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Bilateral asynchronous chronic Achilles tendon tear in lupus erythematosus: a case report

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Abstract Systemic lupus erythematosus is a multisystem and multior-gan disease that, in some patients, causes tendon and ligament degeneration that may lead to rupture. Early treatment is generally preferred but sometimes good results are achieved in chronic lesions. Bilateral failure of the same structure has been described very rarely.

We report the case of a woman with chronic lupus and bilateral asynchronous Achilles tendon lesions. Good short-term clinical results were obtained despite late surgery and lupus chronicity.

Key words Lupus • Injury • Achilles tendon

Introduction

Systemic lupus erythematosus is a multifactorial chronic and progressive inflammatory disease. Female gender is mainly affected. The annual incidence in Italy has been estimated to be between 1.15 and 2.6 cases/100 000 (increasing in recent years) [1]. Viral, hormonal, genetic and pharmacological causes are considered as starting or promoting factors of the autoimmunization process [2]. Hematological alterations are typical: complement factors (C4) and T-suppressor lymphocyte deficits are common [3]; lymphocytotoxic antibodies are present. Lupus involves multiple organs and systems, causing specific injuries. The osteo-articular apparatus is frequently involved: patients complain of pain (hand, wrist and knee are the most commonly affected) and myalgia [4]. Tendon involvement may cause rare structural ruptures, as deduced from medical literature where few cases were reported [5–10], needing surgical treatment. We present the case of a woman with chronic lupus who suffered bilateral Achilles tendon rupture

walking. She was undergoing therapy for lupus with steroids and cyclosporine. Ultrasonography showed a subtotal Achilles tendon rupture. After a few months, the left Achilles tendon also ruptured. Due to serious family problems, she chose conservative treatment.

In January 2005, she came to our observation. She had been treated all those years with prednisone (12.5 mg/day) and cyclosporine (200 mg/day) for lupus. She complained of mild pain but serious difficulties in walking and in performing daily activities, and had strength loss and articulation problems in plantar flexion bilaterally (the plantaris gracilis tendon was intact). Loss of tendon continuity was perceived upon palpation. Ultrasonography examination showed tendon rupture (Fig. 1). Deambulation was performed with small, crawling steps.

Surgery was proposed and performed after a few days. Selective spinal anesthetic technique was employed. Prone position was used. The incision was median on the tendon course (alternative internal para-median approach is possible in order to better visualize the plantaris gracilis tendon). The sural nerve was protected. The Achilles tendon turned out to be completely interrupted and involved in a lardaceous fibromatous tissue with tendon sheath retraction. Release of the two extremities was accomplished, removing the fibrosis and exposing the tendon tissue in a safety zone for suturing and plasty. Release required pathological tissue ablation for about 3.5 cm, slightly wider than the

Case report

In 1997, a 71-year-old overweight woman with lupus (diagnosed in 1988) experienced sudden right heel pain during

Fig. 1a, b Ultrasonography evaluation of the right Achilles tendon in a woman with chronic lupus. **a** Transverse view of tendon in distal part shows tendinous fibril discontinuity and perilesional hypoechoic fluid halo. **b** Longitudinal view shows tendon breakage with surrounding scar tissue

pathological tendon gap. We then performed tendon plasty according to Lindholm (reverse proximal flap from the proximal portion of Achilles tendon distally sutured just proximal to the tendon's calcaneous insertion) along with terminoterminal reabsorbable suturing and a plantaris gracilis intra- and subtendinous "8" sutured loop bracing (a tendon stripper during this procedure was needed). The paratenon, partially degenerated, was sutured only where possible (Fig. 2). This technique enabled the anatomical reduction of the gap and recovered strength for support during ankle movements. The plantaris gracilis, even if a small tendon, was employed as a local graft (it remained distally inserted, having been at about 10 cm proximally dissected) providing structural reinforcement. Antibiotics (for 6 days) and antithromboembolic therapy (30 days) were prescribed.

Fig. 2a-c Surgical technique. **a** Sampling of the distal plantaris gracilis tendon and set up of reverse Achilles proximal tendon strip. **b** Suture of the reverse tendon strip and "8" loop from the plantaris gracilis. **c** Tensioning and tightening of the sutures

The postoperative leg cast was removed the day after surgery because of the patient's psychological intolerance. Soft leg bandaging was then positioned, granting limited pain-free mobilization. Even if not tolerating the cast, the patient showed good compliance; such an attitude allowed early but controlled mobilization after the reconstruction. This option decreases, in our opinion and clinical experience, the risk of peritendinous adhesences, stimulates appropriate tissue fibril orientation, reduces scar complications and increases vascular contribution, facilitating the healing process. Gradual and progressive weight bearing with two crutches was allowed only after 30 days, using an equinus heeled position for 15 days. A physiotherapy program was started for progressive and full recovery of range of motion and muscular trophism, steps and walking coordination control, and finally proprioceptive exercises. Total weight bearing (crutches free) was obtained 60 days after surgery. Normal gait was achieved 30 days later. Histologic examination showed fibril degeneration with sclerohyalinosis and capillar neoangiogenesis. Ultrasonographic examination on the right and left tendons was performed (Fig. 3). Right tendon continuity was good but local calcifications and granulomatosis corresponding to the sutures were observed. The left tendon was broken as expected.

In March 2005 (the patient was 79 y.o.), the same surgical procedure was performed on the left side. Postoperative care was prescribed as for the right side. Hematochemical evidence was: increased mucoprotein, chronic anaemia and complement (C3 and C4) alterations. Follow-up visits were made at 5, 10 and 15 days and, then every 15 (initially) and 30 days. Ultrasonography at 30 days showed good anatomic results on a chronic tendinous degeneration. The latest follow-up was performed 23 months after the right-side surgery and 21 months after the left. Clinical results are good. No pain, laxity or instability was found; complete active and passive articularity was obtained bilaterally (Fig.

Fig. 3 Ultrasonography examination of the right Achilles tendon two months after surgery. Anatomical integrity is observed. Tendinous fibrils are degenerated. Subcutaneous suture stitches causing shadow cones will later induce granulomatous calcifications

Fig. 4 Follow-up examination 8 months after left ankle surgery and 10 months after right ankle surgery. Good clinical plantar flexion and extension are seen. Suture stitch granulomatosis, involving the subcutaneous tissue, is also observed

4). A mild strength loss was seen in plantar flexion on the left side. Deambulation is correct, with only minimal pain after prolonged standing. Subcutaneous multiple granulomatous suture stitches reaction (Fig. 4), characteristically related to the basic chronic pathology, was treated with topical solutions after a dermatological consultation.

Discussion

About 40 cases of spontaneous tendon rupture in lupus have been reported [5, 6]. Lupus is a clear predisposing factor, sometimes causing erosive deformities (Jaccoud syndrome), myopathic syndromes and subcutaneous calcifications. Initial symptoms are often fractional [7]. A fibromyalgic syndrome may always be excluded. Tendon lesion is rare [6] and is mainly directly correlated with temporal pathology and steroid therapy duration, lupus activity grade and hand arthropathy presence. Pritchard and Berney reported a 2.2% rate of tendon lesion [8]. Males and older people are the most affected as a lupus late complication. Lower limb and loaded tendons are more affected; patellar tendon is frequently bilaterally injured as reported [7, 9, 10]. Sometimes injury occurs in multiple districts. Atypical cases are possible [5]. Bilateral Achilles tendon injuries as observed in our patient are very rare. We found in international literature one paper [6] exposing an achilles tendon injuries (in a very young patient, aged 20) in lupus erythematosus, moreover monolateral and caused by chortisonic pulsed therapy. Our patient was treated with immunosuppressor drugs also and this may have contribute, in our opinion, to increase rupture probability and to help bilateral injury. Other Authors [7-10] did report injuries related to patellar tendon (in 2 cases bilateral and in two others monolateral; Wener [10] described a simultaneous one), but Prasad's case [9] had a previous Achilles injury; Aparad reported [4] an injury localized to extensor pollicis tendon. Tendon may be progressively and in sequence injured and in time clinical situation, in most cases, get frequently worsening". Prolonged chortisonic therapy (9 years in our case at first observation), also causing pharmacological side effects, is reported in Literature [6,8], as a very high and an essential risk factor for ruptures. Etiology is unclear: chronic ultrastructural and inflammatory degeneration may cause the breakage [6]; anatomopathologic alterations are localizable years before the injury and a trauma may be decisive. Sometimes injured patients are in lupus remission phase [7].

We think that a correct life style could reduce the risk of tendon rupture. Prolonged steroid therapy is an important causal factor damaging tendon tissue [5, 6]. Chiou et al. in 2005 [6] described an ischemic necrotic tendon injury following steroid pulse therapy. Preoperatively, a

skilled radiologist can localize the lesion using ultrasonography and find ultrastructural characteristics, allowing correct surgical planning. A better but more expensive visualization is possible with MRI. Zoma [4] and Chiou et al. [6] reported MRI to be a useful examination for structural and degenerative visualization. Doppler ultrasonography and angiography could further show flow and vascularization of the tendon and lesion characteristics (especially in chronic cases) and allow localization of critical avascular regions (keeping in mind that surgery allows revitalization with granulation tissue). The surgical procedure should be individualized, considering the injury type and site, treatment chronicity, patient expectation and preoperative planning. An assessment of treatment options and benefits should always be performed with patient involvement. Chronic injuries (more than 4 weeks after trauma) represent about 25% of the total; they are more difficult to repair because of tendon retraction and scarring fibrosis. In these cases, four topics are to be considered: fibrous tissue should always be excised, the tendon may require lengthening plasty, suture tension influences the healing process, and physiotherapy is essential for full recovery. Patient and injury (direct and instrumental) evaluation allows the surgeon to choose between direct repair inside the Achilles tendon (with reinforcement plasty) or tendon/fascial graft. Achilles strips may be performed with Lindhom or Bosworth technique (proximally or distally tipped over), "Z" lengthening, "V" flaps or proximal slipping flaps. The plantaris gracilis is employed for plasty reinforcement or as a local graft. Autologous tendon grafts can be drawn (with or without bone graft) from the fascia lata, from the same Achilles tendon or from the knee extensor tendons [11] (in cases with an extensive loss of substance), employing a gastrocnemius central flap or shift plasty eventually associated with a "grip-box" plasty in those patients with a loss of tendinous substance up to 6 cm [12]. Flexor hallucis longus and peroneus brevis muscle flap have also been described [13] and have been used alone or with other techniques. Percutaneous tendon repair surgery is preferred in acute and small or middle dimension injuries [14]. Scar tissue has been proposed to be used in chronic lesions for direct repair in patients with tendinosis [15]. Direct repair is a primary option (eventually associated with reinforcement plasty) but in chronic cases is not

always possible and grafts techniques should be required. The first option is a local and less aggressive surgery but in some cases it does not suffice to obtain anatomical and functional reduction of the injury; grafting allows the surgeon to fill up the gap and to improve the suture strength, but sometimes it requires a prolonged and more complex rehabilitation.

Functional conservative therapy may be tried in selected cases, and provides good residual clinical mobility and good adaptative rate, in older patients or in cases with contraindications to surgery, but re-rupture risk is high [14, 16] (varying between 8% and 39%) and the tendon heals generally with lengthening. New materials have been studied outlining clinical employment in repair surgery [17]. Clinical results will have to be considered for results validation. In our opinion, reconstructive surgery may be indicated in these patients after a wide and exhaustive clinical, instrumental, objective and subjective evaluation. Patients with low functional demand, active-phase (with multisystem involvement and documented serious tendon degeneration) unstable and non-responsive lupus, limited daily activities, or multiple serious pathologies should initially (or exclusively) be conservatively treated. In other cases, surgery (multiple technical options are possible) may offer clinical and functional improvement even in chronic cases. Preventive and accurate discussion with the patient is always of primary importance.

Postoperative continuous, early (no cast is employed), prolonged (several weeks are requested) and single patient-related physiotherapy is mandatory in order to obtain and speed up a correct healing process, to quick up overall recovery and to decrease postoperative morbidity, thus providing better results [14, 16, 18, 19].

In conclusion, bilateral tendinous breakage in lupus erythematosus is a rare but possible event. Chronic steroid therapy and progressive ultrastructural damage due to the pathology are mainly involved in the pathogenesis of the lesion. Early and customized surgical approach is preferable but good results are possible in chronic lesions. The patient must always be concerned about enhanced complications rate. Prevention may be done individually tailoring effective steroid therapy and fitting life style to the lupus-related limitations.

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