CASE REPORT

Diabetic Muscle Infarction, A Rare Complication of a Common Disease

Shaza A. Samargandy, MD and Amani M. Alhozali, MD, SSC-Med
Department of Medicine, Division of Endocrinology, Faculty of Medicine,
King Abdulaziz University, Jeddah, Saudi Arabia

ABSTRACT

Diabetes mellitus is a multifaceted metabolic disease with several serious complications. Diabetic muscle infarction is a rare musculoskeletal diabetic complication that can be frequently misdiagnosed as it shares clinical manifestations with multiple other more common clinical disorders. We present in this report, a case of diabetic myonecrosis with brief discussion about this unusual complication along with its management and expected outcomes.

Keywords

Diabetes mellitus; Muscle; Imaging.
BACKGROUND

*Diabetes mellitus* (DM) is one of the most common metabolic disorders world-wide. It is associated with many complications involving almost every vital structure in the body. We herein report a rare musculoskeletal complication of this common disease which is diabetic muscle infarction (DMI), also known as diabetic myonecrosis. The usual presentation of DMI is acute limb pain and swelling, which might be confused with other more common differential diagnoses.

CASE REPORT

A 51-year-old female patient presented to the emergency department with a 2-month-history of left thigh pain and swelling. The pain was progressive to the point where it started interfering with the patient's sleep and mobility. The patient described the pain as crampy, diffuse with no radiation, and it poorly responded to over-the-counter analgesics including non-steroidal anti-inflammatory drugs. There was no history of skin redness or fever. She also denied any history of trauma. The patient was diagnosed with DM 20 years ago and currently on insulin. Her blood sugar was poorly controlled as implied from her reported home blood sugar readings. She also stated that she was not following up regularly with her primary care physician and that she was recently diagnosed with hypertension but wasn't compliant to treatment.

On examination, she was an albino patient and apart from hypertension she was vitally stable. Lower limb examination demonstrated bilateral swelling and pitting edema up to the mid of the thighs, but more on the left side. The difference in the circumference was 2 cm (left > right) at the thigh region. There was tenderness upon palpation of the left thigh muscles, but no erythema or warmth or dilated veins detected. Palpation of the left calf did not elicit any tenderness and there was no tenderness upon palpating the right lower limb. Neurological examination showed evidence of proximal myopathy with restricted flexion and extension of the left knee due to pain. Glove and stocking neuropathy were also evident. There was mild periorbital puffiness but no swelling at other sites beyond those mentioned. Fundoscopy examination revealed severe non-proliferative diabetic retinopathy with bilateral macular edema.

Investigations showed normal white cell count of 11000 cell/microliter (4.5-11.5), impaired creatinine of 124 umol/L (53-115). Her hemoglobin A1C was 11.2 and 24-hour urine protein was 4.5 g/day. Her creatine phosphokinase level upon admission was normal at 40 IU/L and the maximum reached during her hospital stay was 211 IU/L. Erythrocyte sedimentation rate was high 100 mm/hr. Lipid profile showed evidence of hyperlipidemia. Blood cultures were negative. Doppler US of the left thigh was done and excluded deep venous thrombosis, and only revealed muscle edema. An x-ray of the left femur did not show evidence of fracture or Osteomyelitis. She was admitted to the hospital and empirical antibiotic coverage for cellulitis and necrotizing fasciitis was started. Subsequently a magnetic resonant imaging (MRI) was ordered and showed evidence of increased signal intensity within the left vastus intermedius, vastus lateralis, adductor longus, adductor brevis and the proximal part of rectus femoris with heterogenous enhancement post gadolinium administration suggesting myonecrosis (Fig. 1) and (Fig. 2). Bone scan and gallium scan ruled out osteomyelitis infection at the left femur (Fig. 3).

**FIGURE 1.**
Magnetic resonant imaging of the left thigh, T1 image showing evidence of increased signal intensity within the left vastus intermedius, vastus lateralis, adductor longus, adductor brevis and the proximal part of rectus femoris.
FIGURE 2.
Magnetic resonant imaging of the left thigh, showing heterogenous enhancement post gadolinium administration suggesting myonecrosis. There was no soft tissue collection or evidence of osteomyelitis.

FIGURE 3.
Bone scan showing no localized uptake in the femur bone to suggest osteomyelitis.
Based on the aforementioned information, the diagnosis of DMI of the left thigh was established. Therefore, antibiotics were stopped and the patient was managed with aspirin 81 mg daily and bed rest. She was started on an angiotensin-converting enzyme inhibitor and statin for nephrotic syndrome. During her hospital stay, she started to show improvement in the form of decreased pain at the affected site, but still needed support for mobilization. Her blood sugar was controlled upon discharge.

**DISCUSSION**

In this report, we illustrate one of the rare complications of DM which is DMI or diabetic myonecrosis. The first case report of DMI was in 1965[1]. MEDLINE English-language literature review identified less than 200 cases world-wide. The exact prevalence of DMI is unknown as it might be under-diagnosed and only case reports and case-series have been reported thus far. DMI can be confused with other more common clinical disorders including necrotizing fasciitis, pyomyositis, deep venous thrombosis, calciphylaxis and muscle hematomas. As in this vignette, all the aforementioned clinical differentials are usually ruled out before a confirmed diagnosis of DMI can be reached.

The condition often occurs in patients with long-term, poorly controlled diabetes, especially patients with type1 diabetes. Nevertheless, it was also reported in a 9-year-old boy with Maturity Onset Diabetes of Youth and interestingly, when his blood sugar came under control, he developed another episode of DMI[2].

In a systematic review by Trujillo-Santos[3] of 166 episodes of DMI occurring in 115 patients, it was noted that the average age at presentation was 42.6 years. The mean duration of diabetes at the time of DMI diagnosis was 14.3 years. The exact pathophysiology is not well-understood, but most of the available literature suggests muscle necrosis resulting from arteriolar occlusion. Also, the rise of erythrocyte sedimentation rate and creatinine phosphokinase (CK) seen in the majority of cases indicates a possible inflammatory process. In our patient, CK was normal upon presentation, but rose over the following days. In contrast to rhabdomyolysis, diabetic myonecrosis usually presents with modestly elevated muscle enzymes or even normal levels as seen in this case.

The presence of positive antiphospholipid antibodies in some cases has raised the suspicion of a thromboembolic underlying mechanism, but a direct relationship has not yet been confirmed[4]. The usual presentation of DMI is an acute unilateral lower limb pain and swelling and the thigh is the most common involved site followed by the calf. Bilateral limb involvement has also been reported[5]. During our literature review, we came across less than 10 case reports involving DMI in the upper extremities[6]. The course of the disease evolves over days and sometimes weeks. Fever is uncommon with this diabetic complication and reported only in 10% of the cases[7].

Diagnosis is usually made by appropriate imaging studies after excluding other more common differential diagnoses. Magnetic resonant imaging is the best imaging modality to diagnose DMI and it frequently shows diffuse enlargement of the affected muscle groups and loss of the fatty intermuscular septa along with subcutaneous edema. T2-weighted and gadolinium-enhanced images show hyperintense muscles indicating infarction[7]. Muscle biopsy is supportive of the diagnosis but rarely required. The biopsy usually demonstrates necrotic muscle tissue with hemorrhagic changes and inflammatory cellular aggregates, along with thickened arterioles that are occluded by fibrin[8]. Diabetic muscle infarction can be differentiated from diabetic amyotrophy clinically and by investigations. The latter often occurs in patients with controlled diabetes and presents with muscle wasting. In contrast, patients with DMI are usually patients with poorly controlled diabetes and present with muscular swelling. Electromyographic and MRI examinations can readily discriminate between the two conditions. Magnetic resonant imaging can also help in ruling out other conditions such as necrotizing fasciitis and muscle hematoma or abscesses that can mimic DMI. A simple doppler ultrasound test can also help to exclude deep venous thrombosis.

Considering the rarity of the DMI cases, no clear consensus regarding the management of such cases exists or if controlling blood sugar will prevent further episodes. Treatment is often conservative with bed rest and low dose aspirin daily. The average period till resolution of symptoms was six weeks. Aspirin use and bed rest were associated with an earlier recovery in comparison to bed rest alone (5 weeks and 8 weeks, respectively). Surgical debridement worsens the outcome, prolongs the recovery time and was associated with higher morbidity from local post-operative complications. Data regarding physiotherapy for DMI are contradictory as some reports showed prolonged recovery time while others revealed possible benefits[9].

Prognosis of DMI is poor since increased adverse cardiovascular outcomes were noted in those patients. In a case series of six patients with end-stage renal disease who developed DMI, three died within 2-20 months of diagnosis with a median duration of 10 months. Death was related to infection in 2 of the 3 fatalities and one due to a cardiovascular event[10]. In another case series of six patients, five patients died within 4 years of follow up due to major cardiovascular disease. Interestingly, prognosis in those patients was comparable to patients who suffered a myocardial infarction[11]. Mortality was not directly related to the DMI, but related to macrovascular complications of diabetes and infection. Recurrence is common occurring in approximately half the patients in either the affected or contralateral limb[11].

In conclusion, diabetic myonecrosis is a rare but serious complication that has a presentation that might mimic a lot of common diseases. Recognition and cardiovascular risk adjustment are warranted as it is associated with poor outcome. Adding DMI to diabetes prognostic risk assessments is suggested.

**Conflict of Interest**

The authors have no conflict of interest.

**Disclosure**

The authors have not receive any type of commercial support either in the form of compensation or finances for this study. The authors have no financial interest in any of the products devices, or drugs mentioned in this article.

**Ethical Approval**

Obtained.

**REFERENCES**


الاحتشاء العضلي السكري، مضاعفة نادرة لمرض شائع

شذى أحمد سمرقندى و أماني معتوق الهزلي
قسم الطب الباطني و الجهاز الصمام
جامعة الملك عبد العزيز
جدة - المملكة العربية السعودية.

الخلاصة

مرض السكر هو مرض استقلالي متعدد الأوجه، وله العديد من المضاعفات الخطيرة، والاحتشاء العضلي السكري هو مضاعفة استقلالية عضلية نادرة لمرض السكر. ومن الارد الخطأ في تشخيصها، لأنها تتشابه مع الأعراض الإكلينيكية مع العديد من الأمراض الأكثر شيوعًا، وقد تقدم تقرير تقدم حالة من نفر العضلات السكري النادرة، مع مناقشة مختصرة عنها مصحوبة بطريقة علاجها والنتائج المتوقعة لهذه الحالة.