

Case Report

Misdiagnosed spontaneous rupture and contralateral laxity of patellar tendons in a patient with systemic lupus erythematosus and long-term steroid use: a case report

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Abstract: Spontaneous patellar tendon rupture is a rare event and is often associated with chronic systemic disorders, such as systemic lupus erythematosus (SLE) and long-term steroid use. As there is usually no history of obvious injury, spontaneous patellar tendon rupture is easily misdiagnosed, which delays treatment, increases the complexity of surgery and reduces long-term functionality. The present study reports a case of misdiagnosed spontaneous rupture and contralateral laxity of patellar tendons in a patient with SLE and long-term steroid use. The case had been misdiagnosed by several clinicians for 5 months prior to the final diagnosis. The ruptured tendon was repaired and reconstructed using an ipsilateral hamstring tendon autograft. After 1 year, the patient had regained the full range of motion and experienced no difficulty in daily activities. Careful history-taking, examination and imaging are crucial to limit the number of misdiagnoses that are made. This report suggests that repair and reconstruction with an ipsilateral hamstring tendon autograft is a safe and effective treatment option for misdiagnosed spontaneous patellar tendon rupture in patients with SLE and long-term steroid use.

Keywords: Patellar tendon, spontaneous rupture, misdiagnosis, systemic lupus erythematosus, steroid, reconstruction, hamstring

Introduction

Patellar tendon rupture requires prompt and correct diagnosis and treatment. Spontaneous patellar tendon rupture is a rare event that is only occasionally reported in the literature. It is usually associated with chronic systemic disorders, including rheumatoid arthritis (RA) [1], hyperparathyroidism [2], morbid obesity [3], Ehlers-Danlos syndrome (EDS) [4], systemic lupus erythematosus (SLE) [5], or the use of certain medications, including steroids or fluoroquinolones [6, 7]. Patellar tendon rupture has also been previously observed in apparently healthy patients [8]. In SLE, the most frequently reported predisposing factor is the use of steroids [9]. Because the patients do not typically have a history of trauma, spontaneous patellar tendon rupture is easily misdiagnosed, which delays treatment, increases the complexity of surgery and reduces long-term functionality.

We have found only four cases of misdiagnosed spontaneous patellar tendon ruptures reported in the literature [5, 10-12]. In this study, we report a case of misdiagnosed spontaneous rupture and contralateral laxity of patellar tendons in a patient with SLE and long-term steroid use, in order to increase attention from clinicians, decrease the rate of misdiagnosis, and improve the understanding and management of this rare event. The present case had been misdiagnosed for 5 months, and was subsequently successfully repaired and reconstructed using an ipsilateral hamstring tendon autograft.

Case description

A 29-year-old woman was admitted to the Department of Orthopaedics of West China Hospital, Sichuan University (Chengdu, China) complaining of inability to actively extend her

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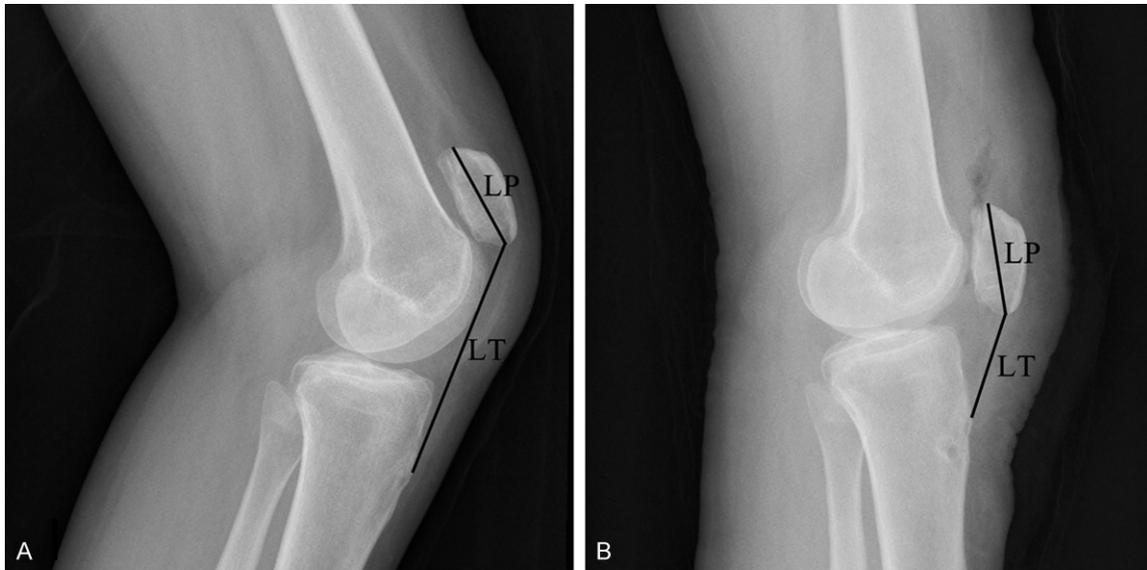


Figure 1. Lateral X-rays of the left knee. A. Preoperative X-ray demonstrating patella alta (Insall-Salvati ratio = 0.46). B. Postoperative X-ray showing normal patellar position (Insall-Salvati ratio = 1.07). LP, length of the patella; LT, expected length of the patellar tendon.

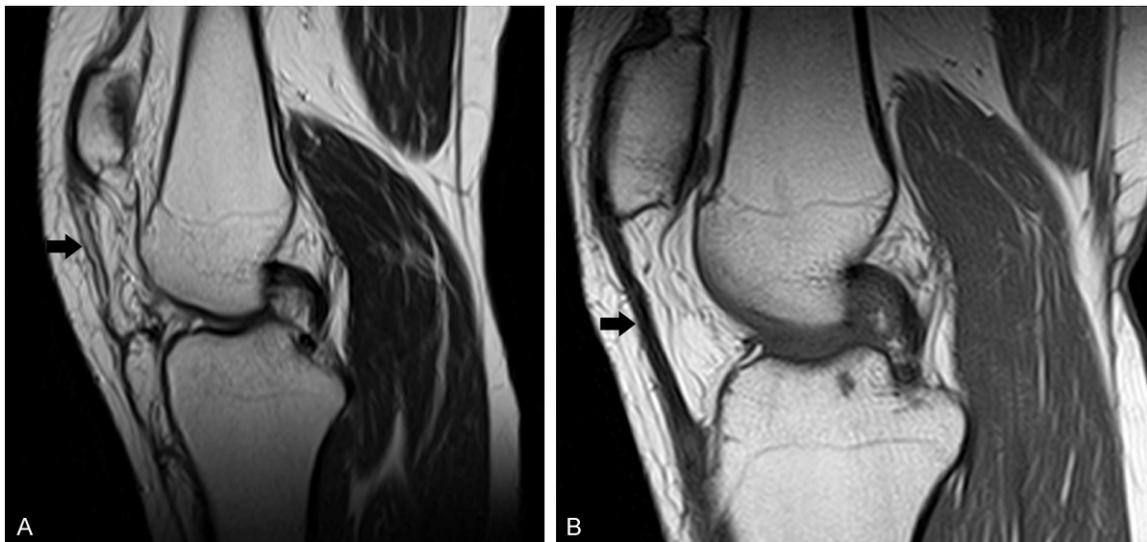


Figure 2. Preoperative MRI of both knees. A. Preoperative MRI of the left knee revealing midsubstance patellar tendon rupture (arrow). B. MRI of the contralateral knee showing that the patellar tendon was continuous but elongated (arrow), with an Insall-Salvati ratio of ~0.67. MRI, magnetic resonance imaging.

leg, and pain and swelling of her left knee following a minor injury 5 months previously. She had been diagnosed with SLE 12 years previously, and had since taken prednisone (10-15 mg/day orally) and hydroxychloroquine sulfate (400 mg/day orally). At 6 months before the injury, she exhibited loss of appetite and weight, and experienced palpitations and dyspnea; the patient subsequently increased her dose of prednisone to 20 mg/day for the next 8 months.

At 5 months before admission to our hospital, the patient had missed a step, braced herself on the next step with her left knee flexed and suffered from a slight external force. She immediately experienced unbearable pain in her left knee and was unable to actively extend it or walk. She was admitted to the orthopaedic clinic of the local primary care facility, where she received an X-ray that revealed 'no osseous injuries and position changes' (**Figure 1A**). Th-

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Figure 3. Physical examination. A and B. Preoperative physical examination of the left knee demonstrated a defect below the patella (arrow) and inability to actively extend the leg or maintain leg extension against gravity, with a 70° loss of active extension. C. Two weeks after the surgery, the patient could perform straight leg raising exercises.

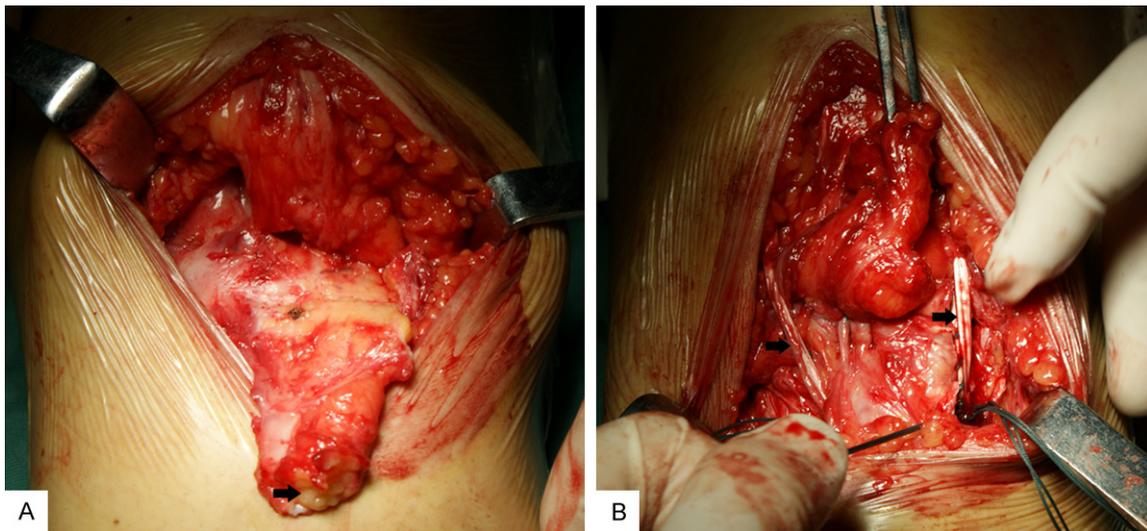


Figure 4. Intraoperative images. A. The left patellar tendon had ruptured transversely through the midsubstance, and the ruptured ends were contractural, absorbed and fat-like (arrow). B. The graft was passed through a transverse tunnel in the tibia and the patella and tightened (arrow), and the ends of the ruptured patellar tendon were minimally debrided to remove the degenerated tissues, then joined and sutured with Ethibond.

us, 'soft tissue injury' was diagnosed and no special treatment was given, except oral non-steroidal anti-inflammatory drugs and external medication. At 1 month after the injury, the patient still could not actively extend her left knee and experienced difficulty climbing stairs. The patient returned to the same primary care facility and magnetic resonance imaging (MRI) was performed on the left knee (**Figure 2A**). The MRI revealed 'no ligament and osteochondral injury'. The doctor suggested that the patient should be observed over a period of time. More than 3 months later, the patient experienced no improvement in knee extension, and was subsequently seen by the director of the primary care facility. A detailed physical examination was performed, revealing a palpable defect below the patella. Examination

of the previous X-ray and MRI scan led to a diagnosis of 'left patellar tendon rupture'. The director therefore suggested that the patient should be transferred to the Department of Orthopaedics at our hospital to undergo surgery at 5 months after the initial injury.

At the Department of Orthopaedics, a physical examination of the left lower extremity revealed a defect below the patella (**Figure 3A**). The patient was unable to actively extend her leg or maintain leg extension against gravity, with a 70° loss of active extension (**Figure 3B**). We also observed that her contralateral knee had a 10° loss of active extension. The previous lateral X-ray of her left knee at 30° of flexion demonstrated the presence of a high-riding patella (patella alta), according to the Insall-Salvati

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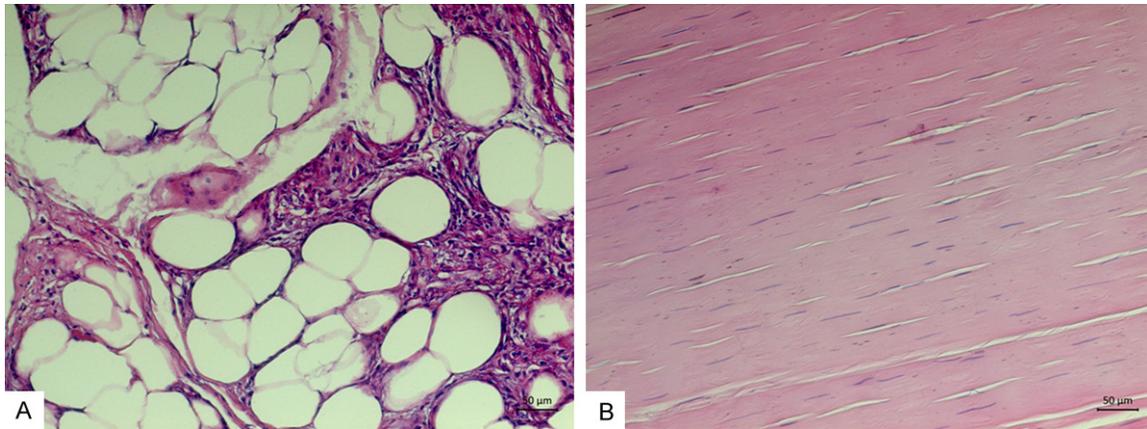


Figure 5. Postoperative histological findings. A. Tendolipomatosis and chronic inflammation of the ruptured patellar tendon, with a lack of normal tendon structure. B. Normal gracilis tendon. Haematoxylin and eosin staining; magnification, $\times 40$.

evaluation method [13], with an Insall-Salvati ratio of 0.46 (**Figure 1A**). MRI of the left knee revealed a midsubstance patellar tendon rupture (**Figure 2A**). Additionally, MRI was performed on the right knee and revealed patella alta, with an Insall-Salvati ratio of ~ 0.67 , and the patellar tendon was observed to be continuous but elongated (**Figure 2B**). Left patellar tendon rupture and right patellar tendon laxity were diagnosed.

The patient was admitted for repair and reconstruction of the ruptured left patellar tendon at 5 months after the initial injury. Preoperative examinations were normal. We repaired and reconstructed the tendon using an ipsilateral hamstring tendon autograft. During surgery, we observed that the left patellar tendon had ruptured transversely through the midsubstance, with disruption of the retinaculum. The ruptured ends were contractural, absorbed and fat-like (**Figure 4A**). The hamstring tendons were combined as a double strand graft, which was then passed through a transverse tunnel in the tibia and the patella in a figure-eight pattern, as previously described [14]. As this was tightened, the ends of the ruptured patellar tendon were minimally debrided to remove the degenerated tissues, then joined and sutured with Ethibond (**Figure 4B**). Postoperative histological examination revealed tendolipomatosis and chronic inflammation of the ruptured patellar tendon, with a lack of regular tendon structure (**Figure 5A**), in contrast with the normal gracilis tendon (**Figure 5B**). Surgical intervention was not required for the right patellar ten-

don laxity, considering that the right knee exhibited relatively good function and that surgery on bilateral knees would cause difficulty during rehabilitation and increase the risk of postoperative complications.

The postoperative period was uneventful. After 2 weeks, the patient was able to perform straight leg raising exercises (**Figure 3C**) and walk without bending the knee under the protection of a hinged brace. X-ray of the left knee revealed normal patellar location with an Insall-Salvati ratio of 1.07 (**Figure 1B**). The patient was subsequently discharged. At the 6 week follow-up, the knee flexion range of motion was 0-90°, and the patient was ambulating independently on crutches. At 1 year after the operation, the patient had regained a full range of motion without experiencing difficulty with daily activities, and was discharged from follow-up.

Discussion

The causes of patellar tendon rupture can be traumatic or spontaneous. Spontaneous ruptures have been defined as “ruptures that occur during movements and activities that should not, and usually do not, damage the musculo-tendinous units” [15]. The usual mechanism for patellar tendon rupture is contraction of the quadriceps muscle against sudden flexion of the knee joint [16]. In a biomechanical analysis of human patella rupture, Zernicke *et al* observed that a force of 17.5-times the body weight was required to rupture a healthy patel-

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Table 1. Misdiagnosed spontaneous patellar tendon ruptures described in the literature

Authors/year	Patient gender	Patient age (years)	Interval between misdiagnosis and correct diagnosis (weeks)	Predisposing factors	Reconstruction approach	Outcome
Present	F	29	21	SLE, steroid use	Hamstring tendon autograft	Excellent
Cherrad T/2015	M	40	12	Idiopathic chronic renal failure, haemodialysis	Ipsilateral quadriceps tendon autograft, wires	Good
Lu M/2012	M	18	1.5	SLE, steroid use	Hamstring tendon autograft	—
Siwek CW/1981	M	36	13	RA, steroid use	Fascia lata autograft, pins and wires	Unsatisfactory
Cooney LM/1980	F	51	24	SLE, steroid use	Sutures	—

F, female; M, male; SLE, systemic lupus erythematosus; RA, rheumatoid arthritis.

lar tendon in a young individual [17]. It is unlikely that this force alone is created in the majority of patellar tendon rupture cases, especially in the case of spontaneous rupture, suggesting that there must be some structural changes that cause weakening of the tendon. Kannus *et al* reviewed biopsy specimens from 891 cases of spontaneous tendon rupture from 1968 to 1989, including 53 patellar tendon ruptures. Around 97% of all ruptured tendons exhibited degenerative changes, including hypoxic degeneration tendinopathy, mucoid degeneration, tendolipomatosis, and calcifying tendinopathy [18]. In the present case, tendolipomatosis and chronic inflammation were observed at the end of the ruptured patellar tendon. Patellar tendons tend to rupture in patients with SLE due to chronic degenerative and reparative changes and chronic inflammation [19]. In SLE, the most frequently reported predisposing factor for spontaneous patellar tendon rupture is the use of steroids [9]. Alves *et al* reviewed 55 cases of spontaneous tendon rupture in SLE and observed that the majority of patients described were receiving various doses of steroids [20]. Therefore, we suggest that, together, SLE and steroids promote patellar tendon rupture.

We have found only four cases of misdiagnosed spontaneous patellar tendon ruptures reported in the literature [5, 10-12] (Table 1). Including the present case, misdiagnoses were made in three male and two female patients, ranging in age from 18 to 51 years old. The intervals between misdiagnosis and correct diagnosis ranged from 1.5 to 24 weeks, with our case misdiagnosed for the second longest time (~21 weeks). Two of the misdiagnosed patients had bilateral injuries [10, 11], and all patients had predisposing factors, including SLE [5, 10], ste-

roid use [5, 10, 11], RA [11], and idiopathic chronic renal failure and haemodialysis [12].

Regarding the rate of misdiagnosis of patellar tendon rupture, only data demonstrating traumatic patellar tendon rupture could be found. In a previous review, 10 out of 36 traumatic ruptures (28%) were misdiagnosed on initial examination and 7 (19%) were repaired >2 weeks after the injury [11]. In another report involving 11 cases of traumatic patellar tendon rupture, all of the cases were misdiagnosed upon initial examination [21]. We speculate that the rate of misdiagnosis of spontaneous patellar tendon rupture will be higher than that of traumatic rupture, because patients with spontaneous patellar tendon rupture do not usually have a history of obvious injury, and because the detection of positive physical symptoms is difficult due to severe swelling of the injured knee during the acute stage. Furthermore, certain clinicians, particularly young clinicians at primary care facilities, lack understanding of this rare event, as demonstrated by the current case report.

Careful history-taking, examination and imaging are crucial to limit the number of misdiagnoses [14]. Misdiagnosis delays treatment, increases the complexity of surgery and reduces long-term functionality [11]. Surgical repair of the patellar tendon is required following a complete tendon rupture to restore optimal function. Simple reduction of the torn tendon ends is often difficult when repair has been delayed by >6 weeks [22]. Ideally, it should be repaired shortly after the injury to minimize tendon retraction and adhesion. Chen *et al* reported a case series in which each patient received surgical treatment within 72 h after injury. All 7 patients were athletes, and returned to weight-

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lifting training within 1 year and to competition within 18 months after the surgery [7]. Conversely, when treatment of patellar tendon rupture was delayed, due to tendon retraction and absorption, reconstruction was often required, and worse outcomes were observed. Consequently, prompt and correct diagnosis and management is crucial.

In order to avoid misdiagnosis, clinicians should pay close attention to patients complaining of dysfunction of knee extension with minimal or no trauma. Patients with spontaneous patellar tendon rupture usually have a history of systemic diseases, such as SLE, long-term steroid use and an increase of steroid dosage at several months before the rupture. Careful physical examination may detect patellar alta, palpable defects over the patellar tendon, inability to actively extend the knee and inability to stand or walk. Lateral X-ray should be taken routinely; although patellar tendon rupture is often misdiagnosed as 'no osseous injuries and position changes', more careful reading and interpretation of the X-ray may detect patellar alta and a low Insall-Salvati ratio. MRI or ultrasonography can also be useful when the diagnosis is unclear.

Several types of graft have been used for the reconstruction of ruptured patellar tendons, including autografts (hamstring tendons [4, 5, 14, 23], ipsilateral quadriceps tendon [12] or fascia lata [11]), artificial ligaments [24] and allografts, such as Achilles tendon [1] and bone-patellar tendon-bone [25]. However, a standard methodology has not yet been developed. In this case, reconstruction using artificial ligaments or allografts presented a high risk of breakage and infection due to the long-term steroid use and the poor soft tissue condition of the patient because of the 5-month delay in diagnosis. Additionally, the sources of allografts are limited and artificial ligaments are expensive. Consequently, we reconstructed the ruptured patellar tendon using an ipsilateral hamstring tendon graft and achieved satisfactory effects. Gilmore *et al* reviewed 503 cases of patellar tendon rupture from 1947 to 2013, in which 149 chronic reconstructions used 8 different operative techniques, with autologous grafts (using either gracilis, gastrocnemius, quadriceps, patellar or semitendinosus tendons) being the most popular technique,

and achieved significantly improved results compared with primary repair [26]. Takata *et al* previously reported that repair and reconstruction with an autologous hamstring tendon was an effective treatment option for patellar tendon rupture in a patient with EDS [4]. Maffulli *et al* reported that patellar tendon reconstruction with use of the ipsilateral hamstring tendon graft was safe, and clinically and functionally effective, and allowed the patient to return to preoperative daily activities, with very satisfactory outcomes in 17 of 19 patients with chronic patellar tendon ruptures [23]. We believe that ipsilateral hamstring tendon grafts are ideal for the reconstruction of the patellar tendon as they are easy to handle, strong, and harvested routinely in tendon and ligament reconstruction.

Conclusion

In conclusion, spontaneous patellar tendon rupture in patients with SLE is a rare event and is easily misdiagnosed. Misdiagnosis delays the appropriate treatment, increases the complexity of reconstruction and reduces long-term functionality. Clinicians must keep this diagnosis in mind and pay close attention to patients that complain of extension dysfunction of the knee with minimal or no trauma. Careful history-taking, examination and imaging are crucial to limit the number of misdiagnoses. The present case report demonstrates that repair and reconstruction with ipsilateral hamstring tendon autograft is a safe and effective treatment option for misdiagnosed spontaneous patellar tendon rupture in patients with SLE and long-term steroid use.

Disclosure of conflict of interest

None.

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