Case Report

Diabetic Myonecrosis: Rare Complication of a Common Disease
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Abstract
We report a case of sudden left thigh pain in adductor muscles of medial compartment, known as diabetic myonecrosis, in a 50-year-old patient with poorly controlled diabetes mellitus. Diabetic muscle infarction is a rare end-organ complication seen in patients with poor glycemic control and advanced chronic microvascular complications. Proposed mechanisms involve atherosclerotic microvascular occlusion, ischemia-reperfusion related injury, vasculitis with microthrombi formation, and an acquired antiphospholipid syndrome. The clinical presentation is swelling, pain, and tenderness of the involved muscle, most commonly the thigh muscles. Management consists of conservative measures including analgesia and rest.

Key words: Diabetic Myonecrosis, Diabetic Mellitus, Complication

Introduction
The term diabetic muscle infarction also known as spontaneous diabetic myonecrosis is used to refer to a rare complication of diabetes mellitus manifesting as spontaneous ischemic necrosis of skeletal muscles. It causes acute or subacute pain, swelling, and tenderness, typically in the thigh or calf. It affects both type 1 and type 2 diabetic patients who have long duration of diabetes and frequently have microvascular complications.\textsuperscript{1} Diabetic myonecrosis most commonly affects the lower limbs involving the following muscle groups- quadriceps (~60%), hip adductors (~15%), hamstrings (~10%), hip flexors (~2%).\textsuperscript{2} Radiographic features in Computed Tomography are diffuse muscle enlargement with decreased attenuation, hyperattenuating subcutaneous fat and Magnetic Resonance Imaging displays a mass-like area of muscle necrosis.\textsuperscript{3} The diagnosis is confirmed by muscle biopsy with histopathology. It normally responds well to conservative treatment and is self-limiting. High (>50%) recurrence rates are reported and in these cases prognosis is poor with most patients dying within five years.\textsuperscript{4}

Case Report
A 50-year-old man with type 2 diabetes for 15 years on insulin therapy presented with swelling of right lower leg for 15 days, generalized weakness for 1 month and uncontrolled diabetes for 1 year, burning sensation in micturition for 10 days and no previous history of trauma or similar episode.

On examination his pulse 84 bpm regular, blood pressure was 140/80 mmHg, pulse 84 bpm regular, anaemic and there was pedal edema bilaterally, generalized scaly lesions. Right thigh was swollen, tender without fluctuation. He had bilateral non proliferative diabetic retinopathy and no signs of polyneuropathy. His total leucocyte count was 12090/cmm, hemoglobin 5gm/dl, ESR 41mm at 1\textsuperscript{st} hour. His Hemoglobin A1c was 9.9%, serum creatinine 4.7mg/dl, CRP 136mg/L, Urine R/M/E –glucose (nil), protein (+), acetone (nil), pus cell (4-8), epithelial cell(0-2), red blood cell-occasional. Urine culture and sensitivity- Growth of Escherichia coli, D Dimer- positive, Serum lactate dehydrogenase 666U/
L. Diabetic nephropathy, hypertension, retinopathy were clinically evident. Our provisional diagnosis was acute kidney injury on chronic kidney disease due to cellulitis (Right thigh) with urinary tract infection. He was given intravenous antibiotic and for anemia 6 units of red cell concentrate was transfused and improved clinically. During hospital course he developed sudden left sided thigh pain and described as aching, visual analogue scale 7 out of 10 during maximal intensity with restricted knee flexion and no alleviating factors. Ultrasonogram of left thigh showed small linear subcutaneous collection at the medial aspect of lower third of the left thigh, Doppler study both lower limb vessels showed bilateral mild atherosclerotic change and about 30% flow reduction in anterior tibial, posterior tibial and arteria dorsalis pedis on both sides. MRI of the left thigh showed myonecrosis/ inflammatory lesion of adductor muscles in the medial compartment of left proximal thigh. The patient was treated conservatively with insulin, analgesics (paracetamol), antibiotics, antihypertensives, and physiotherapy. He was recovering and discharged on request.

Discussion
Diabetic muscle infarction is a rare and uncommon manifestation of long standing poorly controlled diabetes. The exact pathogenesis is not known but different hypothesis propose it as hypoxia-reperfusion injury, atherosclerotic occlusion, or vasculitis with thrombosis, atheroembolism of small vessels and hypercoagulable state. But most accepted hypothesis is thromboembolic events resulting from tissue ischemia leading to oxidative stress and finally tissue necrosis. Reperfusion injury causes further damage associated with endothelial dysfunction; manifested as impaired endothelium-dependent dilation in arterioles along with increased oxygen radicals, with less nitric oxide, following reperfusion. Following imbalance between superoxide and nitric oxide in endothelial cells leads to production and release of inflammatory mediators (Tissue necroting factor, platelet aggravating factor) and increased biosynthesis of adhesion molecules. The inflammatory cascade increases intra-compartmental ischemia and further necrosis. Some authors have found evidence consistent with a hypercoagulable state as the
etiology of diabetic myonecrosis involving increased factor VII activity, increased levels of plasminogen activator inhibitor and thrombomodulin. Presence of antiphospholipid antibodies in diabetic patients may increase the risk of diabetic myonecrosis.

Angervall and Stener first described this condition in 1965. A recent systematic review of the literature up to August, 2001 identified a total of 116 patients and showed that this complication was more common in women (61.5% of all cases) and in longstanding diabetes (mean 14.3 years). It is frequently unilateral with occasional bilateral involvement with predilection of the thigh muscles, the calf being the second most commonly affected site. Vascular complications of diabetes were present in the majority of cases, particularly nephropathy (71%), retinopathy (59%), and neuropathy (55%).

Clinical features are acute or sub-acute pain, swelling and tenderness typically the thigh muscles also including calf muscles. Diabetic myonecrosis usually presents unilaterally but 8.4% cases reported as bilateral. Similar clinical features may be found in pyomyositis, necrotizing fasciitis, deep vein thrombosis, soft tissue abscess, cellulitis, hematoma, and acute compartment syndrome.

Laboratory and imaging studies aimed at excluding other disorders and routine laboratory investigations are relatively nonspecific. There may be leukocytosis and serum creatine kinase levels may remain normal or slightly elevated. MRI is the imaging test of choice since it is noninvasive, with a better sensitivity and anatomic definition. The gold standard for the diagnosis is muscle biopsy that may be necessary for confirmation when the presentation is atypical as in our case. Needle biopsy is preferred over excisional and incisional biopsies given the potential complications.

The characteristic feature is an increased signal from the affected muscle area (intramuscular and perimuscular tissues) in T2-weighted, inversion-recovery, and gadolinium-enhanced images and isointense or hypointense areas on T1-weighted images, secondary to increased water content from edema and inflammatory changes that accompany the infarction. On biopsy it grossly appears as nonhemorrhagic pale muscle tissue. Histologically, there are large areas of muscle necrosis and edema, phagocytosis of necrotic muscle fibres, granular tissue, and collagen. This is followed by eventual replacement of necrotic muscle fibers by fibrous tissue, myofiber regeneration, and mononuclear cell infiltration.

Diabetic myonecrosis is a self-limiting disease that responds well to conservative management. Good glycemic control should be achieved and patients usually recover within few weeks with supportive treatment like bed rest and analgesics for pain.

Although the short-term prognosis of diabetic myonecrosis is good, the long-term prognosis is poor and most patients die within 5 years. Recurrence has been reported in 47.82% cases involving either the same or the other limb.

Conclusion

Though diabetes mellitus is a common disease worldwide, diabetic myonecrosis is still a rare disease. It should be considered in patients with long standing diabetes mellitus with microvascular complications who present with acute-subacute onset severe focal muscle pain in the absence of any systemic symptoms. MRI is the most sensitive test for diagnosis. Strict glycemic control, analgesics and bed rest are mainstay of treatment.

Conflict of interest: Nothing to declare.

References