Diabetic myonecrosis: An underreported complication of diabetes mellitus

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ABSTRACT
Diabetic myonecrosis is an underreported complication of long-standing, poorly controlled diabetes mellitus which is usually self-limiting and responds well to conservative management. Patients frequently have microvascular complications, and although short-term prognosis is good, the long-term prognosis is poor. We report four cases of diabetic myonecrosis admitted in a tertiary care hospital.

Key words: Diabetes mellitus, magnetic imaging resonance, myonecrosis

INTRODUCTION
Diabetic myonecrosis is a rare complication of diabetes mellitus which usually present with sudden onset pain of the involved muscle. It affects both type 1 and type 2 diabetic patients who have long duration of diabetes and frequently have other microvascular complications.[1] We report four cases of diabetic myonecrosis admitted in a tertiary care hospital.

CASE REPORTS
Case 1
A 60-year-old male, with history of diabetes for 4 years, regularly on oral hypoglycemic agents, presented with insidious onset pain and swelling of right thigh for 3 months. There was no history of injury or any constitutional symptoms. His body mass index (BMI) was 18.2 kg/m², blood pressure 150/90 mm Hg, and there was pedal edema bilaterally. Right thigh was hot, swollen with induration of medial and posterior part which was tender. There was no fluctuation. He had bilateral nonproliferative retinopathy and signs of sensorimotor polyneuropathy of both lower limbs. His total leucocyte count was 4370/mm³, hemoglobin 9.5 gm/dl, and erythrocyte sedimentation rate (ESR) 60 mm at the end of first hour. His HbA1c was 8.1%, serum creatinine 0.5 mg/dl, and 24 hours urinary albumin 30.8 mg. Ultrasonography of the thigh including doppler study showed subcutaneous edema only. Magnetic imaging resonance (MRI) of the right thigh revealed altered signal intensity involving the muscles of medial and anterior compartment of the thigh. There was predominant involvement of the adductor muscles showing heterogenous increased enhancement and multiple nonenhancing areas in the adductor magnus muscle suggesting myositis with myonecrosis [Figure 1]. The patient was treated conservatively with insulin, analgesics, antibiotics, antihypertensives, and physiotherapy. He recovered well and was discharged after 3 weeks.

Case 2
A 56-year-old male, with history of diabetes for 10 years who was on oral hypoglycemic agents irregularly, presented with pain and swelling of lower portion of left thigh since 3 months. He had pallor, bilateral pedal edema, and hypertension. Lower part of the left thigh was swollen, hot, tender, and firm with pitting edema [Figure 2]. He also had signs of polyneuropathy and proliferative diabetic retinopathy. Glycemic status was poor (RBS on admission 460 mg/dl, HbA1c 17%) with elevated serum creatinine...
level (1.6 mg/dl). MRI of the left thigh revealed altered signal intensity and swelling of vastus medialis and lateralis muscles with increased heterogenous enhancement of the vastus medialis muscle suggesting muscle infarction [Figure 3]. There was also effusion of the femoropatellar joint. He was treated with insulin, broad-spectrum antibiotics, and analgesics besides other supportive treatment. He recovered within 3 weeks of hospital stay.

**Case 3**

A 45-year-old female presented with complaints of insidious onset pain over lower part of right thigh. She gave history of diabetes for 4 years and was on oral hypoglycemics very irregularly. She had hypertension (BP 160/90 mm Hg), distal symmetrical mixed sensorimotor polyneuropathy, and nonproliferative diabetic retinopathy. There was a localized tender and firm oval-shaped lump (5 × 4 cm) on the medial aspect of the right thigh. Glycemic status was poor (RBS at admission 523 mg/dl, HbA1c 12.6%), serum creatinine 1.4 mg/dl, and her 24 hrs urinary albumin was 734 mg. Ultrasonography of the thigh showed ill-defined hypoechoic areas involving vastus group of muscles and there was increased vascularity around the hypoechoic areas [Figure 4]. She was treated with insulin, analgesics, and antihypertensives. She responded well and was discharged after 3 weeks.

**Case 4**

A 40-year-old male presented with pain and swelling of the left thigh for 3 months. He had history of diabetes for 5 years and was on oral hypoglycemic agents with good glycemic control (HbA1c 7.3%). His BMI was 22.3 kg/m², blood pressure 130/80 mm Hg, was afebrile, and had pallor. Left thigh was swollen, tender, and edematous with a diffuse firm feeling. There was no significant inguinal
Diabetic myonecrosis or diabetic muscle infarction is an uncommon manifestation of long-standing and poorly controlled diabetes mellitus.\[1\] It was first described by Angervall and Stener in 1965.\[2\] Since then more than a hundred such cases have been reported. A.J. Trujillo-Santos, in a systemic review of all reported cases of diabetic myonecrosis, found that it was more common in women (61.5% of all cases), in type 1 diabetes (59% of all cases), and in long-standing diabetes (mean duration of disease 14.3 years).\[3\] We are reporting four cases of diabetic myonecrosis admitted in a tertiary care hospital over a period of 2 years. Three of the cases are male and all are type 2 diabetes mellitus with a mean duration of diabetes of 5.75 years. The exact pathogenesis is not known but may involve hypoxia-reperfusion injury, atherosclerotic occlusion, or vasculitis with thrombosis.\[4\] Atheroembolism of small vessels has also been proposed as the possible mechanism. Another proposed theory is the participation of the coagulation cascade leading to hypercoagulability.

The usual presentation is sudden onset pain of the involved muscle. The thigh muscles (usually the vastus group) are most commonly affected but calf muscle can also be affected. One case of upper limb involvement has also been reported.\[5\] Bilateral involvement has been reported in 8.4% cases.\[6\] The affected area is swollen, tender, firm, and edematous with a feeling of a mass. Constitutional symptoms are usually absent. Similar presentation may also be seen in pyomyositis, necrotizing fascitis, deep vein thrombosis, soft tissue abscess, cellulitis, hematoma, and acute compartment syndrome.\[7\] Most patients have long duration of diabetes (mean duration 14.3 years) and poor glycemic status.\[8\] But it has also been reported as the first manifestation of diabetes mellitus in some cases.\[9\] About 97% cases have other microvascular complications, most often being nephropathy.\[10\]

Routine laboratory investigations are not helpful in diagnosis. Total leucocyte count, creatine phosphokinase, and ESR may be slightly elevated. Lack of correlation between muscle involvement and creatine phosphokinase level has lead to delay in medical advice by approximately 4 weeks. MRI is the best investigation for diagnosis. The characteristic features of diabetic myonecrosis in MRI are an increased signal from the affected muscle area in T\textsubscript{1}-weighted, inversion-recovery, and gadolinium-enhanced images and isoointense or hypointense areas on T\textsubscript{2}-weighted images.\[11\] Sonographic findings of diabetic muscle infarction include internal linear echogenic structures coursing through the lesion; an absence of internal motion or swirling of fluid with transducer pressure; and lack of predominantly anechoic areas.\[12\] Computed tomography scan shows diffuse muscular enlargement with diminished attenuation of the affected muscle, increased attenuation of the subcutaneous fat, and thickening of the subcutaneous fascial planes and of the skin.\[13\] Although biopsy has been done in many cases for diagnosis, currently it is not recommended because of potential complications and delayed recovery.\[14\]

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. Kapur et al found that patients who underwent surgery had an average recovery period of 13 weeks compared to 5.5 weeks for those received conservative treatment only.\[15\] Although the short-term prognosis of diabetic myonecrosis is good, the long-term prognosis is poor and most patients die within 5 years.\[16\] Recurrence has been reported in 47.82% cases involving either the same or the other limb.\[17\]

In conclusion, diabetic myonecrosis is an uncommon complication of long-standing poorly controlled diabetes presenting as sudden onset pain over the affected muscle. Patients frequently have microvascular complications. MRI is the investigation of choice. Short-term prognosis is good with most patients recovering within weeks with conservative treatment, but long-term prognosis is poor. Diabetic myonecrosis should be considered in the differential diagnosis for diabetic patients who present with painful swollen muscle.

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