Internal Medicine
The Japanese Society of Internal Medicine
doi: 10.2169/internalmedicine.9299-17
Intern Med 57: 717-720, 2018
http://internmed.jp

【 CASE REPORT 】

Emergency Radiotherapy for Spinal Cord Compression due to Bone Sarcoidosis

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Abstract:
Sarcoidosis is an inflammatory granulomatous disease that is systemic, but bone involvement is uncommon. A 68-year-old man was referred to our hospital complaining of right shoulder pain with numbness. Computed tomography revealed systemic lymphadenopathy and multiple bone lesions. Because malignant lymphoma with a mass lesion protruding into the vertebral canal was considered, he underwent urgent radiotherapy. Thereafter, a needle biopsy of the left parasternal node was performed and showed epithelioid granulomas, confirming a diagnosis of sarcoidosis. Since his neurologic symptoms improved, the patient was not given systemic corticosteroids. Radiotherapy may be useful for local control of bone sarcoidosis.

Key words: bone disease, CT-guided biopsy, radiotherapy, sarcoidosis

(Intern Med 57: 717-720, 2018)
(DOI: 10.2169/internalmedicine.9299-17)

Introduction

Sarcoidosis is a granulomatous disease that can affect multiple organ systems. The lungs, eyes, and skin are commonly affected, but bone involvement is uncommon. In this report, we describe a case of sarcoidosis with vertebral lesions presenting as right shoulder pain and numbness. Due to the presence of a mass lesion on the seventh cervical vertebra (C7) that compressed the spinal cord, the patient was treated with urgent radiotherapy, which mitigated the neurologic symptoms.

Case Report

In November 2015, a 68-year-old man was referred to our hospital for the assessment of systemic lymphadenopathy and bone involvement. Since April 2015, he had been experiencing right shoulder pain that had been attributed to osteoarthritis. In August 2015, he had ocular pain and was diagnosed with uveitis. Aside from uveitis, there were no findings meeting the diagnostic criteria for ocular sarcoidosis. His shoulder pain persisted despite treatment with acetaminophen.

On a physical examination, the patient had bilateral hyperreflexia of the biceps, triceps, brachioradialis, patellar, and Achilles tendons. The grip of his right hand was weak, and he had a sensory deficit on the fifth digit of the right hand. Laboratory tests showed mild liver dysfunction and elevated levels of soluble interleukin 2 receptor (sIL-2R) and sialylated carbohydrate antigen (KL-6), as shown in Table. There were no significant increases in the serum levels of calcium, alkaline phosphatase, or angiotensin-converting enzyme (ACE). Chest roentgenogram revealed no abnormal findings. Computed tomography (CT) showed lymphadenopathy of the parasternal, left axillary, lower paratracheal, subcarinal, right hilar, and abdominal paraaortic nodes and a bone lesion on C7, which was confirmed by magnetic resonance imaging to have extended into the vertebral canal and compressed the spinal cord. The vertebral lesion was isointense on T1-weighted imaging and hyperintense on T2-weighed imaging (Fig. 1). Fluorodeoxyglucose-positron emission tomography (FDG-PET) showed an abnormal uptake in the vertebral lesion, with a maximum standardized

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Received: April 3, 2017; Accepted: July 4, 2017; Advance Publication by J-STAGE: November 20, 2017
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Table. **Laboratory Test Results.**

Blood counts		Biochemistry		Ca	9.6 mg/dL
WBC	6,590 /mm^3	TP	7.1 g/dL	ALP	256 U/L
Neutro	77.0 %	Alb	4.6 g/dL		
Lymph	17.0 %	BUN	17 mg/dL	Serology	
Mono	3.0 %	Cre	0.86 mg/dL	CRP	0.110 mg/dL
Eosino	1.0 %	Na	141 mEq/L	sIL-2R	1,980 U/mL
RBC	4.88×10^6 /mm^3	K	4.1 mEq/L	KL-6	526 U/mL
Hb	15.0 g/dL	Cl	99 mEq/L	ACE	20.7 IU/L
Ht	45.1 %	AST	40 U/L		
MCH	30.7 pg	ALT	51 U/L		
MCV	92.4 fL	LDH	186 U/L		
Plt	196×10^3 /mm^3	Glu	86 mg/dL		

WBC: white blood cell, RBC: red blood cell, Hb: hemoglobin, Ht: hematocrit, MCH: mean corpuscular hemoglobin, MCV: mean corpuscular volume, Plt: platelet, TP: total protein, Alb: albumin, BUN: blood urea nitrogen, Cre: creatinine, AST: aspartate aminotransferase, ALT: alanine aminotransferase, LDH: lactate dehydrogenase, Glu: glucose, ALP: alkaline phosphatase, CRP: C-reactive protein, sIL-2R: soluble interleukin-2 receptor, KL-6: Krebs von den Lungen-6, ACE: angiotensin converting enzyme

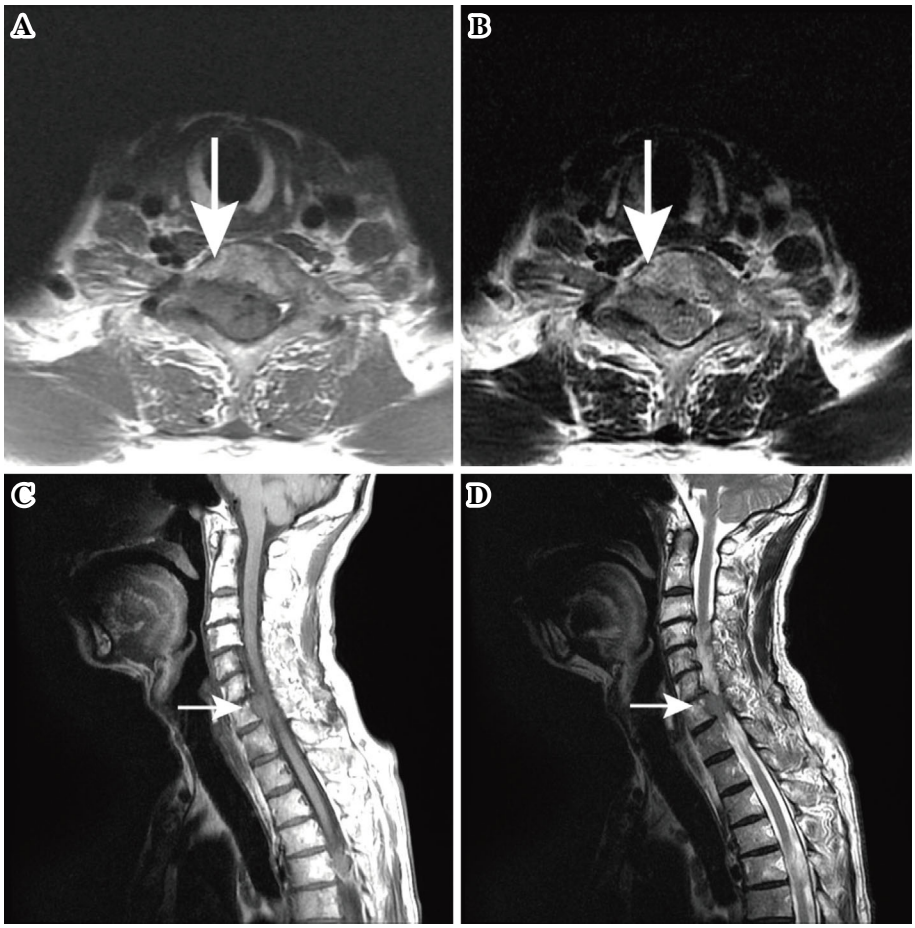

Figure 1. **Magnetic resonance imaging reveals a mass on the seventh cervical vertebra extending into the vertebral canal. It has an isointense signal on T1-weighted imaging (A) and a hyperintense signal on T2-weighted imaging (B). The sagittal section also shows a mass on T1-weighted imaging (C) and T2-weighted imaging (D).**

uptake value of 11.8 (Fig. 2), as well as in the other lymph nodes. Although we did not examine the cerebrospinal fluid, it would have been informative and helpful in ruling out neurosarcoidosis.

Based on the presence of systemic lymphadenopathy without a pulmonary lesion and the markedly elevated level of serum sIL-2R, we suspected malignant lymphoma with a vertebral lesion compressing the spinal cord and started ur-

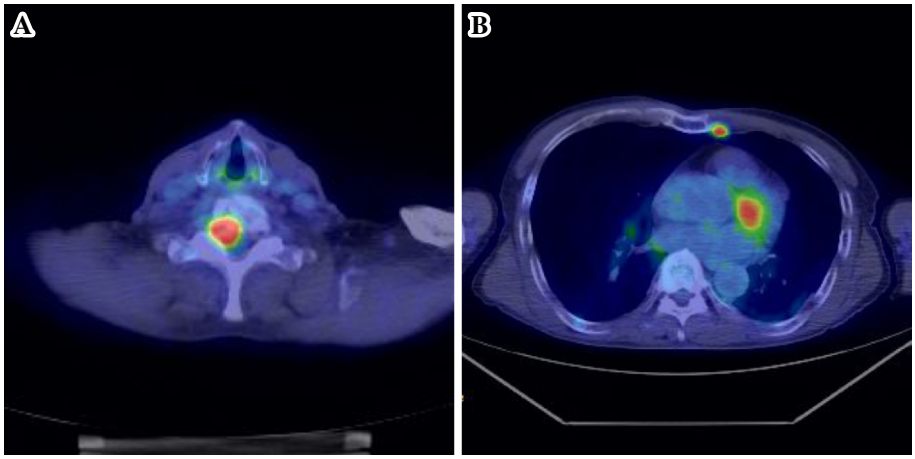

Figure 2. **Positron emission tomography shows the intense accumulation of ^{18}F-fluorodeoxyglucose in the vertebral lesion (A) and in the left parasternal lymph node (B).**

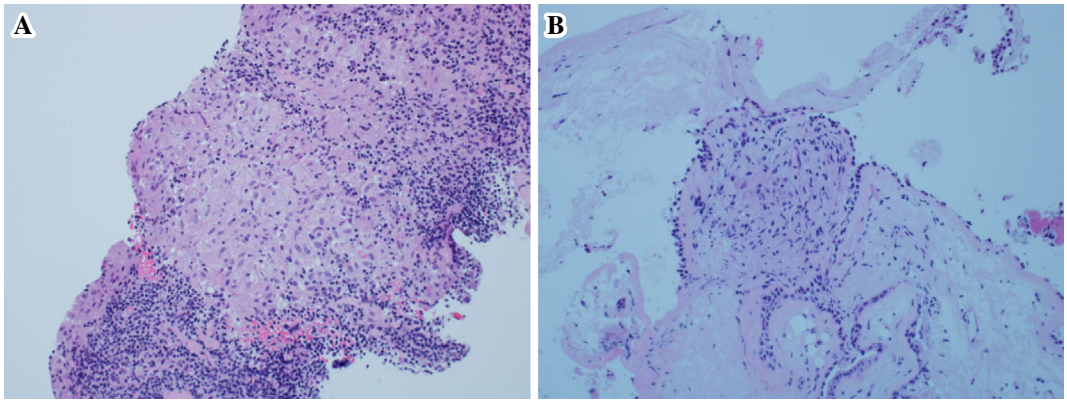

Figure 3. **A photomicrograph of a lymph node biopsy specimen shows non-caseous epithelioid granuloma (A). A transbronchial lung biopsy reveals a similar epithelioid granuloma (B).**

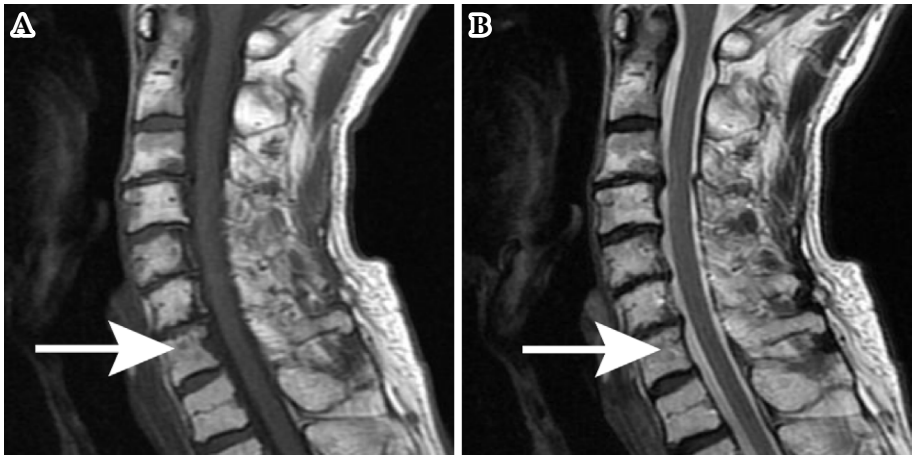

Figure 4. **Magnetic resonance imaging after radiotherapy shows a reduction in the size of the seventh vertebral lesion. The lesion disappeared almost completely on T1-weighted imaging (A) and T2-weighted imaging (B).**

gent radiotherapy. FDG-PET showed an abnormal uptake in the parasternal node, a safe location for a biopsy, with a maximum standardized uptake value of 7.0. Therefore, a CT-guided biopsy was performed on the parasternal node and revealed epithelioid granulomas (Fig. 3). We then conducted a transbronchial lung biopsy and demonstrated the presence of immature epithelioid granulomas. We also obtained a sample of the bronchoalveolar lavage fluid, which

showed lymphocytic predominance cells (47%). The CD4-CD8 ratio was elevated (2.07), although not to a significant degree. A diagnosis of sarcoidosis was made based on these findings.

After receiving 20 Gy of radiotherapy, the pain and numbness of the right shoulder resolved, and the mass lesion on C7 decreased in size (Fig. 4). Since there was no heart involvement or progressive lung disease, the patient was not treated with systemic corticosteroids, but was only followed up instead. The administration of ophthalmic corticosteroids for uveitis was continued.

Discussion

This case highlighted two important clinical issues. First, sarcoidosis can be associated with bone involvement with mass effects. Second, radiotherapy may be useful for the local control of granulomatous mass lesions that compress surrounding organs.

Bone involvement of sarcoidosis, which is characterized by granuloma formation, was reported to occur in 1% to 13% of patients (1) and is often associated with bone destruction and resorption that can result in pathologic fractures. To date, there has been only one report of bone sarcoidosis that formed a mass lesion compressing the surrounding organs (2). Bone involvement of sarcoidosis commonly appears in the proximal and middle phalanges and rarely affects the vertebrae (1, 3). In this case, there were no bone lesions of the extremities nor skin lesions, such as lupus pernio. Because this patient presented with neurologic symptoms, radiotherapy was urgently started; this successfully decreased the size of the C7 lesion and resolved the symptoms. There have so far been few reports of radiotherapy to treat neurosarcoidosis (4, 5). To our knowledge, this was the first report of bone involvement in a case of sarcoidosis that was successfully controlled by radiotherapy. There have been some reports of emergency radiotherapy for other granulomatous diseases (6). It may be acceptable to treat benign lesions with radiotherapy to reduce compression urgently.

In the present case, a biopsy of the parasternal lymph node enabled us to distinguish between sarcoidosis and malignant metastasis. Because of the absence of significant lung lesions on chest CT, the elevated level of sIL-2R, and the normal serum ACE level, we initially considered malignant lymphoma and initiated urgent radiotherapy. In the absence of significant chest CT findings, sarcoidosis is difficult to diagnose correctly; in such cases, FDG-PET might be useful for identifying more discrete abnormalities that can be sampled for a pathologic confirmation of the presence of non-caseating epithelioid granulomas (7). Because a bone biopsy was difficult to perform, the patient underwent a biopsy of the parasternal node and the lung instead to confirm the diagnosis of sarcoidosis.

In this case, a diagnosis of sarcoidosis was made after the urgent radiotherapy had finished. If we had been able to make the diagnosis earlier, then radiotherapy would have been discontinued and switched to systemic corticosteroid therapy.

In conclusion, sarcoidosis can be associated with bone involvement that may compress the surrounding organs. Sarcoidosis should be considered in the differential diagnosis of systemic lymphadenopathy with bone lesions. The present case suggested that radiotherapy might be useful for the local control of bone sarcoidosis which compresses the surrounding organs.

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 upon request.

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

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