Recurrent diabetic muscle infarction, a rare complication of diabetes: a case report

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Received 7 October 2016
Accepted 17 January 2017

Egyptian Rheumatology & Rehabilitation 2017, 44:181–184

Diabetic muscle infarction is a rare complication of diabetes mellitus that presents as a localized, exquisitely painful swelling and limited range of motion of the involved extremity. The onset is usually acute, persists for several weeks and resolves spontaneously over several weeks to months without the need for intervention. However, as diabetes mellitus is an immunocompromised state and any painful swelling in the limbs is often taken as infectious in aetiology, the patient is inadvertently investigated with invasive procedures and is started on unnecessary antibiotics, adding to the burden of management. Keeping in view the low threshold for starting antibiotics in painful limb swelling in diabetes mellitus in our setting, we hereby describe a case of recurrent painful diabetic muscle infarction, first involving the right upper and later the right lower limb, managed with physical rest and analgesics. This case emphasizes that the treating physician keep this rare complication of diabetes mellitus in consideration in the respective clinical scenario and adopt a less aggressive (a noninvasive method like ultrasound) rather than a more aggressive (an invasive method like muscle biopsy) approach in diagnosis and take a similar approach towards management.

Keywords:
diabetes mellitus, diabetic muscle infarction, diabetic myonecrosis

Introduction
Diabetic muscle infarction (DMI) is a rare cause of acute severe muscle pain in patients with long-standing diabetes mellitus, with other microvascular and macrovascular complications in many of them, presenting to rheumatologists, endocrinologists, orthopaedic surgeons and physicians. It was first reported in 1965. A systematic review of the literature through August 2001 identified a total of 116 patients [1]. The differential diagnosis includes focal or systemic myositis, localized abscess, haematoma, deep venous thrombosis (DVT), muscle tumour and osteomyelitis. Although the diagnosis can easily be established by ultrasound and/or MRI, definitive diagnosis requires biopsy of the affected area of the muscle. We describe a case of recurrent unexpected acute muscle pain in the right forearm and the right thigh due to DMI in a long-standing type 2 diabetic female patient, with tripathy successfully managed by conservative treatment.

Case report
A 50-year-old woman with a history of type 2 diabetes mellitus of 10 years’ duration complicated by diabetic nephropathy (24h urinary proteins 1 g/24h), nonproliferative diabetic retinopathy and diabetic peripheral neuropathy presented to us with a painful swelling on the right forearm just below the elbow joint. On general examination, the patient was a thin lean woman with a BMI of 18.9 kg/m², haemodynamically stable and afebrile. Systemic examination revealed nonproliferative retinopathy and sensorimotor neuropathy. On local examination she was seen to have a swelling on the right forearm, which was warm and exquisitely tender. She had normal counts, raised acute-phase reactants, erythrocyte sedimentation rate and C-reactive protein, and mild increase in muscle enzyme creatinine phosphokinase. A liver function test revealed mild hypoalbuminaemia, whereas lipid profile and kidney function tests were normal. Serial blood cultures, cryoglobulins, coagulogram, antinuclear antibody and rheumatoid factor were negative.

Doppler ultrasound showed no evidence of DVT; however, musculoskeletal ultrasound of the forearm showed evidence of muscle necrosis with diffuse increase in echogenicity of forearm muscles with tiny necrotic areas. Oedema was also seen extending into the adjoining myofascial planes and vascularity was also increased showing low-resistance flow. An MRI scan of the right forearm showed hyperintensity within muscle...
fibres on T2-weighted image (Fig. 1). Contrast-enhanced MRI showed patchy heterogeneous enhancement of the right forearm muscle fibres. Small peripherally enhancing areas of fluid were also seen within the muscle fibre (Fig. 2). Axial T1-weighted image with contrast of right forearm showed patchy heterogeneous enhancement (Fig. 3). STIR axial images of the right forearm showed diffuse muscle oedema (Fig. 4). The patient did not give her consent for muscle biopsy. The patient was initially started on intravenous antibiotics and analgesics in addition to the treatment for diabetes and hypertension, but later antibiotics were stopped in view of the ultrasound and MRI findings suggestive of muscle necrosis. Her symptoms improved gradually over a period of 2 weeks, and she was discharged after she had been pain-free without analgesics.

Five days later she re-presented with a painful well-defined swelling in the right thigh above the knee joint. On examination she was found to be afebrile and

Figure 1

T2-weighted image showing hyperintensity within muscle fibres

Figure 2

STIR axial images of the right forearm showing diffuse muscle oedema

Figure 3

Contrast-enhanced MRI showing patchy heterogeneous enhancement of the right forearm muscle fibres. Small peripherally enhancing areas of fluid are also seen within the muscle fibre
haemodynamically stable and had a discrete swelling on the anterior aspect of the right thigh, which was warm and tender. Her counts were normal, erythrocyte sedimentation rate and C-reactive protein were increased with a mild elevation of creatinine phosphokinase 219, and plain radiograph of the right thigh was normal. Doppler ultrasonography (USG) of the right thigh showed no evidence of DVT, inflammatory arthritis or abscess. Musculoskeletal ultrasound of the right thigh showed evidence of muscle necrosis consistent with previous ultrasound findings of the right forearm. The patient could not afford a repeat MRI scan and was also deferred because of diabetic nephropathy. However, this time the patient finally consented to muscle biopsy of the thigh. Muscle needle biopsy was performed under USG guidance and the microsection showed viable muscle fibres, few myofibres with necrosis and areas replaced by mature adipose tissue in a background of sparse chronic inflammatory cell infiltrate suggestive of myonecrosis (Fig. 5). The patient was continued on insulin; antihypertensive drugs and analgesics were added. The patient was not started on antibiotics during this admission. Her symptoms gradually improved, and she was discharged and could walk with support.

Discussion
The term DMI also known as spontaneous diabetic myonecrosis is a rare complication of diabetes and is used to refer to spontaneous ischaemic necrosis of skeletal muscles. It causes acute or subacute pain, swelling and tenderness, typically in the thigh or calf, and should be suspected in any diabetic patient. Patients may have mild fever [2,3]. Bilateral involvement occurs in nearly one-third of cases and recurrence at the same or different site(s) in nearly one-half [4]. Rarely, the upper limb may be involved. Neck muscles (levator scapulae) involvement, complicated by staphylococcal sepsis, has been described in an immunosuppressed diabetic transplant patient [5].

It usually affects patients with long-standing and poorly controlled diabetes mellitus and is more common in type 1 diabetes with multiple microvascular complications [2,3]. Various pathogenic mechanisms have been proposed. Diabetic microangiopathy atheromatosis and embolisation of atheromatous material from ulcerated aortic plaques were proposed as the cause of muscle infarction in earlier reports [6,7]. However, only a minority of cases had a vascular occlusion corresponding to the extent of muscle necrosis in later reports, suggesting that initial ischaemic events lead to muscle oedema, which increases the pressure within facial compartments and causes further ischaemia [8]. Hypoxia–reperfusion injury may have an important role in the pathogenesis with the following sequence of events [9].

Compartment syndrome precipitated by small thrombotic/embolic events, producing ischaemic muscle damage and leading to a potent inflammatory response, hyperaemia and reperfusion with
the generation of reactive oxygen species, results in further muscle damage, creating a vicious cycle with extensive muscle necrosis. Creatinine kinase levels may be normal or increased depending on the stage of condition the sample is taken from [4]. USG and MRI have been used to assess patients with DMI, with contrast-enhanced MRI being the most useful diagnostic technique. The presence of linear echogenic structures, the absence of a predominant anechoic area and no evidence of internal motion with transducer pressure discriminate a DMI from an abscess [10]. MRI may show high intensity in the involved muscle on T2-weighted sequences, as well as subcutaneous oedema and subfacial fluid with loss of normal fatty intramuscular septa on a T1-weighted image [3,4,11].

Administration of gadolinium-containing MRI contrast agents distinguishes the nonenhancing infarcted muscle from the surrounding inflammation or oedema, and the contrast should be avoided in patients with impaired renal function to prevent nephrogenic systemic fibrosis [12]. Arteriography, generally not performed for diagnostic purposes, may reveal atherosclerotic luminal narrowing [13]. Muscle biopsy may show muscle necrosis and muscle oedema. Occlusion of arterioles and capillaries by fibrin may also be seen [2,14].

Awareness of the syndrome and the presence of clinical features suggest the diagnosis. Laboratory and imaging studies are aimed to exclude other disorders of acute pain and tenderness, such as pyomyositis, spontaneous gangrenous myositis, clostridial myonecrosis, necrotising fasciitis, venous thrombosis, intramuscular haematoma, calciphylaxis and muscle tumours.

Optimal treatment is uncertain, and rest and analgesics result in recovery within weeks; antplatelets and/or anti-inflammatory drugs are also effective within weeks. Surgical excision may also be needed in some cases [4]. Physiotherapy may cause a worsening of the condition and routine daily activity may often be painful but is not harmful [2,8]. Addition of low-dose aspirin is suggested. NSAID may speed up recovery and should be considered if not contraindicated. Narcotics may be considered in patients with high risk for NSAID adverse effects. The condition resolves spontaneously over a few weeks to months in most patients. Long-term outlook is poor due to the underlying arteriopathy leading to death from a major vascular event occurring within a few years in the majority of patients [2].

The clinical features of our patient closely resemble those of previously reported cases. She had long-standing uncontrolled diabetes with chronic microvascular complications, and she responded to conservative therapy.

**Conclusion**

DMI is an uncommon complication of a common disease and it should be suspected in a patient with long-standing diabetes who presents with a painful swollen limb. Ultrasound and/or MRI can be utilized as an imaging modality without the need for muscle biopsy to establish the diagnosis. Although muscle biopsy is required for definite diagnosis of DMI, it can sometimes complicate its course, and hence it should be utilized only in rare cases. It resolves spontaneously over a few weeks to months with conservative management, including rest to the involved limb and analgesics in most of the patients. The long-term outlook is likely to be poor because of the underlying arteriopathy.

**Financial support and sponsorship**

Nil.

**Conflicts of interest**

There are no conflicts of interest.

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