CASE REPORT

Diabetic Myonecrosis as a Rare Cause of Acute Onset Severe Leg Pain in a Patient on Hemodialysis - Case Report and Review of Literature

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Abstract

Diabetic muscle infarction (DMI) or diabetic myonecrosis (DMN) is a rarely reported and commonly underdiagnosed microvascular complication of longstanding uncontrolled diabetes mellitus. The authors report a case of elderly male with type 2 diabetes mellitus, in his mid-fifties who presented with complaints of acute onset, sharp and persistent pain in lateral aspect of the left thigh associated with marked tenderness. The routine laboratory investigation is often nonspecific and do not provide much value in diagnosis of DMI. Magnetic resonance imaging in combination with a proper history can help in diagnosis. Muscle biopsy can help in diagnosis but is usually not recommended as it prolongs the disease process and delays recovery. Treatment is rest, analgesics, and maintenance of optimal glycemic control.

Keywords

Diabetic myonecrosis, Diabetic muscle infarction, Muscle biopsy

Introduction

Diabetic myonecrosis or diabetes muscle infarction (DMI) is a rarely recognized complication of longstanding uncontrolled diabetes mellitus, especially in conjunction with diabetic kidney disease (DKD). Various hypothesis like microangiopathy, ischemic-reperfusion injury, and vasculitis have been variably proposed to describe the pathogenesis. In the absence of specific diagnostic modalities, magnetic resonance imaging (MRI), along with clinical history, recognition of associated risk factors, and muscle biopsy in selected cases can help in diagnosis. Adequate bed rest, proper glycemic control, and judicious use of analgesics can help in management of this condition. Here, we report a case of diabetic myonecrosis and a short review of the literature.

Case Report

A 56-year-old male patient admitted with 10-days history of pain in lateral aspect of left thigh. Pain was constant and sharp in nature, which worsened with movement and was localized to lateral aspect of thigh with marked tenderness. There was no evidence of any skin rash or edema. There was no history of any trauma. Patient had a significant medical history of hypertension and poorly controlled type 2 diabetes mellitus (T2DM) due to incompliance with prescribed treatment. After hospitalization, bilateral proliferative diabetic retinopathy was detected on fundoscopic examination. He was a non-smoker and a non-alcoholic. The blood workup was within normal limits apart from a decreased hemoglobin (Hb) of 7.4 gm% (reference range 13-17 gm%), elevated urea of 128 mg/dl (reference range 13-45 mg/dl), creatinine of 8.5mg/dl (0.5-1.5 mg/dl), creatinine phosphokinase (CPK) of 285 IU/L (reference range 46-171 IU/L), C-reactive protein (CRP) of 160 mg/dl (reference range < 0.3 mg/dl), HbA1c of 13.5 (reference range 6-6.5). During the course of hospitalization, the patient received 2packed red blood cell (PRBC) transfusion and underwent hemodialysis thrice. Left lower limb atrial and venous doppler study showed evidence of iso to hypoechoic lesion involving the entire length of vastus lateralis muscle with internal vascularity on doppler.

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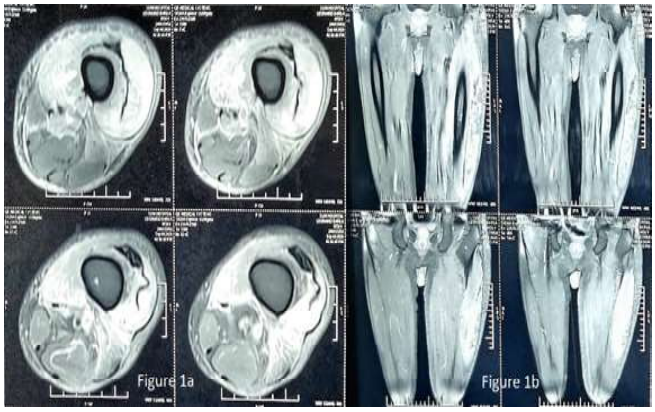

Fig 1. Magnetic resonance imaging (MRI) showing thickened vastus lateralis, intermedius, medialis, adductor longus and brevis muscle of left thigh (Figure 1a) with altered signal intensity and heterogenous post-contrast enhancement without any collection or bone involvement (Figure 1b).

Magnetic resonance imaging (MRI) revealed thickened vastus lateralis, intermedius, medialis, adductor longus and brevis muscle of left thigh (*Fig 1a*) with altered signal intensity and heterogenous post-contrast enhancement without any collection and underlying bone was normal (*Fig 1b*). The disease was managed with adequate rest, non-steroidal anti-inflammatory drugs (NSAIDs), strict glycemic control, and aggressive dialysis.

Discussion

Diabetic muscle infarction (DMI) or spontaneous diabetic myonecrosis is a rarely reported complication of diabetes, associated with ischemic necrosis of skeletal muscles typically of calf or thighs in the absence of occlusion or atheroembolism of major arteries, associated with acute onset painful swelling and tenderness that usually evolve over days to weeks.[1] It is often present with other microvascular complications with nephropathy being the most common association.[2] Exact prevalence is unknown as most of the information has only been obtained from the reported cases.

A systematic review published in 2015 have identified less than 200 cases, with average age of presentation being 45 years.[1] Although the number of patients with type 1 and type 2 diabetes were similar, but the mean age of presentation was lower for patients with T1DM as compared to T2DM (36 years vs 52 years). [1] Also, the mean duration of diabetes at the time of presentation was longer for patients with T1DM vs T2DM (19 years vs 11 years). [1] Most common associated microvascular complication was nephropathy (>75% of cases). An earlier review in 2003 involving 116 patients since the time of first case report found DMI to be more common in type 1 DM patients. [3] A recent systematic review published in 2018 had similar observations, although 81 % patients were on dialysis and recurrence rate was around 43.9%.[4] Although the variable theories of atherosclerosis, diabetic microangiopathy, vasculitis, ischemia-reperfusion injury and hypercoagulable state has been proposed, but none of these factors clearly explain the pathophysiology. [5,6]

Patients usually present with acute onset of swelling and tenderness over muscles of lower limbs (thigh- 55%, calf- 15%) and rarely in the muscles of upper limb in the absence of trauma. [1] Pain may be associated with erythema, induration, persistent mass like swelling and other signs of inflammation. Bilateral involvement is rare (8%), although recurrence is common (61%) and is usually seen involving different muscle groups.[1] When associated with nephropathy or end stage renal disease (ESRD), bilateral presentation is more common (19.5%) and involves multiple muscle groups (39%).[4] Symptom persisted for 2-17 weeks with a mean of 4 weeks. Compartment syndromes, secondary bacterial infections, osteomyelitis are rare presentations. [7]

Lab workup exhibits elevated creatine kinase (CK) levels, usually >700 U/L (reference range <150 U/L), leukocytosis, elevated acute phase reactants (ESR & CRP), and high HbA1c. [8] Magnetic resonance imaging (MRI) is often the investigation of choice. MRI may show high intensity on T2-weighted images as well as subcutaneous edema, and sub-fascial fluid and hypointense or isointense on T1-weighted images because of loss of normal fatty intramuscular septa. [9] Gadolinium contrast can help in distinguishing low signal, non-enhancing infarcted muscle from surrounding edema and inflammation, but is generally avoided in patients with eGFR <15-30 ml/minute, because of increased risk of development of nephrogenic systemic fibrosis (NSF). If gadolinium contrast is indispensable for diagnosis, dialysis must be considered alongside. Rim enhancement can be seen around the necrotic areas within the areas of ischemic muscle. [9,10] Ultrasonography and arteriography have limited role in diagnosis. Muscle biopsy provides definite diagnosis but not currently recommended as it can prolong or complicate the disease process. [11] Biopsy is reserved only for few cases where diagnosis cannot be ascertained, infection cannot be excluded from blood cultures (both aerobic and anaerobic) or the case is refractory to conventional treatment. CT-guided core needle biopsy is preferable to excision biopsy. Biopsy from affected muscles show necrosis and edema along with fibrinous occlusion of affected capillaries and arterioles. [12] Venous doppler ultrasound with compression can be done to rule out deep vein thrombosis. Plain film radiography of the affected area to rule out clostridial myonecrosis is essential.

The common differential diagnosis includes pyomyositis,

spontaneous gangrenous myositis, clostridial myonecrosis, necrotizing fasciitis, venous thrombosis, intramuscular hematoma, neoplasms and calciphylaxis. [13] Patients with pyomyositis usually present with fever, leukocytosis, and well-defined intramuscular fluid collection. Spontaneous gangrenous myositis is more toxic and associated with a worse prognosis and is usually characterized by gangrenous necrosis in the absence of abscess formation. Venous thrombosis is characterized by skin discoloration, and palpable cord-like thrombosed vein in the extremities, and usually diagnosed with compression ultrasonography. Calciphylaxis (calcific uremic arteriolopathy) in a background of end stage renal disease is identified by livedo reticularis, violaceous painful plaque like subcutaneous nodules on trunk, buttock and proximal extremities affecting dermis and subcutaneous fat and very rarely muscles.

In the absence of randomized trials comparing various treatment approaches, optimal glycemic control, low-dose aspirin (75 to 162 mg daily) with adequate bed rest and analgesics are the current recommended treatment for DMI / DMN. Non-steroidal anti-inflammatory drugs (NSAIDs) like naproxen (375 mg twice daily) or ibuprofen (400 to 600 mg three times daily), non-NSAID analgesics like acetaminophen, or opioids like tramadol and codeine can be used for symptom relief, in the lowest dose needed and for the least duration required, because of the associated renal impairment in most of the cases and increased risks of bleeding. [1,8] Physiotherapy should be ideally avoided during acute phase of the disease which usually lasts several weeks. [14] The time to recovery ranges from 8 weeks with rest and analgesics to 5.5 weeks with pharmacological treatment and up to 13 weeks in complicated cases requiring surgical excision. 8 Patient of diabetic myonecrosis (DMN) especially with renal disease are at high risk of recurrence (40%). The long-term prognosis is often poor (2-year mortality of 10%) and death usually results from a major vascular event because of underlying arteriopathy. [8]

Conclusion

Myonecrosis is an uncommon microvascular complication of diabetes that requires a high index of suspicion for diagnosis to prevent the unnecessary biopsies and overzealous use of antibiotics. It should be considered in diabetes mellitus patients who present with acute-subacute onset severe focal muscle pain without systemic symptoms and lengthy duration of the disease with associated microvascular consequences. The most accurate test for diagnosis is an MRI. Only few situations would necessitate a muscle biopsy. The core components of treatment include strict glycemic control, bed rest, and adequate analgesia.

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