A Japanese Case of Vertebral Sarcoidosis

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Abstract

We herein report the first Japanese case of vertebral sarcoidosis diagnosed using multiple imaging modalities and a biopsy. CT, MRI and (18)F-fluorodeoxyglucose positron emission tomography (18F-FDG PET) detected multiple vertebral lesions, and a vertebral biopsy guided by the PET findings confirmed the diagnosis of vertebral sarcoidosis. Although the disease was refractory to corticosteroids, treatment with methotrexate (MTX) achieved a good response. Our case suggests that MRI and 18F-FDG PET are useful for determining the site for a biopsy and that MTX is effective for treating vertebral sarcoidosis.

Key words: multiple imaging modalities, MRI, 18F-FDG PET, biopsy, vertebral sarcoidosis

(Intern Med 52: 2825-2829, 2013)  
(DOI: 10.2169/internalmedicine.52.0074)

Introduction

Sarcoidosis is a systemic inflammatory disease that predominantly affects the lungs and intrathoracic lymph nodes. In Caucasian patients with sarcoidosis, the rate of skeletal involvement ranges widely from 5% to 39%, according to previous reports (1, 2). However, the estimated rate is much lower (1% to 3%) in Japanese sarcoidosis patients (3). The small bones of the hands and feet are most frequently affected; however, involvement of the vertebral bones (vertebral sarcoidosis) is extremely rare.

This report is the first to document a case of vertebral sarcoidosis in a Japanese patient diagnosed using multiple imaging modalities and confirmed with a skeletal biopsy. A literature review of imaging and treatment is also provided.

Case Report

In June 2012, a non-smoking 73-year-old woman was referred to our hospital with increasing back pain and abnormal findings on spinal magnetic resonance imaging (MRI). She had a history of sarcoidosis that had been diagnosed on a skin biopsy in 2004 and early-stage right breast cancer that had been surgically treated in 2010. There was no family history of sarcoidosis or autoimmune diseases. At that time, contrast-enhanced computed tomography (CT) scans performed to search for metastases showed numerous enlarged mediastinal, mesenteric and para-aortic lymph nodes, multiple low-density areas in the liver and spleen, and small parenchymal and subpleural nodules in both lungs. The masses were considered to be lesions of sarcoidosis. During the patient's annual follow-up, the abnormalities on CT were unchanged compared with those observed on the previous CT scans. In July 2011, she began to experience lower back pain that was not associated with any neurologic symptoms. Six months later, she underwent spinal MRI, which revealed abnormal signal intensity (Fig. 1); however, the levels of tumor markers for breast cancer (carbohydrate antigen 15-3 and carcinoembryonic antigen) were unchanged. Although the MRI findings were considered to be highly suspicious for metastatic disease or vertebral sarcoidosis, the patient decided not to undergo further studies and treatment. However, over the next six months, she experienced an unexplained increase in lower back pain that did not respond sufficiently to nonsteroidal anti-inflammatory drugs or opioids.

She was then referred to our hospital, where a physical examination found lower vertebral tenderness. Laboratory tests were all within the normal limits, apart from increased angiotensin-converting enzyme (ACE) (38.7 IU/L; normal range: 7.7-29.4 IU/L) and lysozyme (18.3 μg/dL; normal range: 7.7-29.4 IU/L) and lysozyme (18.3 μg/dL; normal range: 7.7-29.4 IU/L) and lysozyme (18.3 μg/dL; normal range: 7.7-29.4 IU/L).
No active abnormalities were found on an ophthalmic examination. Liver and renal function tests and the levels of C-reactive protein, muscle enzymes, serum calcium and urine calcium were within the normal limits. No active abnormalities were found on an ophthalmic examination, electrocardiography or echocardiography. A (18) F-fluorodeoxyglucose positron emission tomography (18F-FDG PET) scan demonstrated multiple areas of intense uptake of the radiotracer in the liver, lungs, lymph nodes and vertebral bodies (Fig. 2). On plain radiographs of the hands, no lesions were detected. Spinal MRI showed progressive multiple lesions in the lower vertebral bodies and iliac bones with multiple compression fractures (Fig. 1). These lesions exhibited low signal intensity on T1-weighted images (T1 WI) and high signal intensity on T2-weighted images (T2 WI). Since the patient did not have a recent history of trauma, the multiple compression fractures were thought to be caused by the progressive multiple lesions in the vertebral bodies. CT images showed that the enlarged lymph nodes and lung lesions were unchanged, while the lesions in the liver and spleen had improved compared with that observed on the previous CT scans. However, the osteosclerotic lesions in the lower vertebral bodies were progressive (Fig. 3), which was suggestive of metastatic breast cancer or vertebral sarcoidosis. A needle biopsy of the L2 vertebral body demonstrated granulomatous lesions of epithelioid and giant cells without necrosis, pointing to a diagnosis of sarcoidosis (Fig. 4).

The patient received 30 mg/day of prednisolone for two weeks and experienced immediate pain relief in the lumbar spine. However, the lumber pain relapsed when the predni-
Osseous involvement usually causes solitary sarcoidosis is low and appears to be even lower in Japanese patients. The incidence of skeletal sarcoidosis can be charted using multiple imaging modalities and confirmed with a skeletal biopsy.

To the best of our knowledge, this is the first report to describe a case of vertebral sarcoidosis in a Japanese patient that was diagnosed using multiple imaging modalities and confirmed with a skeletal biopsy. In general, sarcoidosis predominantly affects the lungs, skin, lymph nodes, eyes and liver. The incidence of skeletal sarcoidosis is low and appears to be even lower in Japan. Osseous involvement usually causes solitary cystic osteitis of the phalangeal bones of the hands and feet. Vertebral sarcoidosis is extremely rare, and most cases involve known sarcoidosis. The thoracic spine is the most frequently affected site in patients with vertebral sarcoidosis, although any area of the spine can be involved. Patients with vertebral sarcoidosis nearly always present with radicular back pain both during activity and at rest. Our patient also experienced lumbar pain with immediate pain relief after treatment. Furthermore, the lumbar pain appeared to be due to the activity of vertebral sarcoidosis. Our case is rare because multiple vertebral lesions were affected, which were progressive despite the stability of the various other sarcoidosis lesions in the lungs, lymph nodes, liver and spleen.

The results of various laboratory tests and MRI examinations in our case were in accordance with the findings of previous reports. Laboratory tests frequently show elevated ACE levels in sarcoidosis patients with skeletal involvement. Imaging modalities, such as CT and MRI, are reported to be equally good at detecting sarcoidosis lesions in patients with vertebral involvement. In our case, CT was able to capture images of sclerotic lesions, which are generally rare. Lesions in the spine and long bones are frequently lytic with or without a rim of sclerosis, and detecting sclerotic lesions is rare. On MRI, vertebral sarcoidosis usually presents as hypointense lesions on T1WI and hyperintense lesions on T2WI, as observed in our case. However, the MRI appearance of vertebral sarcoidosis can be similar to that of multiple myeloma or osteolytic bone metastases. Therefore, performing a biopsy of the active lesions is important in order to confirm the diagnosis of vertebral sarcoidosis, as previously reported. In patients with vertebral sarcoidosis, MRI abnormalities resolve following successful therapy, which reflects the resolution of granulomas and replacement with fibrous elements. Therefore, MRI findings may be used to chart the progression of vertebral sarcoidosis.

Furthermore, vertebral sarcoidosis lesions were also detected in our patient on MRI and 18F-FDG PET, which...
were useful for determining the diagnostic biopsy site and assessing the active lesions. For many years, nuclear imaging using 67gallium (67Ga) has played an important role in the assessment of sarcoidosis (22). In recent years, 18F-FDG PET has been proposed to be a useful modality for the diagnosis and management of patients with inflammatory processes, particularly sarcoidosis (23, 24). According to a previous review, whole-body 18F-FDG PET has been reported to be more sensitive than 67Ga scans for assessing the activity of sarcoidosis and demonstrates great value in detecting occult diagnostic biopsy sites (25). Our findings suggest that 18F-FDG PET is a potential modality for assessing vertebral sarcoidosis.

Corticosteroids, such as prednisolone, are often used as first-line treatment for skeletal sarcoidosis and can achieve a good response (10, 13, 17, 26). The prednisolone dose is usually 15-40 mg/day, which is tapered according to the clinical response. In patients who are refractory to corticosteroids, treatment with MTX, infliximab and adalimumab has been reported to be potent (10, 12, 21, 27). In our case, MTX was found to be effective. Although fracturing of the vertebral lesions is a direct adverse effect of vertebral sarcoidosis, no treatment options to decrease this risk are currently available. If neurologic involvement occurs secondary to vertebral destruction, surgical stabilization and decompression may be required (28).

In conclusion, we herein reported the first case of vertebral sarcoidosis in a Japanese patient diagnosed using multiple imaging modalities and a biopsy. This case report demonstrates that both MRI and 18F-FDG PET are potential tools for detecting vertebral sarcoidosis lesions and monitoring the therapeutic response. Furthermore, performing a biopsy is important for differentiating between vertebral sarcoidosis and malignant lesions.

The authors state that they have no Conflict of Interest (COI).

References