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CASE REPORT

Acute sarcoid myositis with unusual radiologic findings

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ABSTRACT
A 59-year-old man presented with bilateral calf pain and swelling for two weeks. Ultrasound and magnetic resonance imaging examination showed multiple bilateral, nodular, and spindle-shaped lesions in the gastrocnemius and soleus muscles. On physical examination, hyperpigmented, papular lesions were noticed; biopsy of the skin of his right elbow showed granulomatous inflammation. His angiotensin converting enzyme level was markedly elevated. Computed tomography showed diffuse interstitial thickening, miliary nodules, and traction bronchiectases throughout the lung parenchyma. Ophthalmologic examination showed uveitis in his left eye. Based on the lung, eye, and skin findings, a clinical diagnosis of sarcoidosis was made. After two months of corticosteroid treatment, his muscle lesions largely resolved.

Key words: • *radiology* • *myositis* • *sarcoidosis*

Sarcoidosis is a granulomatous inflammatory disease of unknown origin affecting multiple organ systems. It mostly affects the hilar lymph nodes, eyes, and heart. Symptomatic muscle disease is rare and is seen in less than 0.5% of cases. There are three types of symptomatic muscle disease: chronic myopathy, palpable nodules, and acute myositis (1–4). Herein we report the ultrasound (US) and magnetic resonance imaging (MRI) findings of a patient with sarcoidosis who initially presented with acute myositis.

Case report

A 59-year-old man with a two-week history of bilateral calf pain and swelling was referred to our radiology department for the evaluation of his lower extremities. Presumptive clinical diagnosis was deep vein thrombosis in his lower extremities. Doppler US findings were normal. In the axial US examination, there were multiple bilateral, hypoechoic nodular lesions in the gastrocnemius and soleus muscles. Some of these lesions were spindle-shaped and were seen along the muscle fibers in sagittal US examination. They were avascular in the color and power Doppler US (Fig. 1). The creatine kinase (CK) level was 58 U/L (normal, 22–240), aspartate aminotransferase (AST) was 21 IU/L (normal, 8–40 IU/L), alanine aminotransferase (ALT) was 10 IU/L (normal, 4–40 IU/L), lactate dehydrogenase (LDH) was 33 IU/L (normal, 124–232 IU/L), C-reactive protein (CRP) was 10.8 mg/dL (normal, 0–10), and the erythrocyte sedimentation rate (ESR) was 1 mm/hr. Serum calcium level was within normal limits. For further examination of muscle lesions, we performed extremity MRI. There were multiple nodular and spindle-shaped lesions in the gastrocnemius and soleus muscles bilaterally. When compared with muscle fibers, these lesions were isointense on T1-weighted images and hyperintense on T2-weighted images. They showed diffuse enhancement after contrast medium was injected (Fig. 2). On physical examination, we noticed hyperpigmented papular lesions measuring 2 mm to 3 cm in the skin of his elbow and both calves. The biopsy of the skin of his right elbow showed granulomatous inflammation. There was no necrosis. Because of his complaint of mild productive cough, high resolution computed tomography (HRCT) of the lung was performed. This showed diffuse interstitial thickening, miliary nodules, and traction bronchiectases throughout the lung parenchyma. Angiotensin-converting enzyme (ACE) level was 115 U/L (normal, 7–25). Ophthalmologic examination showed uveitis in his left eye. Based on the lung, eye, and skin findings, the presumed clinical diagnosis was sarcoidosis. Prednisolone was administered at 12 mg/day. Afterwards, azathioprine (Imuran) 100 mg/day was given. After two months, muscle weakness was significantly decreased. ACE level (20 U/L) returned to normal, and uveitis regressed. On US examination, his muscle lesions had largely resolved.

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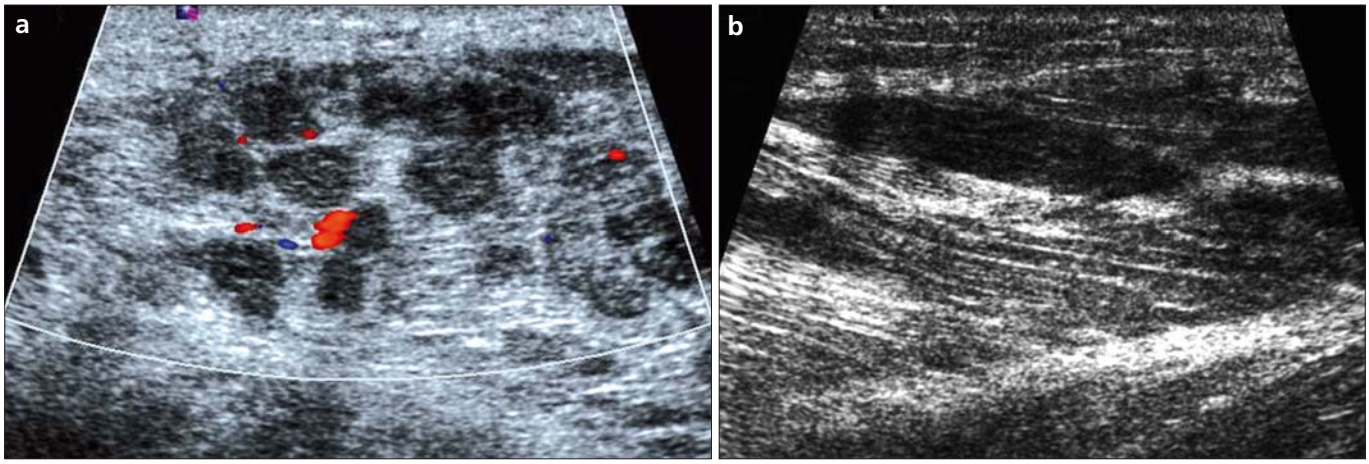
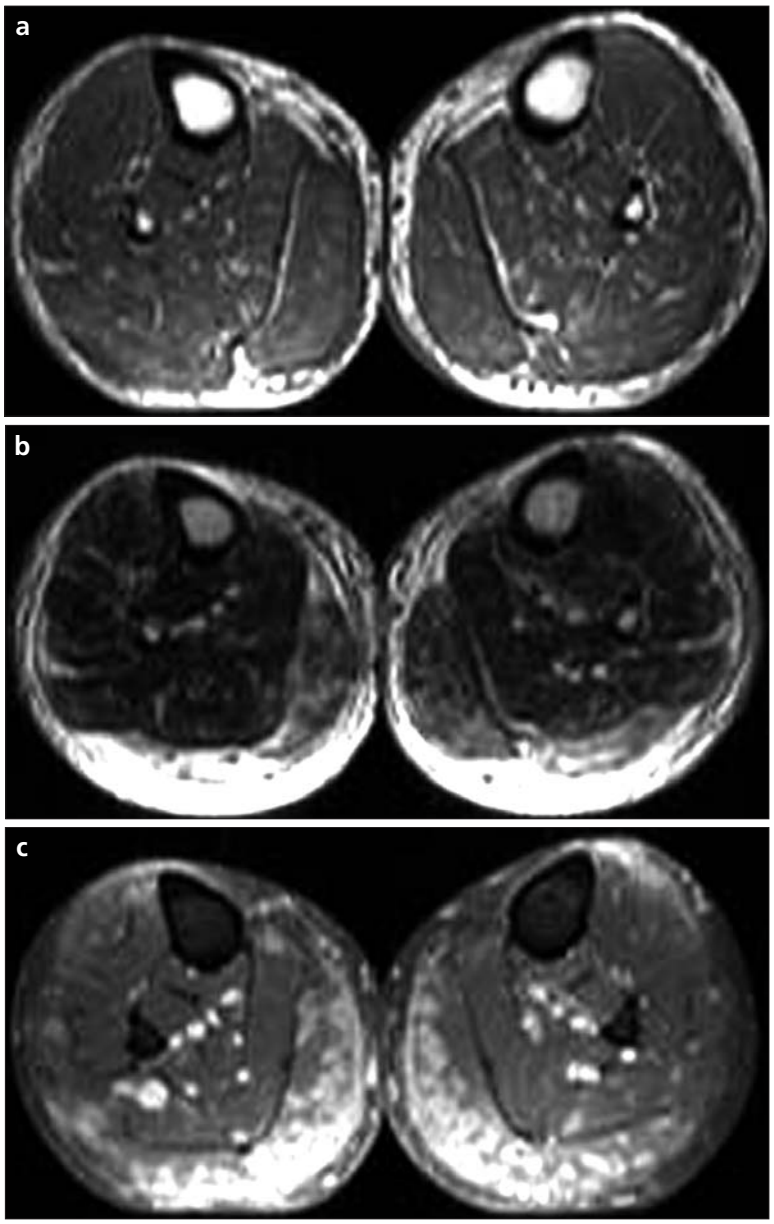


Figure 1. a, b. Axial color Doppler US **(a)** and sagittal US **(b)** images show multiple hypoechoic lesions in the gastrocnemius and soleus muscles which are avascular in Doppler examination **(a)**.



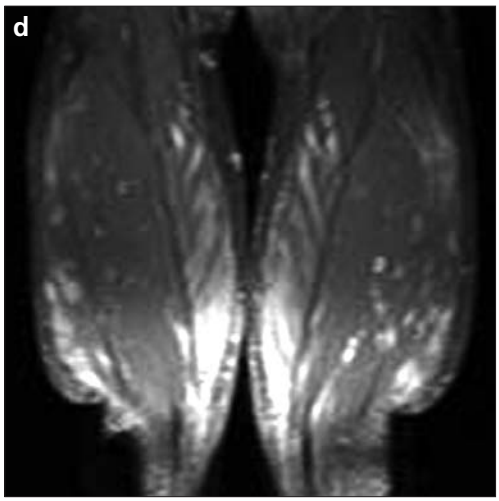


Figure 2. a–d. Extremity MRI shows multiple lesions which are isointense on axial T1-weighted **(a)** and hyperintense on axial T2-weighted **(b)** MR images compared to muscle. The lesions show homogenous contrast enhancement on T1-weighted fat-suppressed axial **(c)** and coronal **(d)** MR images and are nodular and spindle shaped on the coronal image **(d)**.

Discussion

According to recommendations of Baughman et al., a diagnosis of sarcoidosis can be established by means of the following criteria (5): (i) the presence of granuloma in a biopsy specimen without evidence of tuberculosis, fungus, malignancy, or other cause of granuloma, together with clinical features suggesting sarcoidosis, such as bilateral hilar adenopathy on chest radiography, erythema nodosum, uveitis, and maculopapular skin lesions; (ii) in the absence of biopsy material, the presence of previously mentioned clinical features and additional features highly consistent with sarcoidosis, such as raised concentra-

tion of ACE, bronchoalveolar lavage fluid lymphocytosis, abnormal gallium scan, and lupus pernio. Evidence of multiple organ system involvement has also been emphasized (5, 6). Sarcoid myopathy was first described in 1908 by Licharew, who discussed the case of a 17-year-old girl with lupus pernio, splenomegaly, and multiple nodules in muscles (7).

Symptomatic muscle involvement in sarcoidosis has three clinical types: chronic myopathy, palpable nodulosis and acute myositis (3). The most common type is chronic myopathy, characterized by slowly progressive weakness and disability over months or years. It is usually found in chronic stages of the disease. Patients with chronic myopathy are usually middle-aged women with symmetrical proximal muscle disturbance. The trunk and neck muscles may be involved. Palpable nodulosis is an unusual type. These nodules are initially soft and small, but they increase in size and become painful. The most commonly involved site is the lower extremity (90%), followed by the upper extremity (43%). Masses vary in size from several centimeters to 22 cm in length (8). The rarest form of muscle involvement in sarcoidosis is acute myositis (9). Patients with this form have tenderness, myalgias, and proximal muscle weakness. Additional findings may include acute polyarthritis or erythema nodosum. Acute myositis is seen in patients younger than those with palpable nodulosis and chronic myopathy, with onset typically before 30 years of age (3). Acute myositis may be the initial manifestation of sarcoidosis or be part of the chronic progressive form. The patients with acute myositis usually have elevated CK and ESR, but normal levels have also been reported. Patients with palpable nodulosis also have nonspecific laboratory parameters (e.g., there have been several reports of normal serum calcium and CK; others have had high ESR, peripheral blood eosinophil counts and hypergammaglobulinemia). However, in most cases, chest radiographs were abnormal. Laboratory studies in chronic sarcoid myopathy may be normal, but ESR and serum immunoglobulins are usually increased (10).

The age of our patient is older than defined in the literature (3). Generally, sarcoidosis is identified clinically in patients with foggy vision or mediastinal lymphadenopathy, but muscular weakness may be infrequently observed as an initial symptom (11). The interesting point is that the presenting complaint of our patient was muscle pain, although pulmonary computed tomography (CT) examination showed grade III sarcoidosis. In addition, our patient had distal muscle weakness, which is an unusual finding in acute sarcoid myositis. Myopathic sarcoidosis generally causes symmetrical proximal muscle weakness and wasting in the lower limbs (12). In the laboratory studies of our patient, only the ACE level was significantly increased.

MRI and CT examinations are useful diagnostic tools in sarcoidosis. Otake et al. reported MRI findings of 28 patients with sarcoid myopathy (20 nodular, 8 with chronic myopathy) (13). Sarcoid nodules were clearly identified on MRI in 18 of 20 (90%) patients with nodular involvement, whereas MRI findings were normal in all patients with chronic myopathy. According to MRI, these nodules were well-demarcated oval lesions consisting of a star-shaped area of low signal intensity surrounded by an area of higher signal intensity. This finding has been described as a "dark star" appearance (14). MRI findings of musculoskeletal sarcoidosis lesions may reveal systemic involvement and a greater burden of granulomas than previously thought, which may have therapeutic and prognostic significance. When musculoskeletal manifestations are detected, MRI can also guide biopsy in patients with suspected but unproven sarcoidosis and can be used to follow response to treatment (15). The sensitivity of CT is less than that of MRI in evaluating sarcoid nodular myopathy. Otake et al. had CT scans of 10 patients with nodular myopathy. Hypodensity in central areas of the nodules and slight peripheral enhancement were seen in all patients (13). Otake performed ultrasonography on six patients; studies showed a central zone of increased echogenicity and a peripheral area of decreased echogenicity (14). In another report, MRI showed increased signal intensity on T2-weighted images (16).

Our US findings of avascular, hypoechoic multiple nodules in the gastrocnemius and soleus differ from those reported in the literature. Our case showed multiple nodular lesions that were isointense on T1-weighted MR images and hyperintense on T2-weighted MR images compared to muscle fibers. The nodular lesions showed enhancement after contrast medium was injected. On sagittal images, some of the lesions were spindle shaped, which had not previously been described in the literature. In addition, contrast enhancement was evident. Unlike MRI findings of Otake et al. (13), we detected diffuse, rather than peripheral, enhancement in these nodular lesions.

Muscle biopsy is not routinely used to diagnose chronic sarcoidosis, because pathological findings are similar in acute and chronic myopathy. Noncaseating granulomas associated with inflammatory component are evident (9, 17). We did not perform muscle biopsy because thorax CT, clinical, and laboratory findings of our patient were consistent with sarcoidosis.

Corticosteroids are usually administered in the treatment of acute sarcoid myositis (9). Response to corticosteroids in chronic myopathy is unpredictable, whereas acute myositis responds well (12). In our case, because of decrease in the size of lesions after medical treatment on US examination, we diagnosed acute sarcoid myositis.

In summary, sarcoid myositis should be considered in the differential diagnosis of muscle lesions. Correlation with clinical and laboratory findings is essential for correct diagnosis because US and MRI findings are nonspecific in most cases.

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