Scaphoid instability caused by a giant cell tumour of the tendon sheath: A case report

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Abstract

Giant-cell tumour of the tendon sheath (GCTTS) is a soft tissue tumour that may invade bone, causing an intrinsic osseous lesion or instability on radiographs. A case with scaphoid instability caused by a histologically-confirmed neighbouring GCTTS has rarely been described in the literature. No definite and radical method is available for the treatment of GCTTS. This report describes an unusual case of a 22-year-old woman who previously experienced a GCTTS in her right elbow, which was removed 10 years earlier. Currently, she presented with an enlarged painless right wrist mass with focal swelling. The mass has been present for 5 years. During the previous 6 months, she felt something pop and experienced pain with limited motion in her right wrist. Magnetic resonance imaging demonstrated a well-circumscribed soft tissue mass. Under general anaesthesia, complete surgical resection of the mass was undertaken. Histopathological examination revealed that the mass was a GCTTS. Less invasive leverage reduction with external fixator support and iliac crest bone autologous graft for treatment of carpal instability were performed. Radical resection combined with external fixator support and bone grafting can provide a new option for the treatment of carpal instability.

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Introduction
A giant-cell tumour of the tendon sheath (GCTTS), an uncommon soft tissue tumour with a pathophysiological process that has been well documented by recent literature,1–4 is a benign tumour that typically presents as a localized, painless, slow-growing, nodular or polylobular mass that potentially interferes mechanically with tendon or joint function.2–5 GCTTS has a high rate of recurrence (up to 30–45%).6,7 This case reports on a female patient with a GCTTS in her right elbow that was excised adequately 10 years ago. The patient revisited the hospital 5 years later and a neoplasm was found in her right wrist with right scaphoid instability or dislocation when the wrist was actively flexed. This case report presents a review of the radiographic, histopathological, and magnetic resonance imaging (MRI) findings of this case.

Case report
A 22-year-old right-hand dominant woman presented to the Department of Orthopaedics, The First Affiliated Hospital of Sun Yat-sen University, Huangpu District, Guangzhou, Guangdong Province, China in February 2016 with an enlarged painless right wrist mass with focal swelling that had persisted for 5 years (Figure 1A). During the previous 6 months, the patient had felt something pop and experienced pain with limited motion in her right wrist. Initially, she took no action; and the pain and discomfort settled after a period of 3 days. After 1 month, the wrist still ached and felt weak, and the range of motion was limited. The patient reported that a GCTTS in her right elbow had been removed 10 years previously. No recent injury, antecedent trauma, local neurological symptoms, fevers, or changes in weight were found in the patient.

Physical examination revealed a non-tender, 3 × 3 × 3 cm firm, mobile mass mostly overlying the dorsal aspect of the scaphoid and trapezium. The swelling was non-bony. No rash or other identifiable overlying skin changes were noted, and the lesion did not transilluminate. The patient had normal strength. The range of motion of the wrist was limited compared with the left wrist, with a 20° loss of flexion, 25° loss of extension and 8° loss of radial and ulnar deviation. Full pronation and supination were noted. Sensation to light touch remained intact in all three nerve distributions. A negative Tinel’s sign was noted, and the mass was non-pulsatile with a normal vascular examination. Laboratory tests, including erythrocyte sedimentation rate, C-reactive protein and white blood cell, were normal.

The simple radiographs revealed a 3-cm solitary soft tissue shadow dorsal to the scaphoid and trapezium. The mass contained fluffy bone thickening and an almost complete destruction of the distal radius over a distance >0.3 cm without associated periosteal reaction, mineralization, extrinsic osseous erosion, or cystic change in the adjacent bone (Figure 1B). An MRI indicated that the mass was mainly located dorsal to the scaphoid and trapezium without communication with the radioulnar joint. The mass appeared hypointense in T1-weighted sequences and hyperintense in T2-weighted sequences (Figures 1C and 1D).
Under general anaesthesia, complete surgical resection of the mass was undertaken. The mass on the wrist joint level appeared firm and was multi-nodular with a yellowish-tan to mottled brown colour (Figure 2A). Intraoperatively, the mass was attached firmly to the surrounding tissue (Figure 2B). Less invasive leverage reduction with external fixator support and iliac crest bone autologous graft for treatment of carpal instability were performed (Figures 2C and 2D). The overall results of immediate postoperative radiographs and ultrasonic examination were satisfactory.

Microscopic examination revealed that the tumour was primarily composed of broad dense collagen bundles with inflammatory cells and round cells scattered throughout. A few areas of acellular, dense eosinophilic material were noted. The tumour contained foci of multinucleated giant cells, round synovial-like cells, inflammatory cells, xanthoma cells, and siderophages with haemosiderin pigment (Figure 3A). An immunohistochemistry analysis revealed that the internalized cells as well as the cannibal cells expressed cluster of differentiation (CD) 68, CD45,
and Ki-67 (granular cytoplasmic expression), which are presented in Figures 3B, 3C, and 3D. Histopathological examination revealed that the lesion was a GCTTS.

The study was approved the Investigational Ethical Review Board of the First Affiliated Hospital, Sun Yat-sen University and written informed consent was obtained from the patient.

Discussion

Immunohistochemical analysis of the present tumour indicated that it was a GCTTS. GCTTS is commonly found next to joints, including the ankle, knee, hip, elbow, and shoulder, and spine. Wrist occurrence is relatively rare. A previous study described 19 cases of GCTTS, of which one occurred in the wrist. Another published study reported 14 cases of GCTTS, of which one case occurred in the wrist. Neoplastic cells account for less than 16% of total cells, and a frequent chromosomal translocation [t(1:2)(p13;q37)] is responsible for the overexpression of colony stimulating factor 1 (CSF1) RNA/protein (fusion CSF1:COL6A3). CSF1 leads to a massive invasion of macrophages, stimulating increased phagocytic and chemotactic activity, and causes tumour cell cytotoxicity. Although the hypothesis of a true neoplasm resulting from sesamoid bones, synovial membrane, or primitive mesenchymal cells has been proposed, some researchers believe that GCTTS is an inflammatory disease that may be associated with inflammation and trauma (including chronic strain). Tendon or synovial injuries lead to fibrous hyperplasia of the synovium. Approximately 50% of patients have a history of trauma. In this present case, there was no definite history of trauma. Progressive enlargement or

Figure 2. Yellowish-tan to mottled brown-coloured masses with typical multi-lobulated appearance are excised from the lesion (A). Intraoperative view showing obvious adhesion to surrounding tissue (B). Postoperative radiographs with external fixator supported and iliac crest bone autologous graft (C, D). The colour version of this figure is available at: http://imr.sagepub.com.
recent enlargement of the tumour was the first-episode symptom. GCTTS is usually a benign tumour of still uncertain origin. However, malignant degeneration can occur in rare cases. According to histological criteria, GCTTS can be locally aggressive, with reports of the most frequent hand or foot sites and the possibility of multiple recurrence. Surgical resection is an effective treatment, but the recurrence rate is high.

Giant-cell tumour of the tendon sheath was diagnosed in the current case based on the history of a painless slowly growing wrist mass combined with distinctive radiological and histological findings. Plain radiographs revealed a soft tissue mass without associated periosteal reaction or bony erosion. MRI has the advantages of multi-dimensional imaging; high resolution of soft tissue; clearly depicting the shape, size, range and adjacent tissue structure of the tumour; and high sensitivity and specificity for diagnosing GCTTS. In the T1-weighted image of this case, the GCTTS signal, which is similar to the skeletal

Figure 3. Representative high-power photomicrographs of the tumour tissue. The tumour contained foci of multinucleated giant cells (red arrow), which are present in the centre of the field (haematoxylin and eosin) (A). Ki-67 immunostaining of the peripheral zone, which was relatively high (B). Cluster of differentiation (CD) 45 immunostaining of the peripheral zone. The histiocytes were positive for CD45. CD45-positive lymphocytes were scattered (C). CD68 immunostaining of the cellular zone. The internalized cells showed cytoplasmic expression of CD68 (D). The colour version of this figure is available at: http://imr.sagepub.com. Scale bar 100 μm.
muscle signal, was hypointense. On T2-weighted imaging, the signal intensities of the GCTTS tended to be between those of skeletal muscle and fat. Most of the lesions showed non-homogeneous signal intensities, including foci of low-signal intensities, which might be associated with haemosiderin deposits on T2-weighted imaging. These current findings are consistent with those of previous reports. Previous studies reported inconsistent MRI signals for GCTTS and a variety of signals were noted. Some authors have noted that the GCTTS signals on T1-weighted and T2-weighted imaging were equal. A small number of GCTTS lesions exhibited low signals on T1-weighted imaging, whereas most signals were similar to skeletal muscle signals. However, on T2-weighted imaging, equal, high or low signals were reported, and most of the signals were not uniform. GCTTS exhibited a variety of MRI signals, which may be related to the pathological composition of GCTTS and duration of the disease.Positron emission tomography (PET)/computed tomography (CT) imaging appears to have promise in predicting long-term outcome. For example, an increased radionuclide uptake of 18F-fluorodeoxyglucose (FDG) PET/CT caused by inflammation-induced hypervascularity and heterogeneously distributed hypercellularity is the premise of imaging diagnosis. From this limited study, 18F-FDG PET/CT, the most effective predictor of long-term outcome following treatment, helps to diagnose the inflammation resulting from a pathological influx of activated macrophages, delineate the local extent of the disease, and monitor the efficacy of treatment.

Giant-cell tumour of the tendon sheath is composed of haemosiderin, collagenous tissue and fat. Given the paramagnetic effect of haemosiderin, the local magnetic field is obviously nonhomogeneous. Thus, the characteristic low signal can be observed on T1-weighted and T2-weighted imaging. The tumour is commonly iso-intense relative to the muscle on T1-weighted images. The variable intensity on T2-weighted images can be detected because of the variable content of haemosiderin, liquid, lipids, fibrous tissue, and haemorrhage. Gradient echo sequences cause a lower signal because the low paramagnetism further amplifies the low signal of the tumour. A published study showed that the tumour in 10 of 20 GCTTS patients appeared as a soft tissue mass with homogeneous, predominantly low-signal intensity on T1- and T2-weighted images. In the current study, MRI enhanced scans exhibited markedly nonhomogeneous enhancement. Histological analysis revealed that the mass was composed of proliferative round synovial-like cells that differed microscopically accompanied by a variable number of multinucleated giant cells, siderophages, inflammatory cells, and xanthoma cells. Furthermore, the underlying disease process of GCTTS involves inflammation. GCTTS has clinicopathological features, including a nodular growth pattern and propensity for recurrence after incomplete excision, suggesting that the mass is a neoplastic proliferation of synovial histiocytes and fibroblasts. Although some uncertainty remains concerning its histogenesis, karyotypic abnormalities isolated in GCTTS cells have largely been accepted as evidence of a neoplastic phenomenon.

During 3-year follow up, the patient had normal wrist joint function, normal range of motion compared with the contralateral wrist, and no clinical evidence of recurrence based on MRI evaluation. We plan to continue a long-term follow-up for any signs of disease recurrence.

In conclusion, a GCTTS with a predilection for juxta-articular locations can cause carpal instability. Radical resection
combined with external fixator support and bone grafting can provide a new option for the treatment of carpal instability.

**Declaration of conflicting interests**

The authors declare that there are no conflicts of interest.

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