CASE REPORT

Spontaneous biceps tendon rupture in a patient with mixed connective tissue disease

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Summary

Tendon rupture is a rare complication that occurs in patients receiving corticosteroid therapy. We report a case of a middle-aged man with mixed connective tissue disease who presented with spontaneous biceps tendon rupture 5 weeks after initiation of high-dose corticosteroid therapy. Musculoskeletal ultrasonography was performed at the clinic and helped to confirm the diagnosis. It is a new imaging modality that is increasingly used in rheumatology clinics in Malaysia as it serves as an extension to physical examination. Musculoskeletal ultrasonography is preferred by patients as it is noninvasive, does not involve ionising radiation, painless, relatively inexpensive and can be performed readily at the clinic.

Case Summary

A 58-year-old retired man presented to the Rheumatology Unit with progressive skin tightening for the past 5 months. This had resulted in limited range of motion of the hands with difficulty in gripping objects. He also complained about difficulty in rising from a sitting position and reaching to objects from a height. On further questioning, he experienced epigastric discomfort and dyspepsia. There was, however, no Raynaud's phenomenon, dyspnoea, reduced effort tolerance, dysphagia or diarrhoea. There were no symptoms to suggest thyroid disease and he denied consumption of statins or supplements.

Physical examination revealed a thin middle-aged man who had sclerodactyly, microstomia and generalised skin induration involving the trunk and the limbs. There was proximal muscle weakness with a power of 4/5 in shoulder abduction and hip flexion bilaterally, as well as weak neck flexor muscles. He was diagnosed with mixed connective tissue as he had clinical features of both systemic sclerosis and polymyositis.

Laboratory tests showed the following results: Positive antinuclear antibody (ANA)(titre of 1: >2560), positive rheumatoid factor, extractable nuclear antigen (ENA) panel showed presence of anti-ribonucleoprotein (RNP) antibody, anti-Sm and anti-SSA antibodies, but anti-Scl70 antibody was negative. Both creatine kinase and lactate dehydrogenase (LDH) levels were high at 3329 U/L and 1755 IU/L, respectively. ESR was elevated at 92 mm/h.

Our patient underwent muscle biopsy, which confirmed the presence of polymyositis. He was promptly started on high-dose oral prednisolone and azathioprine. In the meantime, he was screened for internal malignancy for which the results were all negative.

Figure 1: "Popeye" deformity depicting prominent swelling at the distal portion of the right upper arm which is due to distal retraction of the muscle belly of biceps.

Upon review 5 weeks later, he complained of a sudden onset of pain over the anterior aspect of the right shoulder associated with difficulty in raising his right arm. He could not recall any preceding history of trauma or lifting of heavy loads. He was on prednisolone 40 mg daily. Physical examination demonstrated a depression at the region of the bicipital groove accompanied by a non-tender, soft tissue swelling over the mid-portion of the right upper arm (Figure 1). Proximal muscle weakness was

still present in both upper and lower limbs, similar to the initial presentation. Elbow flexion was weak with power of 4/5 on the right but 5/5 on the left. However, muscle power of forearm supination was 5/5 bilaterally. There was remarkable improvement in creatine kinase level though, with a significant reduction to a level of 512 U/L.

Biceps tendon tear was suspected, which was subsequently confirmed on musculoskeletal ultrasound performed by the rheumatologist using a portable ultrasound machine at the clinic. There was a complete tear of the long head of biceps tendon with retraction of the inferior portion of the tendon till the musculotendinous junction. Biceps tendon was not visualised in the bicipital groove. The other rotator cuff tendons (i.e., supraspinatus, infraspinatus and subscapularis) were normal. The dynamic abduction test to assess for shoulder impingement showed a negative finding.
Patient was referred to the orthopaedic surgeon who then advised for conservative therapy as surgical intervention was deemed unnecessary. He continued with physiotherapy and the prednisolone dosage was gradually tapered while azathioprine was optimised to a level of 2 mg/kg/day.

Within 8 weeks of prednisolone therapy, serum creatine kinase level had normalised. He continued to show improvement in terms of muscle power and by 6 months, he had regained full power of all muscle groups including the neck flexor muscles. In particular, he had also managed to regain full power of elbow flexion. He was indeed very pleased with his recovery.

Discussion

A vast majority of biceps tendon rupture involves the proximal long head. Cases of biceps tendon rupture are frequently secondary to injuries, which are the result of repetitive microtrauma and overuse.[1]

Biceps tendon rupture occurs when there is a sudden or prolonged contraction of the muscle against resistance in middle-aged and elderly individuals with pre-existing chronic bicipital tendinitis. Biceps tendon rupture can also be due to insidious inflammation of the tendon from impingement in the subacromial region resulting in chronic microtrauma.

Our patient was very certain that there was no history of trauma prior to the biceps tendon rupture. Radiography of the shoulder was normal. In addition, ultrasonography did not reveal any features suggestive of chronic tenosynovitis or shoulder impingement syndrome. Despite the rupture, our patient only had mild weakness of elbow flexion with preserved forearm supination. This diminution of strength is minimal due to functional brachialis and supinator muscles.

We believe that biceps tendon rupture in our patient is most likely the consequence of corticosteroid therapy. Several drugs have been reported to be associated with increased risk of tendon rupture, in particular fluoroquinolones[2] and corticosteroids. Among patients on corticosteroid therapy who had tendon ruptures, the rupture sites that have been reported include Achilles tendon, patellar tendon, biceps tendon and extensor pollicis longus.[3-8] These patients were receiving oral preparations of corticosteroids for rheumatic and nonrheumatic diseases, and the doses ranged from 5 mg daily to 30 mg daily. The duration of corticosteroids when tendon ruptures occurred varied from 3 months to 23 years.

The exact mechanism by which corticosteroids cause tendon rupture is not clearly understood. Haines suggested that corticosteroids could possibly suppress repair of degenerated or partially ruptured tendons to a point that complete rupture can arise even after a trivial incident.[8] The fact that our patient has concomitant polymyositis may suggest that muscle inflammation probably weakens the muscle fibres, thus augmenting the risk of rupture. Tendon rupture as a consequence of systemic sclerosis per se has not been reported previously.

In most cases, biceps tendon rupture can be diagnosed based on the history and physical examination alone. Distal retraction of the tendon within the arm produces a "Popeye" deformity. Imaging studies can be employed as an extension of physical examination to confirm the diagnosis and rule out other disorders.

This case clearly illustrates the importance of ultrasonography in the evaluation of the patient's problem.[9,10] Its advantages include painless examination, non-invasiveness, lack of ionising radiation, lower cost compared to MRI, dynamic imaging capability and more importantly it gives rapid interpretation and can

be performed at the point-of-care. In spite of these advantages, ultrasonography poses some challenges and limitations. The cost of acquiring an ultrasound machine can be quite considerable and this is largely dependent on the resolution required. Acquisition of ultrasound skills takes time and it may not be possible for a trainer to be available throughout the whole training period. Ultrasonography is operator-dependent and inter-observer variability is not unusual.

Of late, there has been an increasing use of ultrasonography for musculoskeletal indications among rheumatologists in Malaysia. Notably, training in musculoskeletal ultrasonography has now been incorporated into the rheumatology subspecialty training programme in the Ministry of Health Malaysia. Conservative management with physical therapy[11] was considered appropriate in this patient given his age and the fact that he was a retiree and his lifestyle wass not physically demanding. Thus, his impairment would not affect his activities of daily living. It is therefore important that treatment is individualised.

Conclusion

Spontaneous biceps tendon rupture is an uncommon condition. However it can be diagnosed clinically and confirmed by ultrasonography. Conservative management is the norm with surgical intervention reserved for younger, active individuals, especially those who require maximum supination strength.

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