


Pelvic and Lumbar Pain Revealing Bone Sarcoidosis: A Case Report

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Yahya Charifi¹ , Younes Dkhissi¹, Meryem Haloua¹,
Youssef Alaoui Lamrani¹, Ghizlane Berrady², Meryem Boubbou¹,
Afaf Amarti³, Mustapha Maaroufi¹ and Badreddine Alami¹

¹Radiology and Imaging Department, Centre Hospitalier Universitaire Hassan II, University Sidi Mohamed Ben Abdellah, Fez, Morocco, ²Internal Medicine Department, Centre Hospitalier Universitaire Hassan II, University Sidi Mohamed Ben Abdellah, Fez, Morocco,

³Anatomopathology Department, Centre Hospitalier Universitaire Hassan II, University Sidi Mohamed Ben Abdellah, Fez, Morocco

ABSTRACT: A 54-years-old woman suffered from a bilateral pelvic and lumbar pain. She had a systemic investigation shows a diffuse bone lesions. A biopsy of iliac bone was done and revealed sarcoidosis. Bony manifestations of sarcoidosis are rare and challenging to diagnose. Our report shows that MRI with diffusion weighted sequences is a simple and non invasive technique to evaluate bone infiltration. It helps to distinguish inflammatory disease from metastatic lesions and to guide the osseous biopsy. Moreover, fluorodeoxyglucose PET scan, are useful in establishing the extent of damage and measuring disease activity.

KEYWORDS: Sarcoidosis, CT scan, biopsy, iliac bone

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CORRESPONDING AUTHOR: Yahya Charifi, Centre Hospitalier Universitaire Hassan II, Fez 30050, Morocco. Email: y.charifi92@gmail.com

Introduction

Our case is about a patient who was diagnosed with sacroiliac involvement of osseous sarcoidosis, which had some similarities with myeloma or metastases, rejected by immunohistochemistry.

The treatment of bone manifestations of sarcoidosis is not specific. Some cures of moderate doses of corticosteroids or hydroxychloroquine have been used in most cases.

In the other hand, other patients have benefited from immunosuppressive treatment, especially methotrexate, with a good response and no recurrence of symptoms.

Case Presentation

Fifty-four years old female, without any medical history, suffered from a bilateral mechanical and inflammatory pelvic and lumbar pain, developing since the last 2 years. Firstly, lumbar arthritis or ankylosing spondylitis were suspected.

The patient was treated with non-steroidal anti-inflammatory drugs, with minor improvement but persistence of the symptoms.

The clinical evolution resulted on a profound asthenia which also suggested tuberculosis given the high incidence on local context.

Biological tests results showed a normal C-Reactive-Protein, without hyperleukocytosis, which excluded an inflammatory syndrome. Sputum samples analysis to identify mycobacterium tuberculosis complex has been done, and completed by an RT-PCR which were negative.

On the first line of investigation, a CBC test, Protein electrophoresis, vitamin D dosage, and serology were done without abnormality.

On the second line of investigation, since lumbar pain has not resolved by the treatment, imaging was carried out.

A whole body computerized tomography (CT)-scan showed para aortic and retro peritoneal lymphadenopathy with no other abnormalities (Figure 1) suggesting a secondary lymphatic node localization or lymphoma. A 18F-fluorodeoxyglucose positron-emission tomography showed a nodular uptake in adenopathy and a diffuse non-specific uptake in iliac bone (Figure 2).

In the other hand, magnetic resonance imaging (MRI) showed a pelvic bone lesions in T1 contrast sequence and diffusion perfectly correlated to the bone uptake zones which were accessible to osseous biopsy (Figure 3).

The histological results revealed the presence of intense lymphoid infiltrates, possibly of a reactive nature, associated with a few epithelioid cells without caseous necrosis compatible with a granulomatous lesion (Figure 4).

The treatment decision was a long-term systemic corticosteroids (prednisone) combined with an adjuvant treatment based on vitamin D and calcium with biological supervision. A 6-month treatment has been initiated, it is still in progress. A spectacular recovery was noted within the first month.

Discussion

Sarcoidosis is a multisystemic inflammatory disease without a clear causal etiology, it characterized by the presence of epithelioid and giganteo-cellular granulomas without caseous necrosis. This disease frequently appears as tracheobronchial axis-parallel hilar lymphadenopathy associated with interstitial lung involvement that may lead to pulmonary fibrosis.²¹



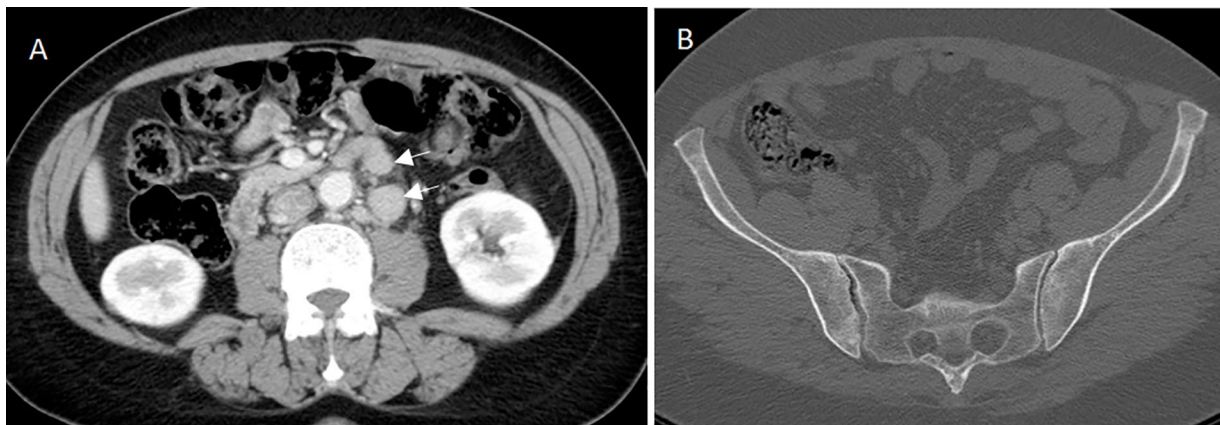


Figure 1. (A) Voluminous para aortic and retro peritoneum lymphadenopathy (arrowhead). (B) No visible bone lesion on the CT scan that could motivate a biopsy.

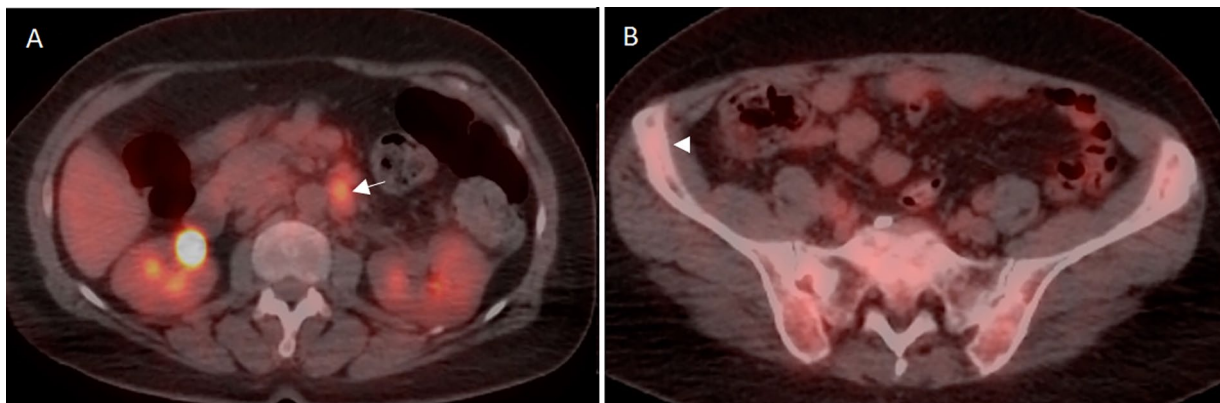


Figure 2. 18F-fludeoxyglucose positron-emission tomography: (A) Uptake in adenopathy (arrow), (B) diffuse and non-specific uptake on iliac bone (arrowhead).

Bony involvement is uncommon,¹ mostly affects peripheral small bones such as the phalanges of the hands and feet, it is observed in 5% to 7% of patients with sarcoidosis. This localization is common in black-skinned people and women aged 30 to 50 years with chronic multisystemic disease activity. These skeletal manifestations of the disease, may be unilateral or bilateral and usually asymmetrical.² However, in some studies, it may affect axial skeleton, skull, long bones, and ribs.³

The pelvic form as in our case is not classical and tends to be similar to inflammatory sacroiliitis secondary to spondyloarthropathy or infectious sacroiliitis (favored by long-term corticosteroid therapy). In the literature, bone sarcoidosis and inflammatory sacroiliitis might be associated in young patients. But in our study, the patient age did not allowing us to evoke spondyloarthropathy.⁴⁻⁶

In axial forms of bone sarcoidosis, sclerotic lesions are the most common, may be confused with metastases.⁷ In a study by Zhou et al,⁸ the spine and pelvis were the site of sarcoid bone lesions in 68.8% and 35.9% of cases, respectively.

Sarcoidosis bone lesions in the area of the hands are found in 15% of cases and sometimes give a “moth-eaten” appearance with soft tissue swelling.²²

Clinically, pain is mostly seen in skeletal sarcoidosis and may be the initial sign of the disease,⁸ it can be moderate or disabling.⁹ The spinal location may result in neurological symptomatology in most cases. However, some authors have stated that bone damage may be asymptomatic. On the other hand, bone symptoms may identify sites for an imaging guided biopsy.⁸

Targeted biopsy is the key to diagnosis bone sarcoidosis. Imaging, while insensitive and non-specific, is often helpful for guiding the biopsy. Positron emission tomography (PET) with 18F-fludeoxyglucose is proposed as a valuable method for the detection of bony manifestations that cannot be visualized by radiography or CT scan.^{10,11} It is difficult to differentiate lesions of disseminated (or solid) sarcoidosis from metastases or other malignant infiltration such as myeloma. Indeed, some authors have described that false positives of bone metastases can be mimicked by multifocal sarcoidosis on 18F-FDG PET/CT,¹²⁻¹⁴ due to granulomatous bone marrow infiltration which may show 18F-FDG uptake.^{15,16}

MRI is very useful and it can be performed as a second line of diagnosis when CT and PET scans are non-conclusive. In our case, the diffusion sequence was able to highlight target lesions

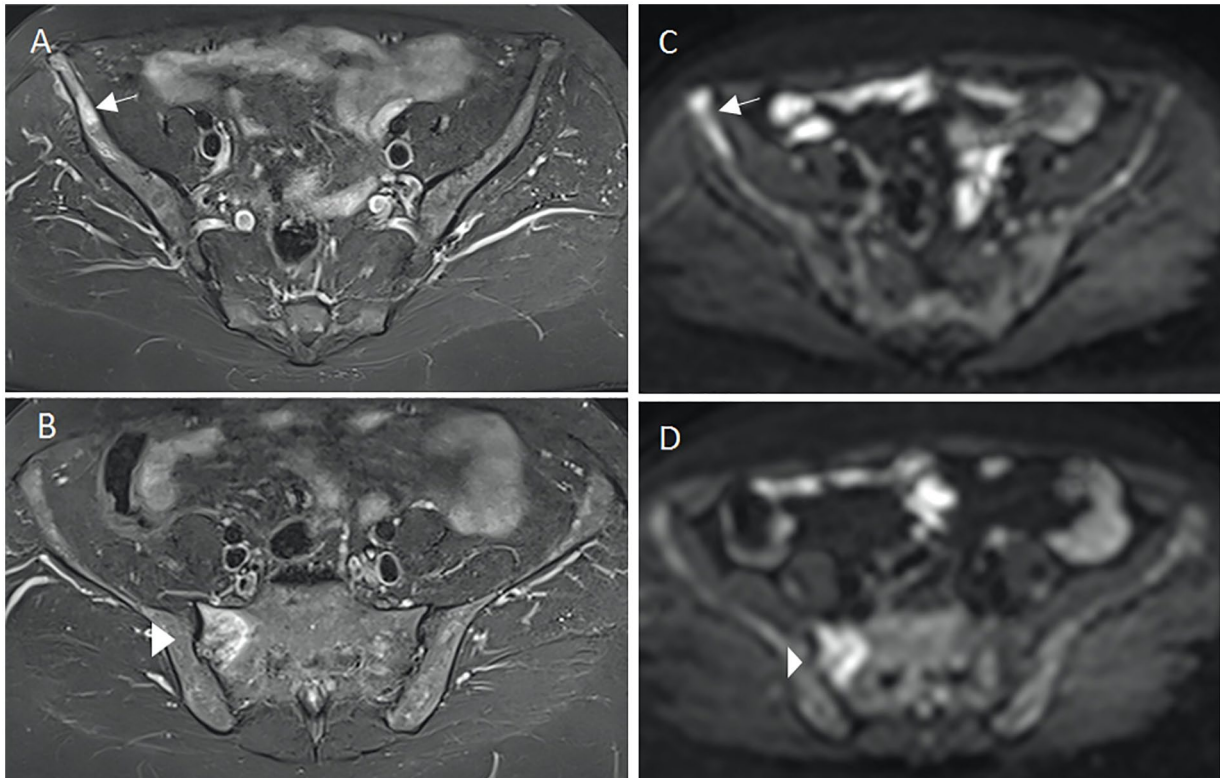


Figure 3. (A and B) T1 contrast sequence with fat suppression: multiple polymorphic lesions of the pelvic bone structure, involving the iliac spine (arrow) and sacroiliac joint (arrowhead). The cortical bone is continuous, suggesting a benign, granulomatous origin. (C and D) Diffusion weighted sequence shows their active character on the iliac bone (arrow) and sacroiliac joint (arrowhead).

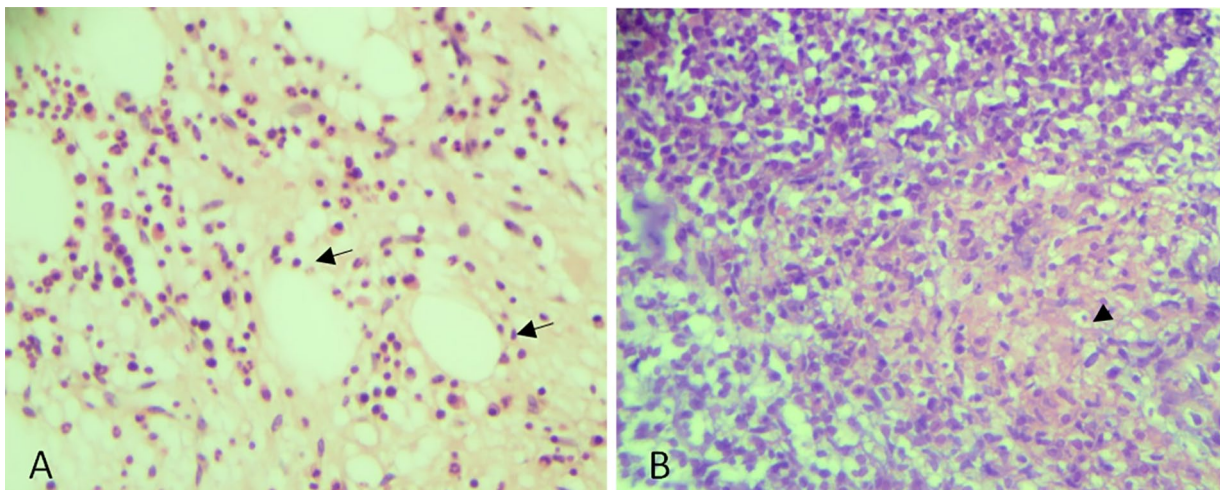


Figure 4. Few epithelioid cells (arrow) (A) associated with granuloma and lymphoid cells (arrowhead) (B).

for bone biopsy (right iliac wing), the value of diffusion sequence was demonstrated in such cases of difficult diagnosis.¹⁷ Indeed, it is used to evaluate the average microscopic movements of water in tissues at the millimeter scale of MR images. Thus, the hypercellular zones present a restriction of the diffusion of water molecules with a hypersignal on the MRI images.¹⁸

In our study, MRI shows a continuous cortical bone suggesting a benign granulomatous origin but we were unable to make a formal decision on benign or malignant nature of the bone lesions. This is why a biopsy was performed on an active hypercellular area of the right iliac wing, as it presented a

diffusion restriction with a high signal on the T1 with contrast injection sequence.

Regarding the therapeutic management, no consensus exists on the treatment of bone sarcoidosis. Although asymptomatic subjects do not seem to need treatment.¹⁹ Since bone involvement is often a sign of a multisystemic disease, many patients will be treated for other organ manifestations. Zhou et al⁸ described that the majority of patients with bone sarcoidosis had associated organic lesions, as in our case, where lymph node involvement was observed. Therefore, 79.7% of patients in his series were users of

prednisone at doses ranging from 20 to 40 mg daily to <10 mg daily. Corticosteroids are however given as first line therapy for bone sarcoidosis, but such therapy is often limited by adverse events.

Methotrexate, was considered to be useful for organic manifestations,¹⁹ and may prevent a recurrence of symptoms. Tumor necrosis factor inhibitors are also used for other systemic manifestations of sarcoidosis as described in an earlier case report.²⁰ In rare cases, temporary treatment with moderate-dose corticosteroids or hydroxychloroquine have been most commonly used.⁷

Conclusion

Bone sarcoidosis is still an unusual diagnosis and is more common in women. Musculoskeletal lesions are usually associated with a multi-systemic condition, as in our case with extra-thoracic lymph nodes.

Biopsy is considered the key examination, but diffusion MRI has proven to be a non-invasive and radiation-free technique, useful for the evaluation of bone infiltration to distinguish inflammatory bone marrow disease from metastatic disease. It is overcoming the limitations of 18F-FDG PET/CT.

Infliximab plays an important role in the treatment of resistant forms of bone sarcoidosis and is often administered as first or second line therapy.

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Author Contributions

Alami Badreddine and Yahya Charifi performed diagnosis and biopsy, Afaf amarti's service provided the anatomopathological analysis. All authors read and approved the final manuscript.

Ethics Approval and Consent to Participate

Our institution does not require ethical approval for reporting individual case or case series.

Consent Information

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

ORCID iD

Yahya Charifi  <https://orcid.org/0000-0002-7008-7346>

Availability of Data and Materials

The data sets are generated on the data system of the CHU hassan II of Fes, including the biological data, the operative report, and the data of the anatomopathological analysis

REFERENCES

- Newman LS, Rose CS, Maier LA. Sarcoidosis. *N Engl J Med.* 1997;336:1224-1234.
- Thelie N, Allanore Y. Localizaciones osteoarticulares de la sarcoidosis. *EMC-Aparato Locomotor.* 2009;42:1-11.
- Kucharz EJ. Osseous manifestations of sarcoidosis. *Reumatologia.* 2020;58:93-100.
- Kremer P, Gallinet E, Benmansour A, Despau J, Toussiro E, Wendling D. Sarcoidosis and spondylarthropathy. Three case-reports. *Rev Rhum Engl Ed.* 1996;63:405-411.
- Jansen TLTA, Geusens PPM. Sarcoidosis: joint, muscle and bone involvement. *Eur Respir Monogr.* 2005;32:210.
- Cadiou S, Robin F, Guillin R, et al. Spondyloarthritis and sarcoidosis: related or fake friends? A systematic literature review. *Joint Bone Spine.* 2020;87:579-587.
- Sparks JA, McSparron JL, Shah N, et al. Osseous sarcoidosis: clinical characteristics, treatment, and outcomes—experience from a large, academic hospital. *Semin Arthritis Rheum.* 2014;44:371-379.
- Zhou Y, Lower EE, Li H, Farhey Y, Baughman RP. Clinical characteristics of patients with bone sarcoidosis. *Semin Arthritis Rheum.* 2017;47:143-148.
- Bechman K, Christidis D, Walsh S, Birring SS, Galloway J. A review of the musculoskeletal manifestations of sarcoidosis. *Rheumatology.* 2018;57:777-783.
- Akaike G, Itani M, Shah H, Ahuja J, Yilmaz Gunes B, Assaker R, Behnia F. (2018). PET/CT in the diagnosis and workup of sarcoidosis: focus on atypical manifestations. *Radiographics,* 38(5), 1536-1549.
- Makis W, Palayew M, Rush C, Probst S. (2018). Disseminated multi-system sarcoidosis mimicking metastases on 18F-FDG PET/CT. *Molecular imaging and radionuclide therapy,* 27(2), 91.
- Baldini S, Pupi A, Di Lollo S, Marchionni N, Shraim R, Bosi A. (2008). PET positivity with bone marrow biopsy revealing sarcoidosis in a patient in whom bone marrow metastases had been suspected. *British journal of haematology,* 143(3), 306-306.
- Aberg C, Ponzio F, Raphael B, Amorosi E, Moran V, Kramer E. FDG positron emission tomography of bone involvement in sarcoidosis. *AJR Am J Roentgenol.* 2004 Apr;182(4):975-7. doi: 10.2214/ajr.182.4.1820975. PMID: 15039174.
- Talmi D, Smith S, Mulligan ME. Central skeletal sarcoidosis mimicking metastatic disease. *Skeletal Radiol.* 2008;37:757-761.
- Gotthardt M, Blecker-Rovers CP, Boerman OC, Oyen WJ. Imaging of inflammation by PET, conventional scintigraphy, and other imaging techniques. *J Nucl Med.* 2010;51:1937-1949.
- Mostard RL, Prompers L, Weijers RE, et al. F-18 FDG PET/CT for detecting bone and bone marrow involvement in sarcoidosis patients. *Clin Nucl Med.* 2012;37:21-25.
- Moore SL, Kransdorf MJ, Schweitzer ME, Murphey MD, Babb JS. Can sarcoidosis and metastatic bone lesions be reliably differentiated on routine MRI? *Am J Roentgenol.* 2012;198(6):1387-1393.
- Woolf DK, Padhani AR, Makris A. Assessing response to treatment of bone metastases from breast cancer: what should be the standard of care? *Ann Oncol.* 2015;26:1048-1057.
- Sweiss NJ, Lower EE, Korsten P, Niewold TB, Favus MJ, Baughman RP. Bone health issues in sarcoidosis. *Curr Rheumatol Rep.* 2011;13:265-272.
- Hasni SA, Kunz D, Finzel K, Gruber BL. Osseous sarcoidosis treated with tumor necrosis factor-inhibitors: case report and review of the literature. *Spine (Phila Pa 1976).* 2010;35:E904-E907.
- Costabel U, Hunninghake GW. ATS/ERS/WASOG statement on sarcoidosis. Sarcoidosis Statement Committee. American Thoracic Society. European Respiratory Society. World Association for Sarcoidosis and Other Granulomatous Disorders. *Eur Respir J.* 1999;14:735-737.
- Aptel S, Lecocq-Teixeira S, Olivier P, Regent D, Teixeira PG, Blum A. Multimodality evaluation of musculoskeletal sarcoidosis: imaging findings and literature review. *Diagn Interv Imaging.* 2016;97:5-18.