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Five-decade-delayed closed flexor tendon rupture due to Galeazzi dislocation fracture associated with Behçet syndrome: A case report

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ABSTRACT

INTRODUCTION: Closed flexor tendon rupture after a malunited distal radius fracture is rare and usually becomes apparent early after the fracture. Most cases are accompanied by a severe distal radio-ulnar joint capsule injury, wherein bone protrusion (as a spur) directly stresses the tendons. We experienced a nonspecific flexor tendon rupture associated with an old fracture and the presence of collagen disease.
PRESENTATION OF CASE: A 63-year-old woman presented with delayed complete rupture of the flexor digitorum profundus (FDP) of the fifth digit. Her history included closed fracture on the left wrist at age 13 years. At 27 years, she was diagnosed with Behçet syndrome and commenced oral prednisolone 10 mg/day. At the current admission, physical examination revealed that she was incapable of fifth finger flexion after minor passive extension. The fifth digit FDP rupture appeared to be due to damage at the wrist-level fracture site. A tiny capsule rupture was seen on the volar side of the distal radio-ulnar joint. We resected ulnar head osteophytes protruding from the capsule hole and transferred tendon from the fifth FDP to the fourth FDP.
CONCLUSION: Reportedly, metalloproteases weaken tendon structure by acting as a collagenase in patients with Behçet syndrome. Also, vasculitis next to a tendon and steroid intake are considered to impede the tendon repair process. Hence, even minor trauma may lead to complete tendon rupture. Although an injury seems slight, we should take into account the possible history of bone and joint trauma.

1. Introduction

Closed flexor tendon rupture, which is relatively rare, is caused mainly by direct trauma, direct attrition, inflammatory disease, or iatrogenically [1]. Among these causes, direct attrition is usually due to bone protrusion, caused by a malunited distal radius fracture. It is accompanied by severe damage to the distal radio-ulnar joint (DRUJ) capsule. Hence, tendon rupture appears early after fracture occurrence in most cases [2–4]. In contrast, when the cause is inflammatory disease, the tendon rupture is often accompanied by surrounding synovial infiltration, raising the question of whether insufficient control of the primary disease caused the rupture [1].

In rheumatoid arthritis patients, we are sometimes confronted with finger extensor tendon rupture at the level of the wrist joint who require operative treatment because of impaired recovery. Although direct repair of the tendon is generally the best choice, we usually have no other choice but tendon transfer or transplantation because of the paucity of tissue. Hence, it is still unclear how much causes, such as synovitis, influence the rupture.

Among patients with collagen diseases such as lupus, scleroderma, and Behçet syndrome, subcutaneous tendon rupture is relatively rare [5,6]. Hosokawa et al. reported spontaneous tendon rupture in lupus, but in that case nonunion of an old hamate hook fracture was the cause [7]. Cases of atraumatic Achilles tendon rupture have been reported, with long-term corticosteroid usage possibly being a risk factor [8–10]. Here, we report a case of delayed closed flexor tendon rupture in the patient with Behçet syndrome and a wrist fracture experienced five decades previously. This case report was in line with the SCARE criteria [11].

2. Presentation of the case

A 63-year-old woman visited our hospital complaining of an inability to flex the little finger of her left (dominant) hand. Her history included a closed fracture of the left wrist joint at age 13 years,

Abbreviations: BS, Behçet syndrome; CT, computed tomography; DRUJ, distal radio-ulnar joint; FDP, flexor digitorum profundus; FDS, flexor digitorum superficial; MMP, matrix metalloprotease.

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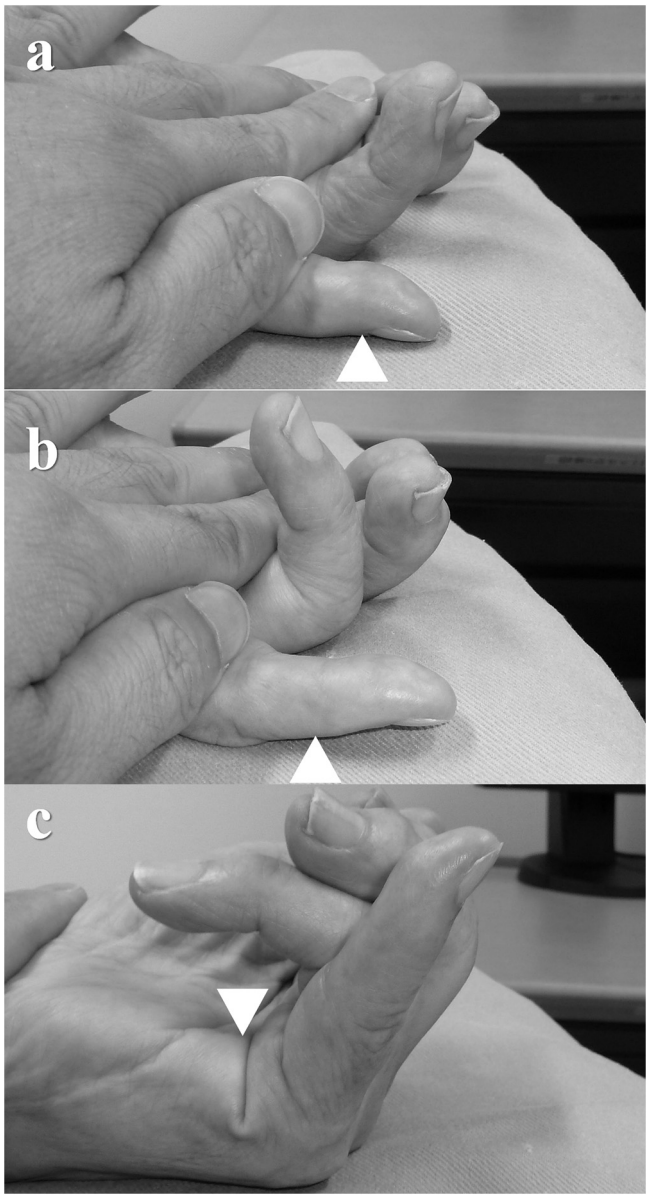

Fig. 1. Physical examination. The patient cannot flex her distal (a) or proximal (b) phalangeal joint actively because of the flexor tendon rupture. She is able to flex the metacarpophalangeal joint (c) by using intrinsic muscles.

for which she received conservative therapy with a forearm-to-hand plaster. Radiographs were not currently available for review. At age 27 years, she was diagnosed with Behçet syndrome, for which she was prescribed oral prednisolone 10 mg/day. One month prior to the visit of our unit, her left little finger had been extended passively when she tried to pick up something but could not be flexed afterward without help (Fig. 1).

Physical examination revealed erythema nodosum on her arms, legs, and trunk due to Behçet syndrome. Also, even though some finger joints were swollen due to Behçet syndrome, she did not claim any impairment of the upper extremities. She had an obvious deformity of the left wrist joint accompanied by a volar prominence of the distal end of the ulna. Additionally, there was no active flexion of the distal or proximal interphalangeal joints of her left little finger.

Radiographs of the left wrist joint exhibited an old malunion in which the distal part of radial shaft tilted 11°, and the ulnar head had dislocated toward the volar side (Fig. 2a). These findings suggested that the previous injury could have been the Galeazzi fracture dislocation occurred during childhood. As on the radiographs, computed tomography (CT) showed a dorsal tilt of the radius and DRUJ osteoarthritis with a bone spur (Fig. 2c). There were also no lesions on the carpal bones. Magnetic resonance imaging revealed that the distal stump of the flexor digitorum profundus (FDP) of the fifth digit was on the volar aspect of the fifth metacarpal diaphysis (Fig. 2b) with mild proliferation of synovial tissue around the injured wrist joint.

Based on these findings and the duration of the post-traumatic period (five decades), we diagnosed the patient as having a complete rupture of the FDP of the left little finger in zone V caused by tendon degeneration resulting from Behçet syndrome and long-term oral prednisolone intake.

Surgical treatment started with a single longitudinal incision over the volar aspect of the left wrist and longitudinal incision of the transverse carpal ligament. The fifth FDP was identified and found to be completely ruptured in zone V, and its flexor digitorum superficial (FDS) was denatured (Fig. 3a). The fourth FDP was somewhat frayed but maintained its continuity. The distal stump of the FDP of the fifth digit was accompanied by moderate synovitis. The proximal stamp was not found within the width of the skin incision (Fig. 3b). Contrary to our preoperative inspection, there was pinhole-size discontinuity of the DRUJ capsule and no more than mild spur protrusion at the discontinuation of the capsule (Fig. 3c).

Because the body of the ulnar head itself did not have contact with the flexor tendons during forearm supination, we decided that it would be sufficient to proceed with osteophyte resection and DRUJ capsule repair, thereby avoiding further mechanical stress to the tendons. While carrying out that surgery, we found only slight damage to the fourth FDP. Hence, the fifth FDP was transferred to the fourth FDP at the level of metacarpophalangeal joint using interlacing sutures (the fourth and fifth A1 pulleys were incised longitudinally) (Fig. 3d).

Due to incision of the transverse ligament, the repaired tendon was on the tip of the hamate hook because of distal radius angulation. We therefore decided to perform a hamate hook resection as well, in view of possible rupture of other tendons. The flexor retinaculum was not repaired, and the wound was closed in layers. The dorsal splint was applied postoperatively, aiming to keep the wrist and metacarpophalangeal joint flexed.

Postoperatively, only passive flexion was allowed for the fifth digit during the first 3 weeks after surgery. Afterward, active flexion was started in the wrist flexion position. The dorsal splint was removed at the beginning of the fifth week, when active finger flexion in dorsiflexed position at the wrist was also allowed.

At the final 12-month follow-up, the patient showed improved motion and function. She could now flex the proximal interphalangeal joint 80° and the distal interphalangeal joint 70°. She was thus satisfied with the surgery. Regarding activities of daily living, she could perform light work, such as folding paper, knitting a sweater, or cutting food (Fig. 4).

3. Discussion

Closed flexor tendon rupture due to malunited distal radius fracture is usually associated with severe DRUJ capsule disruption and becomes apparent a short time after the original trauma (except in one report) [4,12]. In such cases, it has been assumed that direct mechanical stress due to protrusion of the ulnar head causes the tendon's rupture. In our case, five decades had passed since the primary trauma, and there was only a pinhole-like discontinuity of the DRUJ capsule. Therefore, we considered that protrusion of ulnar head did not independently cause tendon rupture.

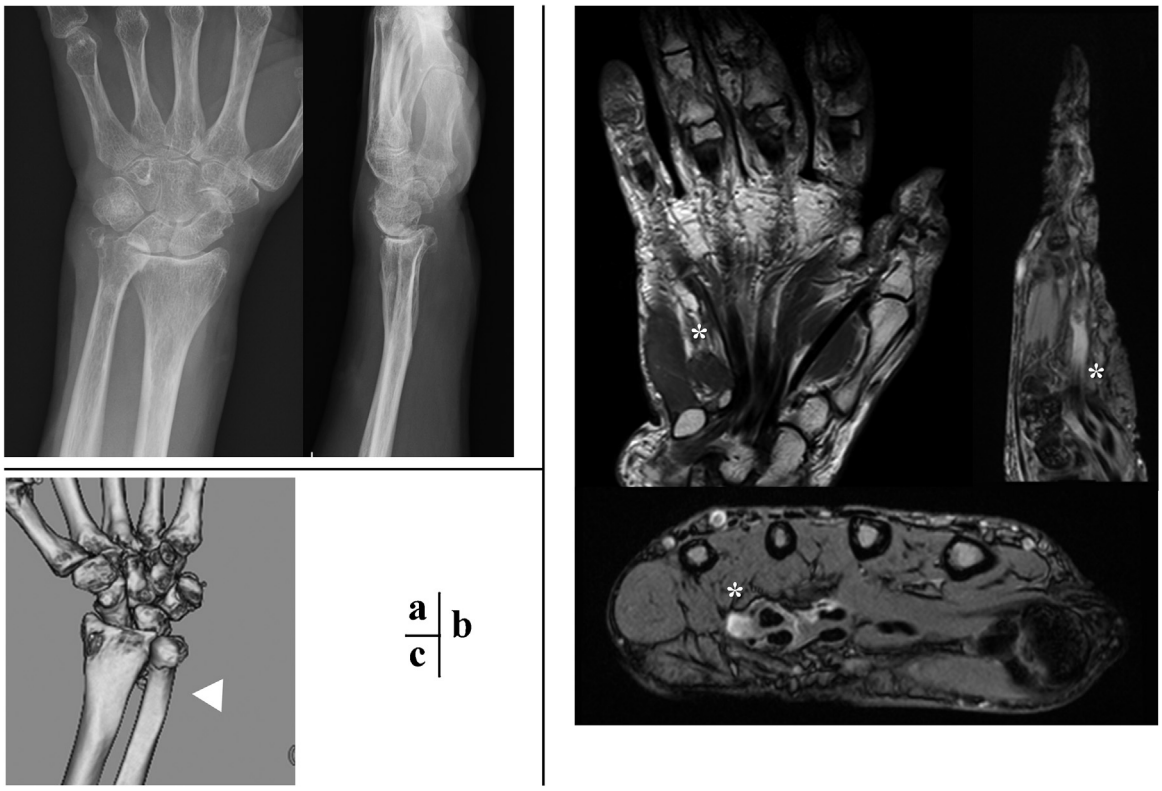

Fig. 2. Clinical examination. Dorsal tilt at the distal radius and volar dislocation of the ulnar head are seen on plain radiography (a) and computed tomography (arrowhead) (c). (b) Magnetic resonance image indicates discontinuity of the flexor tendon of the fifth finger (*).

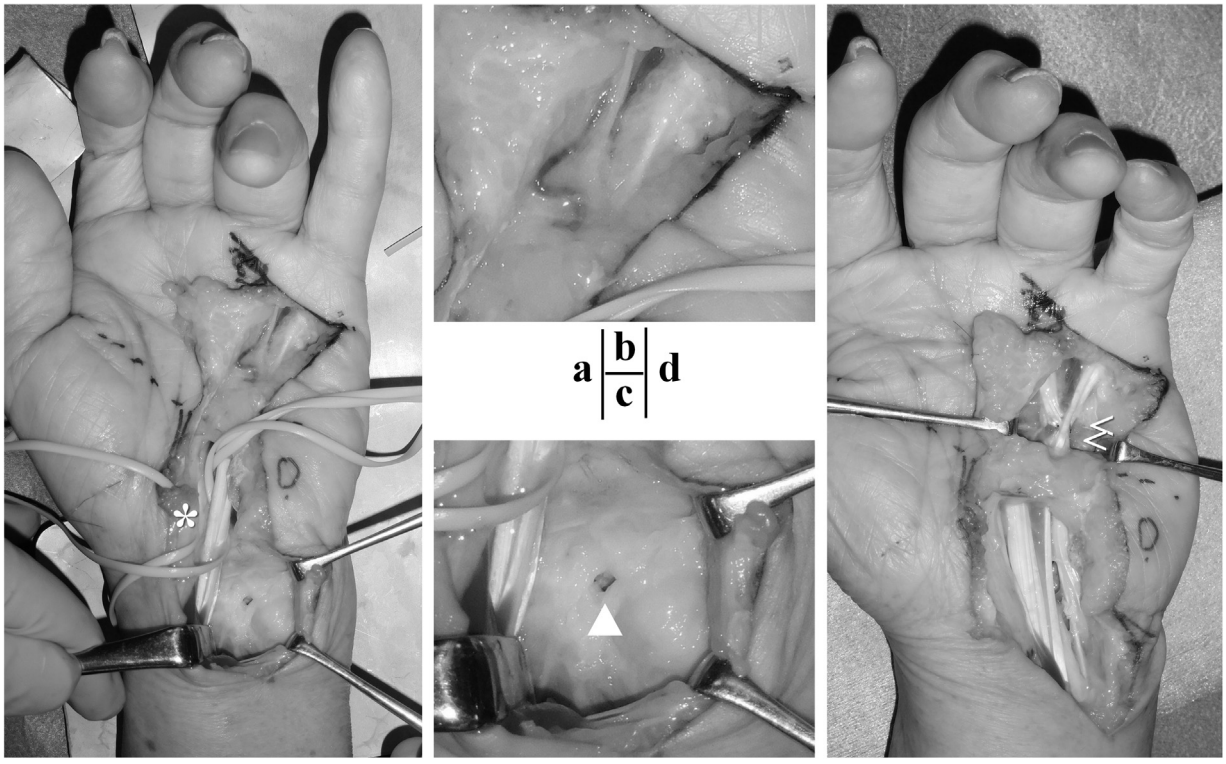

Fig. 3. Intraoperative findings. (a) The fourth flexor tendon was not damaged (*), but the fifth tendon was not seen in the carpal tunnel. (b) The distal stump of the fifth flexor tendon (profundus) had degenerated and was involved with synovial tissue. (c) There is a pinhole-like discontinuity at the volar aspect of the joint capsule (arrowhead). (d). We transferred the distal stump of the fifth flexor tendon to the fourth flexor tendon (<).

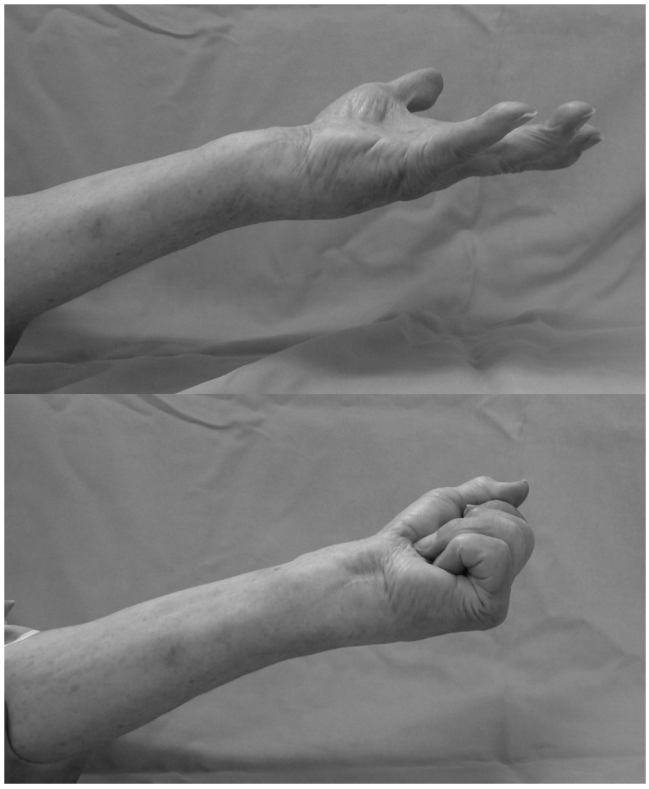

Fig. 4. At the 12-month (final) follow-up, the patient actively achieved full flexion.

In collagen diseases (e.g., Behçet syndrome), tissue repair could be inhibited by corticosteroid usage. Even idiopathic Achilles tendon rupture has been reported [8]. We also noted that corticosteroid ingestion affects capillary blood flow around tendons. In our case, this mechanism could have been related to the ruptured tendon.

Histopathologically, there was a non-specific infiltration of inflammatory cells in the tendon stump. However, we found intra-articular synovial proliferation in the DRUJ and radiocarpal joint during surgery, so we considered the influence of matrix metalloproteases (MMPs) in the joint fluid. It has been reported that MMP-1, MMP-8, and MMP-13 degrade type I collagen, which is abundant in skin tissue, bones, and tendons [13]. It has been also reported that MMP-8, which is expressed in synovial fibroblasts, is related to idiopathic tendon rupture in rheumatoid arthritis patients. MMP-2, MMP-3, and MMP-9 are not believed to act as collagenase of type I collagen [14,15].

The serum MMP-9 level is reported to be increased in patients with Behçet syndrome [14]. Even though MMP-9 does not degrade type I collagen by itself, it can exacerbate soft tissue damage—such as tendon fibrillation or capsule pinhole in our case—because it enhances collagenase (MMP-1,MMP-8, MMP-13) activity [16].

Thus, in our case, at first there was chronic irritation of the volar capsule of wrist joint by the malunited bones. Next, following the onset of Behçet syndrome and oral corticosteroid intake, the joint capsule started to degenerate and become thinner, resulting in a perforation. Then, joint fluid containing MMPs leaked into and around the tenosynovium. The MMPs (as collagenase) and direct irritation from the ulnar head spur encouraged degeneration of the flexor tendons. Finally, although creating only a small external force on the fifth finger, this degenerated flexor tendon was disrupted five decades after the initial trauma.

4. Conclusion

We report a case of flexor tendon rupture that occurred after both malunion of a Galeazzi dislocation fracture and the fragility of soft tissue due to collagen disease. At first, we thought that the malunion had no impact on the tendon rupture, but it has been proven that even a tiny bone lesion may affect the joint capsule when some conditions interact. Thus, when encountering a case of spontaneous tendon rupture in a patient with collagen disease or known oral steroid usage, we should pay attention to the patient's traumatic history regarding bones and joints, even though the patient has no obvious complaint—even if it goes back five decades.

Conflicts of interest

This research did not have any conflict of interests.

Sources of funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Ethical approval

This case report was in line with the Declaration of Helsinki 2013 and was line with institutional acceptable practice (approval No: ERB-C-818).

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Masato Ohara: write this paper.
Shogo Toyama: write this paper in part, surgeon of this case.
Ryo Oda: surgeon in chief.
Yusei Katsuyama: data collection.
Hiroyoshi Fujiwara: interpretation, chief of upper limb surgery.
Toshikazu Kubo: total management.

Registration of research studies

UIN 3702.

Guarantor

Ryo Oda MD. PhD.

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