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Case Report

Focal myositis: a rare case report

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ABSTRACT

Focal myositis is an uncommon, self-limiting, benign skeletal muscle disease, which is generally determined as an inflammatory pseudotumor. The etiology is not fully known, although it has been suggested that subclinical damage may play a role. As it leads to a tumoral mass it may be confused with several diseases leading to incorrect evaluations. Definitive diagnosis is made by biopsy of the skeletal muscle. In radiologic diagnosis, magnetic resonance imaging is the most important modality. In this paper we presented the imaging findings of a 58-year-old female patient with focal myositis who was admitted with complaints of forearm swelling.

Keywords: Magnetic resonance imaging; myositis; ultrasonography.

Focal myositis is an uncommon, self-limiting, benign skeletal muscle disease, which is generally identified as an inflammatory pseudotumor. It was first described by Heffner et al.[1] in 1977. The disease occurs over a wide age range of 7-94 years (mean 41 years) and there is equal involvement between both sexes. It generally occurs in the lower extremities. Atypical localization of lesions has been shown in the neck, tongue, hand, and eyelid and paraspinal muscle.[2-5] As it leads to a tumoral mass, there may be confusion between several diseases leading to incorrect evaluations.[6]

The etiology is not fully known, although it has been suggested that subclinical damage may play a role in the occurrence of this disease.[7] Cases of focal myositis have been reported in literature associated with ischemia due to vascular malformations. Therefore, ischemia has been considered as one of the responsible mechanisms.[8]

An absolute diagnosis is made by taking a biopsy of the skeletal muscle. There is histological resemblance to skeletal muscle myopathies and dystrophies. Sometimes concomitant eosinophil accumulation is seen.

The clinical, laboratory and imaging findings are here presented of a female with focal myositis. She presented with complaints of forearm swelling.

CASE REPORT

A 58-year-old female patient presented with complaints of pain and swelling of the right forearm, more evident on the medial side, which had been ongoing for 10-15 days. Use of the arm had started to become increasingly difficult because of the pain. There was nothing remarkable in the patient or family history. In the physical examination, peripheral pulses were obtained in both upper extremities; blood pressure was measured as 130/80 mmHg, body temperature as 36.5 °C and pulse as 90/min and rhythmic. The swollen area was mildly sensitive and painful. Muscle strength and neurological examination results were normal. A written informed consent was obtained from the patient.

The full blood count values were normal. Erythrocyte sedimentation rate was 24 mm/hr, C reactive protein was 8 mg/dL (normal, 0.2-5) and

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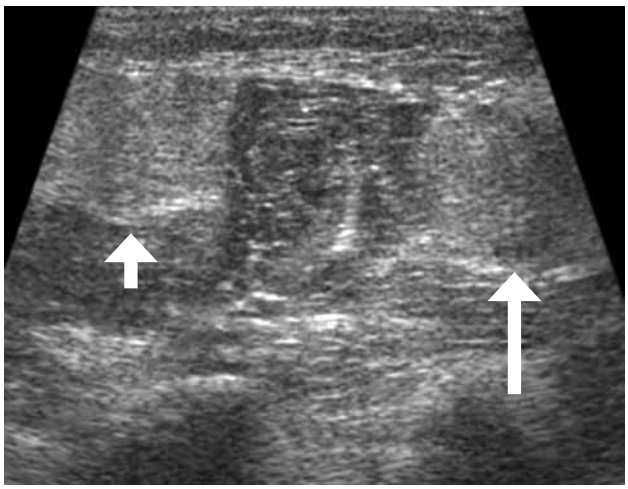

Figure 1. Increased echogenicity is observed in the flexor carpi ulnaris (long arrow) and the flexor carpi radialis (short arrow) on the axial ultrasonography image.

romatoid factor was 7.19 IU/mL (normal, 0-50). In the biochemical tests, very high values were determined of lactate dehydrogenase (LDH) at 561 IU/L and creatinine kinase (CK) at 7888 IU/L. Other biochemical test results were normal.

During the B-mode ultrasonography examination, increased diffuse homogenous echogenicity was observed in the flexor carpi radialis and flexor carpi ulnaris muscles (Figure 1). During the magnetic resonance imaging (MRI), there were high signals in the short TI inversion recovery (STIR) series in these two muscles, isointense diffuse areas on the T_1 series and in the adjacent superficial facial plane appeared edema and minimal fluid (Figure 2). With contrast dye, irregular, amorphous and weak involvement was observed in the muscles (Figure 3). Inflammatory myositis was considered based on the MRI findings.

Serological tests for Cytomegalovirus, Epstein-Barr virus, Coxsackie virus, Parvovirus, Hepatitis viruses, Human immunodeficiency virus, Toxoplasma and Brucella were negative. Anti-nuclear antibodies, anti-double-stranded DNA antibodies, anti-centromere antibodies, anti jo-1, anti-Scl 70, anti-histon antibodies, anti-cardiolipin-immunoglobulin M and immunoglobulin G, anti-Sjögren's syndrome A and anti-Sjögren's syndrome B antibodies were found to be negative.

The patient was given an appointment for a biopsy and a course of nonsteroidal anti-inflammatory drug commenced. At the follow-up examination which took place one month after the symptomatic treatment, the complaints in the arm had completely disappeared and the sedimentation, LDH and CK values were observed to have returned to normal. As the findings had returned to normal in a short time, no histopathological examination was made.

DISCUSSION

Although the etiology of focal myositis is not fully understood, subclinical trauma or viral infections have been suspected.[7] In the differential diagnosis, rather

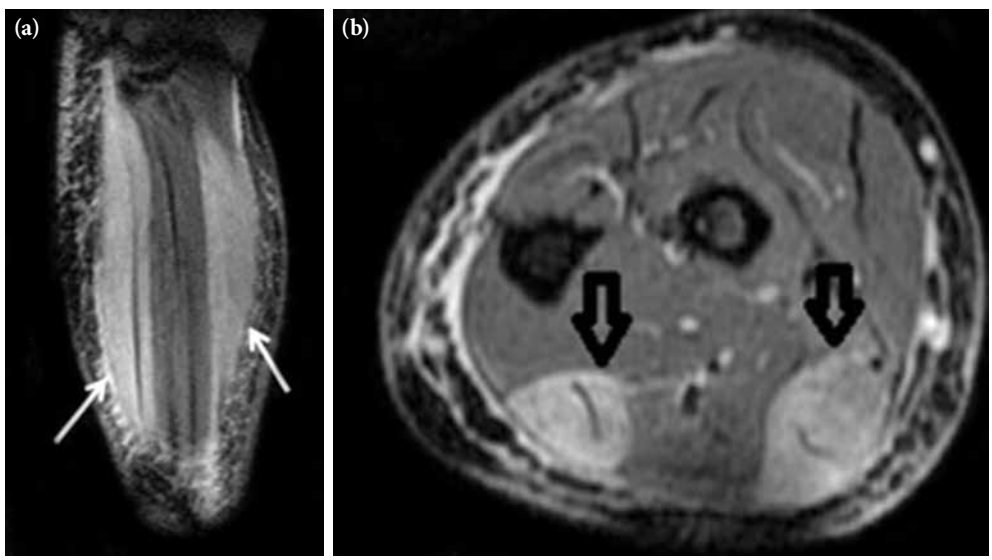

Figure 2. Evident signal increase observed in the flexor carpi ulnaris and the **(a)** flexor carpi radialis on the coronal (white arrows) and **(b)** axial (black arrows) T_2-weighted magnetic resonance images.

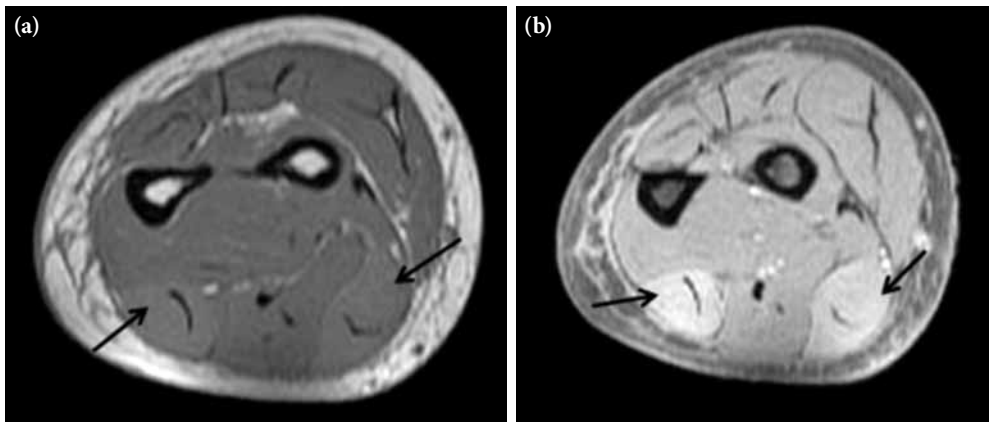

Figure 3. Axial non-contrast T_1-weighted magnetic resonance image showing **(a)** slight signal increase in both muscles (black arrows) and contrast T_1-weighted magnetic resonance image showing **(b)** contrast involvement in both muscles (black arrows).

than infectious myositis being first considered, patients are tested for diabetes or immune system problems. In the current case, there was no immunosuppressive disease. Serological examinations directed to infectious agents were found to be negative, just as there were no skin findings or raised temperature which could have been related to systemic infection. As the patient recovered without antibiotic treatment, the lesion in this current case was not thought to be of infectious origin.

In the differential diagnosis, myositis related to autoimmune disorders was excluded as there was no Reynaud's phenomenon which could be considered connective tissue disease in the current case, no clinical findings such as oral aphtae, arthritis, serositis and alopecia and negative antibodies indicating immune diseases. Another disease considered in the differential diagnosis was eosinophilic fasciitis. In eosinophilic fasciitis, diffuse thickening and contrast involvement in the deep and superficial fascia are typical. Muscle involvement is observed secondarily in the superficial sections adjacent to the fascia.[9] In the current case, as there was dominant involvement in the muscle tissues and there was no possibility of secondary effect in the adjacent fascia, eosinophilic fasciitis was excluded.

Primary and secondary tumors must be considered in the differential diagnosis. Most cases in presented in the literature had a biopsy have had biopsy applied because of this concern. In the current case, as the complaints were resolved in a short time, and there were no indications of any other organ being involved. The likelihood of a tumor was not a priority in the differential diagnosis.

In cases with focal myositis, sonographic image has been reported to vary and may be hypoechoic or hyperechoic.[2,10] In the current case, the lesions were seen to be widespread, homogenous and hyperechoic. During the computed tomography, the appearance of focal myositis is non-specific. Compared to the muscle, it may be slightly hypodense or isodense. An increase in volume may be observed in the affected muscle.[6,11] Therefore, MRI is the most optimal evaluation method in focal myositis.[12] On MRI, lesions are observed as hypointense compared to the muscle on T_1-weighted series and a homogenous high signal is observed in the involved muscle on fat-suppressed T_2-weighted series. During the contrast examination in some cases, homogenous contrast involvement can be observed.[13] The STIR MRI of the current case had a widespread, hyperintense appearance. On the contrast MRIs, irregular, amorphous and weak contrast was seen. The evident high signal on the STIR series and no observation of mass-like contrast involvement suggested that the lesion was a strongly inflammatory pseudotumor.

In conclusion, although focal inflammatory myositis is a rarely seen benign disease, it can be confused with tumors and in particular, with autoimmune diseases and as well as several other conditions. In addition to clinical data, MRI is the most important imaging method in the diagnosis of focal myositis.

Declaration of conflicting interests

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