Myositis in Behcet's Disease after Tonsillectomy

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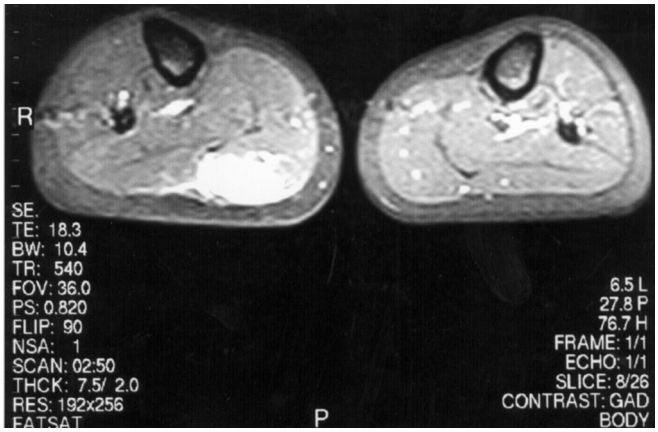
Behcet's disease is a chronic, relapsing, inflammatory disease characterized by recurrent oral aphthae, genital ulcers, ocular disease, skin lesions, neurologic manifestations, vascular disease, arthritis, and gastrointestinal involvement. Muscular involvement is rare and there are only a few case reports in the literature of such a manifestation. In this communication we report a patient with Behcet's disease-associated myositis that was related temporally to tonsillectomy. We also describe the unique magnetic resonance imaging features of the myositis.

Patient Description

A 27 year old Jewish woman of North African ancestry presented with pain and swelling of her right calf. The patient stated being generally healthy aside from recurrent episodes of streptococcal tonsillitis. She was unaware of any chronic illness. A week before her admission she underwent an elective tonsillectomy uneventfully. Five days after the operation she began to complain of pain and swelling of her right calf which became progressively worse, to the point that she was unable to walk. She had a fever of 38.2°C, and the right calf was swollen, slightly erythematous, exquisitely tender to touch, and warm. On palpation, stiffness was noted along the vertical axis of the calf. The rest of the physical examination was normal. Laboratory examinations revealed a white blood cell count of 15,980, erythrocyte sedimentation rate 85 mm/hour, and C-reactive protein 6.1 mg/dl. Creatine kinase was normal, as were kidney and liver function tests. Ultrasonography of the calf showed muscle and subcutaneous fat thickening, and excluded deep vein thrombosis. Magnetic resonance imaging of the calf was performed: on T1-weighted images the muscle appeared normal, T2-weighted and STIR images of the calf revealed increased signal intensity in the medial head of the gastrocnemious muscle, along 20 cm of its length, with enhancement after administration of intravenous gadolinium [Figure]. The features of the lesion were compatible with an inflammatory reaction. Fine-needle aspiration of the calf yielded scant bloody fluid, without pus. Cytology examination showed only red blood cells.



MRI of the calves: Post-contrast FAT-SAT axial image demonstrates hyperintense signal along the medial head of the right gastrocnemious muscle, with contrast enhancement of the lesion.

Because of the suspected diagnosis of bacterial myositis, treatment with cefazolin was begun. In spite of the antimicrobial treatment the patient's symptoms did not subside. Blood cultures, as well as cultures from the muscle aspiration, were negative. The patient was reevaluated and additional medical history details were elicited. She recalled that she suffered from recurrent oral ulcers. In addition, the patient recalled recurrent events of painful genital ulcer, as well as recurrent subcutaneous erythematous, raised and painful skin lesions on the anterior surface of the thigh, very suggestive of erythema nodosum. This information initially was not mentioned by the patient since these symptoms had been experienced several years before the present admission, without exacerbation related to the myositis. Re-inspection of venipuncture sites revealed lesions resembling a pathergy reaction. HLA B5 was positive. Ophthalmologic examination was normal.

With this new information, a presumptive diagnosis of myositis associated with Behcet's disease was made. Treatment with prednisone, 40 mg/day, was started with a rapid improvement in symptoms and signs. The patient was discharged several days later and steroid therapy was gradually tapered over several weeks without a recurrence of the myositis.

Comment

We describe a patient with chronic symptoms suggesting Behcet's disease, and an acute presentation of myositis that did not improve despite antibiotic treatment. Treatment with steroids resulted in prompt resolution of the patient's acute symptoms.

A Medline search with the key words: "myositis" and "Behcet's disease" revealed several case reports of localized myositis associated with Behcet's disease, of which

four are mentioned below [1-4]. All the patients in these reports had myositis of the legs. The majority of patients had indices of systemic inflammatory response including fever, elevated erythrocyte sedimentation rate and C-reactive protein. In the majority of cases, patients were treated successfully with prednisone. These features are very similar to the clinical characteristics of our patient. Histologic features of muscle biopsies in the prior reports included myocyte degeneration, and interstitial and perivascular inflammatory infiltrate.

As mentioned, the patient underwent a tonsillectomy one week prior to admission. A causal relation between tonsillectomy and Behcet's disease-associated myositis may exist. Some studies suggest a possible pathogenetic role of certain streptococcal antigens that have cross-reactivity with human peptides [5]. Hypothetically, exposure to streptococcal antigens may occur post-tonsillectomy due to the local inflammatory process after the surgery. Antigens of streptococci, which are part of the normal flora of the mouth, may induce an inflammatory response with cross-reactivity against self-antigens, and this may lead to a cascade of events evolving to exacerbation of Behcet's disease, including myositis in this specific case.

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