Mod Rheumatol (2006) 16:324–326
DOI 10.1007/s10165-006-0508-0

CASE REPORT

Kaoru Murakoshi · Shigeki Momohara · Katsunori Ikari
Takuji Iwamoto · Kouichirou Kawamura
Taisuke Tomatsu · Noriko Iikuni · Hiroshi Okamoto

Neglected spontaneous rupture of the Achilles' tendon in patients with systemic lupus erythematosus

Received: February 2, 2006 / Accepted: June 23, 2006

Abstract Spontaneous Achilles' tendon rupture associated with systemic lupus erythematosus (SLE) is rare complication in literature review. We encountered two patients with neglected spontaneous ruptures of Achilles' tendons who had been on corticosteroid therapy to treat SLE. The ages of these patients were 43 and 49 years, and both were women. One of them was a case of bilateral Achilles' tendons rerupture. Achilles' tendons of both patients were reconstructed by surgery because of delay in their diagnosis. Histological section of the both ruptured Achilles' tendon revealed fibrotic scar tissue and little existence of inflammatory change. We concluded that careful diagnosis, surgical suture, and careful treatment after operation are necessary for Achilles' tendon rupture in those patients with SLE.

Key words Achilles' tendon · Corticosteroid · Spontaneous rupture · Systemic lupus erythematosus (SLE)

Introduction

Spontaneous Achilles' tendon rupture associated with systemic lupus erythematosus (SLE) is rare complication in the English and Japanese literature, but the etiology is not well understood. Furie and Chartash reported that tendon rupture in SLE was a rare but potentially disabling complication.[1] Khan and Ballou reported spontaneous rupture of Achilles' tendon in a patient with SLE, and noted it as a late complication of SLE, more likely to occur in older patients.[2] Furthermore, Prasad et al. reported an SLE case of spontaneous, bilateral patellar tendon rupture in a woman with previous Achilles' tendon rupture.[3]

The etiology of Achilles' tendon rupture in SLE is not well understood. In some cases it is secondary to trauma and in others it is related to inflammatory changes in and/or around the tendon as a result of the underlying disease process.[1] Many important issues still remain unanswered concerning the cause, pathogenesis, diagnosis, and management of the Achilles' tendon disorders in SLE. Corticosteroid therapy may be responsible for Achilles' tendon rupture.

Here we report two patients with SLE who had neglected spontaneous rupture of the Achilles' tendons. Moreover, one had injury to both Achilles' tendons. None of our cases experienced any major trauma. Furthermore, they complained little of any symptoms in their ankles for a while after the rupture, and the lack of severe symptoms prevented the early diagnosis of Achilles' tendon rupture. In this article, we report clinical features and management of neglected spontaneous rupture of Achilles' tendon in our two SLE patients.

Case reports

Case 1

Neglected spontaneous, rerupture of bilateral Achilles' tendon occurred in a 43-year-old woman with an 8-year history of SLE. She was treated with 40 mg/day prednisolone (PSL) from the onset of the disease. However, because disease activity remained high, steroid pulse therapy was also performed once, and therapy using selective adsorption of anti-DNA antibody was performed three times for the treatment of lupus nephritis. Ten days after the steroid pulse therapy, she fell down on the ground and afterwards felt a light pain around her left Achilles' tendon on walking. However, she was able to walk by herself even with some pain. Around that time, she had complained of worsening pain especially in the bilateral wrists, knees, and ankles. Therefore, she

K. Murakoshi · S. Momohara (✉) · K. Ikari · T. Iwamoto · K. Kawamura · T. Tomatsu
Department of Orthopedic Surgery, Institute of Rheumatology, Tokyo Women's Medical University, 10-22 Kawada-cho, Shinjuku-ku, Tokyo 162-0054, Japan
Tel. +81-3-5269-1711; Fax +81-3-5269-1726
e-mail: smomohara@ior.twmu.ac.jp

N. Iikuni · H. Okamoto
Department of Internal Medicine, Institute of Rheumatology, Tokyo Women's Medical University, Tokyo, Japan

noticed that pain in her left ankle was caused by her disease activity. At this time the total amount of PSL was 33 820 mg. Three weeks after the injury, she still felt a slight disturbance around her left ankle joint, and decided to come to our department. Rupture of the Achilles' tendon was diagnosed on palpation and an observation of a positive Thompson's squeeze test. Surgical suture for tendon was performed, because 3 weeks had already passed after the injury. There were no definite degenerative changes in the Achilles' tendon macroscopically, and the tendon was simply sutured with a Bunnel procedure.

When she stood up from bed on day 10 after the operation, she felt pain around her contralateral ankle joint. She was immediately diagnosed with rupture of the right Achilles' tendon and was treated conservatively, using casting with steroid dose reduction.

Afterwards the condition of both Achilles' tendons improved, and she was able to walk on her own some weeks later. However, her left Achilles' tendon reruptured because she fell down 15 weeks after the first operation. In addition, 2 weeks later her right Achilles' tendon also reruptured when she stood up. Surgical repair was performed on both Achilles' tendons. There were definite degenerative changes at the end of the Achilles' tendon macroscopically (Fig. 1A). The tendon was sutured with the modified Lindholm procedure,[4] and reinforced at the site of the rupture with the Turco procedure using the peroneus brevis tendon for augmentation.[5]

Histological section of both ends of the reruptured Achilles' tendon revealed fibrotic scar tissue and little existence of inflammatory change (Fig. 1B). The patient returned to regular activities after discharge and made a complete recovery.

Case 2

Spontaneous left Achilles' tendon injury occurred in a 49-year-old woman with a 10-year history of SLE. She treated with 20 mg daily PSL at the onset of the disease, PSL was continued at a dose from 2.5–10 mg per day according to disease activity for some years. However, there was a recurrence of disease activity in September 1998, and steroid pulse therapy followed by a dose of PSL 20–30 mg per day was given for the treatment of lupus nephritis. She felt pain and had limited range of motion in her left ankle. At this time, she had a complaint of just slight pain in her other joints, e.g., fingers and wrists. She was able to walk and was diagnosed with Achilles' tendonitis on October 19. At the end of November she sprained her left ankle again. Therefore, she was referred to our department complaining of left ankle pain on December 14. The patient had difficulty walking alone and had plantar flexion of the left ankle. On physical examination, a defect in her Achilles' tendon was palpated and the Thompson test was positive. From these findings, she was diagnosed as having left Achilles' tendon rupture.

Surgical suture for tendon was performed, because 2 months had already passed after the initial injury. There was definite degenerative change at both ends of the Achilles' tendon macroscopically (Fig. 2A). The tendon itself was sutured with the Kirchmayer procedure, and the rupture site was repaired with the modified Lindholm procedure[4] and reinforced with the Turco procedure.[5] Histological section of the ends of the Achilles' tendon revealed fibrotic scar tissue as seen in Case 1 (Fig. 2B). After the operation, she returned to regular activities and made a full recovery.

Discussion

A complete rupture of the Achilles' tendon usually occurs in sports that require jumping, running, and quick turns.[6] However, these two patients we encountered with SLE had no or little trauma. Moreover, they thought that the pain was due to arthritis of their ankle joints, and they were able to walk for a while even after injury. Therefore, there may be specific causes of ruptured Achilles' tendons in SLE and there may be a possibility of pre-degenerative changes before the incidence in the Achilles' tendons. In the case of rheumatoid arthritis (RA), there are some reports of spon-

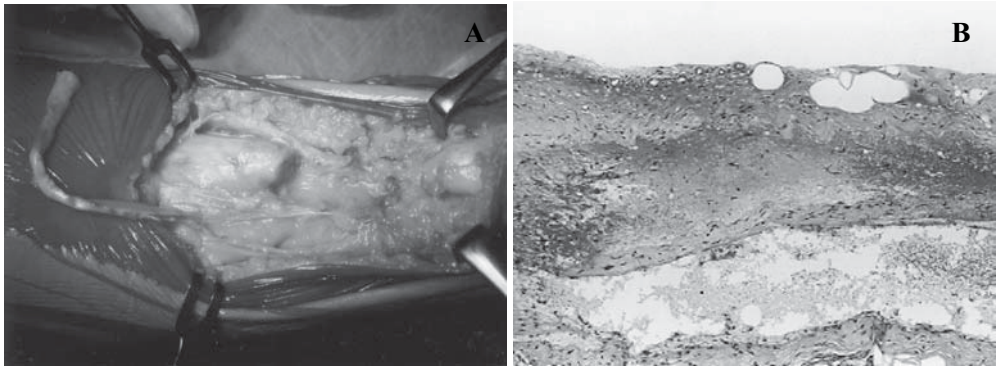

Fig. 1A,B. Case 1. **A** Macroscopic findings at reoperation. *Left side* (suture side previously): There was a large gap between the end of rupture of Achilles' tendon. There was definite degenerative change macroscopically in operation. Tendon was sutured with the modified Lindholm procedure,[5] and the site of rupture was reinforced with the Turco procedure using the peroneus brevis tendon for augmentation.[6] **B** Histological section of the end of the both reruptured Achilles' tendon revealed only fibrotic scar tissue, and little existence of inflammatory change (Hematoxylin and eosin, ×40)

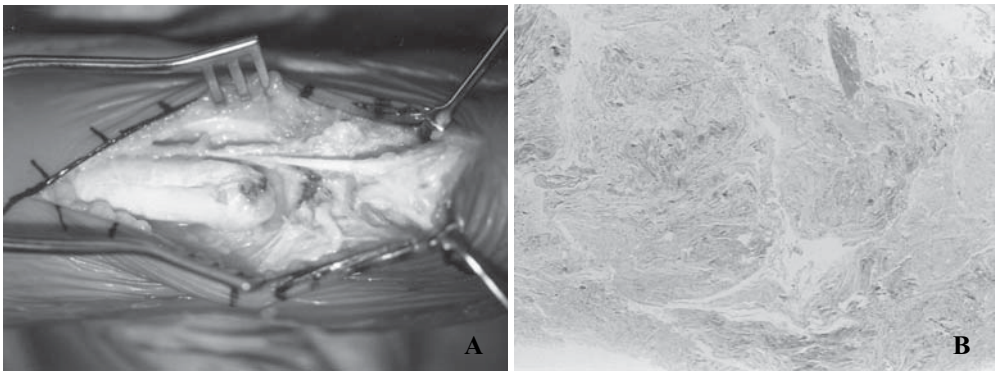

Fig. 2A,B. Case 2. **A** Macroscopic findings during operation. There was definite degenerative change at the end of Achilles' tendon observed macroscopically. Therefore, the tendon itself was sutured with the Kirchmayer procedure, the rupture site repaired using the modified Lindholm procedure,[5] and reinforced with the Turco procedure using the peroneus brevis tendon.[6] **B** Histological section of the end of the Achilles' tendon revealed fibrotic scar tissue as for Case 1 (Hematoxylin and eosin, ×40)

Table 1. Neglected spontaneous rupture of Achilles' tendon in two patients with systemic lupus erythematosus

	Case 1	Case 2
Age (years)	43	49
Disease duration (years)	8	10
Duration from onset of symptoms to diagnosis for Achilles' tendon rupture	3 weeks	2 months
History of oral steroid (years)	8	10
Total amount of steroid at injury (mg)	33 820	10 135
Daily amount of steroid at injury (mg/day)	40	20
Steroid pulse therapy	One time	One time

taneous ruptured Achilles' tendon. Matsumoto et al. reported that histological section of the ruptured Achilles' tendon revealed the existence of rheumatoid granulation that consisted of lymphocytes, histiocytes, and small vascular proliferation within the tendon tissue in RA.[7] In both of our cases, histological section of the ruptured Achilles' tendon revealed only fibrotic scar tissue and little existence of inflammatory change. This may cause only minor symptoms, leading to the delay of diagnosis for Achilles' tendon rupture. In cases of no succinct traumatic event, as in our patients, symptoms such as decreased range of motion on the affected joints may occur weeks or up to months after the rupture. Therefore, the delay of the diagnosis was due to the relatively long course from the rupture to the appearance of the symptoms.

Steroid therapy for SLE may also be responsible for Achilles' tendon rupture. Hersh and Heath reported that spontaneous Achilles' tendon rupture associated with long-term oral steroid use is not uncommon, particularly in older patients who use these drugs daily to treat systemic diseases.[8] In other cases, Achilles' tendon rupture was reported to be associated with local steroid use.[8,9] Taken together, the cause of spontaneous rupture of Achilles' tendon may be due to the pathology of SLE or steroid, or both. In this clinical setting, we think that minor injury may trigger Achilles' tendon rupture.

In our cases, we undertook to reconstruct the neglected ruptured Achilles' tendon with the modified Lindholm procedure,[4] and reinforced this with the Turco procedure.[5] Neglected Achilles' tendon ruptures are debilitating injuries, and the optimal management is surgical procedure. We performed repair of these neglected spontaneous Achilles' tendon ruptures using a combination of peroneus brevis transfer and plantaris tendon augmentation. The management by peroneus tendon transfer is safe but technically demanding. Therefore, it afforded good recovery, even in SLE patients with a long-term neglected rupture as in our cases.

In conclusion, spontaneous Achilles' tendon rupture associated with SLE is very rare. In clinical features they complained little of symptoms in their ankles for some time even after the rupture because of their arthritis. Therefore, the diagnosis may be delayed and rupture of the Achilles' tendon might be neglected. We recommend that careful diagnosis, surgical suture, and careful treatment after operation are necessary for Achilles' tendon rupture in patients with SLE.

References

1. Furie RA, Chartash EK. Tendon rupture in systemic lupus erythematosus. Semin Arthritis Rheum 1988;18(2):127–33.
2. Khan MA, Ballou SP. Tendon rupture in systemic lupus erythematosus. J Rheumatol 1981;8(2):308–10.
3. Prasad S, Lee A, Clarnette R, Faull R. Spontaneous, bilateral patellar tendon rupture in a woman with previous Achilles tendon rupture and systemic lupus erythematosus. Rheumatology (Oxf) 2003;42(7):905–6.
4. Kirschenbaum SE, Kelman C. Modification of the Lindholm procedure for plastic repair of ruptured Achilles tendon: a case report. J Foot Surg 1980;19(1):4–11.
5. Turco VJ, Spinella AJ. Achilles tendon ruptures–peroneus brevis transfer. Foot Ankle 1987;7(4):253–9.
6. Jarvinen TA, Kannus P, Paavola M, Jarvinen TL, Jozsa L, Jarvinen M. Achilles tendon injuries. Curr Opin Rheumatol 2001;13(2):150–5.
7. Matsumoto K, Hukuda S, Nishioka J, Asajima S. Rupture of the Achilles tendon in rheumatoid arthritis with histologic evidence of enthesitis. A case report. Clin Orthop Relat Res 1992;280:235–40.
8. Hersh BL, Heath NS. Achilles tendon rupture as a result of oral steroid therapy. J Am Podiatr Med Assoc. 2002;92(6):355–8.
9. Bedi SS, Ellis W. Spontaneous rupture of the calcaneal tendon in rheumatoid arthritis after local steroid injection. Ann Rheum Dis. 1970;29(5):494–5.