

Cytodiagnosis of Multicentric Giant Cell Tumour of Tendon Sheath: A Rare Case Report

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ABSTRACT

Giant Cell Tumour of Tendon Sheath (GCTTS) is a benign soft-tissue tumour that commonly arises from the tendon sheath complex and periarticular soft tissue of small joints. These tumours are usually localised and solitary, with multiple occurrences being rare. A multicentric origin is considered unusual and very few cases have been reported to date. Hereby, the authors present a case report of a 40-year-old female with a history of multiple swellings over the bilateral tendo-Achillis region, knees, left great toe and dorsum of the right middle finger since, 10 years ago. Biochemical parameters were normal and radiological investigations did not reveal any bony abnormalities. Fine needle aspiration from all sites revealed features consistent with GCTTS. Histopathology also showed findings typical of GCTTS. The present case highlights the unusual multicentric presentation of GCTTS.

Keywords: Benign tumour, Tendon sheath complex, Tendo-Achillis region

CASE REPORT

A 40-year-old female presented with a history of multiple swellings over the bilateral tendo-Achillis region [Table/Fig-1a,b], left great toe [Table/Fig-1c], bilateral knees [Table/Fig-1d] and dorsum of the right middle finger [Table/Fig-1e] for the past 10 years. On examination, the sizes of the swellings were 3×3 cm each on the bilateral tendo-Achillis, 4×3 cm on the left great toe, 1.5×1.5 cm each near both knee joints and 3.5×2 cm near the right middle interphalangeal joint. The swellings were firm and mildly tender in nature. She was clinically diagnosed with gout.

The patient was active and healthy. She denied any history of trauma, fever, chills, or weight loss. The patient had no significant medical history and was not taking any medications. She provided a past history of similar soft-tissue swelling on the bilateral tendo-Achillis 25 years ago, for which she underwent local surgical excision; however, no documents are available. Family history also revealed that her brother is experiencing similar soft-tissue swelling in the bilateral tendo-Achillis region, but the diagnostic work-up has not yet been performed.

Laboratory investigations revealed a serum uric acid level of 2.1 mg/dL and an Erythrocyte Sedimentation Rate (ESR) of 13 mm/1st hour. No bony involvement was detected on X-rays of all joints [Table/Fig-2,3]. Fine Needle Aspiration Cytology (FNAC) was performed on all the swellings. Cytosmears from all the swellings revealed similar morphology. The cytosmears were cellular, showing a mixed population of mononuclear cells, multinucleated giant cells and foamy macrophages [Table/Fig-4]. A cytodiagnosis of multicentric GCTTS was made. Surgical excision followed by histopathological



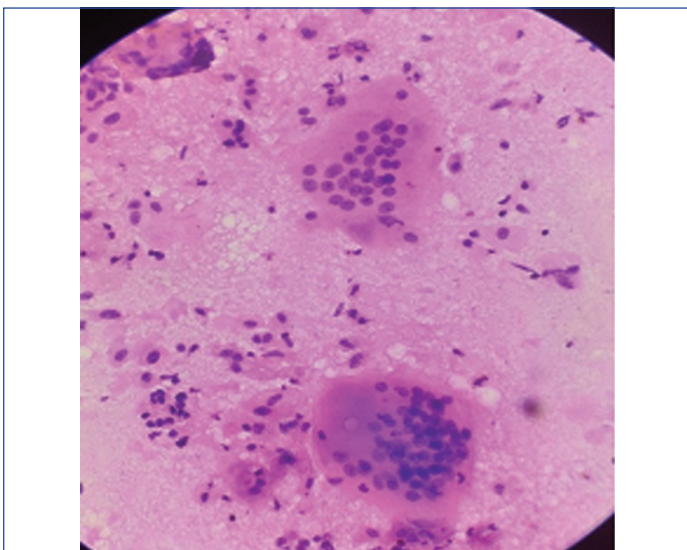
[Table/Fig-1]: a) Swelling over right-side tendo-Achillis region; b) Swelling over left tendo-Achillis region; c) Swelling over left-side great toe; d) Swelling over bilateral knee; e) Swelling over right-side middle finger; f) X-ray left ankle joint shows soft-tissue mass over the tendoachilles and over great toe.



[Table/Fig-2]: X-ray of the right hand showing soft-tissue mass over the middle finger.

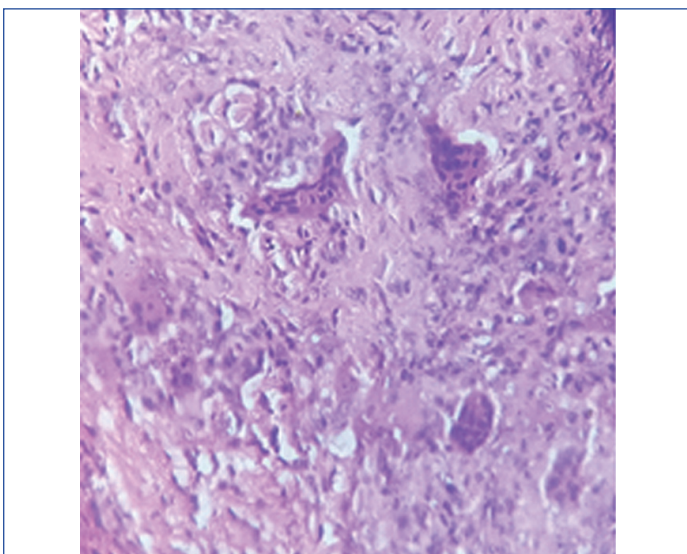


[Table/Fig-3]: X-ray right leg showing soft-tissue mass over the knee and the tendo-achilles.



[Table/Fig-4]: Cytosmear showing multinucleated giant cells and mononuclear stromal cells admixed with foamy macrophages (H&E, 400X).

evaluation by Haematoxylin and Eosin (H&E) staining showed mononuclear cells and giant cells in a collagenous background, confirming the diagnosis of GCTTS [Table/Fig-5].



[Table/Fig-5]: Histopathology showing giant cells and mononuclear stromal cells in a collagenous background (H&E, 400X).

As there were no haemosiderin-laden macrophages in the cut section, this rules out Pigmented Villonodular Tenosynovitis (PVNTS). Additionally, PVNTS predominantly affects larger joints, causing pain, swelling and stiffness. However, in present case,

the patient was asymptomatic with involvement of both smaller and larger joints. It was also differentiated from other soft-tissue lesions containing giant cells, such as reparative granuloma, by the absence of trauma, its uncommon location (reparative granulomas commonly occur in the mandible) and the presence of only a few giant cells.

DISCUSSION

Giant Cell Tumours (GCTs) of the tendon sheath were first described as fibrous xanthoma in 1852 by Chaissaignac. It is the second most common soft-tissue tumour affecting the hand, following ganglion cysts. The incidence of GCT of the tendon sheath is estimated to be around 1 in 50,000 individuals. This tumour typically occurs as a solitary mass in a finger; simultaneous multicentric lesions in the hand are very rare [1,2]. Some case reports and reviews have identified the presence of multiple GCTTS. Pathade SC et al., reported the occurrence of multiple GCTTS in the hand of a 26-year-old female [2]. Park JW reported a case in a 33-year-old male with two separate GCTTS that developed simultaneously in the same thumb [3]. Novick SD et al., reported a patient with localised multifocal GCTTS affecting the tendon sheath of the Flexor Pollicis Longus (FPL) on the volar surface of the right thumb [4]. Similarly, Singh T et al., reported multifocal GCTTS at different sites in a finger [5].

The GCTTS are rarely present in the palm, wrist, foot, knee, ankle, elbow, or hip. Ushijima M et al., reported that 87.9% of cases occurred in the digits (fingers and toes) and 12% in larger joints (knee, elbow, ankle and hip) [6]. Zhang Y et al., reported 20 cases of GCT-TS in the foot and ankle [7]. The occurrence of multiple giant cell tumours arising at different sites is exceptionally rare, with limited documentation available in the published literature. Our case report highlights the occurrence of GCTTS in multiple sites of a 40-year-old female. The atypicality in our case lies in the involvement of both small and large joints and shows multicentricity. Bhuyan P et al., reported a single case similar to ours, involving bilateral elbow and tendo-Achillis regions in a 21-year-old female [8].

The peak incidence is generally observed in individuals in their 30s and 40s, with a slight predominance in females. Giant cell tumour of the tendon sheath is now considered a benign neoplasm rather than a reactive lesion. The key pathogenic event is a chromosomal translocation $t(1;2)(p13;q37)$, leading to the overexpression of Colony-stimulating Factor 1 (CSF1) by a minority of neoplastic synovial-like stromal cells. This overexpressed CSF1 attracts non neoplastic macrophages, foam cells and multinucleated giant cells via the CSF1 receptor (CSF1R), forming the bulk of the tumour. Although trauma and inflammation were once thought to be causal, they are now viewed as secondary contributors [9,10]. The mutation of the nm23 gene has been linked to an increased chance of recurrence of GCT. This tumour is classified into two types based on location and behaviour: Type I (Nodular/Localised, typically in hands with a low recurrence rate) and Type II (Diffuse, often in larger joints). The diffuse form tends to be hypercellular with fewer giant cells, whereas the localised form displays numerous giant cells and is less cellular [1,2,11].

CONCLUSION(S)

Multicentric GCT of the tendon sheath is a rare condition. Although the hand is the most common site of involvement, one should keep in mind that the lower extremities can also be affected. Radiological investigations and cytodiagnosis are helpful for preoperative diagnosis and planning for surgical management. However, histopathological examination is also important to rule out malignancy in cases of recurrence. The patient should be advised to follow up at regular intervals due to its high recurrence rates.

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