

Case Report

Myonecrosis Masquerading at an Unusual Site: A Diagnostic Dilemma

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ABSTRACT

Background/Objective: Diabetic myonecrosis is a rare complication of diabetes. Thigh and hip muscles are the most common sites while our patient presented with the involvement of calf muscles. The objective of this report is to describe a patient with diabetic myonecrosis of the calf muscles highlighting the need to suspect it at atypical sites also.

Case Presentation: A 65-year-old male, a known case of type 2 diabetes which was poorly controlled, presented with subacute onset right calf swelling and pain for 1 month without any history of trauma or fever. On local examination, right calf was swollen, tender, and indurated with a girth difference of around 3 cm as compared with that of the left calf. Ultrasonography was suggestive of marked muscle edema, whereas magnetic resonance imaging was suggestive of myonecrosis in the calf muscles (gastrocnemius and soleus). Muscle biopsy confirmed the diagnosis of myonecrosis. The patient improved with conservative management and a good glycemic control.

Discussion: Diabetic myonecrosis is a complication of a longstanding uncontrolled diabetes mellitus. This case highlights the fact that though the most common sites of presentation of diabetic myonecrosis are the thigh and the hip muscles but it can present at atypical sites also.

Conclusion: Diabetic myonecrosis is an uncommon but serious complication of diabetes mellitus that requires high clinical suspicion for making the diagnosis. Clinicians should also be vigilant of the atypical sites of presentation and the high chances of recurrence and the long-term mortality seen in such patients.

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Introduction

Diabetic myonecrosis is a rare and often underdiagnosed complication of diabetes mellitus (DM) especially seen in those with poor glycemic control and multiple microvascular complications.¹ It presents with acute or subacute nontraumatic muscle

pain and swelling and is often underdiagnosed and missed. Muscles of the thigh and the hip region are most commonly involved although uncommonly other muscle groups can get affected.^{1,2}

We report a case of diabetic myonecrosis of calf muscles in an elderly male with poorly controlled DM, highlighting the importance of having a high clinical suspicion in cases presenting at atypical sites.

Case Presentation

A 65-year-old male presented with subacute onset right calf swelling for 1 month associated with pain. There was no history of any trauma. It was not associated with fever or any other systemic

Abbreviations: CPK, creatinine phosphokinase; DM, diabetes mellitus; LDH, lactate dehydrogenase.

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symptoms. He is a known case of type 2 DM for 16 years and is on oral antidiabetic drugs and pre-mixed insulin twice a day with a poor glycemic control along with having diabetic nephropathy and neuropathy. On examination, pulse rate was 78/min, blood pressure of 124/76 mm Hg, a respiratory rate of 14/min, and an oral temperature of 98.5°F. On local examination, right calf was swollen with a girth difference of around 3 cm as compared to that of the left calf. The swelling was tender and indurated with a well-defined demarcation of the indurated area.

On biochemical evaluation, patient had a hemoglobin of 13.2 g/dL with a total leukocyte count of 11,500 (65% neutrophils and 22% of lymphocytes) and a platelet count of 2.5 lakhs. Renal function tests showed a creatinine of 1.0 mg/dL and urea of 28 mg/dL. HbA1c was 9.2%. Routine urine examination showed a 4+ glycosuria and traces of protein. Urinary albumin to creatinine ratio was 364.0 µg/mg. C-reactive protein was 5.5 mg/L (normal <5 mg/L), lactate dehydrogenase (LDH) was 480 IU/L (normal 140–280 IU/L) and creatinine phosphokinase (CPK) level of 92 IU/L (normal 20–200 IU/L). Ultrasonography with Doppler study was suggestive of increased muscle echogenicity and marked muscle edema. Magnetic resonance imaging (MRI) of the right calf region showed altered signal intensity

Highlights

- Diabetic myonecrosis is a rare and underdiagnosed complication of diabetes mellitus
- It is seen commonly with poor glycemic control and multiple microvascular complications
- Thigh and hip muscles are the most common sites
- Rarely it can present at atypical sites like the calf muscles and upper limb muscles
- Clinicians should have a high clinical suspicion to diagnose these cases

Clinical relevance

Diabetic myonecrosis is a complication of longstanding uncontrolled diabetes mellitus. This case highlights the fact that though the most common sites of presentation are the thigh and the hip muscles but it can present at atypical sites also and is associated with a high chance of recurrence and long-term mortality.

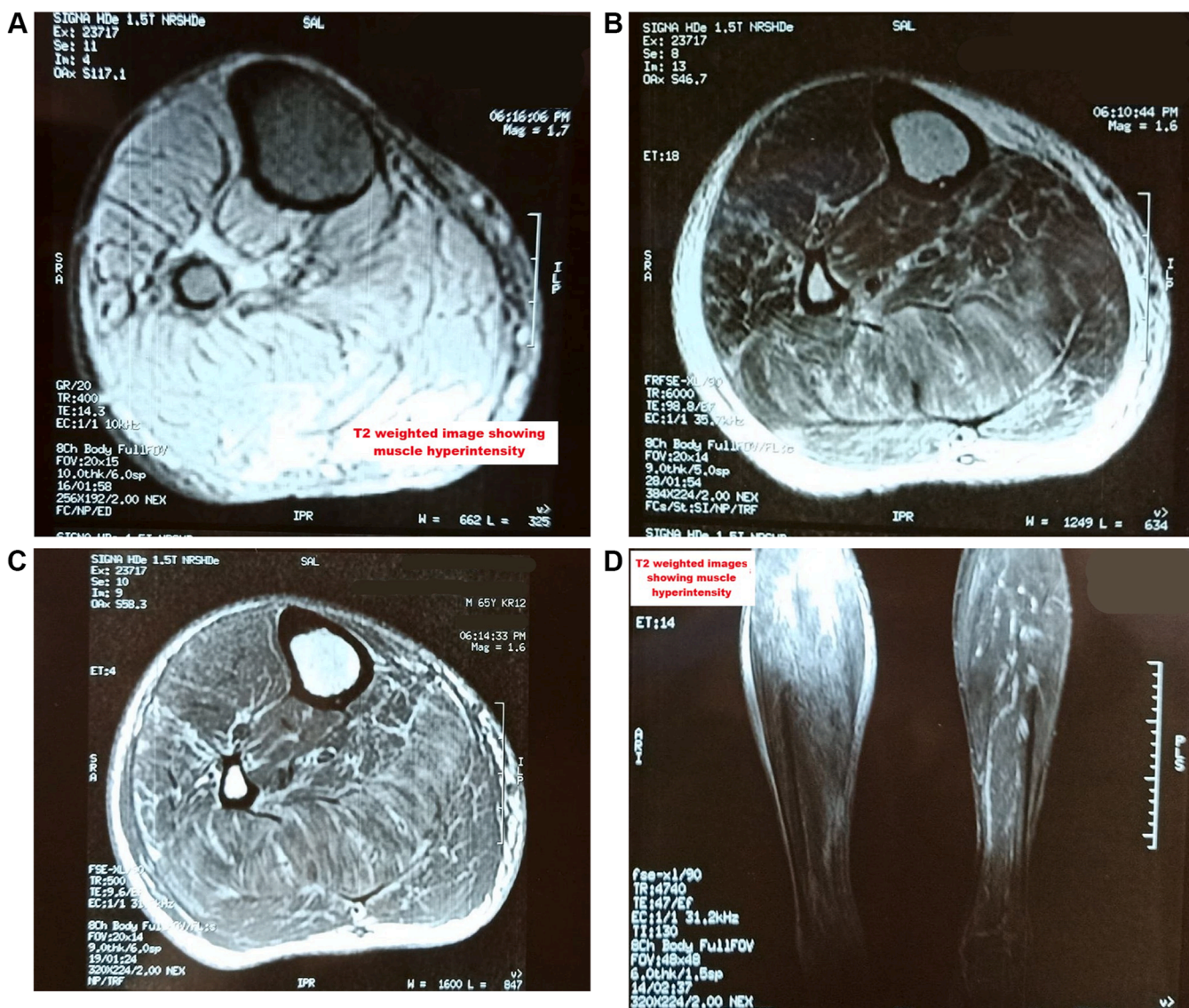


Fig. 1. Magnetic resonance imaging (MRI) images of the right leg.

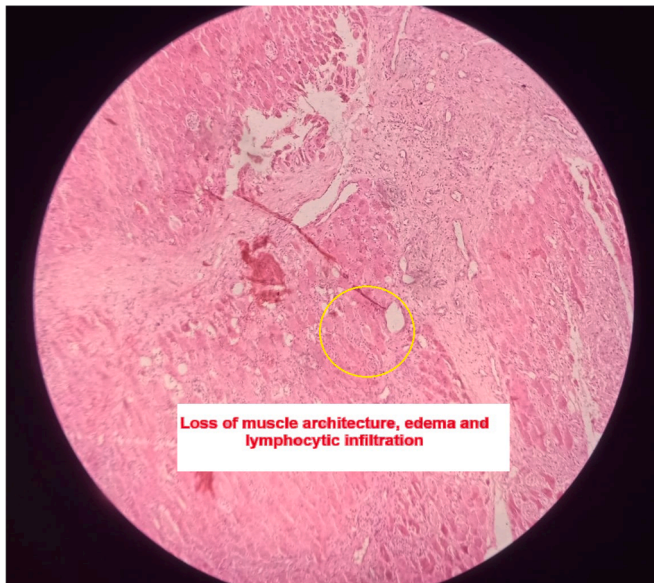


Fig. 2. Histopathology of the muscle biopsy specimen (low power microscopy) showing interstitial edema and loss of normal muscle architecture.

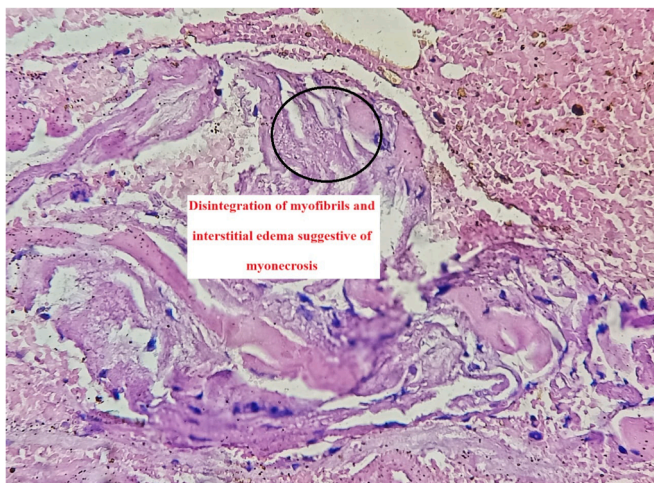


Fig. 3. Histopathology of the muscle biopsy specimen (high power microscopy) showing disintegration of myofibrils and interstitial edema suggestive of myonecrosis.

(hyperintense in T2 and short tau inversion recovery sequences) in both the heads of gastrocnemius and also the soleus muscles with associated hypointense areas on T1 sequence and loss of intramuscular septae suggestive of myonecrosis (Fig. 1). Muscle biopsy was taken and showed features of interfascicular lympho-histiocytic infiltration with presence of few necrotic muscle fibers (Figs. 2 and 3), suggestive of myonecrosis. Gram staining and culture from the suture site were negative.

The patient improved with conservative management, bed rest, and a good glycemic control with multiple subcutaneous injections of insulin and oral antidiabetic drugs.

Discussion

We report a case of diabetic myonecrosis presenting at an uncommon and atypical site in a patient with poorly controlled DM which is an underrecognized complication of diabetes and should

Table

Frequency of Involvement of Various Muscles/Muscle Groups in Diabetic Myonecrosis

Muscles affected	Percentage of involvement (%)
Quadriceps	60-65
Hip adductors	13
Hamstrings	8
Hip flexors	2
Calf muscles	15
Bilateral involvement	10

be differentiated from other similar presenting pathologies, thereby preventing unnecessary invasive procedures.

Diabetic myonecrosis is also known as *diabetic muscle infarction*, *ischemic myonecrosis*, and *aseptic myonecrosis*. It was first described in 1965 by Angervall and Stener as “tumoriform focal muscle degeneration.”¹

It is a rare complication of an uncontrolled, longstanding DM (usually > 15 years) and is therefore frequently ignored and underdiagnosed. The mean age of presentation is around 40 years and is more commonly seen in females and has a higher incidence in patients of type 1 DM, though it can also be seen in patients of type 2 DM. It is often present in patients with multiple micro- and macrovascular complications of DM. Our patient, a known case of type 2 DM, had multiple microvascular complications.²

The patients usually present with an acute or subacute onset of pain, swelling, and tenderness of the involved muscle groups. There is usually no history of any trauma. Signs of systemic infection or inflammation like fever are absent. The pain may be severe enough leading to restriction in movement.³ Our patient also had a similar presentation with involvement of the right calf without any history of fever or any trauma. Most commonly, the muscles of the thigh and hip region (in around 85% of the cases) are involved with quadriceps being the most frequent. Calf region is uncommonly involved (around 15%) and only a very few cases have been reported. The approximate percentages of the frequency of the muscles involved are summarized in the Table.⁴⁻⁶

The differential diagnosis which should be considered in all the cases of diabetic myonecrosis includes deep vein thrombosis, pyomyositis, abscess, neuropathic pain, statin-induced myalgia, fasciitis, and malignancy. Deep vein thrombosis usually presents with a painful and tender swelling associated with edema. Pyomyositis or abscess present with systemic signs and symptoms along with leukocytosis along with the imaging features of fluid/pus collection. Diabetic neuropathy is usually symmetrical and in the form of gloves and stocking distribution while mono-neuropathy presents with a focal sensory and/or motor weakness in the distribution of that particular nerve. Statin-induced myalgia is usually bilateral, gradual in onset without any swelling and responds to statin discontinuation. These clinical features help in differentiating myonecrosis from these differentials.

The exact pathogenesis of diabetic myonecrosis is not yet known, although some theories have been put forward. These include hypoxia-reperfusion injury, diabetic microangiopathy, atherosclerosis, hypercoagulability, arteriosclerosis, and vasculitis among others. The theory which seems to be most relevant and is widely accepted is arteriosclerosis.⁷ Chester and Banker in their study suggested the major role of atheroembolism in the pathophysiology of diabetic myonecrosis. However, in the cases which they had reviewed, no emboli were found and the main protagonist driving the disease was arteriosclerosis obliterans.⁸ The role of antiphospholipid antibodies has also been suggested. Galtier-Dereure et al.⁹ in their study showed that there is a higher prevalence of these antibodies in patients of diabetes with

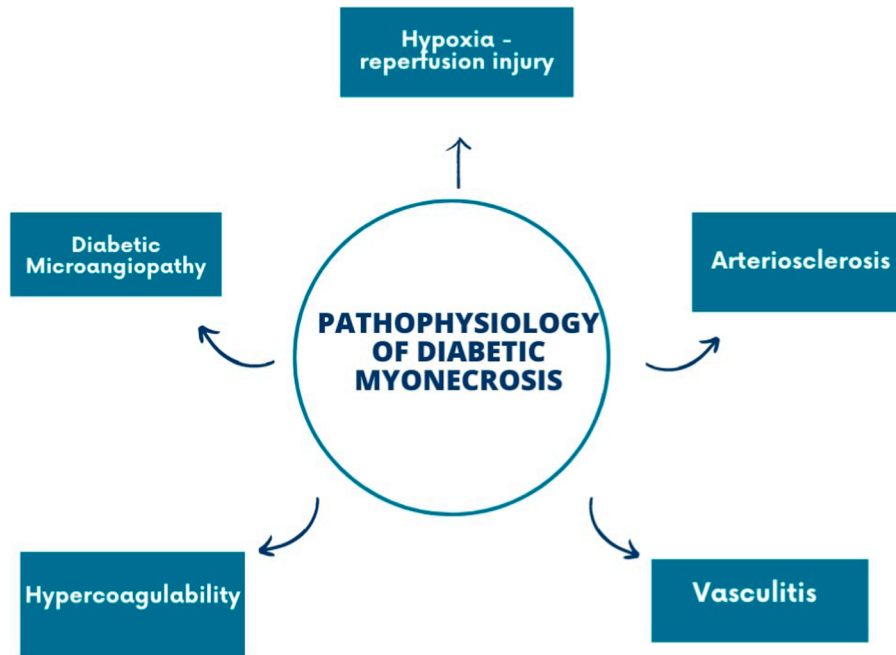


Fig. 4. Pathophysiologic processes involved in the development of diabetic myonecrosis.

macroangiopathy and microvascular complications and as these antibodies create a procoagulable state, they make the patients more susceptible to develop thrombotic complications and diabetic myonecrosis. The main processes involved in the pathogenesis of diabetic myonecrosis are summarized in Figure 4.

Laboratory investigations in patients of diabetic myonecrosis may show mild nonspecific leukocytosis. Erythrocyte sedimentation rate might be increased in up to half of the cases.¹⁰ Muscle enzymes such as LDH and CPK might be mildly elevated in some of the cases. LDH and CPK were within the normal range in our case. First-line radiological investigation is ultrasonography. It helps not only in suggesting a diagnosis of diabetic myonecrosis but also in ruling out other differential diagnosis such as abscess and hematoma.³ Doppler imaging should be done to rule out deep vein thrombosis. The radiological modality of choice is MRI which shows a diffuse enhancement of the involved muscle/muscle groups (best seen on T2-weighted images) with focal areas of low intensity signal suggesting the necrosed part of the muscle.¹¹ Muscle biopsy is not recommended in all the cases as it may be associated with delayed wound healing or chances of hematoma formation or infection.¹² It is reserved in only those cases where there is a diagnostic uncertainty, any atypical presentation or when there is no improvement in the symptoms after proper management. Biopsy findings in cases of myonecrosis show intrafascicular lymphocytic infiltration, necrotic muscle fibers, loss of normal muscle architecture, and interstitial edema. In our case, we decided to go for muscle biopsy because the presentation was at an atypical site and being evaluated at an academic institution. The features seen in muscle biopsy are lymphocytic infiltration, edema, necrosis of the muscle, and fibrosis.²

Management of diabetic myonecrosis is mainly conservative comprising bed rest, aggressive glycemic control, and analgesia. The recovery is spontaneous within 4 to 6 weeks but due to commonly associated diabetic complications and associated role of microvascular thrombosis and hypercoagulability, a high 5-year mortality rate is seen.^{2,10–14} Up to 40 % of the patients may have recurrence.¹⁵

In conclusion, diabetic myonecrosis is an uncommon complication of DM. Although most commonly the muscles of the thigh

are involved, clinicians should also be aware of the atypical sites of presentation, its high chance of recurrence, and the high rate of long-term mortality seen in these patients. Management is mainly conservative and a good glycemic control is a must.

Disclosure

The authors have no conflicts of interest to disclose.

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Statement of Patient Consent

Informed consent was obtained from all the patients.

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