LETTERS

Necrotising myositis in Behçet's disease: characteristic features on magnetic resonance imaging and a review of the literature

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Myositis is rarely associated with Behçet's disease. We report such a case with characteristic magnetic resonance imaging (MRI) findings, and review the literature.

CASE REPORT

A 29 year old man was first admitted to Matsunami General Hospital because of high fever and muscle pain of both lower legs, finally resulting in him being unable to walk. Painful multiple subcutaneous nodules of both lower legs and the left arm were seen. There was no history of trauma. Total leucocytes, erythrocyte sedimentation rate, and C reactive protein were raised. The serum creatine kinase value was normal. An MRI study of the lower legs (fig 1) showed a focal mass-like lesion, about 3 cm in diameter, in the left gastrocnemius muscle with a decreased intensity on a T_1 weighted image compared with that for normal muscle. Gadolinium enhanced T_1 weighted images showed a well defined rim of contrast enhancement and a hypointense central area. An axial T_2 weighted image showed bright signal intensity in and around the focal mass-like lesion. The same MRI findings were seen in the other nodules of the lower legs. Computed tomography (CT) did not disclose the focal mass-like lesion. Antibiotics were not effective. The symptoms and multiple nodules resolved spontaneously about one month after admission, and the patient was discharged.

One month after discharge, he was admitted to our hospital because of a relapse, with similar symptoms. Painful multiple subcutaneous nodules of both lower legs, in different areas from those of his previous admission, were found. MRI findings of the mass lesions were similar to those of the previous admission.

On admission the patient had polyarthritis and skin lesions. Recurrent aphthous ulcerations had been noted over the previous two years. Pathergy testing was positive. A skin biopsy was performed and showed thrombophlebitis. HLA-B51 was positive. From these results, Behçet's disease was diagnosed. A biopsy of a nodule from the left gastrocnemius muscle was carried out. Examination of the muscle biopsy specimen obtained from the nodular lesion showed an inflammatory granulation predominantly with an infiltration of neutrophils and macrophages, associated with focal central necrosis of the muscle and perivasculitis in the surrounding muscular tissue. A culture of the tissue specimen was negative for bacteria. These findings were consistent with necrotising myositis.

The symptoms and multiple nodules of the legs resolved spontaneously. After discharge, colchicine was given, and no painful multiple subcutaneous nodules have reappeared.

DISCUSSION

We reviewed nine cases of Behçet's disease with myositis reported in English[1–9] and the present case (table 1). Three were generalised and seven were localised myositis. Painful multiple nodules were not described in the cases. All of the localised cases involved the legs. In our case the histological findings were similar to most of the other reported localised cases; it seems possible that vasculitis as a component of Behçet's disease may participate in the pathogenesis of myositis.

MRI has proved to be better than CT scans for the detection of soft tissue diseases—notably, muscle disorders, but was not described in the cases reviewed above. In diabetic muscle infarction and pyomyositis, a gadolinium infusion showed a slightly enhanced rim and a dark central area in T_1 weighted images. Our case suggests that radiological differentiation among these lesions is difficult. A prompt biopsy and a cell culture should be carried out.

Colchicine may be useful for treating genital ulcers, erythema nodosum, and arthritis of Behçet's disease, especially in women.[10] In the cases reviewed here, only one patient

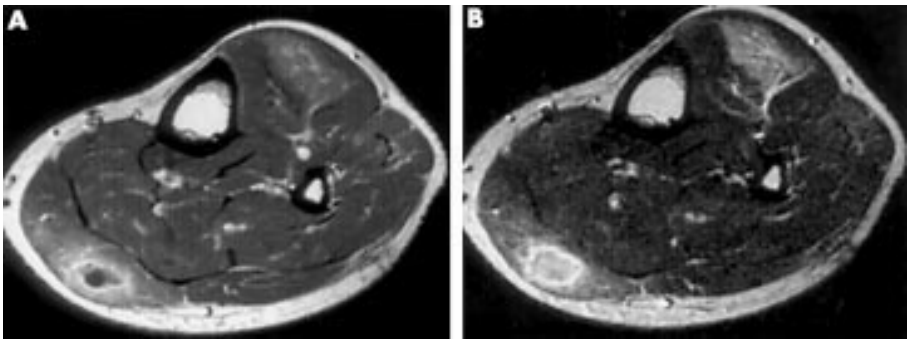

Figure 1 MRI of the lower legs was performed on the patient's first admission. An axial T_1 weighted image showed a focal mass-like lesion, about 3 cm in diameter, in a left gastrocnemius muscle with decreased intensity relative to that of normal muscle. After administration of gadolinium, the T_1 weighted image showed a well defined rim of contrast enhancement and a hypointense central area (A). An axial T_2 weighted image showed bright signal intensity in and around a focal mass-like lesion (B).

Table 1 Review of Behcet's disease with myositis in the English literature

Patient/ age (years)/sex	Muscle symptom	General symptoms*	Serum CK	Myositis	Recurrence	Histological finding of affected muscle	Treatment	Ref
1/23/M	Nodule of right quadriceps femoris	–	Normal	Localised	–	Muscle fibre degeneration and mono- and polymorphonuclear cell infiltration with accentuation at perivascular areas	Spontaneous resolution	1
2/42/M	Myalgia of both legs	+	ND	Localised	–	Severe muscle fibre degeneration with diffuse mononuclear infiltration; the vessels have the same type of vasculitis and perivasculitis	Corticosteroid	2
3/55/M	Myalgia of right arm	+	High	Generalised	+	Diffuse muscular necrosis and inflammation	Corticosteroid	3
4/25/M	Myalgia of both legs, neck	+	ND	Localised	+	Inflammatory granulation tissue with fragments of striated muscle fibres	Spontaneous resolution	4
5/14/F	Myalgia uni- or bilaterally in the calves	+	High (only three days)	Localised	+	Myositis with an inflammatory infiltrate in the interstitial and perivascular regions and focal necrosis of myocyte	Corticosteroid; colchicine not effective	5
6/19/M	General progressive muscle pain and weakness	+	High	Generalised	–	Diffuse mononuclear round cell infiltration and scattered muscle fibre degeneration	Corticosteroid; cyclosporin	6
7/22/M	Myalgia of both legs	+	ND	Localised	–	Severe granulocytic-monocytic inflammation with abundant myophagocytosis and degenerating muscle fibres	Corticosteroid	7
8/68/F	General progressive muscle pain and weakness	+	High	Generalised	–	Diffuse mononuclear round cell infiltration in the perimysial and endomysial connective tissue	Corticosteroid	8
9/12/M	Myalgia of right calf	+	Normal	Localised	+	ND	Corticosteroid	9
10†/29/M	Nodules of both legs and right arm	+	Normal	Localised	+	Infiltration of neutrophils and macrophages with focal central necrosis of myocytes and perivasculitis	Spontaneous resolution	

M, male; F, female; ND, not done.
* Unspecific alterations like an increase in erythrocyte sedimentation rate, C reactive protein, body temperature; †present case.

received colchicine during the acute phase of myositis, with no striking effect on the myositis. In our case, necrotising myositis did not recur after the administration of colchicine. The usefulness of colchicine for prevention of myositis in Behçet's disease needs to be further studied.

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