Spontaneous bilateral quadriceps rupture in a patient with ankylosing spondylitis: A case report

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Abstract
Ankylosing spondylitis is a prototype rheumatoid factor-negative spondyloarthropathy that causes the vertebrae to fuse making the spine less flexible, resulting in a hunched-forward posture.

A 47-year-old male who was a known case of ankylosing spondylitis presented with left quadriceps rupture after raising his right foot to lightly hit a football. He was fitted with a knee immobilizer as an emergency measure. The extent of the rupture was not yet established when patient requested leave to go home. The right quadriceps was ruptured as well when he took his immobilizer off to go the bathroom.

We report herein an extremely rare case of ankylosing spondylitis with atrophy in the quadriceps to the extent of causing pathological muscle rupture. Prolonged period of immobilization results in muscle wasting which leads to knee joint instability so the muscle should be repaired as soon as possible.

Keywords: Stress test, Patellofemoral pain syndrome, Ankylosing spondylitis, Case report.

Introduction
Ankylosing spondylitis is a chronic inflammatory disease of the axial skeleton that is normally characterised by pathological changes that begin in the ligamentous attachment to the bone. Involvement of the sacroiliac joint in the form of sacroiliitis is almost always present and the disease is known to have association with human leukocyte antigen (HLA) B27 in 90% of the cases, showing a strong genetic predisposition. There is abnormal adhesion and rigidity in the intervertebral joints resulting in a bamboo spine that develops as the outer fibres of the fibrous ring of the intervertebral disc ossify which results in the formation of marginal syndesmophytes between adjoining vertebrae. Inflammation in other parts of the body such as the eyes is also seen in 40% of the cases.1

On the other hand, rupture of the quadriceps tendon is quite uncommon itself and usually occurs in patients over 40 years being more common in men than women. This injury can occur during falls, direct blows, laceration injuries or due to iatrogenic causes.2,3 Bilateral ruptures are even more uncommon and are highly correlated with systemic diseases that cause pathological degeneration of the quadriceps tendon.4 Weakness and rupture of this tendon have also been reported due to steroid injections or fluoroquinolone use such as ciprofloxacin and levofloxacin.5,6

Ankylosing spondylitis is not normally associated with quadriceps degeneration to such an extent that it can cause rupture as was seen in the case reported herein.

Case Report
A 47-year-old married Indian male orthopaedic surgeon presented in the emergency department at the Jeddah National Hospital on 7th August 2015 with the complaint of sudden onset of suprapatellar pain and inability to extend the knee of the left lower limb. The patient explained that he had just lifted his right foot to pass a football back to a group of kids playing in the park during his morning walk, when he experienced pain in his left suprapatellar region, lost his balance and fell down. The patient was provisionally diagnosed with ankylosing spondylitis at the age of 39 years during a routine x-ray which showed changes in the spine and the sacroiliac joint. He had no peripheral signs of the illness such a uveitis, skin rashes, any gastrointestinal illness, or joint pain other than that at the sacroiliac joint. His back and neck pain would be severe in the morning and relieved when doing exercise. He had transient hearing loss due to an unknown aetiology in the left ear at the age of 41 years which eventually recovered on its own. The patient explained that he had never taken any medications for his illness but did take corticosteroids

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once after he felt numbness in his hands while performing a surgery.

Examination was performed and a provisional diagnosis of a ruptured quadriceps was made but the extent of the rupture was not yet established. A complete workup of laboratory tests was performed. HLA-B27 was found to be positive and slight hyperuricaemia was detected. Other findings were unremarkable.

The patient was fitted with an immobilizer on the left knee and scheduled a for series of investigations, including a magnetic resonance imaging (MRI), as the doctors were sceptical as to how such a serious injury occurred spontaneously, and was listed for surgery in two days. The patient insisted on going home for the night. He was given permission considering that being a doctor he understood the risks and precautions that he must take. Before going to bed that night, he went to the bathroom, kept his crutches on the side and as he started to limp towards the toilet, he experienced another spontaneous episode of suprapatellar pain, now in his right lower limb, lost balance and fell down. He was brought back to the hospital. An MRI was performed to confirm the diagnosis, informed consent was obtained along with basic work up and investigations before surgery was done and the muscle repair was performed in two layers with strong Ethibond ® 5 sutures. During surgery, the edges of the rupture were found to be rather smooth and there was no profuse bleeding indicating a previously atrophied muscle due to pathological degeneration (Figure-1).

The patient was advised bed rest for a month even though he was capable of walking few hours after surgery. He was recommended for a physiotherapy regimen immediately to speed up the process of recovery. After two months, the patient was able to walk normally but there was considerable quadriceps lag, that is, lack of full knee extension, in both legs. After four months, the quadriceps lag in the right limb recovered but some still existed in the left limb. There was hollowing noted in the supra-medial aspect of the left knee indicating wasting of the vastus medialis. Due to some intra-operative muscle loss, the knee joint is not fully stable so the patient has to walk keeping his knee joint in the locked, fully extended, position which is troublesome when climbing up or coming down the stairs. The patient also complained of patellofemoral joint pain since after surgery but these problems are improving with time. After six months of surgery, the patient was performing all his professional activities normally. Currently, his spine is fully ankylosed but there is some movement at the atlanto-axial and atlanto-occipital joint, more so at the former.

Discussion

Systemic arthropathies such as rheumatoid arthritis has been reported to result in bilateral complete rupture of the quadriceps but rupture due to ankylosing spondylitis is extremely rare, and even more so when the patient has not been taking any systemic steroids for the ankylosing spondylitis as steroid abuse has also been implicated with muscle rupture. The various systemic diseases that predispose people to quadriceps tendon rupture include arteriosclerosis, diabetes mellitus, systemic lupus erythematosus (SLE), primary and secondary hyperparathyroidism, gout, tuberculosis, vasculitis, and steroid injections to the tendons. Furthermore, the patient did not report any pain in the knee joint prior to the muscle injury, so the degeneration of muscle overlying the knee joint is also unlikely.

Degenerative changes commonly occur in the tendons as the aging process causes their architecture to change. However, quadriceps tendon rupture is rare even among older people. Thus, other underlying factors, and not...
A study conducted in 1983 concluded that muscle changes do occur in ankylosing spondylitis and more specifically, muscle atrophy in the quadriceps was seen in eight out of the twenty subjects. Following this, we can, with some degree of confidence, conclude that ankylosing spondylitis can indeed be one of the systemic causes of quadriceps rupture.

Pathological muscle weakness in ankylosing spondylitis to the extent of causing a spontaneous bilateral quadriceps rupture is extremely rare. Prognosis is good for surgical repair if treatment begins immediately. However, delayed surgical repair of ruptured quadriceps tendons may result in increased complications due to the retraction of the tendon by quadriceps muscle spasm. This may result in permanent fibrosis of the muscle, resulting in limited power and mobility. Many individuals are unable to return to their pre-injury activity levels and report chronic strength deficits in the affected leg. "Stress test" at the knee joint, up to 90 degrees flexion, should be performed after suturing (Figure-2). Quadriceps lag, knee joint instability and patellofemoral joint pain were the most prominent complications seen in this case. Post-operative physiotherapy should be advised.

**Figure-2:** Stress test after suturing.

**Conclusion**
Pathological muscle weakness in ankylosing spondylitis to the extent of causing a spontaneous bilateral quadriceps rupture is extremely rare. As the prognosis after surgery is good, the procedure should not be delayed. Post-operative physiotherapy should also be recommended.

**Disclaimer:** Written informed consent was obtained from the patient for the publication of this case report and any accompanying images.

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**References**