CASE REPORT

Acute exacerbation of previously undiagnosed chronic focal myositis in an Aboriginal patient on maintenance haemodialysis

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SUMMARY

We describe a haemodialysis patient who presented with an exacerbation of previously undiagnosed chronic focal myositis during a hospital admission for missed dialysis and chronic foot osteomyelitis. The association of focal myositis with haemodialysis has been reported once previously, but we report the third case in our experience and argue that it is probably more common than previously appreciated. We consider a focused differential diagnosis for a diabetic dialysis patient with leg pain and discuss important features of this rare condition.

BACKGROUND

Leg pain is a common symptom in haemodialysis patients and has a broad differential diagnosis. We present a maintenance haemodialysis patient with focal myositis of the right hip. This case adds to a previously reported case of focal myositis associated with haemodialysis, and two other unreported cases in our department's experience.

CASE PRESENTATION

A 49-year-old Aboriginal Australian woman with end-stage renal failure (ESRF) secondary to biopsy proven diabetic nephropathy presented with dyspnoea, having missed three episodes of maintenance haemodialysis. She had recently taken leave from the hospital, where she was being treated for osteomyelitis of her right foot, in order to attend a funeral some 900 km away. This infection was an ongoing complication of a forefoot amputation 12 months earlier.

Three days into her admission, following otherwise uneventful stabilisation on haemodialysis, she complained of a sore right hip and buttock. The pain was acute in onset, and was of variable severity. Consequentially she was reticent to walk and spent increasing time in bed. She reported no arthralgia or any systemic symptoms except fevers. She denied incurring any injury. At this time she was taking her usual medications, which included atorvastatin for hypercholesterolaemia, in addition to piperacillin/tazobactam for the ongoing infection, which was restarted on admission. Other than a painful hip, her admission was complicated by the development of a lower respiratory tract infection due to extended spectrum β lactamase *Escherichia coli*, which resolved after treatment with meropenem.

On examination, the proximal region of the right upper leg and the right buttock were warm and indurated. There was no spreading erythema,

dissuading us of infective cellulitis, and other than a generalised swelling, no circumscribed masses. The range of movement of the right hip joint was normal and a neurological examination of the lower limbs was unremarkable. On further questioning, she had experienced mild localised right hip pain for slightly over a year, for which she had not sought medical advice. This acute episode of pain was treated with paracetamol and oxycodone, when required.

Initial investigations revealed anaemia (76 g/L), mild leucocytosis ($11.1 \times 10^9/L$), raised C reactive protein (89.9 mg/L), and uraemia (31.1 mg/dL), consistent with chronic osteomyelitis in the context of ESRF. The creatine kinase (CK) level was normal. The sodium and potassium concentrations were normal.

An ultrasound scan of the hip revealed oedema of the subcutaneous tissues, with patchy areas of increased echogenicity in the musculature without any focal collections or evidence of an abscess.

Subsequently, MRI of the hip demonstrated extensive signal abnormality of the right gluteal musculature consistent with an inflammatory process (figure 1A, B). Concurring with the ultrasound results, there was no abscess or collection.

To establish a firm diagnosis, a biopsy of the right buttock musculature was arranged. This showed a mild interstitial mononuclear inflammatory cell infiltrate with features of oedema, consistent with focal myositis. There was no neutrophilic infiltrate in the musculature, arguing against the presence of a bacterial infection (figure 2A, B).

Prior to the start of prednisolone, we conducted an autoimmune screen, which demonstrated a weakly positive antineutrophil cytoplasmic antibodies immunofluorescence, which stained with a cytoplasmic pattern. Anti-myeloperoxidase, anti-PR3, anti-double-stranded DNA, antismooth muscle, antinuclear antibody and rheumatoid factor titres were not elevated. Furthermore, complement levels were within the normal range.

Treatment with prednisolone 50 mg daily led to a gradual improvement in pain, but was complicated by poor glycaemic control. The steroid dose was tapered, and the patient underwent rehabilitative physiotherapy making a good recovery.

DISCUSSION

A patient with diabetes with ESRF presenting with leg pain bears a broad differential diagnosis. This has been discussed in detail elsewhere.¹ Particularly

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Figure 1 MRI of the right hip and thigh demonstrating extensive signal abnormality involving the gluteal musculature and extending superiorly along the posterior aspect of the gluteus maximus attachment. There is overlying inflammation of the subcutaneous tissue. Arrows indicate inflamed musculature: (A) coronal section and (B) axial section.

important to this case was the consideration of infective myositis or abscess formation, given the presence of distal osteomyelitis. In this context, unilateral leg swelling should prompt the consideration of two additional very important diagnoses, first, a proximal deep vein thrombosis and second, a neoplastic lesion. Neither of the diagnoses were present on investigation with ultrasound and MRI in this case. Interestingly, however, a case of focal myositis has been reported in which inflammation surrounded metastatic lesions to the muscle tissue.² Given that our patient exhibited renal as well as retinal microangiopathy, diabetic muscle infarction was also an important consideration.^{3 4} Calciphylaxis is another microangiopathy to consider. A rare complication of haemodialysis, it is characterised by necrotising, ulcerating cutaneous lesions. Unusually it can present with subcutaneous non-ulcerating plaques.⁵

Focal myositis is an uncommon diagnosis in a patient with leg pain. It is a distinct and rare inflammatory myopathy usually confined to one muscle, typically of the leg. It presents clinically as a painful, swollen muscle mass. It is largely a diagnosis of exclusion characterised histopathologically by relatively non-specific inflammatory changes with additional interstitial fibrosis and myofibre degeneration.⁶

Focal myositis is an infrequent side effect of statins, and it is notable that our patient was taking atorvastatin throughout the course of her illness. Muscle pain is a fairly frequent side effect of statins occurring in 1–5% of patients.⁷ Statins typically produce elevated CK levels, even without clinically significant myositis as a result of myolysis.⁷ Given that our patient's CK

Figure 2 Histological examination of a biopsy from the right buttock showing inflammatory changes consistent with myositis. There is muscle fibre separation due to oedema and a mild interstitial mononuclear inflammatory cell infiltrate, with occasional eosinophils. Immunohistochemical staining was not undertaken: (A) H&E stain, $\times 20$ and (B) H&E stain, $\times 10$.

level was not markedly elevated, we consider it unlikely that atorvastatin was a significant contributory factor.

The clinicopathological entity of focal myositis has previously been reported in one haemodialysis patient. Revaz *et al*¹ described focal myositis affecting the peroneus longus and brevis muscles in a haemodialysis patient, which responded rapidly to a short course of prednisolone and which did not progress to a systemic inflammatory myopathy. Two further cases involving myositis have been reported in haemodialysis patients: a case of dermatomyositis treated with intravenous immunoglobulins⁸ and a case of septic myositis.⁹

The experience of our department is that haemodialysis-associated focal myositis is more common than reflected in the diminutive extent of this literature. We have managed two similar cases prior to the present case, one of whom required treatment with azathioprine and mycophenolate, and therefore believe that focal myositis in association with haemodialysis may be an under-reported condition. Our patient presented with a flare of chronic focal myositis following missing haemodialysis. It is difficult to identify a trigger for this flare.

Focal myositis is a benign condition that often resolves spontaneously. This course is likely accelerated by a short period of corticosteroid treatment. In very rare cases focal myositis is a misdiagnosis of another inflammatory myopathy such as

polymyositis with focal onset. It is, therefore, important to follow these patients closely, bearing in mind the risk of progression to polymyositis.^{10 11}

In summary, we report a case of chronic focal myositis in a patient with ESRF on haemodialysis, which flared up shortly after restart of haemodialysis. The association of haemodialysis and focal myositis is likely under-reported, and nephrologists should be vigilant to this unusual diagnosis.

Learning points

- ▶ The differential diagnosis for leg pain in a diabetic haemodialysis patient is very broad and includes some rare but important conditions.
- ▶ Focal myositis is an uncommon condition, and an association with haemodialysis may be under-reported.
- ▶ Focal myositis may progress to systemic inflammatory myopathy, so vigilant follow-up of patients is required.

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Competing interests None.

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REFERENCES

- 1 Revaz S, Theumann N, Lohrman JA, *et al.* Leg pain due to bilateral focal recurrent myositis in a hemodialysis patient. *Am J Kidney Dis* 2005;45:E7–11.
- 2 Uppal SS, Salopal TK, Singh H. Left gluteal focal myositis in a patient with signet ring adenocarcinoma of the stomach: not a paraneoplastic phenomenon. *Rheumatol Int* 2004;24:365–7.
- 3 Madhan KK, Symmans P, Te Strake L, *et al.* Diabetic muscle infarction in patients on dialysis. *Am J Kidney Dis* 2000;35:1212–16.
- 4 Lentine KL, Guest SS. Diabetic muscle infarction in end-stage renal disease. *Nephrol Dial Transplant* 2004;19:664–9.
- 5 Fine A, Fleming S, Leslie W. Calciphylaxis presenting with calf pain and plaques in four continuous ambulatory peritoneal dialysis patients and in one predialysis patient. *Am J Kidney Dis* 1995;25:498–502.
- 6 Smith AG, Urbanits S, Blaivas M, *et al.* Clinical and pathologic features of focal myositis. *Muscle Nerve* 2000;23:1569–75.
- 7 Thompson PD, Clarkson P, Karas RH. Statin-associated myopathy. *JAMA* 2003;289:1681–90.
- 8 Finielz P, Gendoo Z, Chuet C, *et al.* Dermatomyositis in a patient under chronic hemodialysis: treatment with intravenous immunoglobulins. *Ann Med Interne (Paris)* 1994;145:148–9.
- 9 Papoulidou F, Giannopoulos G, Pliakogiannis T, *et al.* Septic myositis in a chronic dialysis patient. *Clin Nephrol* 2001;56:175–7.
- 10 Heffner RR Jr, Barron SA. Polymyositis beginning as a focal process. *Arch Neurol* 1981;38:439–42.
- 11 Flaisler F, Blin D, Asencio G, *et al.* Focal myositis: a localized form of polymyositis. *J Rheumatol* 1993;20:1414–16.

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