



Isolated tuberculous tenosynovitis of the Achilles tendon: a report of two cases

Aşil tendonunda izole tüberküloz tenosinovit: İki olgu sunumu

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Tüberküloza bağlı tenosinovitin ayak tendonlarında görülmesi oldukça nadirdir. Bu çalışmada Aşil tendonunda yerleşim gösteren izole tüberküloz tenosinoviti olan iki olgu sunuldu. Olgulardan biri 19 yaşında kadın hasta idi; sol Aşil tendonu üzerinde şişlik ve ağrı yakınması vardı. Bu olguda komplet eksizyon ve antitüberküloz kemoterapi hastalığın iyileşmesi için yeterli oldu. Ameliyat sonrası dönemde altı aylık tıbbi tedavi sonunda hastanın manyetik rezonans görüntüleme (MRG) sonuçları normal bulundu, 27. aydaki kontrol MRG'de nüks görülmedi. İkinci olgu aynı zamanda romatoid artrit ve diyabet hastası olup yoğun sitostatik ajan kullanan 53 yaşında bir kadındı. Yakınmaları çömelmede, merdiven çıkmada zorluk, sağ ayak bileğinde şişlik ve ağrı idi. Hastada apse drenajı ve kistik kitle eksizyonu yapıldı. Antitüberküloz kemoterapi sonrası takiplerde ayak bileği bölgesinde hastalığa ait bulgu saptanmamış olmasına rağmen hasta miliyer tüberküloz nedeniyle altıncı ayda kaybedildi. Ayağın arkasında geçmeyen ağrı ve şişlik bulunan hastaların ayırıcı tanısında tüberküloz tenosinovit de akılda bulundurulmalıdır.

Anahtar sözcükler: Aşil tendonu/patoloji; tenosinovit/etyoloji; tüberküloz, osteoartiküler/patoloji.

Tuberculous tenosynovitis involving the tendons of the feet is very rare. Isolated primary tuberculous tenosynovitis of the Achilles tendon was detected in two women aged 19 and 53 years, respectively. The younger patient had a swollen and painful Achilles tendon in the left foot. Complete excision of the lesion followed by antituberculous chemotherapy for six months resulted in complete improvement. Magnetic resonance imaging showed normal findings at the end of six months and no recurrence after 27 months of follow-up. The older patient had diabetes and was on cytostatic treatment for rheumatoid arthritis. She had difficulty in squatting and climbing stairs due to swelling and pain in the right ankle. She underwent abscess drainage and excision of the cystic mass. Despite disappearance of symptoms in the affected ankle in the course of antituberculous chemotherapy, she died due to miliary tuberculosis in the sixth month. Tuberculous tenosynovitis should be considered in the differential diagnosis of patients suffering from persistent swelling and pain in the hind foot.

Key words: Achilles tendon/pathology; tenosynovitis/etiology; tuberculosis, osteoarticular/pathology.

Tuberculous tenosynovitis, although rare, is an established condition, with the majority of cases involving the hands and wrists.^[1,2] Even rarer are localizations on the tendons of the feet; very few cases have been reported in the literature.^[1,4,9,11] We present two cases of isolated tuberculous tenosynovitis of the achilles tendon, which has been reported on only three separate occasions before in the English literature.^[4,9]

Case report

Case 1– A 19-year-old female presented in April 2002 complaining of swelling over the left achilles tendon for 5 months. There was neither a history of trauma to the foot, nor any recent illness, fever or weight loss. Walking and climbing upstairs resulted in discomfort and irritation in her left hindfoot. Physical examination revealed mild limping during walking. Active ankle motion was limited because of the



Figure 1. Magnetic resonance imaging of the left ankle. **(a)** Sagittal T_2 -weighted image demonstrating a cystic collection on the achilles tendon, with lobulated contours and without evidence of osseous extension. **(b)** Axial T_2 -weighted image, near to the ankle level, revealing the achilles tendon surrounded by the cystic collection anteromedially.

tenderness, while there was no limitation passively. There was a subcutaneous, 2x3x5 cm cystic swelling and mild tenderness along the course of the achilles tendon. Tenderness was worse upon dorsiflexion of the ankle. The mass had become larger progressively over the 5 months. It was soft in consistency, immobile over the achilles tendon posteromedially, and without any changes in the overlying skin. One month prior to presentation, an aspiration biopsy of the cystic mass, performed in another orthopaedic center, revealed many leukocytes by Gram stain. The EZN (Ehrlich-Ziehl-Neelson) stain did not show any acid resistant bacteria. No growth was observed in aerobic and anaerobic culture media.

Magnetic resonance imaging (MRI) of the left ankle revealed a 2.8x3x5 cm cystic mass with lobulated contours, dissecting the distal portion of the soleus muscle, surrounding the achilles tendon posteromedially that was hypointense on T_1 and hyperintense on T_2 sequences (Fig. 1). In the proximity of the cyst, at the medial and lateral aspect of the ankle, hypointense signals on T_1 and hyperintense signals on T_2 sequences were related to edema in the subcutaneous tissues and observed linear collections were considered as post-injection changes. At the distal level, near

to the ankle, achilles tendon was surrounded by the cyst anteromedially (Fig. 2). The plain radiograph of the ankle was normal. The patient was born in Turkey and had no known history of tuberculosis, but one year prior to her presentation, her father completed nine-month-long medical therapy for pulmonary tuberculosis. The plain chest radiograph of the patient revealed a normal cardiothoracic index, a normal mediastinum, and patent costophrenic sinuses and thorax CT was also normal. Routine blood investigations did not reveal any abnormality, except mild monocytosis. Erythrocyte sedimentation rate was 3mm, 9 mm, and 25 mm at half an hour, one hour, and two hours, respectively. The preoperative CRP was under 3.1 mg/L. The total blood count was as follows: White Blood Cell: 4800/mm³, Lymphocyte: 26.8%, Monocyte: 15.7%, and Granulocyte: 55.9%. The result of the PPD test was 13 mm which was considered positive for tuberculosis. Excisional biopsy of the lesion was planned. At operation, a vertical 20-cm incision medial to the achilles tendon was carried out under general anesthesia, using a tourniquet. The multilobulated, dark brown cystic mass was posteromedially to the achilles tendon, surrounding it circumferentially and extending 5 cm proximal to its distal calcaneal

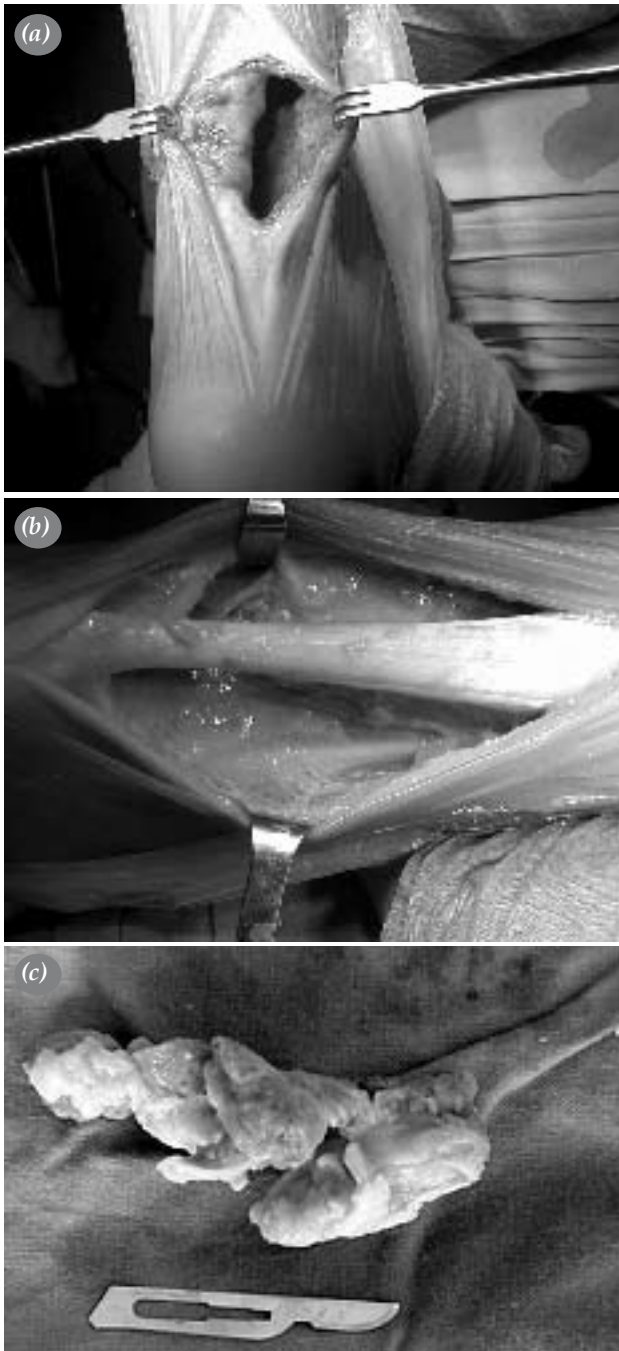


Figure 2.(a) The cystic cavity. After aspiration of the cyst, its fibrotic and thick wall was found to be adherent to the inflamed sheath of the achilles tendon, which also was adherent to the subcutaneous tissue. (b) After complete excision of the cyst, and debridement of the involved soft tissues. (c) Operative specimen of the cyst-wall.

insertion, spreading the distal portion of the soleus muscle apart. Its wall was adherent to the inflamed sheath of the achilles tendon, which was adherent to the subcutaneous tissue (Fig. 3). The aspiration of the cyst revealed a yellow-brown fluid. Anterolateral to

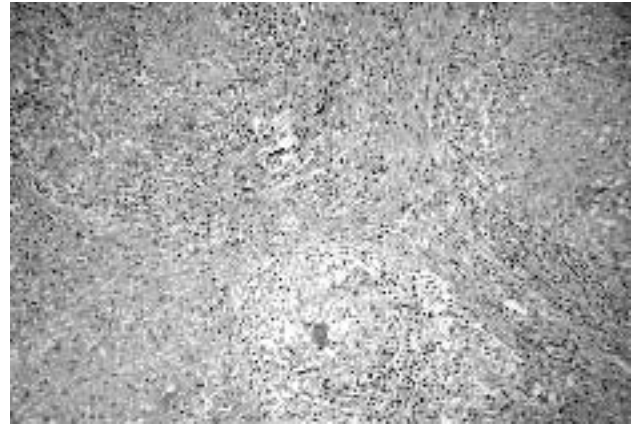


Figure 3. Histologic section: Multiple granulomas with accompaniment of focal necrosis area. (H-E x 200)

the tendon, the wall of the cyst was obviously thickened. The cyst was completely excised without damaging the achilles tendon, and the involved soft tissues were debrided (Fig. 4). The wound was closed over



Figure 4.(a) Clinical appearance two years postoperatively. (b) MRI at 27 months postoperatively reveals normal findings..

a hemo-wac drain, which was removed on the first postoperative day. Operative specimens (Fig. 5) were sent for histological examination, and for aerobic, anaerobic, and fungal cultures, acid-fast bacilli and gram staining. After the operation, the leg and foot were immobilized in a below-knee splint for two weeks. Full weight bearing began three weeks after the operation. Wound detachment complication, seen in early postoperative course, was successfully treated with localized wound care and daily dressings. The microbiological examination was found negative; no bacteria were detected by the gram stain, and only scant amounts of white blood cells were found. No growth was observed in Lowenstein-Jensen culture media. On histopathologic examination, necrotizing granulomatous inflammation was observed (Fig. 6). The presumptive diagnosis was tuberculosis, but the EZN stain failed to show any acid-fast bacteria. After the first postoperative week, the patient received daily antituberculous chemotherapy that consisted of isoniazid (INH) (300 milligrams/day), rifampicine (600 milligrams/day), pyrazinamide (1500 milligrams/day), and ethambutol (1000 milligrams/day). The liver function tests performed on the 21st postoperative day were normal. Ethambutol and pyrazinamide were terminated after the second month of the therapy. The patient had a follow-up examination every three months for twenty-seven months (Fig. 7) In the 6th month of medical treatment the MRI of the patient showed



Figure 5. Preoperative view of the patient showing swelling of the right ankle

normal findings, the patient was freed from tuberculosis and the medical treatment was terminated. In the postoperative 27th month the MRI showed no recurrence (Fig. 8).

Case 2– The second case was a 53 years old female and had complaint of pain and swelling at her right ankle, those were worsened in the last 3 months of a period (Fig. 9). Night sweats, weight loss were other persistent constitutional symptoms. Erythema

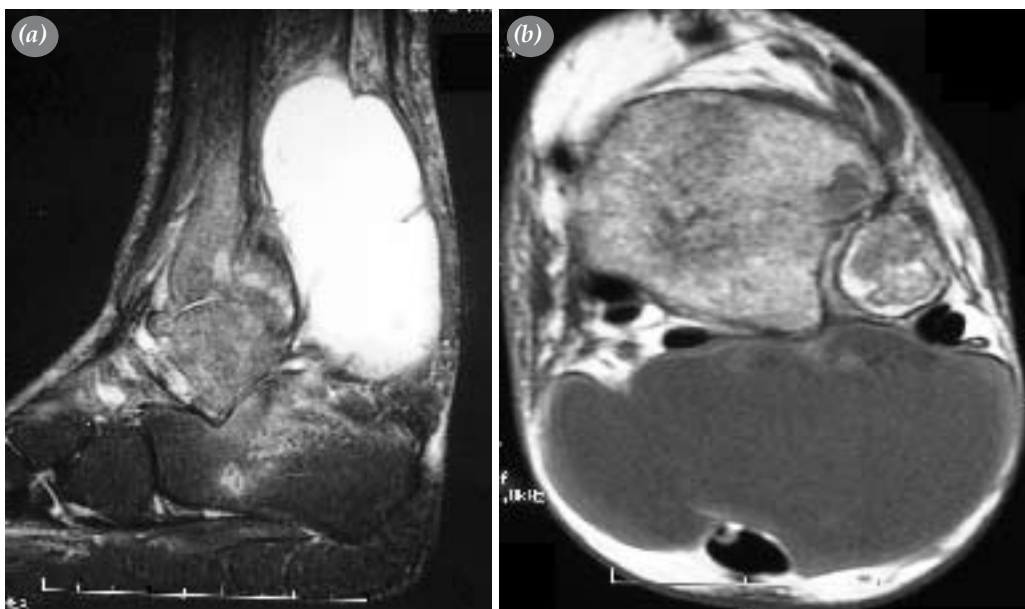


Figure 6. Magnetic resonance imaging of the ankle. (a) Sagittal T_2 -weighted image demonstrating a cystic collection on the achilles tendon. (b) Axial T_2 -weighted image, near to the ankle level, revealing the achilles tendon surrounded by the cystic collection.

was seen two or three times, but heat didn't noticed in this area. She didn't remember any trauma or any other wound. She was diabetic and suffering from rheumatoid arthritis since 15 years and was under remission with methotrexate and steroid treatment. Aspiration and tru-cut biopsy was performed on her medial malleoli at once in an other center. Acid resistant bacteria was seen in microscobic investigation. She was referred to our clinic at 14.11.2005 for further investigation with complaint of discomfort in walking, squatting and climbing upstairs and irritation in her right hindfoot. Antialgic limping was to identify during walking, range of motion was limited because of the tenderness, however there was no limitation passively. Subcutaneous, 7x7x4 cm fluctating swelling, soft in consistency, immobile over the achilles tendon posteromedially, was palpated and tenderness along the course of the achilles tendon was able to provoke with dorsiflexion of the ankle. Magnetic resonance imaging (MRI) of the right ankle revealed 7x7x4cm loculated cystic lesion with signs of inflammation and bursitis. The mass was extending from retrocalcaneal bursae superiorly to Achilles tendon and to the anterior portion of it (Figs. 10-11). The plain chest radiograph, routine blood investigations, tuberculin tests were not specific for tuberculosis.

Surgical treatment was begun with ankle joint arthroscopy and drainage of the so called cold abscess was also performed arthroscopically on extra-articular way of access. The rest of the operative procedure was as the same before, the multilobulated, dark brown cystic mass was seen posteromedial to the achilles tendon, surrounding it circumferentially and close to its distal calcaneal insertion. The inflamed sheath of the achilles tendon was adherent to the tendon, and to the subcutaneous tissue. Proper debridement was performed (Fig: 12). After the third week of immobilization full weight bearing was allowed. Tissue and fluid samples were collected for pathologic and microbiologic investigations. The microbiological examination was found to be positive, gram positive bacterias were detected with great amount of amount of white blood cells and also growth was observed in Lowenstein-Jensen culture media. On histopathologic examination, necrotizing granulomatous inflammation was observed (Figs. 13-14). The presumptive diagnosis was tuberculosis. At the first postoperative week, the patient received daily antituberculous chemotherapy regimen same with



Figure 7. Operative view of the cystic cavity.

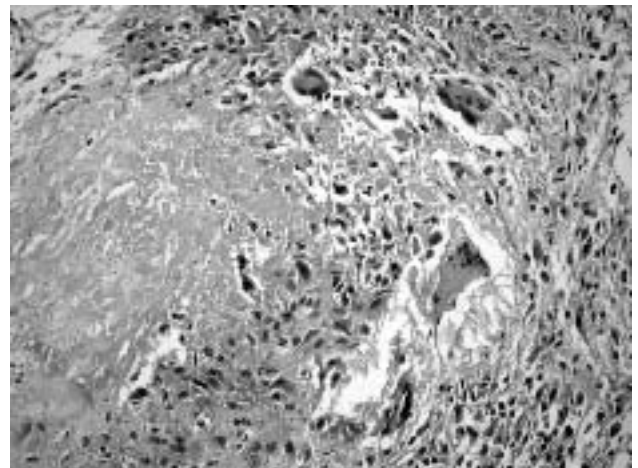


Figure 8. Histologic section: Granulomas with necrotizing multinuclear giant cells in tenosynovial tissue. (H-E x 200)

the other patient (see above). Despite to regression of local symptoms at the ankle, she has died due to miliary tuberculosis and its systemic complications at the 6th month of follow-up time.

Discussion

After the introduction of antituberculous chemotherapy, the prevalence of tuberculosis in Europe and North America declined considerably¹. However, after a period of marked decline of the disease, especially in North America, a gradual increase in the incidence of the disease has been noted in the past two decades.^[7,12,16] This increase has been closely associated with the acquired immune deficiency syndrome (AIDS) epidemic. Abdelwahab et al noted an increase in the number of patients in North America who have tuberculosis, independent of those who have tuberculosis in association with AIDS¹. The incidence of the disease has been projected to increase by 41 percent between 1998 and 2020 if better prevention is

not practiced.^[7] Not too long ago, tuberculosis was the most common disease affecting the skeleton; this is still true in certain areas of the world. Even in economically well-developed countries it is still prevalent. Extrapulmonary tuberculosis is a subacute process that often results in a long delay in diagnosis.^[4] Overall, bone and joint involvement account for 11% to 19% of all extrapulmonary tuberculosis, with the most common sites being the spine and knee.^[3,11] Among extrapulmonary cases, involvement of the hands and feet constitutes only 2% of skeletal disease.^[6] Tuberculous tenosynovitis is an even less common manifestation of bone and joint involvement of the disease. The exact prevalence of tuberculous tenosynovitis is unknown. In 1940, Adams et al. reported the incidence to be one in 7898 admissions. The male-female ratio has been reported to be approximately three to two.^[8,13] Only one study showed an equal sex distribution.^[5] The average age has been reported to be between thirty-five and forty years, changing from four years to eighty-one years.^[4,13] In the literature, small clinical series of tuberculous tenosynovitis have been reported with the tendons of the hand and wrist.^[4,10,13] Involvement of the foot and ankle has always been rare. In the series of Adams et al.^[2], infection involved the foot, and in the series of the Mayo Clinic^[4,21] cases of tuberculous tenosynovitis were present in the foot and ankle. Only two cases of achilles tendon involvement were mentioned, the others involving the extensor and peroneal tendons. Pimm and Waugh reviewed 44 cases with a total of 52 lesions, the lower extremity was involved in only three.^[13] There are only a few reports including involvement of the sheaths of the peroneal, anterior tibial, or posterior tibial tendons.^[1,4,11,13] The unique case presentation of an isolated tuberculous tenosynovitis of the achilles tendon which we could find in the English literature, was reported in 1985 by Goldberg and Avidor.^[9] Nevertheless; in the literature, tuberculous tenosynovitis cases related the mycobacterium *kansasii* is known. However; It is limited amount in comparison with mycobacterium tuberculosis. Therefore mycobacterium *kansasii* should be considered in the microbiological analysis.^[7] Theories regarding pathogenesis of tuberculous tenosynovitis focus on infection via either direct inoculation or secondary hematogenous spread from a primary focus, usually the lung. Patients who are exposed to tuberculosis may or may not become infected, and those who are infected may or may not become diseased. A history of past infection may be absent. Usually,

primary infection heals without clinical evidence of the disease, but the secondary focus may become active, sometimes years later.^[9] In the Mayo clinic study, 44% of patients had other sites of involvement, most commonly the lung, kidney, and spine. One half of patients reported a previous exposure to tuberculosis. Local trauma prior to infection was reported in one third.⁴ Our patients have had an isolated involvement of the achilles tendon, and no other infected focus could be detected, either preoperatively or during the follow-up period. In the literature, tuberculous tenosynovitis is described as having gradual and insidious onset, with slowly progressive enlargement of the involved tendon followed by pain and limitation of motion. The wall of the tendon sheath may be thin and almost translucent, or it may be thick and fibrotic. As the infection progresses, the lining membrane of the tendon sheath is replaced by tuberculous granulation tissue. Surrounding tissues also become inflamed and swelling may contain serous fluid, masses, or fibrinous material. In time, the tendon itself may become involved, with granulation tissue spreading longitudinally among the fibers, and the tendon can rupture spontaneously.^[1,4,11,13] The clinical presentation of our patient was consistent with the literature, in the sense that onset was gradual, and there was slow progressive enlargement of the mass and tenderness during motion. However, she had no limitation of motion. At operation, the wall of the tendon sheath appeared thick and fibrotic (Fig. 3). Surrounding tissues were also inflamed and the multilobulated, dark brown cystic mass contained a yellow-brown, serous fluid and fibrinous material. The tendon itself was not involved.

Laboratory studies have minor diagnostic value and may mislead to an unspecified illness. Patients may present with anemia, leukopenia, thrombocytopenia, monocytosis, basophilia, and eosinophilia. The sedimentation rate can sometimes be elevated, in average of 30 mm/hour and positive the tuberculin skin test, strongly indicates a tuberculous process.^[4,9,11] But in previously immunized or anergic patients, test results are not definitive.^[17] MRI and plain radiographs also have no diagnostic specificity in regards to tuberculosis.^[11] In one of the presented case, the sedimentation rate was 9 mm and 45 mm in the other, considering present of rheumatoid arthritis wouldn't be specific. The only abnormality in the blood investigation was monocytosis. The MRI images,

as with our patients, may have potential to confuse the condition with ganglion and/or tenosynovitis. The diagnosis of tuberculous tenosynovitis is usually based on histological and bacteriological examination of material obtained during the operation.¹ However, culture and histologic diagnosis are not 100% reliable in detecting mycobacterium infection.^[11] Only in the second case growth was observed in Lowenstein-Jensen culture media. Brashear et al. reported negative cultures in two of 10 cases with acid-fast bacilli seen in only five.^[5] Bickel et al also reported similar findings. They stated that typical caseous granulomatous lesions in the tendon sheath are usually tuberculous, and a culture or guinea pig inoculation for tuberculosis may give false-negative results.^[4] On histopathologic examination of our patients, necrotizing granulomatous inflammation was observed; and although the EZN stain was failed in the first case to show any acid-fast bacteria and the culture was negative for mycobacterium tuberculosis. After ruling out the other possible differential diagnoses, the results of the histopathologic examination, the patient's father's history and the prevalence of tuberculosis in Turkey, considered together, led us to diagnose tuberculosis.

In the past, prior to antituberculin chemotherapy, most of the infections were treated only with extensive and complete synovectomy and excision of the surrounding tissue.^[4,13] The addition of systemic streptomycin improved surgical results, even in cases with disease recurrence.^[4,13] However, advances in surgical technique and chemotherapy have not eliminated recurrence of the disease. In case reports by Goldberg, Cramer, and Regnard, recurrences occurred from eight months to 36 years after the initial procedure, making long-term follow-up necessary.^[1,3,9,13,15] Although, in other sites of extrapulmonary tuberculosis chemotherapy alone was as effective as chemotherapy combined with surgical excision,^[14] the treatment of tuberculous tenosynovitis requires surgical debridement. Nevertheless, the extent of surgical debridement is still debatable. Some authors advocate surgical debridement with complete excision of the tendon sheath^[4,8,9], whilst others advocate decompression of the tendon sheath without excision and debridement of the surrounding tissue.^[1,5] Both of the presented cases were treated with complete debridement of involved soft tissue. The first case has resulted in a disease-free interval of more than two years (twenty-seven months). At the second case antituberculous regimen was

continued to the sixth month, clinical, laboratory and radiological findings were all disappeared at the ankle region, when she has died due to millier tuberculosis. In conclusion tuberculous tenosynovitis would be considered in patients with swollen hindfoot and who has an infected family member with tuberculosis close to him/her and in immunosuppressive persons .

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