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Case Report

Clinical and radiological resolution of vertebral sarcoidosis mimicking metastatic disease ^{☆,☆☆}

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ABSTRACT

Sarcoidosis is a disease that exhibits extreme heterogeneous clinical manifestations. Bone involvement in sarcoidosis is rare (1%–13%), and involvement of the vertebrae is even rarer. Usually, it is a diagnosis of exclusion with nonspecific characteristics in imaging.

A 35-year-old male, who has no significant medical history. He came to clinical examination for lower back and associated bilateral lower extremity pain. Magnetic resonance imaging (MRI) was performed to exclude disc-related pathology. It demonstrates left paramedian lumbar disc herniation at L4–L5 level. Multiple small enhancing lesions throughout the lumbar vertebrae were discovered as an incidental finding.

An 18F-labeled fluorodeoxyglucose was performed to evaluate for metastatic disease that shows hypermetabolic apical right nodule of the lung parenchyma with multiple mediastinal and right iliac external adenopathy. Increased uptake throughout the lumbar vertebral lesions was also seen. A CT-guided biopsy of the right apical lung nodule and one of the vertebral lesions (L3) revealed noncaseating granulomas consistent with sarcoidosis. We introduce steroid treatment with favorable evolution of vertebral lesions.

Vertebral sarcoidosis cannot be certainly differentiated from metastatic disease based on imaging only. Accurate diagnosis is only attainable by histopathological verification of the lesions.

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Background

Sarcoidosis is a systemic disease that most commonly affects the lungs. Vertebral involvement is rare and its radiological

features are nonspecific. It is difficult to be differentiated from metastatic disease or vertebral tuberculosis. Therefore, histological confirmation is always recommended. Vertebral lesions rarely resolve without specific treatment and corticosteroids are the first therapy indicated.

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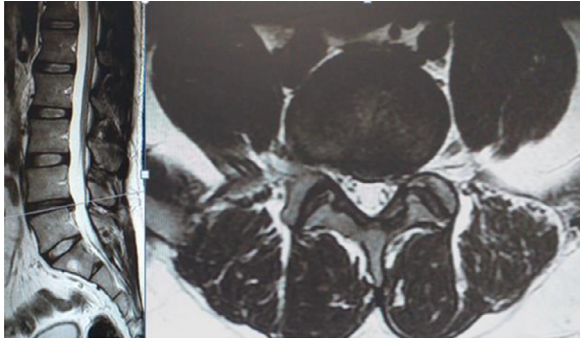


Fig. 1 – T2W MRI in sagittal and axial plans showing left paramedian lumbar disc herniation at L4–L5. The left L5 nerve is compressed.

We report the case of concomitant asymptomatic pulmonary, lymphatic, and vertebral sarcoidosis with favorable evolution of the vertebral manifestations and gradual involution on magnetic resonance imaging (MRI) with steroid treatment.

Case presentation

A 35-year-old male with no significant medical history came to clinical examination for lower back and associated bilateral lower extremity pain. Due to the persistence of lumbar radiculopathy a MRI was performed to exclude disc-related pathology. It shows left paramedian lumbar disc herniation at L4–L5 level that creates disc-nerve root conflict (Fig. 1). However, multiple T1 low-signal small lesions throughout the lumbar vertebral bodies and posterior elements, which were isosignal or bright on T2, and showed enhancement after gadolinium administration were discovered incidentally (Fig. 2). A broad differential diagnosis was considered including metastatic disease, malignant hemopathy, atypical hemangiomas, disseminated infection, and vertebral sarcoidosis. He underwent exhaustive laboratory tests for hematologic and solid tumor malignancies including serum protein electrophoresis, alpha fetal protein hormone levels, and a fecal occult blood test that were all negative. A slightly elevated erythrocyte sedimentation rate was noticed. Since the findings were concerning diffuse metastatic disease, an 18F-labeled fluorodeoxyglucose (18F-FDG) was performed that shows hypermetabolic



Fig. 2 – Vertebral sarcoidosis. (a,b) T1, (c,d) T2, and (e,f) T1 fat-saturated postgadolinium sagittal and axial magnetic resonance images of the lumbar spine demonstrate multiple small T1 hypointense, T2 iso- to hyperintense, and postgadolinium enhancement of lesions in lumbar vertebral bodies and posterior elements.

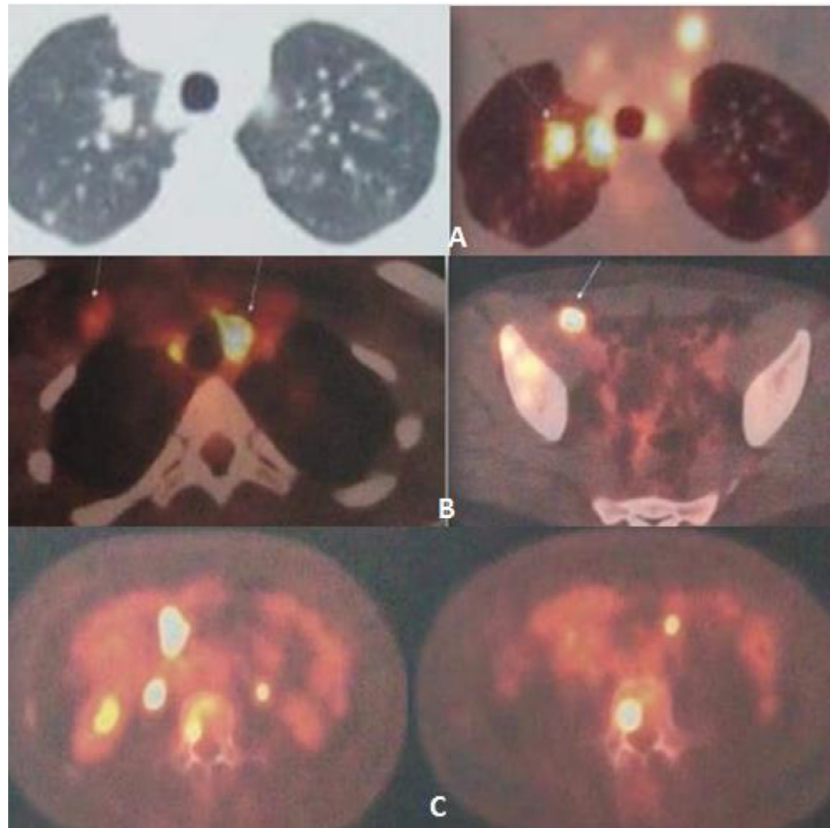


Fig. 3 – FDG-PET scan showed hypermetabolic apical right nodule of the lung parenchyma (A) with multiple enlarged mediastinal and right iliac external adenopathy (B) associated to increased uptake throughout the lumbar vertebrae (C).

apical right nodule of the lung parenchyma with multiple enlarged mediastinal and right iliac external adenopathy associated with increased uptake throughout the lumbar vertebral lesions shown in the MRI. These findings may concern lymphoma or metastatic lung cancer (Fig. 3). The intradermal tuberculin test was negative for tuberculosis. Considering all this, a computed tomography (CT)-guided percutaneous biopsy of the right apical lung nodule was decided. The result was significant for noncaseating and necrotizing granulomas without evidence of metastatic disease or other pathology.

Since the vertebral lesions are not specific and may mimic hematologic or solid tumor metastasis and even in the presence of proven systemic sarcoidosis, another percutaneous CT-guided biopsy of one of the vertebral lesions (L3) revealed noncaseating granulomas, compatible with bone sarcoidosis.

These findings were consistent with asymptomatic concomitant multisystemic sarcoidosis and vertebral involvement. Corticoid therapy was started and the patient was scheduled for a 9-month follow-up with MRI for the spinal lesions, because of their multiplicity and for the heterogeneous aspect. The recommended follow-up MRI showed complete resolution of the areas of marrow signal abnormality (Fig. 4). The pulmonary and lymphatic lesions also favorably responded to therapy.

Discussion

Sarcoidosis is a multisystem disease. The involvement of bone occurs in 5% of patients and it is usually an incidental finding [1]. In a recent review of 22 cases of vertebral sarcoidosis, the lesions were without exception discovered during a workup for back pain [2], and that was even the case in our patient. The lumbar pain was related to disc herniation at L4–L5 level that creates disc-nerve root conflict and not to vertebral lesions.

Vertebral sarcoidosis is difficult to be differentiated from metastatic lesions based on MRI. MRI in vertebral sarcoidosis is nonspecific. The lesions are solitary or multiple and can be predominantly lytic, sclerotic, or mixed. The disc spaces are usually untouched, as in our patient. Extension into the posterior elements has been described, but isolated involvement of the pedicles is rare [3]. The most-reported MRI features are T1-weighted hypointensity and T2-weighted hyperintensity with varying degrees of contrast enhancement depending on nature (osteolytic vs osteosclerotic) and activity.

Osteolytic lesions of sarcoidosis have long T1 and T2 values and pronounced contrast enhancement.

Osteosclerotic lesions tend to be hypointense on T1-weighted, isointense to hypointense on T2-weighted sequences, and nonenhanced after gadolinium administration [2].



Fig. 4 – (A to C) Pretreatment and (D to F) Post-treatment: Sagittal magnetic resonance images of the lumbar spine demonstrate regression of the multiple lesions previously identified. No abnormal enhancement is seen.

In our patient, we found mixed osteolytic and osteosclerotic presentations located in the vertebral bodies of L2, L3, L4, and the posterior arc of L4.

Bone scintigraphy is nonspecific but may be a sensitive indicator of the extent of osseous sarcoidosis [4]. FDG-(18 F) PET-CT has also been described as a sensitive technique [5].

Osseous sarcoidosis tends to have an increased uptake on the 18F-FDG-PET scan since sarcoidosis is a hypermetabolic, inflammatory mechanism, and incriminates the activation of macrophages. Therefore, an 18F-FDG-PET scan may cause false-positive results and confuse the diagnosis of metastatic neoplastic disease which was the case of our patient.

This common appearance makes a distinction from metastatic bone lesions very hard based on imaging alone, and that is why histological confirmation is indicated.

Our case and the cases reported in the literature underline the importance of biopsy confirmation of the bone lesions even do the existence of lung sarcoidosis.

Vertebral sarcoidosis is frequently treated with corticosteroids, but in some controversial cases, other drugs like methotrexate and anti-TNF agents have been used [6,7]. There are some reported cases of improvement without treatment and without leaving a detectable trace [3,7]. In our patient corticosteroids have been used as first-line drugs with spectacular regression of the lesions. The appearance of intraregional fat in osseous sarcoidosis as a viable sign of lesion involution has been reported before [8].

Before making a diagnosis of sarcoidosis, all other causes of noncaseating epithelioid granuloma should be eliminated. Accurate diagnosis is only possible with histopathological

verification. We present this case in the hope to extend the knowledge of vertebral sarcoidosis where the experience with this particular entity is limited because of its rarity.

It is usually secondary to other organ involvement, localized to a few vertebrae, and combined osteosclerotic and osteolytic aspects. Our patient was particular with no symptoms from vertebral involvement, no manifest presence of sarcoidosis in other organs except for incidental pulmonary nodules, and the recognition of this pattern has led to early complete resolution after corticosteroid therapy.

Patient consent

Patient consent obtained.

REFERENCES

- [1] James DG, Neville E, Carstairs LS. Bone and joint sarcoidosis. *Semin Arthritis Rheum* 1976;6(1):53–81.
- [2] Poyanli A, Poyanli O, Sencer S, Akan K, Sayrak H, Acunas B. Vertebral sarcoidosis: imaging findings. *Eur Radiol* 2000;10:92–4.
- [3] Johnson AK, Johnson JM, Ames E, Filippie C. Spontaneous clinical and radiological resolution of vertebral sarcoidosis. *Spine* 2012;37:414–16.
- [4] Rua-Figueroa I, Gantes MA, Erausquin C, Mhaidli H, Montesdeoca A. Vertebral sarcoidosis: clinical and imaging findings. *Semin Arthritis Rheum* 2002;31:346–52.
- [5] Valencia MP, Deaver PM, Mammarrappallil MC. Sarcoidosis of the thoracic and lumbar vertebrae, mimicking metastasis or multifocal osteomyelitis by MRI: case report. *Clin Imaging* 2009;33:476–81.
- [6] Sakellariou GT, Anastasilakis AD, Karanikolas D, Vounotrypdis P, Berberidis C. Central skeletal sarcoidosis: a case report with sustained remission only on methotrexate, and a literature review on the imaging approach, treatment, and assessment of disease activity. *Mod Rheumatol* 2013;23:175–81.
- [7] Salmon JH, Perotin JM, Direz G, Brochot P, Laredo JD, Eschard JP. Vertebral sarcoidosis. Spontaneous favorable outcome: a case report and literature review. *Rev Med Interne* 2013;34:42–6.
- [8] Moore SL, Kransdorf MJ, Schweitzer ME, Murphey MD, Babb JS. Can sarcoidosis and metastatic bone lesions be reliably differentiated on routine MRI? *AJR Am J Roentgenol* 2012;198:1387–93.