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Case Report

Tenosynovial giant cell tumor, localized type, with recurrence, and lung metastases: A case report☆

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ABSTRACT

Tenosynovial Giant Cell Tumors (TSGCT) are the second most frequent benign soft tissue tumor in the hand and wrist after ganglion cysts. The tumor arises from joint synovia and tendon sheaths. It is divided into 2 categories: localized and diffuse type. The well-circumscribed localized type usually responds nicely to single excision surgery. The diffuse type, on the other hand, has a proclivity for becoming an aggressive lesion. We reported a case of a 56-year-old woman who was experiencing shortness of breath as a result of lung metastases. A TSGCT, localized type, was found after radiology, and histology examination of the re-excised surgical lesion was done. This rare case demonstrates the possibility of lung metastases from tenosynovial giant cell tumor, localized type.

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Introduction

Tenosynovial Giant Cell Tumor (TSGCT) or Giant Cell Tumor Tendon Sheath (GCTTS) is a type of tumor that arises from tendon sheaths, synovia, and bursae [1]. The prevalence of TSGCT is 1: 800.000, predominantly affecting women aged 40-50 years old [2]. In 2013, the World Health Organization divided this tumor into 2 categories: localized and diffuse type [1]. The digits and wrists account for two-thirds of cases of localized type (85%), while major joints such as the knee, hip, ankle, and elbow are involved in diffuse type [2].

The localized type is typically benign, whereas the diffuse type is destructive, and aggressive with a malignancy component [2]. TSGCT is a benign tumor that arises from the tendon sheath in most cases. However, in a few cases, the diffuse type has been documented to lung metastases. The purpose of this case report was to confirm that TSGCT, localized type, can become malignant, and spread to the lungs [2,3]. A case of lung metastases and right pleural effusion from right palmar TSGCT, localized type, origin is presented. The tumor was twice verified as a localized type by histopathology and MRI examination.

☆ Competing Interests: The authors have no affiliation with any organization with a direct or indirect financial interest in the subject matter discussed in the manuscript

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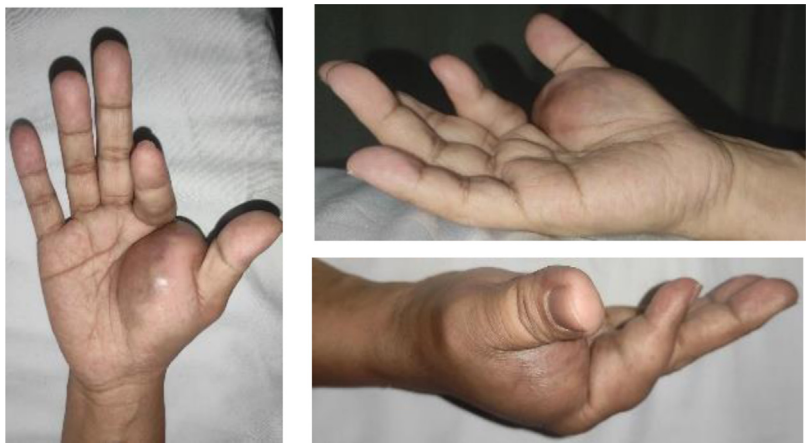

Fig. 1 – Clinical manifestation of patient's right palmar.

Case presentation

A 56-year-old woman came to the emergency room with worsening shortness of breath during activity since the last 8 months. The COVID-19 polymerase chain reaction (PCR) revealed no identified Sars Cov-2 RNA virus. The patient had received all of her vaccinations (2 doses of coronavax). The patient additionally complained of a lump on her right palmar, which restricted her joint movement. Four years ago, the patient had surgery to remove a tumor at the same location. Tenosynovial Giant Cell Tumor was discovered during the latest histopathological investigation. She had no neurologic problems, no abnormal intraabdominal ultrasound, and no abnormalities on her brain CT scan.

A firm, hard, and localized mass on the thenar aspect of the right second digit, measuring 6 × 4.5 × 3 cm, was discovered during a local physical examination. It was mobile, solitaire, painful, and distal sensitivities were normal. Right pleural effusion with metastatic lesions was seen on chest X-ray imaging (Fig. 2). She was stabilized by effusion drainage. The pleural effusion fluid was then investigated (serologic test), and osteoclasts such as large cells, and mononuclear cells were discovered. A biopsy of the left lung nodule revealed atypical mononuclear cells with enlarged nuclei and conspicuous nuclei, as well as a considerable number of osteoclast-type giant cells (Fig. 3).

Based on contrast MRI, a circumscribed, regular border lesion measuring 3.52 × 2.80 × 4.93 cm was found on the right palmar area. It invaded the second caput metacarpal and affected the right flexor pollicis brevis, adductor pollicis brevis, and first lumbricalis muscles. On T1W1, tumor lesions were hypointense and hyperintense. Furthermore, the tumors became enhanced with contrast (Fig 5).

Other visible bones showed no signs of erosion or destruction. She re-excisioned the tumor in the next few days. It was diagnosed as TSGCT, localized type, with osteoclast-like giant cells, and a free tumor margin on histopathology. Based on the clinical, radiological, and histopathological examinations, the patient was diagnosed with a metastatic pulmonary lesion which is associated with TSGCT.

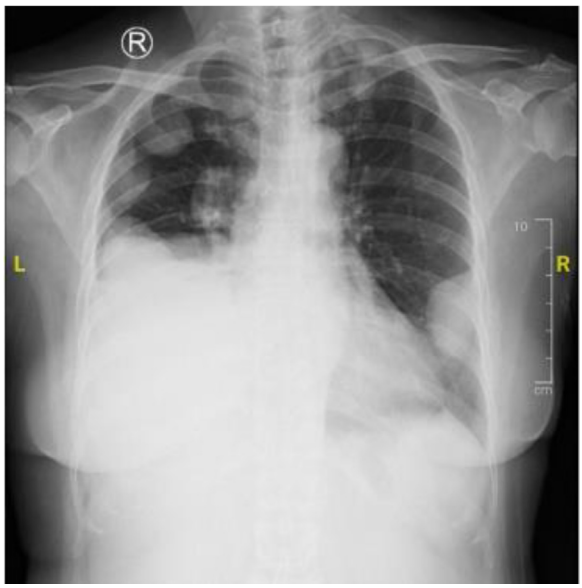

Fig. 2 – Chest X-Ray with multiple metastasis nodules and right pleural effusion.

Discussion

The World Health Organization puts TSGCT into 2 categories; localized and diffuse types [1]. The localized type occurs predominantly in the well-circumscribed tendon sheaths of the hand and foot, meanwhile, the diffuse type develops articular in big joints with a more aggressive growth pattern [1,2]. The involvement of the fossa and extra-articular extension should be assessed for preoperative workup [2]. TSGCT is a benign tumor that arises from the tendon sheath in most cases, but, in a few cases, the diffuse type, which has destructive and aggressive properties, has been found to turn malignant, with lungs metastases [2,3]. There are no case reports or previous research showing the development of TSGCT, localized type, into malignant, and lungs metastases.

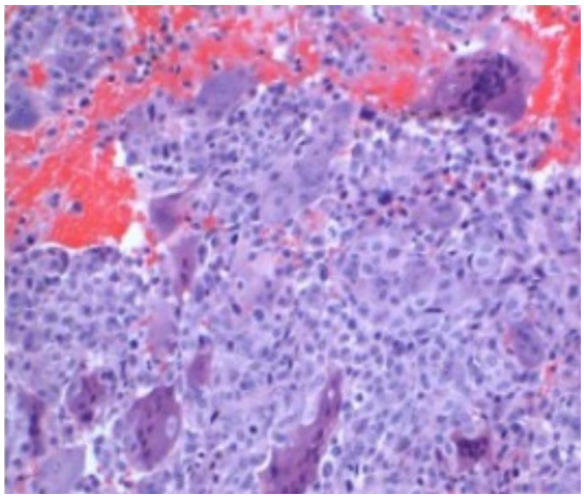

Fig. 3 – Histopathology examination of lung nodule biopsy (hematoxylin and eosin stain, 100x magnification).

The symptoms of the localized form are usually painless and have a minor clinical manifestation. As it grows, it expands, and restricts articular movement. The diffuse type, on the other hand, is painful and progresses over months to years [3]. In this case, patient was unconcerned as to her recurrent hand lump. She went to the ER because she was having difficulty in breathing, which led to the lump being treated. The patient also had soreness in the palm of her hand. However, she could perform her daily routines with some restrictions on her 2 digits, the first, and second. The patient had received all of her vaccinations (2 doses of coronavax). The COVID-19 PCR revealed no identified Sars Cov-2 RNA virus. Dyspneu was stabilized via effusion drainage. In the preparation cells from the right pleural effusion, the serologic test detected osteoclasts like giant cells, and mononuclear cells in the following days. She had no neurologic impairments, no abnormal intraabdominal ultrasound, and no abnormalities on her brain CT scan. The original treatment plan for this patient was chemotherapy. The patient and her family opted to refuse further therapy after receiving a thorough explanation from the oncologist team about the dangers, side effects, and 5-year survival rates of the treatment.

MRI plays an important role in diagnosing and assessing TSGCT type, size, extension, and invasion of adjacent joint and tenosynovial space preoperatively [4]. The localized type is, most of the time, isointense or low-signal intensity in T1- and T2-weighted [3]. It usually possesses uniform enhancement and/or osseous involvement but sometimes exhibits variability in signal intensity on MR images [4,5]. Hemosiderin presence and fibrosis formation raise small, scattered foci of low signal on T1WI, and T2W1 [3,4,6].

The diffuse type has an unclear margin, a multinodular lesion, and vigorous development outside the joint [7]. On T1WI, MRI indicates balance or greater signals than muscle, whereas T2WI has a wide range of characteristics, including hypointense, isointense, and hyperintense signals [8]. In this case, the mass in the right palmar area was discovered to be solid with a clear margin and regular edges, measuring $3.52 \times 2.80 \times 4.93$ cm. It infiltrated the right second metacarpal head and disrupted m. right flexor pollicis brevis, m. right abductor pollicis brevis, first m. right lumbricalis, and provided a hypointense signal on T1WI (Fig. 5). Other bones did not appear to be eroded or being destroyed. The lesion was characterized as a giant cell tumor of the tendon sheath of second proximal digit with tumor-free incision margin, as validated by intra-operative findings (Fig. 6), and confirmed by the second histopathology examination. A well-circumscribed nodular or multinodular cellular mass that may be encapsulated was examined histopathologically for localized type. It contained mononuclear and multinucleated giant cells,

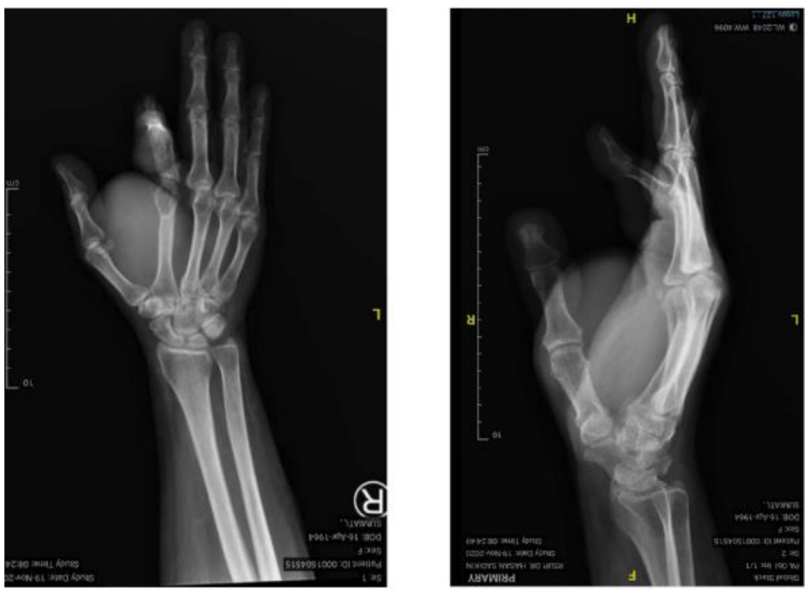

Fig. 4 – Right palmar X-Ray with the presence of soft tissue tumor.

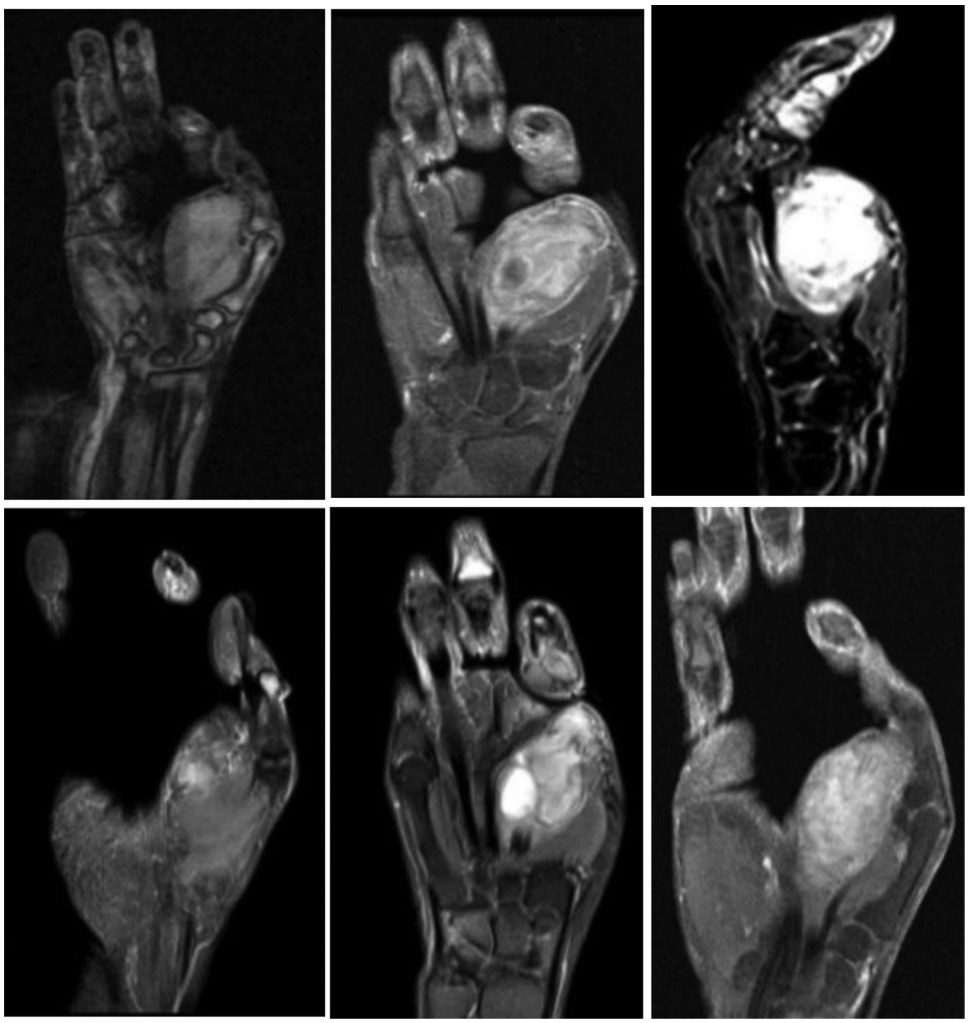

Fig. 5 – Right palmar MRI sequnces: T1W1, T2W1, T1 with contrast and fat saturation, T2 with fat saturation.

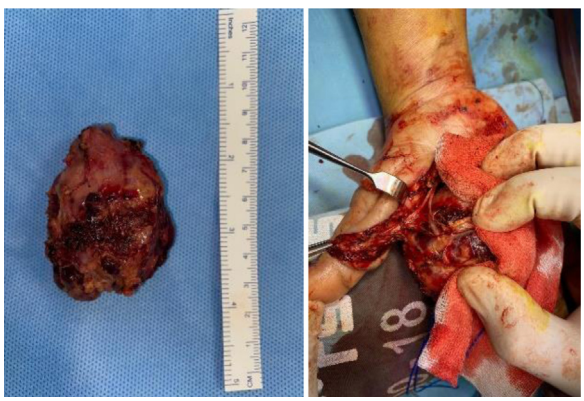

Fig. 6 – Surgical reexicision showed well demarcated, uniloculated and regular surfaced tumor.

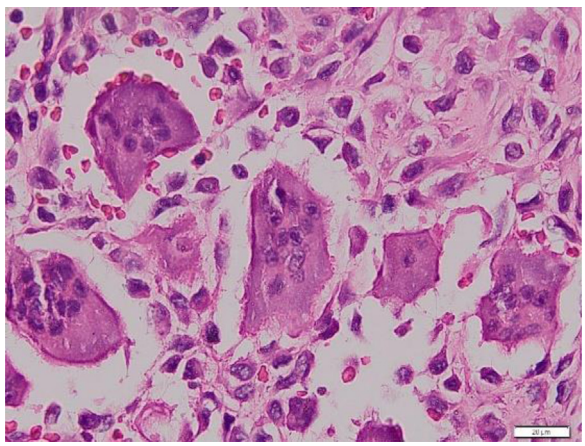

Fig. 7 – Multiple osteoclast giant cells found in patient's histopathology examination (hematoxylin and eosin stain, 400x maginification).

histiocytic and inflammatory cells, and a considerable amount of hemosiderin, as well as nodular proliferation of mononuclear and multinucleated giant cells [3]. Otherwise, the polymorphic population of a mononuclear, and multinucleated

giant cells represented as to the diffuse type [3]. This case is quite unusual because the localized type was previously believed to be benign and had never progressed to malignancy, according to a variety of literature [12].

The conventional treatment for TSGCT is excision. Initial local excision that is adequate may effectively reduce the probability of local recurrence [9]. Furthermore, direct involvement of the extensor tendons, flexor tendons, or joint capsules also play a part in a high-risk recurrence group [10]. The localized type has a low recurrence rate of 0%-6% after surgical therapy, but the diffuse type has a recurrence rate of 14%-92% [11] The diffuse TSGCT recurrence rate after open synovectomy is 14%-67%, and after arthroscopic synovectomy is 40%-92% [14].

Even if it is shown to be benign histologically, the TSGCT, diffuse type, has a tendency to become a metastatic disease after several recurrences. Nonetheless, malignant change with the prospect of metastasis is uncommon [3]. Repetitive operating procedures have been identified as risk factor for that condition [13]. Lung metastases mechanism was still unclear. While opposed to vascular involvement, lymph node involvement was thought to play a higher role in distant metastasis. Furthermore, according to current studies, only TSGCT, the diffuse type, can turn malignant, but this case was localized type [12,13]. TSCGT, diffuse type, metastatic spread to the lungs and/or locoregional lymph nodes is a very rare occurrence [13]. This case is unique since previous research has never shown that TSGCT, localized type, can develop into malignant, and spread to the lungs.

Conclusion

TSGCT required the use of MRI for diagnosis and surgical planning. While all of this type of TSGCT appears to be benign, the localized type should be considered to have a possibility e of lung metastasis. Due to a lack of information in the existing literature, more research on this condition is required. This rare case demonstrates the possibility of lung metastases from tenosynovial giant cell tumor, localized type.

Patient consent

Written informed consent was obtained from the patient family for publication of this case report and accompanying images.

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