A Disconcerting Sarcoidosis, When The Spine Gets Involved...

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Swiss Journal of Radiology and Nuclear Medicine - www.sjoranm.com - SJORANM GmbH - CH-6340 Baar - Switzerland

Abstract

Bone involvement in sarcoidosis is uncommon and may be discovered incidentally when pain appears in affected patients. Indeed, the pelvic-spinal location is the most widespread: It can manifest when the spine is affected by inflammatory pain, sometimes mimicking authentic spondyloarthritis in certain cases, and may even suggest a malignant lesion by its radiological appearance. We describe the original observation of a patient suffering from inflammatory and insomnia-causing spinal pain in whom the diagnosis of mediastinal sarcoidosis was retained in therapeutic abstention on the pulmonary level.

Observation: 52-year-old female patient who had presented for barely a year a chronic cough accompanied by basithoracic pain, leading her to perform a chest CT scan which found bilateral mediastinal-hilar adenopathies and peri-lymphatic pulmonary nodules. The diagnosis of type 2 sarcoidosis was immediately considered. The additional investigations have indeed confirmed the suspected disease on the clinical and radiological level, in particular by converse enzyme positivity, the tuberculin anergy on the IDR, the lymphocyte predominance in the bronchoalveolar fluid but more particularly by the demonstration of the epithelial-giant cellular granuloma on the right iliac lymph node biopsy. However, the patient presents with intense inflammatory back pain, sometimes causing insomnia. A spinal MRI showed nodular bone marrow replacement lesions with pronounced T1 hyposignal and T2 hypersignal enhanced after gadolinium injection. Fortunately, she does not present bone fractures and no osteoporosis on bone densitometry.

Conclusion: Faced with spinal symptoms in a context of sarcoidosis, bone involvement must be considered, especially since there is no correlation between the clinical picture and imaging. The latter most often has a favorable prognosis in the absence of fractures despite sometimes suspicious imaging. The case of our patient clearly illustrates the bone-related nature of the disease; back pain can be particularly intense and should lead to discussion of the use of corticosteroids or even basic treatment for rheumatic diseases.

Keywords: *bone sarcoidosis, inflammatory back pain, spinal MRI*

*Corresponding author: Selma Abdellaoui - received: 21.07.2025 - peer reviewed, accepted and published: 31.07.2025

Introduction

Bone involvement in sarcoidosis is rare, occurring in around 3% according to the literature [1]. All bones can be affected. The spine is the most common location at 47%, followed by the pelvis at 40% [2], and this pelvic-spinal involvement is almost always associated with mediastinal-pulmonary involvement [3, 4]. Back pain is pauci-symptomatic and generally has a good prognosis. Consequently, bone lesions are detected incidentally in half of patients by imaging data [5]. Indeed, MRI or even PET of the spine and pelvis remains a key examination for detecting spinal cord replacement lesions with a high fracture risk and allows for the elimination of other diseases, particularly spondyloarthritis, infection, or malignant lesion [6, 7]. We will describe the case of a patient suffering from atypical bone sarcoidosis with paradoxically intense inflammatory back pain and worrying imaging in the form of staged

lytic lesions in therapeutic abstention due to the pulmonary grading of the sarcoidosis.

Case Report

This is the patient T.N, aged 52, with a history of hypertension under β blockers and operated uterine fibroids, who was hospitalized in pulmonology due to the appearance of a chronic dry cough dating back ten months accompanied by basithoracic pain. Pulmonary auscultation was normal, however a thoracic CT scan had found mediastinal-hilar adenopathies with peri-lymphatic pulmonary nodules, noting some lytic bone lesions of the T4, T5 and T11 vertebrae. Indeed, the patient's main complaint was intense insomnia-causing inflammatory back pain predominantly at the dorsolumbar junction without any notion of trauma or general signs. The possibility of mediastinal sarcoidosis was confirmed: the IDR found tuberculin anergy, the converting enzyme was at 104U/L, the BAL was 20% lymphocytic and the epithelial-giantocellular granuloma was present on the CT-guided iliac biopsy. Regarding the spinal complaint, a malignant cause was eliminated: The tumor markers were normal, the thoraco-abdominopelvic CT revealed scattered pulmonary nodules with irregular contours not exceeding 9 mm associated with bilateral mediastinal-hilar adenopathies, iliac adenopathies grouped ino clusters, the largest of which measured 22 mm in short axis, a condensing lesion of the posterior arch of the 7th left rib and costal fractures of the 3rd, 7th ribs on the left.

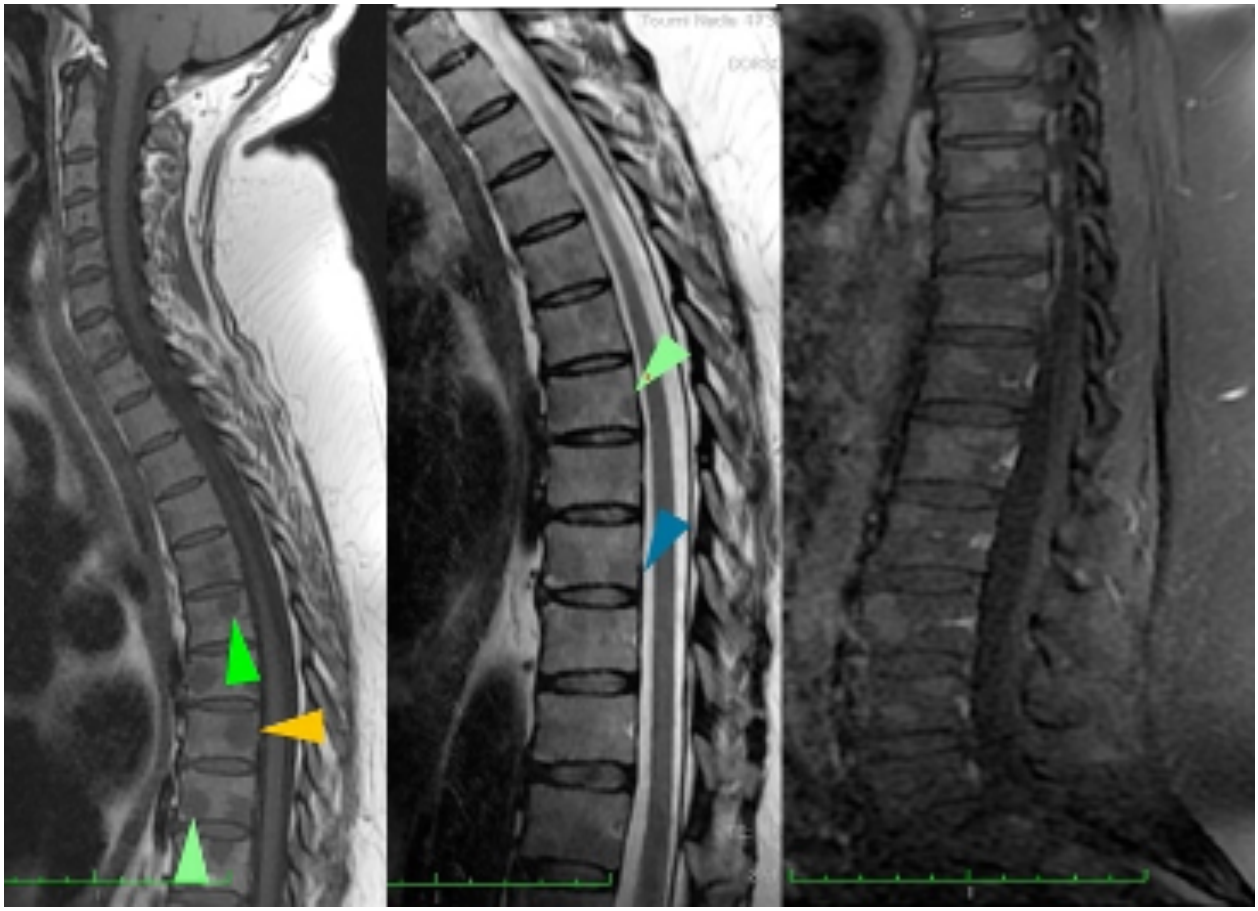

Figure 1: *Appearance of bone lesions on spinal MRI: T1 and T2 hypointense signal and STIR hyperintense signal.*

The scintigraphy found an expression with regard to the vertebral lytic lesions, discrete spots of hyperfixation of the 2nd, 6th left ribs and the 7th right. There was a biological inflammatory syndrome with ESR at 79mm and CRP at 12mg/l. Nevertheless, the calcium and vitamin D levels being normal, the mammography, cervical ultrasound, ENT examination as well as the vaginal smear were without particularity. Given the absence of involvement of the pulmonary parenchyma without repercussions (functional capacity at 80%, normal walking test and normal cardiac ultrasound) the patient was discharged without any treatment. The fracture risk assessment showed a normal densitometry and VFA.

Figure 2a: *enhanced spinal nodular lesions.*

Spinal MRI demonstrated diffuse lesions involving the spine, pelvis, and proximal femora, appearing as nodular lesions with low signal intensity on both T1- and T2-weighted sequences. These lesions were not suppressed on fat-saturated sequences and showed enhancement following gadolinium administration. The lesions varied in size, mostly measuring a few centimeters, with some larger foci: the largest measured 26 mm in the lumbar recess, 20 mm in the thoracic spine, 45 mm at the sacral alae, and 35 mm in the pelvis (*Figures 1, 2a, and 2b*). In contrast, no abnormalities were detected on CT (*Figure 3*).

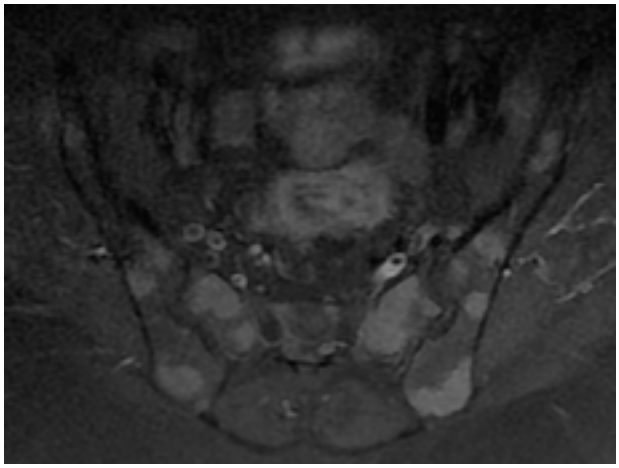

Figure 2b: *Axial T1FS MRI with contrast media: enhanced nodular lesions of the sacral and iliac parts.*

Discussion

Many studies suggest that spondyloarthritis should be investigated in the presence of inflammatory back pain during sarcoidosis. Indeed, the non-fortuitous association between spondyloarthritis and sarcoidosis has already been described in the literature. Kobak S. [8] observed 14.3% of sacroiliitis in 42 sarcoidosis patients. Recently, comparative studies have found a high prevalence of thyroid diseases (OR: 1.3), Sjögren's syndrome (OR: 11.6) and spondyloarthritis (OR: 3.80) in 1237 sarcoidosis patients compared to 4948 subjects without sarcoidosis [9]. Another cross-sectional study of 64 sarcoidosis cases showed that 45.3% had inflammatory back pain, 19% had magnetic sacroiliitis, 7.8% had radiographic findings, and 10.9% had radiological syndesmophytes [10, 11].
Establishing a correlation between clinical and imaging findings remains difficult: Perispinal involvement is asymptomatic in half of cases and has a relatively good prognosis [12], despite sometimes very worrying imaging findings. The radiological appearance is not specific and does not allow the elimination of other diseases such as bacterial infections or malignant lesions [13, 14].

MRI is the key diagnostic examination, it shows nodular, micronodular, or beach-like bone marrow replacement lesions, sometimes combined in T1 hyposignal (often pronouced) and T2 hypersignal, enhanced after gadolinium injection. On PET, we find diffuse heterogeneous hyperfixation [15].
Complications of the neurological compression type requiring surgical stabilization remain exceptional [12] and above all the ab-

Figure 3: *CT sagittal total spine reconstruction.*

sence of abnormality on CT remains an important argument for retaining the diagnosis. Bone biopsy may be indicated in difficult cases. The risk of fracture in this type of injury must be taken into consideration, therefore opportunistic screening and, at best, upstream prevention is required. Moreover, vitamin D supplementation is part of the management of bone fragility while ensuring, however, that it is adapted to the risk of hypercalcemia inherent in the disease.

This fragility can itself be responsible for the alteration of functional respiratory pulmonary performance. This risk is increased when the patient is on corticosteroid therapy or has chronic respiratory failure [13]. For our patient, back pain is symptomatic; a basic anti-rheumatic treatment such as hydroxychloroquine or methotrexate should be discussed, as should anti-osteoporotic treatment (bisphosphonates) for rib fractures.

Conclusion

Bone sarcoidosis remains rare; it should be screened for any spinal pain occurring in the context of this disease. Our case clearly illustrates the atypical nature of the pain, which is a prominent symptom. MRI imaging was of great value; despite its worrisome lesion, it allowed us to rule out serious etiologies and confirm the diagnosis. It would be wise to initiate long-term treatment, ideally after a multidisciplinary discussion, to provide relief to the patient.

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Declarations

Consent for publication: The author clarifies that written informed consent was obtained and the anonymity of the patient was ensured. This study submitted to Swiss J. Rad. Nucl. Med. has been conducted in accordance with the Declaration of Helsinki and according to requirements of all applicable local and international standards.
Competing interests: No competing interests.
Funding: No funding resources.

Conflict of interest:

The authors declare that there were no conflicts of interest within the meaning of the recommendations of the International Committee of Medical Journal Editors when the article was written.

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