mixed cell population of GCTTS can distinguish it from a fibroma.[5,7,8] The primary clinical diagnoses in our patient were ganglion cyst or hemangioma. After radiological evaluation with ultrasonography and MRI we suspected GCTTS, desmoid tumor, or fibroma/fibrosarcoma. After surgical excision, the tumor was diagnosed as a localized type tenosynovial giant cell tumor.

Although there are many treatment modalities, marginal excision is the most commonly used.[1,5,6] The local recurrence rate has been reported between 4 and 45% in most of the cases in the hand.[11-13] There are only a few studies giving information about the recurrence rate of GCTTS after surgery in the foot and ankle. Zhang et al[1] reported a 20% local recurrence rate after marginal excision. Gibbons et al[2] reported no local recurrence after marginal or wide excision of the lesions. In our patient, a marginal excision of the tumor was performed under spinal anesthesia and there was no any signs of recurrence at the last follow-up.

## Conclusions

Giant cell tumor of the tendon sheath should be considered in the differential diagnosis of plantar masses of the foot. It is rare after trauma and usually presents as a solid-solitary soft-tissue mass. But it can be present unexpectedly as a multifragmented mass located at the plantar side of the first MTP joint in association with the FHL tendon as in our case. In addition, GCTTS is frequently seen in females but has not been previously reported in a pregnant woman.

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**Conflict of Interest:** None reported.

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