Case Report

MR imaging appearance of long bone sarcoidosis

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Abstract

Sarcoidosis is a non-caseating granulomatous disease with common involvement of the lungs and lymph nodes. Osseous involvement is rare, with long bone involvement even rarer. One of the characteristic MRI findings that point towards osseous sarcoidosis is the presence of intra- or perilesional fat. Lesions that involve long bones usually show no cortical destruction or periosteal reaction, in contrast to small bones, which show cortical destruction or extraosseous extension. In patients with diagnosed sarcoidosis, bone biopsy could be averted in patients having characteristic findings on MRI. In patients without a diagnosis of sarcoidosis with multiple osseous lesions and aforementioned imaging findings, sarcoidosis can be provided on the differential diagnosis.

Keywords: Bone, MRI, Sarcoidosis.

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Introduction

Sarcoidosis is a multi-system inflammatory disorder with non-caseous granulomas of unknown etiology in the involved organs. It has bimodal distribution with the first peak between ages 20-40 and the second peak being above 50 years and older [1]. Mediastinal lymph nodes and lungs are most commonly affected in approximately 90% of the patients with sarcoidosis [2]. Osseous sarcoidosis (OS) is not very common and involves only 1-13% of the patients with sarcoidosis. The appendicular skeleton, in particular the hands and feet, are commonly affected. The axial skeleton is involved to a lesser degree where it may resemble metastasis [3]. Knowledge of OS may not be well established in clinical practice as the reported prevalence is low. For example, Neville et al. reported OS in 1-15% of patients with sarcoidosis [4]. Similarly, James et al. [5] reported prevalence of 3-13% and Sparks et al. [6] reported around 1.5%. The low prevalence of OS may be due to underdiagnosis as patients with OS may be asymptomatic. However, in patients with diagnosed sarcoidosis, bone biopsy can be averted if there are certain imaging features pointing towards OS, which will be discussed further in the article.

Here we present two cases of OS showing contrasting features on MRI. According to authors' knowledge, there is no reported case of a patient with breast cancer with multiple long bone lesions secondary to OS and was also followed up for 4 years with MRI to establish stability and to exclude the possibility of metastases.

Case summary

Case 1

A 65-year-old man presented with shoulder pain for 6 months. The initial radiograph of the shoulder was negative. Since the symptoms were persistent and rotator cuff tear was suspected, MRI of the shoulder was ordered which demonstrated T1 hypointense and T2 hyperintense intramedullary lesions in the proximal humerus. There was a small focus of the central intrinsic T1 hyperintense signal and the corresponding low signal on fat saturated images, which was consistent with

intralesional fat (Figure 1). There was minimal endosteal thinning of the cortex suggesting a benign and chronic process. Eventually a biopsy was performed to rule out malignancy. The pathology demonstrated multiple non-caseating granulomas consistent with OS. A subsequently performed CT chest demonstrated multiple enlarged calcified hilar and mediastinal lymph nodes with scattered multiple pulmonary nodules. He did not exhibit any pulmonary symptoms. He was given intra-articular steroids for his shoulder pain, which provided significant relief from his pain.

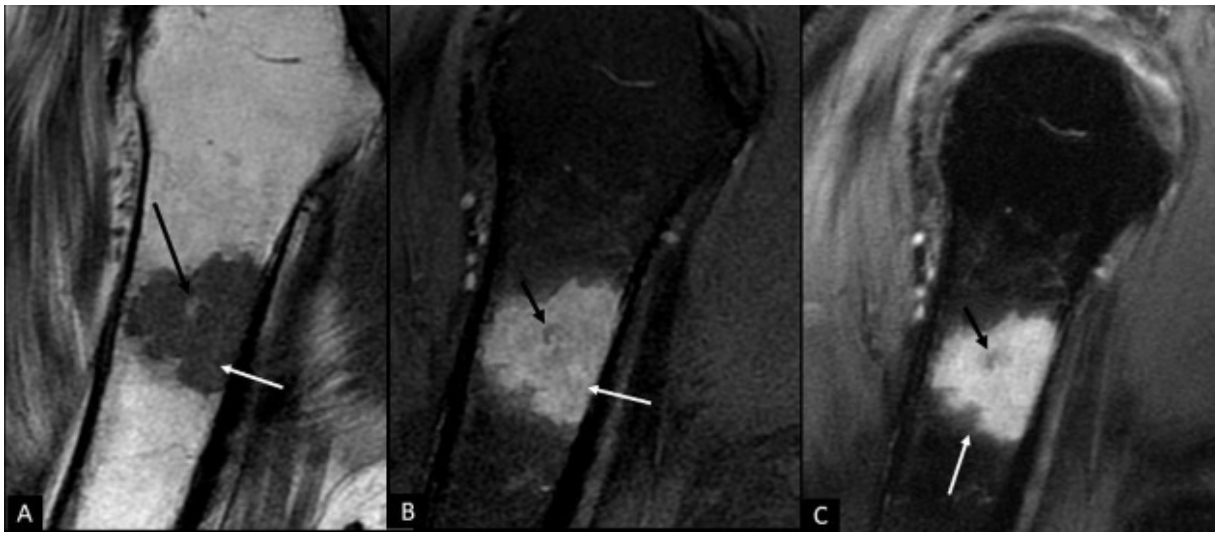

Figure 1. *Sagittal (A) T1 weighted and (B) PD fat saturated MR images of the shoulder. With the proximal humeral diaphysis, there is a well-defined intramedullary lesion, which is hypointense on the T1 weighted image and hyperintense on the PD fat saturated image (white arrow) with intrinsic T1 hyperintense signal and corresponding low signal on fat-saturated image consistent with intralesional fat (black arrow). There is no perilesional edema, cortical destruction, or extraosseous soft tissue component. (C) T1 weighted post contrast fat saturated image demonstrates diffuse enhancement (white arrow).*

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Case 2

A 53-year-old woman presented with severe hip pain with radiation to the knee for 6 months. She had a history of poorly differentiated infiltrating breast cancer treated with lumpectomy followed by adjuvant chemoradiation and was under surveillance. She was also diagnosed with sarcoidosis from cervical lymph-node biopsy and was followed conservatively as she did not have any pulmonary symptoms. On the examination of the hip, there was tenderness to palpation over greater trochanter and a clinical diagnosis of trochanteric bursitis was made. Due to the recurrence of her hip pain, MRI was done, which showed T2 hyperintense lesions with an intrinsic T1 hyperintense signal and a corresponding low signal on short-tau-inversion-recovery (STIR) and fat saturated images (FS) (Figure 2). These imaging findings were in keeping with large bone OS in this patient with sarcoidosis. Of note, she also had a shoulder MRI performed four years ago, which also showed intramedullary lesions but the signal characteristics were different from the lesions in the hip. The lesions were T1 hypointense and hyperintense on PD FS images (Figure 3). However, the intrinsic T1 hyperintense signal was not seen. Due to her history of breast cancer, she was followed closely with repeated MRIs. In the follow-up, there was complete resolution of the lesions on MRI. She was offered intermittent cortisone injections of her shoulder and hip joints, which provided relief to the patient.

The summary of the MRI features of large bone OS lesions in our cases is as follows: multiple intramedullary lesions that are predominately hypointense on T1-weighted imaging and intermediate/hyperintense on T2-weighted imaging with a central intralesional fat signal and variable post-contrast enhancement.

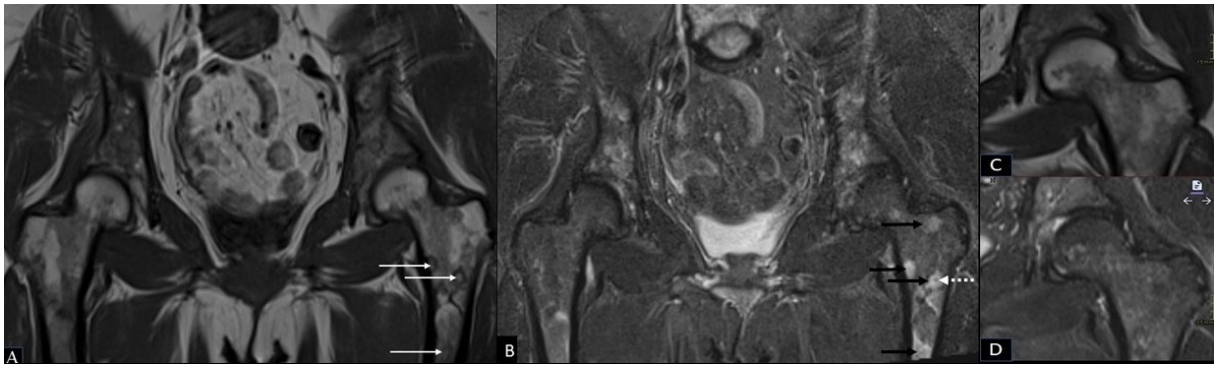

Figure 2. *Coronal T1-weighted (A) and STIR images of the pelvis (B) show extensive multifocal intramedullary lesions in the proximal femurs that demonstrate T2 hypertense signal (black arrows) with intrinsic T1 hyperintense signal (white arrows) and corresponding low signal on STIR (broken white arrow): These findings are in keeping with large bone sarcoidosis in a patient with known history of sarcoidosis. Follow-up coronal T1-weighted (C) and STIR (D) MRI images of the left hip performed 4 years later demonstrates complete resolution of the lesions.*

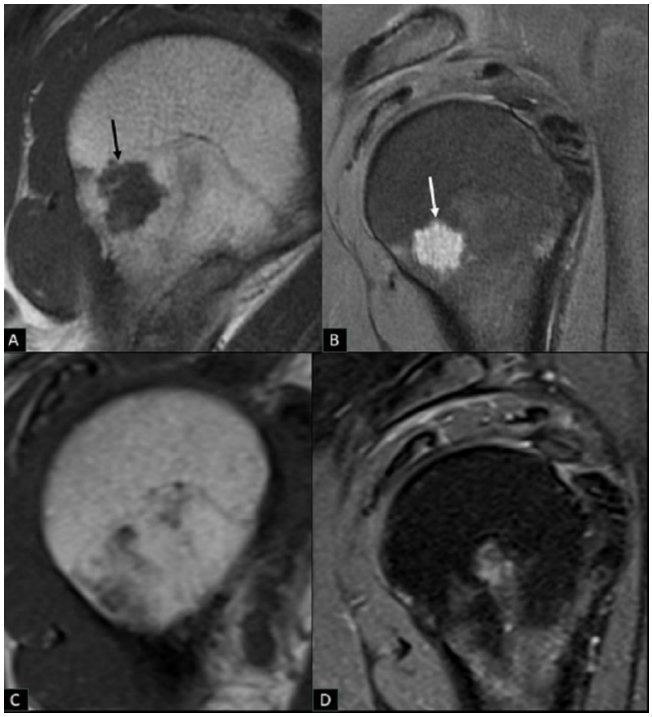

Figure 3. *Sagittal T1-weighted (A) and PD fat saturated images (B) of the shoulder demonstrates well-defined intramedullary lesion in the proximal humeral meta-diaphysis, which is hypointense on the T1 weighted image (black arrow) and hyperintense on the PD fat saturated image (white arrow). There is no perilesional edema or cortical destruction or extraosseous soft tissue component. Follow-up sagittal T1-weighted (C) and STIR (D) MRI images of the left shoulder 8 years later shows complete resolution of the lesion.*

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Discussion

Sarcoidosis is a non-caseating granulomatous disease commonly involving lungs and lymph nodes with osseous involvement in 1-13% of cases [3]. OS could be the initial presentation of undiagnosed sarcoidosis, which may resemble metastasis [7]. The radiographic features of small and large bone OS are variable.

The more common small bone OS has characteristic radiographic features, which are well-described in the literature. Small bone OS may present unilaterally or bilaterally and is usually asymmetric and commonly involves the second or third fingers. The bones demonstrate a characteristic pattern of osteolysis with cystic, lacy and honey-comb like appearance; other features may include cortical destruction, extraosseous extension, soft tissue edema, and sausage finger. Deformities in the hand can occur, which is usually due to a pathological fracture or osseous remodeling [1].

The less common large bone OS is usually multifocal with 50% of the patients being asymptomatic [8]. A study by Mostard et al. demonstrated that one-third of the patients with severe sarcoidosis were found to have multifocal osseous lesions on PET/CT without lesions discernible on low-dose CT [9]. On radiographs, the lesions are often occult or lytic with no cortical destruction or periosteal reaction, similar to our patient. Some lesions may also be mixed or sclerotic [10, 11].

On MRI, large bone OS lesions usually present as multiple, well-defined or indistinct, intramedullary lesions that are hypointense on T1-weighted imaging and intermediate/hyperintense on T2-weighted imaging with variable post contrast enhancement [7]. The lesions may also spontaneously resolve, which was also in our case [3]. A study by Moore et al. [3] concluded that the presence of intralesional or perilesional fat has high specificity but low sensitivity in differentiating OS from metastasis. We postulate that the presence of intralesional fat within the OS lesions is secondary to involution with deposition of fat within the lesion in the setting of chronic inflammation. Islands of red marrow may present similarly to OS on MRI, as red marrow may also contain fat. In both cases

of OS, the OS lesions are mass-like with well-defined convex margins, whereas islands of red marrow have less well-defined margins.

The presence of intralesional fat within an intramedullary lesion would strongly suggest OS in a patient with a known history of sarcoidosis. But there are other differential diagnoses to these osseous lesions. Vascular lesions such as hemangiomas may be difficult to discern from OS based on signal characteristics alone. However, hemangiomas may be distinguished from OS morphologically with its spiculated lattice-like appearance, which may be more obvious with CT. However, if the OS lesion demonstrates a T2 hyperintense signal with a T1 hypointense signal, the OS lesion would be difficult to discern from other bone marrow replacement processes such as metastases, hematogenous spread of osteomyelitis, and brown tumors. In these cases, clinical presentation is important in formulating a differential diagnosis. Histological correlation may also be needed to confirm a diagnosis.

In patients with diagnosed sarcoidosis and intramedullary bone lesions, the presence of intra- or perilesional fat would point towards OS and biopsy could be averted as was the case in our second patient. In patients with no prior diagnosis of sarcoidosis and have all the imaging findings described, sarcoidosis can be provided as a differential in the appropriate clinical context. In conclusion, the knowledge of the imaging characteristics of large bone OS lesions on MRI is important to provide an accurate differential diagnosis, and potentially avert unnecessary biopsy and concern to the patient.

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