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CASE REPORT
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Diabetic Myonecrosis—An Underdiagnosed, Rare Muscular Complication of a Common Disease: A Case Report

ABSTRACT

Diabetic myonecrosis is an uncommonly encountered and underdiagnosed complication of diabetes mellitus. It could be a manifestation of poor glycemic control, and hence, progressive microvascular disease; however, it is usually self-limiting. A known but poorly controlled type 2 diabetes mellitus patient presented with sudden onset and worsening right thigh pain and swelling. Magnetic resonance imaging showed hypertrophy and hyperintensity of the adductor muscles with non-enhanced regions denoting areas of necrosis. Conservative management of bed rest, analgesics, and controlled blood glucose yielded a positive response. Diagnosis of diabetic myonecrosis can be challenging as many diseases mimic this condition, resulting in delayed recognition, diagnosis, and initiation of prompt treatment. Magnetic resonance imaging findings rather than laboratory markers are the mainstay of diagnosis as it localizes the affected muscles and excludes other mimics.

Keywords: Case report, diabetes mellitus, magnetic resonance imaging, muscular infarction, myonecrosis

Introduction

Diabetic myonecrosis (DMN), or muscle infarction, is an uncommon complication of long-standing and poorly controlled diabetes mellitus (DM), a metabolic disorder with an increasing prevalence and a rising burden from its complications.[1] It causes spontaneous ischemic necrosis of skeletal muscles, with sudden onset of severe pain, swelling of the affected limb, and occasionally palpable mass in the absence of trauma and fever. Since it was first described in 1965, only a few cases have been published.[2] Diabetes myonecrosis is frequently unilateral, although bilateral cases have been reported.[3] Commonly affected muscles are the thigh muscles: quadriceps (60%-65%), hip adductors (13%), hamstrings (8%), and hip flexors (2%), although involvement of the calf and upper extremity muscles has been reported.[3,4] An awareness of this condition among physicians is important for prompt diagnosis and treatment, while similarly presenting muscular and vascular pathologies of the limb are excluded.

Case Presentation

A 56-year-old woman presented with a 10-day history of severe and worsening right thigh pain and swelling. The pain was spontaneous, unremitting, and dull aching with no weakness in the right leg but severe enough to restrict her ambulation. There was no history of fever or trauma to the affected limb. She had a similar painful swelling of the same thigh 8 months ago which resolved spontaneously after the use of analgesics. She has been diagnosed with type 2 DM 15 years ago before presentation with poor drug compliance and irregular clinic attendance.

On examination, she was afebrile with normal oxygen saturation. However, she was tachycardic with a pulse rate of 118 bpm and her blood pressure was elevated (178/106 mmHg). She was in severe pain and had difficulty bearing weight with a reduced range of movement in the right leg. The widest circumference of the right thigh (taken about 10 cm from the knee joint) was larger (78 cm) than the left (38 cm), with increased warmth and tenderness. The vibration perception test showed bilateral peripheral neuropathy, although there was no symptom suggestive of nephropathy or retinopathy. The levels of C-reactive protein of (14.1 mg/dL, normal: 1-3 mg/dL), erythrocyte sedimentation rate (75 mm/h, normal: 0-25 mm/h), white blood count (12 000/dL, normal: 4000-11 000/dL), fasting blood glucose (518 mg/dL,

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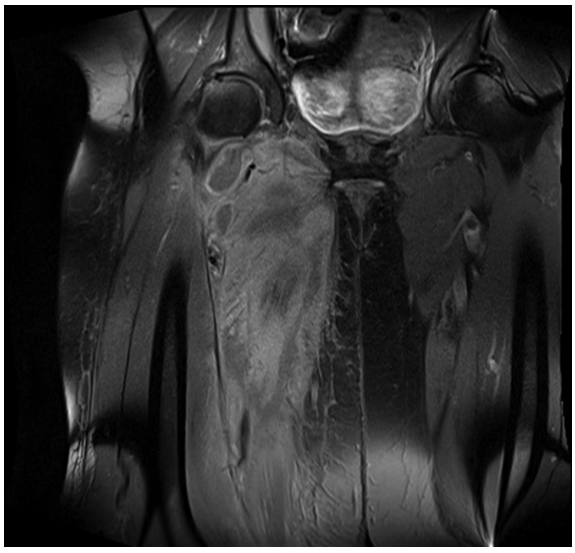

Figure 1. Coronal post gadolinium T1-weighted magnetic resonance imaging of the right thigh showing right adductor muscular swelling with loss of intramuscular fat strands, heterogeneous increased signal intensities, and rim enhancement when compared to the contralateral side. The signal intensity of the femoral bone is normal.

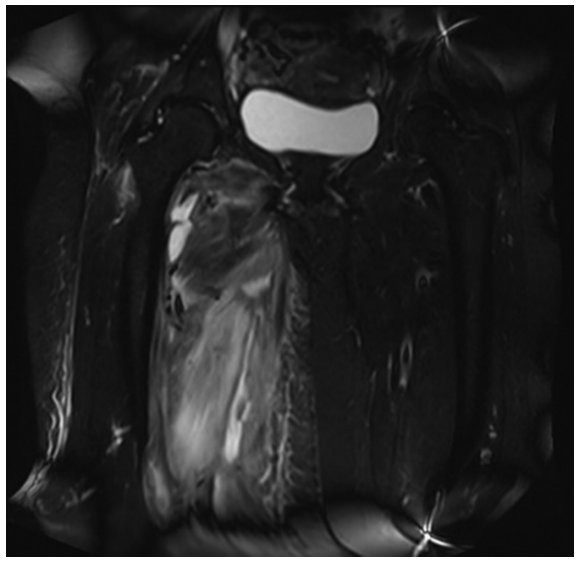

Figure 2. Coronal fat-suppressed T2-weighted magnetic resonance imaging of the right thigh showing diffuse muscle enlargement, loss of intramuscular fat strands, and hyperintensities (edema) in the adductor muscles. The femoral bone intensity is normal.

normal: 70-100 mg/dL), and glycated hemoglobin (18.6%, normal: <6.5%) were elevated. The levels of hemoglobin (10.6 g/dL, normal: 9.5-15.3 g/dL) and creatinine phosphokinase (135 units/L, normal: 38-174 units/L) were within normal range.

B-mode ultrasonography of the right thigh showed enlarged medial thigh musculature and moderate intermuscular fluid collection. No intramuscular abscess collection was seen. Doppler ultrasonography was negative for deep venous thrombosis and arterial thrombus bilaterally. Magnetic resonance imaging (MRI) T1-weighted images showed enlargement of the adductor muscles of the thigh with loss of normal intramuscular septae. T2W MRI revealed marked muscular enlargement and heterogenous signal intensities within the right adductor muscles as well as diffuse surrounding perifascial edema and subcutaneous soft-tissue fluid. Post-gadolinium images showed heterogenous enhancement with non-enhanced and rim-enhanced regions, denoting necrotic areas within the adductor muscles (Figures 1-5). No osseous collection, marrow infarct, or osteomyelitis was seen. Given the history of long-standing and poorly controlled blood glucose and exclusion of infection, a diagnosis of DMN was made. The patient was admitted for bed rest, analgesics (nonsteroidal anti-inflammatory agents) were given, and her blood glucose

MAIN POINTS

- Diabetic myonecrosis is a rare and uncommon muscular complication of long-standing, poorly controlled type 2 diabetes mellitus.
- There is a relative lack of awareness about this condition when compared to other complications of diabetes mellitus. Hence, there is often a delay in its diagnosis, which in turn increases the morbidity associated with the disease.
- Diabetic myonecrosis typically presents with symptoms of compartment syndrome, with many common conditions mimicking the disease. It can be misdiagnosed and treated surgically, hence prolonging patients' morbidity and recovery. However, diabetic myonecrosis is a self-limiting condition that can be easily managed by conservative treatment and supportive therapy.
- Laboratory markers are not definitive in their diagnosis. Magnetic resonance imaging is the investigation of choice, localizing the affected muscles and excluding other mimics of the condition. In addition, it prevents unnecessary and invasive muscle biopsy which is associated with increased patient morbidity and delayed recovery.

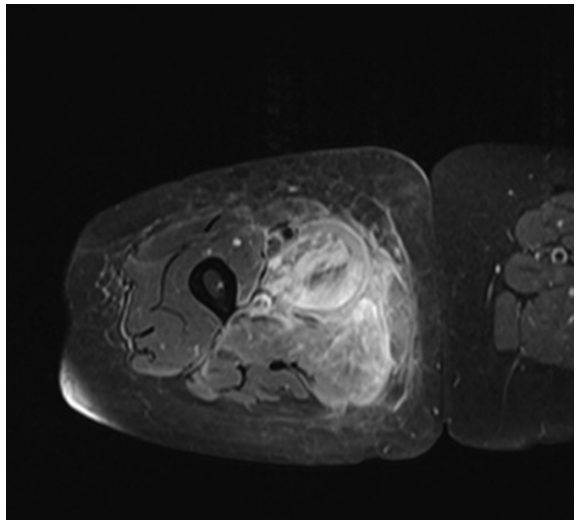

Figure 3. Axial fat-suppressed post gadolinium T1-weighted magnetic resonance imaging of the right thigh showing muscular swelling and hyperintensity in the adductor muscle groups. A small area of non-enhancement suggestive of necrotic tissue was also seen in the adductors.

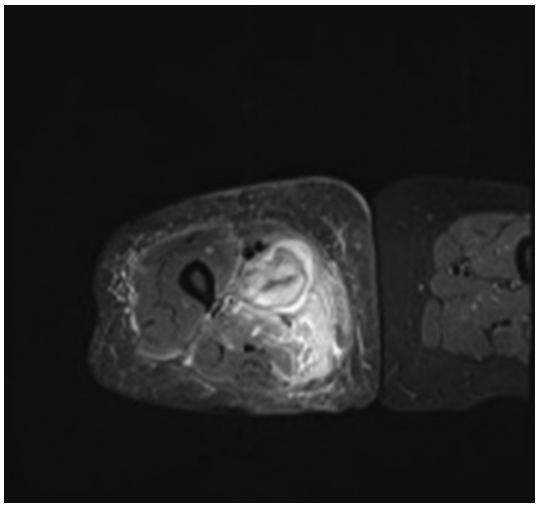

Figure 4. Axial T2-weighted fat-suppressed magnetic resonance imaging of the right thigh showing heterogeneous signal intensities in the adductor muscles. Edema of the adductor muscles and subcutaneous tissue is seen.

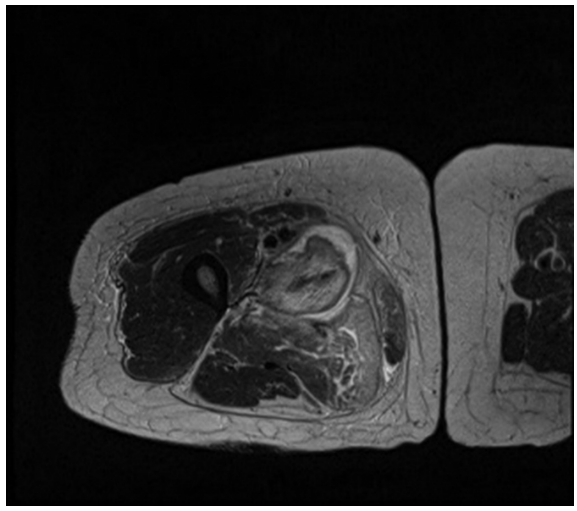

Figure 5. Axial T2-weighted magnetic resonance imaging of the right thigh showing hyperintensities (edema and perifascial fluid) in the adductor muscles.

was controlled. She recovered well and was discharged home after 15 days following this conservative management. Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Discussion

Diabetic myonecrosis is skeletal muscle infarction with clinical presentation of sudden onset of painful swelling of the affected muscle and, occasionally, local swelling. Its pathogenesis is uncertain; however, factors such as diabetic microangiopathy, atherosclerosis, vasculitis with thrombosis, and ischemia–reperfusion injury have been implicated.[3,5] A widely accepted hypothesis is that thromboembolic events from tissue ischemia trigger inflammatory cascades, resulting in tissue necrosis. Reperfusion of necrotic tissues results in reduced nitric oxide and increased oxygen radicals in the endothelial cells. This imbalance results in the production and release of inflammatory mediators such as tumor necrosis factor and platelet-activating factors and increased biosynthesis of adhesion molecules, which causes further damage with endothelial dysfunction.[6] Changes in the coagulation–fibrinolysis cascade have also been implicated in the etiology of DMN as factor VII activity, plasminogen activator inhibitor, and thrombomodulin levels are increased, resulting in hypercoagulability and eventual vascular endothelial damage.[7]

Diabetic myonecrosis has a higher incidence in females with a mean age of presentation between 42.6 and 44.5 years, and in either type 1 or type 2 DM with long-standing and poorly controlled hyperglycemic states.[3,5] A majority of these patients have established multiple microvascular complications of DM such as nephropathy (71%), retinopathy (56%), and neuropathy (54%).[5] Our patient had poorly controlled T2DM complicated with bilateral peripheral neuropathy evidenced by a reduction in her nerve conduction velocity. However, there was no clinical history, signs, or symptoms suggestive of diabetic retinopathy (DR), non-proliferative DR, and nephropathy. Ito et al[8] reported that a reduced nerve conduction velocity may be closely associated with early DR in type 2 DM patients; hence it is a potential independent risk factor and early biomarker of DR. Additionally, the sequential development of neuropathy first, followed by retinopathy, and, finally, nephropathy has been reported in type 2 DM patients[8]; therefore, this patient may develop DR and nephropathy in the nearest future. The differentials of DMN are extensive, and the overlapping symptoms of pain and swelling may easily lead to its misdiagnosis. These include deep vein thrombosis (DVT), superficial thrombophlebitis, pyomyositis, necrotizing fasciitis, hematoma, dermatomyositis, abscess, cellulitis, lymphomas, diabetic amyotrophy, drug-induced myositis, osteomyelitis, muscle rupture, ruptured Baker's cyst, and diabetic lumbosacral plexopathy.[3]

The diagnosis of DMN can be challenging, with laboratory investigations and imaging studies aimed at excluding differentials. Laboratory investigations are inconclusive with no specific marker. There may be leukocytosis, and serum creatine kinase levels may remain normal or slightly elevated as observed in our patient. However, the diagnosis may be possible with a combination of clinical presentation and radiological imaging findings. Findings on ultrasonography and computed tomography scans may be useful in differentiating mimics of DMN but are not diagnostic of DMN. The imaging modality of choice is the MRI, with better sensitivity and anatomical definition and sufficiently excludes other differentials.[9] It shows muscle enlargement and edema, necrosis of the affected muscle in T2W images, and gadolinium enhancement as well as isointense/hypointensity in T1W images as classically observed in this patient.[3,10] Muscle biopsy in DMN diagnosis is usually reserved for uncertain cases with atypical presentation or in treatment failure. This is on account of potential complications that may arise from the invasive procedure.[3] Such complications include post-biopsy infection, poor wound healing, and delayed recovery. Hence, MRI is usually sufficient for a diagnosis.

Diabetic myonecrosis is a self-limiting condition, presenting acutely or evolving over days to weeks. It is usually managed conservatively with bed rest, strict glycemic control, and analgesia, such as low-dose aspirin, and non-steroidal anti-inflammatory drugs if not contraindicated, as seen in this index case.[11] Spontaneous resolution occurs over weeks to months with prompt diagnosis and initiation of therapy. Surgical intervention may be unnecessary, resulting in

exacerbation of symptoms and prolonged hospital stay.[12] Short-term prognosis of DMN is good with poor long-term prognosis due to underlying diabetic microvascular complications. Recurrence is commonly reported in about half of patients either in another group of muscles or the originally affected muscle as seen in our patient.[3,10]

Conclusion

Diabetic myonecrosis is a rare complication of DM and should be considered in poorly controlled long-standing diabetic patients presenting with acute non-traumatic limb pain, swelling, and an occasional palpable mass. It is a reminder that aggressive diabetic control is essential in preventing microvascular complications, which will, in turn, reduce morbidity and mortality. The imaging of choice is MRI, and DMN resolves spontaneously with conservative management.

Informed Consent: Written informed consent was obtained from the patient for publication of this case report and accompanying images.

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