Bilateral lower limb pain and swelling in a young girl with type 1 diabetes mellitus

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CASE REPORT

The patient was a 20-year-old female with poorly controlled type 1 diabetes mellitus of 7 years' duration complicated by nephropathy, background diabetic retinopathy, dense peripheral neuropathy and symptomatic cardiac and gastrointestinal autonomic neuropathy.

In August 1999, she presented with a 3-week history of focal pain and swelling of the inner aspect of her left thigh. Erythrocyte sedimentation rate (ESR) was 105 mm/hr, C-reactive protein (CRP) 56 mg/litre (reference range <5 mg/litre) and white cell count (WCC) 7.5x10^9/litre (reference range 4–11x10^9/litre). Creatine phosphokinase (CPK) was 431 IU/litre (reference range <200 IU/litre). An ultrasound of the swelling showed a 3 cm diameter abnormal collection in the region of the adductor magnus. A magnetic resonance imaging (MRI) scan of the left thigh (*Figure 1*) showed diffuse high signal and swelling in the left adductor magnus muscle and to a lesser extent in the adductor brevis. The subcutaneous tissues were also oedematous. No discrete haematoma or abscess cavity was seen.

A core biopsy of the lesion showed severe degenerative changes with chronic inflammation and occasional giant cells (variant of myocytes) and was reported as showing 'active myositis'. This was therefore treated as focal myositis with prednisolone 30 mg daily, which was gradually weaned down over the course of 2 months, as she continued to improve clinically. Her clinical remission was paralleled by a normalization of ESR and CPK.

In October 2000, she presented with a sudden onset of bilateral shin pain associated with mild pitting oedema, worsening over 2 weeks. There was no history of prior strenuous exercise or trauma. She was apyrexial. The anterior tibial compartments appeared tight. There was no obvious weakness. Her pain worsened on plantar flexion of her feet. Her calves were non-tender. There were no ankle reflexes. Posterior tibial pulses were palpable but not the dorsalis pedis. Sensory testing confirmed the presence of neuropathy. Her blood investigations revealed normal WCC, CPK, ESR and CRP. X-rays of her lower limbs were normal. She was treated as having musculoskeletal pain and sent home with simple analgesia. A week later, she re-presented at casualty with similar complaints. A diagnosis of musculoskeletal pain was again made, and she was sent home with stronger analgesia, with outpatient follow-up at the diabetic clinic to be arranged.

By the following week, she was unable to weight bear because of increasing pain. Clinically, the lower limb oedema appeared marginally worse, otherwise her signs remained unchanged. Her WCC was 9x10^9/litre, CPK 82 IU/litre, CRP 30 mg/dl, but ESR was substantially elevated at 101 mm/hr. Fasting glucose was 23 mmol/litre and glycosylated haemoglobin (HBA1c) 10%. A full connective tissue screen yielded negative results. Doppler sonography and duplex scanning of the lower limb veins and arteries respectively excluded major venous and arterial pathology. After a week of bed rest, leg elevation, regular analgesia and better glycaemic control, there was a noticeable improvement in her condition.

A MRI scan of her lower limbs (*Figure 2*) showed high signal intensity in the long T2-weighted images of both anterior tibial compartments with similar but less prominent features in the gastrocnemius muscles. T1-weighted images showed no significant signal abnormalities.

On clinical (based on previous history of 'focal myositis') and radiological grounds, an initial diagnosis of polymyositis was made, and the patient started on prednisolone 30 mg daily. A muscle biopsy was thought unnecessary, as she continued to make good recovery. The diagnosis of diabetic muscle infarction was reached retrospectively. Her leg pain had resolved completely and oedema subsided 2 weeks after discharge. The steroid was stopped after 2 weeks.

On review of her previous biopsy and MRI scans, and following literature review, it was thought that she most likely had, in 1999, a first presentation of diabetic muscle infarction in the left adductor magnus muscle, which was misdiagnosed as focal myositis, and a year later, a recurrence in different sites.

INTRODUCTION

Acute lower limb pain and swelling may be the consequences of deep venous thrombosis, pyomyositis, myositis, compartment syndrome, haematoma, neoplasm or muscle infarction. This article presents a case of bilateral shin pain in diabetes mellitus resulting from diabetic muscle infarction (DMI).

DISCUSSION

DMI is a rare but recognized complication of diabetes mellitus. It mainly affects longstanding poorly controlled type 1 diabetes mellitus complicated by vasculopathy, such as retinopathy, nephropathy and/or neuropathy (Lafforgue et al, 1999; Grigoriadis et al, 2000). It was also reported in type 2 diabetes mellitus (Scully et al, 1997), but only one case of idiopathic muscle infarction has been reported in a non-diabetic (Lafforgue et al, 1999). This patient, at 19 years of age when she first developed DMI, is among the youngest patients reported.

Patients typically present with acute or subacute atraumatic pain and/or swelling of the lower limb (Lafforgue et al, 1999). The pain can be very severe, worsened by flexion or extension of the limb and improved with immobilization. Although the quadriceps are most often affected (62%), DMI of hip adductors (13%), calves, hamstring, anterior tibial muscles and gastrocnemius-soleus has been reported (Barton and Palmer, 1993; Scully et al, 1997; Grigoriadis et al, 2000), with recurrences occurring in

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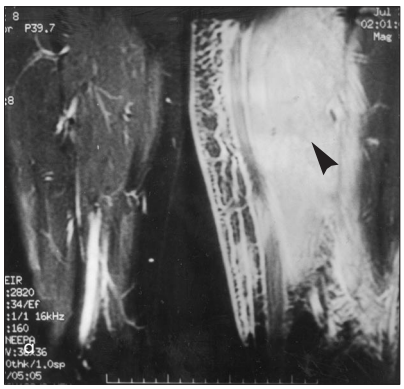


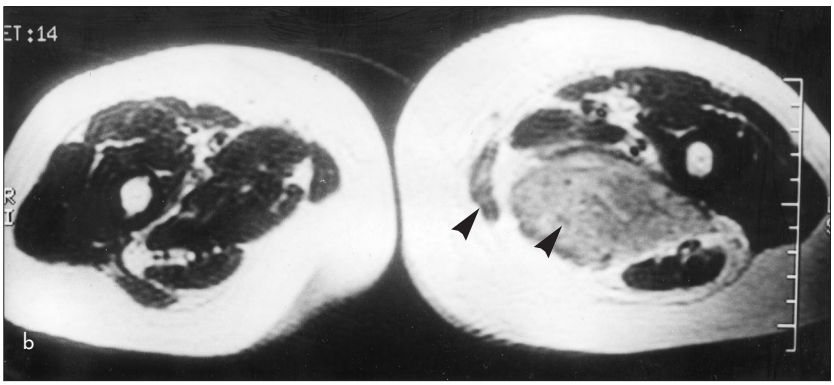
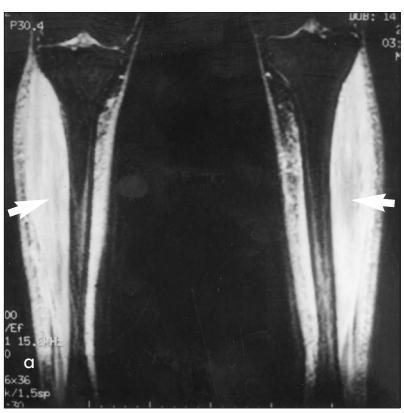


Figure 1. a. Coronal and (b) transverse views of the left thigh by T2-weighted magnetic resonance imaging scan. There is abnormal high signal intensity within the adductor magnus and adductor brevis (arrowhead) without loss of the fascial planes.

the same or contralateral limb in 51% (Grigoriadis et al, 2000).

Laboratory investigations generally show high erythrocyte sedimentation rate, normal white cell count and normal or slightly increased creatine phosphokinase (Scully et al, 1997; Lafforgue et al, 1999). Creatine phosphokinase could be moderately elevated during muscle reperfusion (Van Slyke and Ostrov, 1995) as seen during this patient's first presentation.

Plain radiographs and Doppler ultrasound (apart from excluding deep venous thrombosis) are unhelpful (Lafforgue et al, 1999; Grigoriadis et al, 2000). Radionuclide bone scan may show non-specific increased uptake in the necrosed muscles but should only be performed when a soft tissue tumour or infection with bony involvement is suspected (Lafforgue et al, 1999).



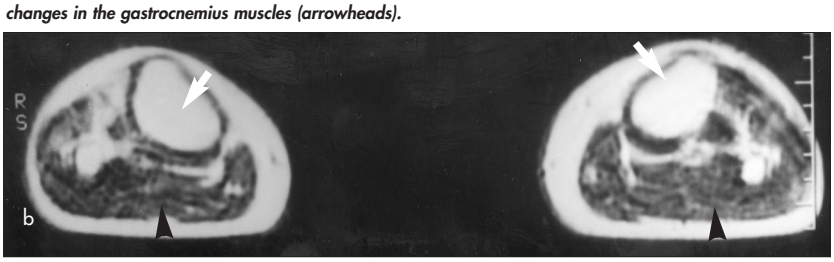
Computed tomography scan could be normal or show enlargement of a muscle group with normal or lower density (Lafforgue et al, 1999; Grigoriadis et al, 2000). Ultrasonography may be helpful in localizing the lesion, with certain characteristics differentiating DMI from abscess or tumour (Delaney-Sathy et al, 2000; Grigoriadis et al, 2000). The imaging modality of choice is magnetic resonance imaging (MRI), which localizes the infarcted muscle(s) precisely (Heureux et al, 1998; Lafforgue et al, 1999; Grigoriadis et al, 2000; Spengos et al, 2000; Silberstein et al, 2001).

Typical MRI findings are increased signal intensity of the enlarged muscle in T2-weighted and short tau inversion recovery (STIR) images, signalling a high water content (Van Slyke and Ostrov, 1995; Scully et al, 1997; Lafforgue et al, 1999; Grigoriadis et al, 2000; Spengos et al, 2000; Silberstein et al, 2001). There are a number of factors which may help to differentiate DMI from polymyositis. These include its localized nature, together with the frequent presence of oedema in subcutaneous and/or perifascial spaces (Lafforgue et al, 1999; Grigoriadis et al, 2000), and rim enhancement around areas of non-enhancement within the infarcted muscle(s) post contrast injection on T1-weighted images (Grigoriadis et al, 2000). The absence of a discrete mass on MRI (Silberstein et al, 2001) would seem to rule out pyomyositis or abscess. MRI may also depict concomitant changes in ipsi- or contralateral muscles which appeared clinically normal (Van Slyke and Ostrov, 1995; Lafforgue et al, 1999).

However, in spite of the potential diagnostic difficulties, the constellation of typical clinical features of DMI in the absence of fever, skin redness, weakness or raised white cell count, together with suggestive MRI findings, are sufficient to secure this diagnosis in a diabetic patient (Van Slyke and Ostrov, 1995; Lafforgue et al, 1999; Grigoriadis et al, 2000).

Although a proper muscle biopsy would be confirmatory, it is unnecessary in patients with typical clinical and MRI features of DMI but recommended for those with atypical presentation or inconclusive MRI findings (Grigoriadis et al, 2000). Histological findings are not totally specific either because of the patchy nature of the lesion. Biopsy may show various stages of muscle necrosis and degeneration, chronic inflammation, oedema,

Figure 2. a. Coronal and (b) transverse views of T2-weighted magnetic resonance imaging of the lower limbs. There is abnormal high signal intensity in the anterior tibial muscles (arrows) with less prominent changes in the gastrocnemius muscles (arrowheads).



haemorrhage, interstitial fibrosis, myofibre regeneration, arteriolosclerosis and active denervation (Lafforgue et al, 1999; Grigoriadis et al, 2000). Indeed, earlier reports commented that excisional biopsy and early ambulation may exacerbate DMI (Barton et al, 1993; Kiers, 1995; Lafforgue et al, 1999; Spengos et al, 2000).

DMI is considered a result of diffuse microangiopathy based on previous autopsy studies and arteriography (Lafforgue et al, 1999; Spengos et al, 2000). A newer concept suggested hypoxia-reperfusion injury to be important in the pathogenesis of DMI (Silberstein et al, 2001).

Correct management involves bed rest, non-weight-bearing, regular analgesia and good glycaemic control (Lafforgue et al, 1999; Grigoriadis et al, 2000; Silberstein et al, 2001). Patients usually recover spontaneously within several weeks.

CONCLUSION

DMI is increasingly recognized as a complication of poorly controlled diabetes mellitus. Important differential diagnosis included muscle neoplasm, abscess, myositis or deep venous thrombosis. Increasing awareness of this syndrome in the right clinical context with typical MRI findings should lead to prompt diagnosis and management. HM

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IN THE PUBLIC'S VIEW...

How long is a piece of string?

An idea isn't a good one just because lots of people think it is. The government has declared its intention to publish figures on the performance of individual doctors. We are told the public want it and that it is their right to know. When this idea was first mooted, UK doctors were almost all against it. Predictably, politicians and the media rejected our criticisms as vested interest.

The atmosphere has changed. Doctors have ceased complaining so loudly. Appraisals have started, and doctors are getting used to having their clinical work examined. More recently, the tone of the British Medical Association (BMA) has softened. We are told that most doctors are changing their view to accept that the public should have information about individual performance. The BMA's tone has shifted from outright objection to grudging compliance, adhering to the pragmatic principle that, if it is inevitable, we must ensure it will be valid.

But the idea is not a good one. Performance cannot be measured in a fair and meaningful way. It matters not a jot what the public want, what the politicians intend or what the BMA (and other organizations of influence) think doctors think. Doctors have ceased complaining because they've got complaint fatigue and patients to treat. But the talk in the postgraduate dining rooms shows we still recognize a bad idea when we're shown one.

Even for the hard outcomes of cardiac surgery, the ranking of individual performance is an invidious process. How to measure the performance of a dermatologist? A child psychiatrist? An anaesthetist? My performance cannot be judged without considering the surgeons I work with, the recovery wards I send my patients to or the wards they come from. What sort of a formula will be needed to correct for all that? What measure would put me in the top flight? The number of cannulae I site at the first attempt? How often I use epidural anaesthesia?

The media response to the Audit Commission's report into accident and emergency (A&E) departments told us all we need to know about whether interpretation of performance data will be fair. Despite a 10% increase in the number of doctors over the last 5 years, A&E departments are performing less well. Not all media commentators mentioned the 7% annual increase in medical admissions, or the increased blocking of beds by immovable medical patients. The response of some of the hospitals criticized by the Commission for Health Improvement is interesting too. Some have justifiably complained that the wrong or invalid data were collected, or that interpretation of the data was flawed. Early on, when the 'drive for quality' mantra was new on everyone's lips, there were warnings that none of the proposals intended to improve quality would succeed unless information was first available and second valid. There is undoubtedly much more information in the NHS now than a few years ago, but too much is of doubtful validity.

We are auditing; we are being appraised; specialist medical societies are keeping an eye. We should cooperate. We should have nothing to do with the publication of individual performance data. They will be meaningless and mismanaged, and our leaders should have the guts to say so. HM

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