

CASE REPORT

Post-traumatic tenosynovial chondromatosis following a triquetrum fracture: a case report

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Abstract

Tenosynovial chondromatosis (TC) is a rare progressive benign tumor from the synovial lining of tendon sheath. The TC mostly affects males between the ages 30 to 50 years old at the ventral side of wrist. There are two different forms of TC that have been proposed in previous studies: an idiopathic cause (primary TC) and a joint related diseases cause (secondary TC). Even though trauma has been written to be a common reason for TC, a case of a secondary TC affecting the dorsal wrist following a triquetrum fracture has never been written before. The aim of this report is to present a rare case of a solitary post-traumatic TC at the dorsal wrist following a triquetrum fracture. We describe the clinical presentation, imaging modalities, histopathological and treatment challenges to manage this difficult lesion.

INTRODUCTION

Synovial chondromatosis (SC) is a rare benign lesion of the synovium found in joints and tendon sheaths that undergo metaplasia leading to cartilaginous nodules [1–3]. The SC in joints usually involves large areas such as the knee and the hip [1]. If SC develops extra-articular in the tendon sheaths, it is referred to as tenosynovial chondromatosis (TC). The TC tends to occur in the fingers, feet and wrist [1–3]. Despite being a benign lesion, the synovium produced in TC can calcify, generate loose bodies and damage the adjacent cartilages or tendons [4]. Maccagnano *et al.* defined TC into primary TC or secondary TC [4]. The primary TC occurs in normal joints by an unknown etiology while the secondary TC is the result of joint diseases such as osteoarthritis, neuropathic arthropathy and osteochondral fractures [4]. Even though trauma has been written to be a common reason for TC, a case of a secondary TC affecting

the dorsal wrist following a triquetrum fracture has never been written before. The aim of this report was to present an unusual case of a solitary post-traumatic TC following a triquetrum fracture.

CASE REPORT

A 62-year-old woman with a past medical history of obstructive sleep apnea, hypertension and no toxic habits, presented with an 18-month history of progressive pain, swelling and enlarging mass on the dorsum of her dominant right wrist. Patient reported that the mass progressively changed from a soft to a hard consistency during this time, producing a restriction on the range of motion (ROM) at the hand. Two years before our initial visit, she had a non-displaced triquetrum fracture after falling from standing height at her home (Fig. 1).

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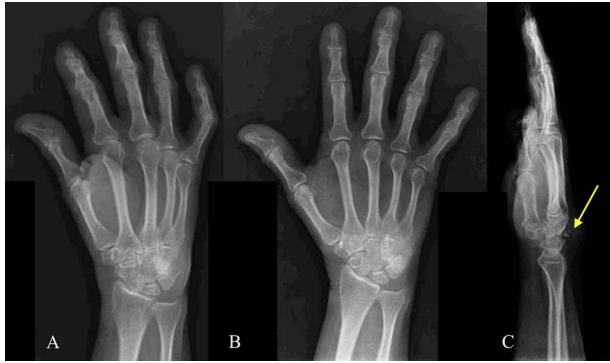


Figure 1: Right (A) oblique, (B) anteroposterior and (C) lateral hand X-rays with a triquetrum fracture (yellow arrow).



Figure 2: Right (A) anteroposterior, (B) oblique and (C) lateral hand X-rays before intralesional biopsy.

At evaluation, a non-obese (body mass index=24) patient with a palpable, soft and non-tender mass on the dorsum area of her right wrist was noted. The right hand had no neurological or vascular compromise. She had swelling and pain in dorsal wrist, with limited ROM of fingers at active and passive extension. Laboratory studies were negative for rheumatologic disease. Radiographs revealed a partially calcified mass in the dorsal aspect of the wrist concerning for a possible osseous or cartilaginous lesion (Fig. 2). A magnetic resonance image (MRI) of the right wrist showed an extensor tenosynovitis and a heterogenous lesion (measuring $2.6 \times 1.3 \times 2.5$ cm) at the level of the first carpal row; suggestive for focal pigmented villonodular synovitis (PVNS), soft tissue chondrosarcoma or TC (Fig. 3).

After MRI results, an initial open intralesional biopsy was performed due to the assessment of an indeterminate soft tissue lesion with high probability of malignancy [5]. The pathology report from initial biopsy revealed a TC lesion. Upon diagnostic confirmation, patient was oriented on the prognosis and scheduled for an open mass excision. At surgery, patient was blocked with a supraclavicular anesthesia. A dorsal surgical incision was done at the previous initial biopsy site, along with a wide dissection of extensor tendons and extensor tenosynovectomy. The extensor retinaculum was surgically opened and a wrist joint arthrotomy was performed. At further exploration, a 2.2×1.3 cm soft tissue tannish trabeculated rubbery mass was excised from the carpus with an extensive curettage (Figs 4 and 5). The joint

capsule was irrigated, and extensor retinaculum and adjacent tissues were rearranged for wound closure. Patient tolerated procedure without major complications. The pathology specimen was sent to National Institute of Health which revealed similar findings to initial biopsy, confirming a well-differentiated lesion consistent with TC.

At 1-week postop visit, patient had significant improvement of associated pain symptoms, wounds healed appropriately, and an adequate hand ROM returned. After 2 years of follow-up, patient continues to do well, and no recurrence or metastasis have been observed (Fig. 6).

DISCUSSION

This report highlights a rare case of TC lesion at the dorsal wrist manifested after a triquetrum fracture. TC is a rare progressive benign tumor from the synovial lining of tendon sheath that mostly affects males between the ages 30 and 50 years old. The TC have been more commonly reported at the dorsal side of hand and wrist [1, 2, 4].

The exact pathogenesis of TC is unknown; however, it has been postulated that tendons undergo an initial neoplastic proliferation of cartilage tissue with hyperplasia in synovial and cartilage nodules [4]. Two different forms can underly the differences of TC etiology [4]. The primary TC relates an unknown etiology while the secondary TC involves previous history of osteoarthritis, osteochondral fractures or neuropathic arthropathy [4].

The diagnosis of a TC is challenging due to the clinical and imaging variability presentation. Clinically, a wrist can present with swelling, pain, finger deformities and reduced ROM caused by compression of nearby structures [6]. A wrist X-ray can reveal calcification or ossification of cartilaginous nodules, bone erosion or have normal findings [1]. Additional imaging tools, such as MRI, play an excellent role in the location of loose bodies, proliferative synovium and extension of TC lesion prior to surgery. However, TC can be mistaken in MRI for other soft tissue conditions such as periosteal chondroma, PVNS and soft tissue chondrosarcoma [4, 7, 8]. In cases where there is an inconclusive diagnosis made by MRI, a biopsy with histological analysis remains the gold standard for diagnosis [4, 5].

The standard treatment for TC is complete excision of the neof ormation and adjacent synovium [9]. Postoperative TC recurrence of the wrist has been reported in 3–23% of cases, requiring additional surgery for excision [10]. In cases where there is a high suspicion for a malignant soft tissue tumor of the wrist, an intralesional biopsy can be performed prior to the complete excision [5]. In our patient, the involvement of loose bodies was not present at initial X-ray and MRIs; and a high suspicion of malignant soft tissue was noticed. Based on the imaging studies and the preference of the principal investigator, an intralesional biopsy was performed prior to an excisional biopsy.

A clinical diagnosis of secondary TC should be suspected in patients with history of prior wrist bone fracture. If clinical and imaging modalities are inconclusive for TC, an initial intralesional biopsy can be performed prior to the complete excision of the mass. After surgery, patients should be monitored regularly for possible recurrence of TC.

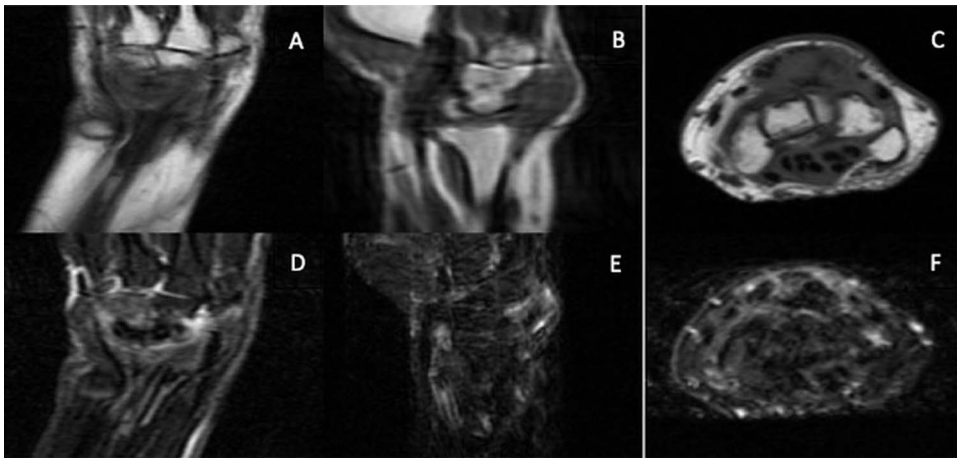


Figure 3: Right magnetic resonance imaging of the right wrist in (A, D) 'coronal,' (B, E) sagittal and (C, F) axial view revealing a dorsal wrist mass.

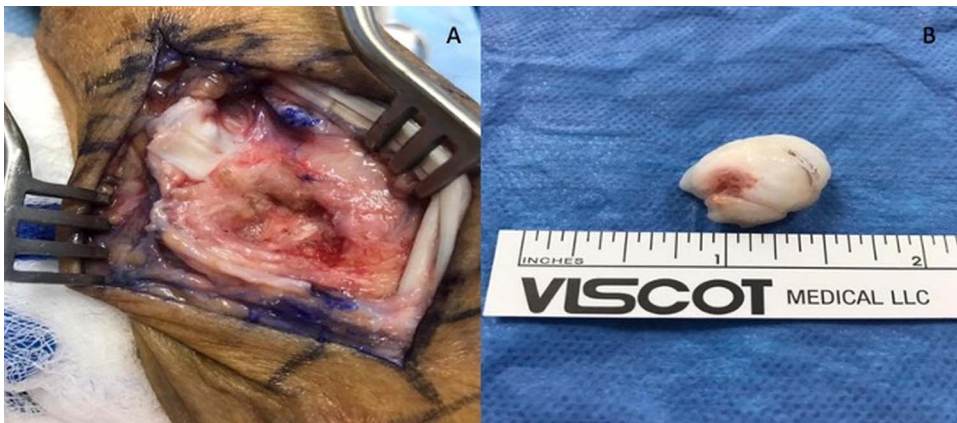


Figure 4: Intraoperative view of right dorsal wrist with (A) mass bed and bony excavation and (B) extracted calcified mass.

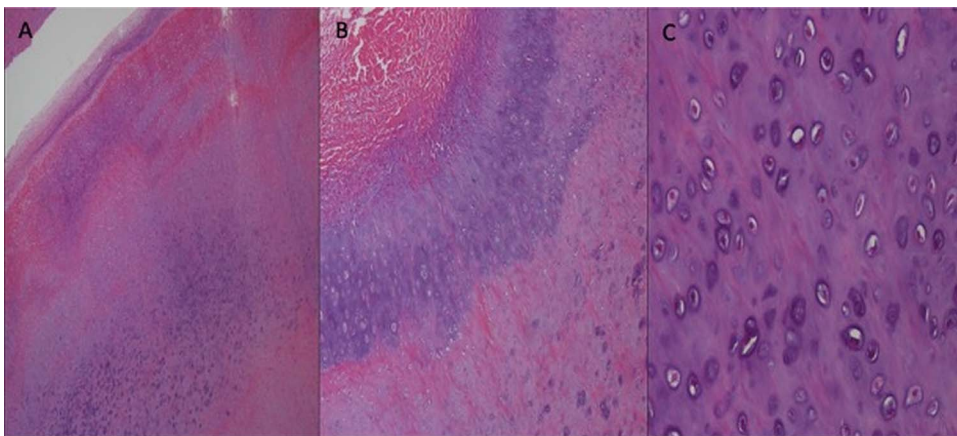


Figure 5: Histopathology of excised mass from (A) low power to (C) high power field showing a well-differentiated cartilaginous lesion of a tenosynovial chondromatosis.



Figure 6: Postop evaluation of intact wrist in (A) flexion, (B) extension, (C) supination and (D) pronation.

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CONFLICT OF INTEREST STATEMENT

None declared.

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This study did not receive any funding.

CONSENT

Written consent obtained.

GUARANTOR

Danny Mangual.

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