

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

Myositis and myopathy of sarcoidosis: A case report [☆]

Tucker v R. Burr, MD*, João F. Kazan-Tannus, MD, PhD

Loyola University Medical Center, Department of Radiology, 2160 S. First Ave, Maywood, IL 60153 USA

ARTICLE INFO

Article history:

Received 13 November 2020

Revised 5 December 2020

Accepted 5 December 2020

Keywords:

Sarcoidosis

Myositis

Myopathy

ABSTRACT

Muscular manifestations of sarcoidosis are commonly found on biopsy but rare on correlated imaging. We present a rare case of a 36-year-old male patient with sarcoid myositis and image findings of active myositis in the lower back and pelvic girdle musculature. This case suggests considering sarcoidosis as a differential diagnosis in the setting of chest findings and new lower back and lower extremity weakness.

© 2020 Published by Elsevier Inc. on behalf of University of Washington.

This is an open access article under the CC BY-NC-ND license

(<http://creativecommons.org/licenses/by-nc-nd/4.0/>)

Background

Sarcoidosis is a well-known noncaseating granulomatous disease. Although first described by Hutchinson in 1877 with dermatologic findings, it was early in the 20th century when sarcoidosis was recognized as a multisystem disease [1]. Sarcoidosis is widely accepted to have idiopathic origin, although it is postulated that there is an antigenic stimulation triggering an imbalanced T-cell inflammatory response [2,3]. Muscular manifestations of sarcoidosis are commonly found on biopsy but rare on correlated imaging. We present a rare case of a 36-year-old male patient with sarcoid myositis and image findings of active myositis in the lower back and pelvic girdle musculature.

Case presentation

A 36-year-old male presents with left lower back and left lower extremity weakness including inability to exercise and heaviness when climbing stairs. He denies pain or myalgia. He has a past medical history of sarcoidosis diagnosed 5 years prior with initial presentation of panuveitis and subsequent findings of enlarged hilar lymph nodes, elevated angiotensin converting enzyme levels, and left lower leg skin biopsy showing granulomatous dermatitis. The patient was treated at that time with steroids and methotrexate, later becoming lost to follow-up. Notably, the patient was not treated with steroids in the interim (removing this as a confounding factor in diagnosis of muscular lesions). Other complaints include chronic

[☆] Consent for publication: All patient data has been removed and informed consent is not required to publish.

* Corresponding author.

E-mail address: tucker.burr@lumc.edu (T.v.R. Burr).

<https://doi.org/10.1016/j.radcr.2020.12.014>

1930-0433/© 2020 Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>)

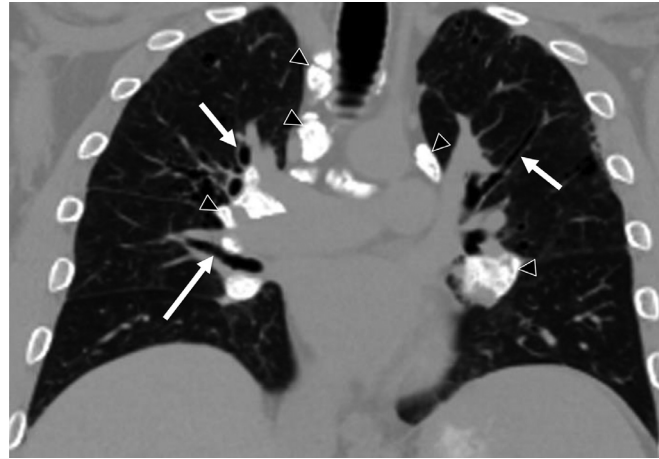


Fig. 1 – Coronal noncontrast CT shows prominent calcified mediastinal and hilar adenopathy (black arrowheads). There is additional prominent bronchiectasis (straight arrows).

dry eyes and dyspnea with exertion. On physical exam, left hip strength was diminished with unsteadiness during the get up and go test.

The initial differential diagnosis from neurology for the patient's lower extremity weakness included inclusion body myositis and limb girdle muscular dystrophy as well as sarcoidosis. A biopsy of the left hamstring and left quadriceps muscles revealed granulomatous myositis with component of neurogenic atrophy. Pathology determined findings to be consistent with sarcoidosis. The patient is currently transitioning from a steroid regimen to methotrexate to control symptoms.

Imaging findings

A noncontrast computed tomography reveals diffuse mediastinal and hilar enlarged lymph nodes with coarse calcification (Fig. 1). The main pulmonary artery is mildly enlarged measuring 3.4 cm (Fig. 2), suggestive of pulmonary hypertension and pulmonary involvement. Lung windows reveal upper lobe predominant subpleural nodularity, bronchiectasis, perihilar honeycombing, and architectural distortion typical of fibrosis seen with sarcoidosis (Fig. 3).

Computed tomography images of the abdomen and pelvis, show patchy multifocal muscular edema and atrophy of pelvic girdle muscles (Fig. 4). Patchy foci of hypodensity may be sequelae of intramuscular nodules. A magnetic resonance imaging of the lumbar spine (Fig. 5) reveals T2 hyperintensity most prominent within the left paraspinous muscles with mild signal within the left psoas, consistent with symptoms of weakened hip flexion and lower back weakness (Fig. 6).

Discussion

Sarcoidosis is a multisystem disease most commonly involving the lungs, although the radiologist may see other signs

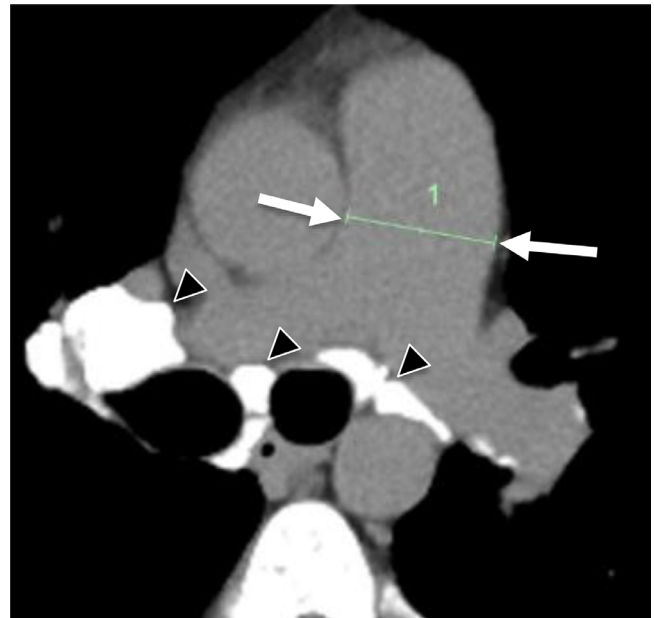


Fig. 2 – Axial noncontrast CT reveals an enlarged main pulmonary artery measuring 3.4 cm (straight white arrows). Marked calcified mediastinal and hilar lymph nodes are present (black arrowheads).

of disease in the liver, skin, brain, and rare active disease in the muscle, as in this case. Most patients with sarcoidosis are young (less than 50 years old). Outside of the United States it is common among Scandinavian people, while in the United States it is common among Black people [4] Prevalence is higher in females than males.

Diagnosis involves clinical or radiographic findings in concert with tissue biopsy showing epithelioid cell granulomas [5]. Biopsy is taken from the most accessible prominent site of suspected disease, often the skin or a superficial lymph node. Sarcoidosis commonly involves the lungs and hilar lymph

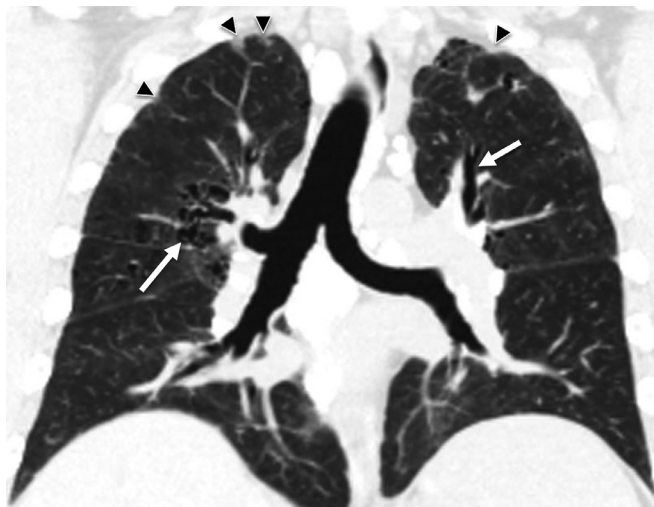


Fig. 3 – Coronal noncontrast CT with lung windows shows signs of pulmonary fibrosis. There is upper lobe predominant architectural distortion, subpleural interstitial nodularity (black arrowheads), and bronchiectasis with peribronchovascular honeycombing (straight arrows).



Fig. 4 – Axial CT image. Multifocal patchy edema and atrophy within bilateral pelvic girdle musculature including gluteus maximus (curved arrows), piriformis (white arrowheads), gluteus minimus and medius (black arrowheads), and minimally within iliopsoas (straight arrows) and piriformis. Calcified iliac lymph nodes are also visible.

nodes – a historical staging system is based on chest radiograph findings. Other notable sites of involvement include the skin, central nervous system, liver and gastrointestinal system, and heart. Various lab abnormalities may support the diagnosis including elevated serum angiotensin converting enzyme, elevated serum calcium, and elevated urine calcium. Spontaneous remission is seen with many patients. Treatment for active disease commonly involves corticosteroids and other anti-inflammatory agents [6].

Noncaseating granulomas may become coalescent to form nodules and masses throughout the body. Muscular manifestations most-commonly occur in the lower extremities, proximal more than distal, also seen in this case. Although

skeletal muscle granulomatous involvement is commonly seen on muscular biopsy (50%-80% of patients), most patients are asymptomatic, discrete muscular lesions are rare, and imaging manifestations of muscle lesions or muscular involvement are found in less than 2% of patients [7,8]. Magnetic resonance imaging is useful in delineating extent of fatty replacement of muscle and determining an optimal site for biopsy. Myopathy and muscle granuloma resolve over time, with or without treatment.

Sarcoidosis is a common disease among certain populations worldwide and within the United States. This is a multi-system disease most commonly involving the lungs, but, as seen in this case, may manifest as myopathy and myositis

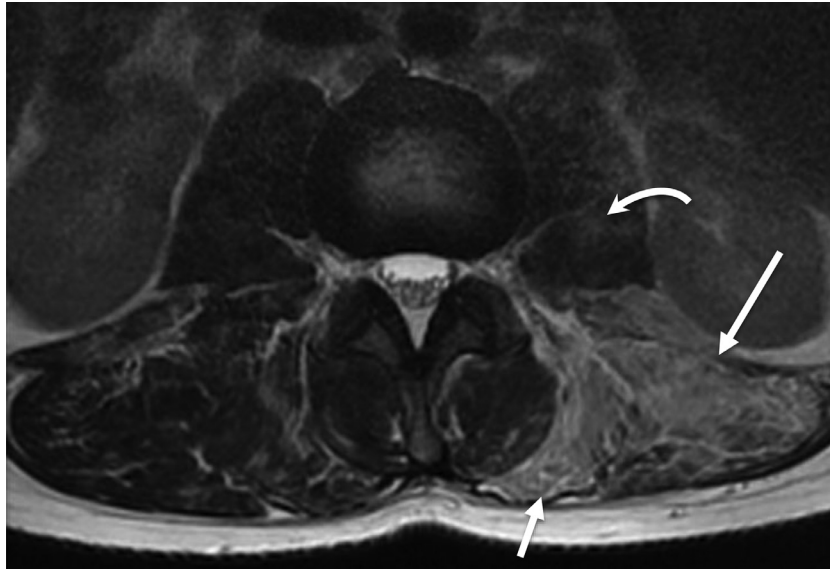


Fig. 5 – T2-weighted MRI of the lumbar spine reveals edema and atrophy most prominent within the left paraspinous muscles (straight arrows) as well as likely mild edema within the left psoas muscle (curved arrow).

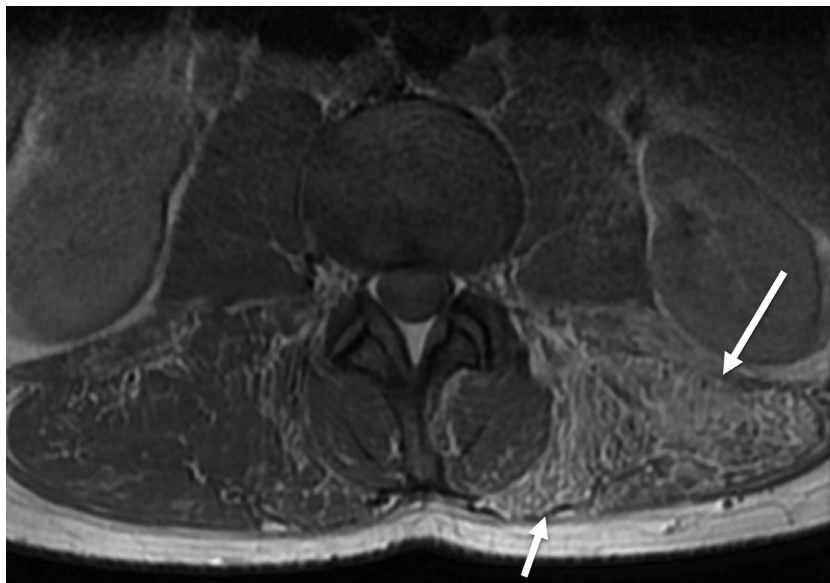


Fig. 6 – T1-weighted MRI with contrast. Left paraspinous musculature showed enhancement with contrast administration (straight arrows).

appearing as edema and enhancement with contrast administration. Other manifestations include granulomatous nodules seen near the musculotendinous junction. Initial signs and symptoms may be nonspecific; diagnosis may require the correlation of clinical, imaging, and pathological findings. By recognizing diverse manifestations and the extent of disease, the radiologist may aid the clinician in early management and better outcomes for patients with sarcoidosis.

REFERENCES

- [1] James DG, Sharma OP. From Hutchinson to now: a historical glimpse. *Curr Opin Pulm Med* 2002;8(5):416–23. doi:10.1097/00063198-200209000-00013.
- [2] Le V, Crouser ED. Potential immunotherapies for sarcoidosis. *Expert Opin Biol Ther* 2018;18(4):399–407. doi:10.1080/14712598.2018.1427727.

-
- [3] Miyara M, Amoura Z, Parizot C, et al. The immune paradox of sarcoidosis and regulatory T cells [published correction appears in *J Exp Med*. 2006 Feb 20;203(2):477]. *J Exp Med*. 2006;203(2):359–70. doi:[10.1084/jem.20050648](https://doi.org/10.1084/jem.20050648).
- [4] Arkema EV, Cozier YC. Epidemiology of sarcoidosis: current findings and future directions. *Ther Adv Chronic Dis* 2018;9(11):227–40. Published 2018 Aug 24. doi:[10.1177/2040622318790197](https://doi.org/10.1177/2040622318790197).
- [5] Moore SL, Teirstein AE. Musculoskeletal sarcoidosis: spectrum of appearances at MR imaging. *Radiographics* 2003;23(6):1389–99. doi:[10.1148/rg.236025172](https://doi.org/10.1148/rg.236025172).
- [6] Belfer MH, Stevens RW. Sarcoidosis: a primary care review. *Am Fam Physician* 1998;58(9):2041–56.
- [7] Resnick D. *Diagnosis of bone and joint disorders*. United Kingdom: Saunders; 2002.
- [8] Otake S, Ishigaki T. Muscular sarcoidosis. *Semin Musculoskelet Radiol* 2001;5(2):167–70. doi:[10.1055/s-2001-15675](https://doi.org/10.1055/s-2001-15675).