

Atypical CT findings of pulmonary sarcoidosis A case report

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Abstract

Rationale: Pulmonary involvement occurs in about 90% of patients with sarcoidosis. However, delayed diagnosis sometimes occurs due to atypical thoracic imaging findings.

Patient concerns: A 52-year-old woman presented with recurrent uveitis and fever of unknown origin. She had been admitted to the hospital due to fever, but its cause was not determined.

Diagnoses: Chest computed tomography (CT) revealed a solitary pulmonary nodule and an enlarged right axillary lymph node. The nodule had showed an interval growth from 0.7 cm to 1.1 cm over 18 months, when compared to the previous chest CT. Mosaic attenuation was also observed.

Interventions: The patient underwent thoracoscopic wedge resection of the nodule and excisional biopsy of the enlarged lymph node to exclude malignancy and non-caseating granulomas consistent with sarcoidosis was confirmed.

Outcomes: Medical treatment with prednisolone and azathioprine was administered. Fever and uveitis no longer recurred after 6 months of medication. We demonstrated that sarcoidosis shows only atypical pulmonary imaging findings, with an enlarged solitary nodule, an axillary lymphadenopathy, and mosaic attenuation, mimicking a malignancy.

Lessons: Awareness on atypical CT manifestations that are correlated with pathologic findings may be helpful for early diagnosis of sarcoidosis.

Abbreviations: ACE = angiotensin-converting enzyme, CRP = C-reactive protein, ESR = erythrocyte sedimentation rate, FUO = fever unknown origin, GGO = ground glass opacities.

Keywords: computed tomography, fever, sarcoidosis

1. Introduction

Sarcoidosis is a multisystemic disease of unknown etiology and characterized by non-caseous epithelioid granulomas.^[1] About 90% of patients with sarcoidosis show thoracic radiologic abnormalities. Pulmonary sarcoidosis can present with various radiologic findings. The most common findings are bilateral hilar lymph node enlargement and micronodules with a perilymphatic distribution.^[2,3] However, atypical radiologic manifestations of pulmonary sarcoidosis can occur in approximately 25% to 30% of patients with thoracic abnormality.^[4] They include several nonspecific imaging findings, such as isolated lymphadenopathy,

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Received: 20 March 2018 / Accepted: 18 June 2018 http://dx.doi.org/10.1097/MD.000000000011456 solitary pulmonary nodule, conglomerated masses, and ground glass opacities (GGO), which may cause delayed diagnosis.^[3]

Here, we present an interesting case of pulmonary sarcoidosis that shows only atypical imaging features mimicking a malignancy on the chest CT in a patient who manifested with recurrent fever and uveitis.

2. Case presentation

A 52-year-old woman was referred to our rheumatology department for further examination of the cause of repeated anterior uveitis, which occurred 3 times in the last year and occurred in both eyes this time. Although no specific comorbidity was found, she had complained of recurrent fever and fatigue in the last 18 months. Regarding fever, she had been admitted to the hospital, but the cause was not identified. Since then, she has been taking acetaminophen whenever she has fever. Laboratory examinations showed increased levels of serum angiotensin-converting enzyme (ACE) (71.6 IU/L), erythrocyte sedimentation rate (ESR) (85 mm/h), and C-reactive protein (CRP) (2.12 mg/ dL). Tests for anti-nuclear antibody, anti-neutrophil cytoplasmic antibody, and rheumatic factor were negative or normal.

We suspected sarcoidosis as the cause of uveitis with fever, and chest CT was performed to confirm the diagnosis. Chest CT revealed a 1.1-cm solitary pulmonary nodule in the left upper lobe (Fig. 1A) and an enlarged right axillary lymph node (Fig. 1B). The nodule had showed slow interval growth from 0.7 to 1.1 cm, when compared with the previous CT result taken for fever of unknown origin (FUO) work-up 18 months ago. Mosaic attenuation with air trapping was also shown on both CT scans

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Figure 1. Atypical manifestations of pulmonary sarcoidosis confirmed as a cause of fever of unknown origin. A, Chest CT shows a 1.1-cm well-defined nodule (arrow) in the left upper lobe. B, An enlarged axillary LN is shown (arrows). C, There is mosaic attenuation with air-trapping (arrows). D, High-power photomicrograph (original magnification, × 100; H&E stain) of a lung specimen obtained at wedge resection shows several small non-caseous granulomas adjacent to the bronchiolar epithelium. E, Pathologic photomicrograph (original magnification, × 200; H&E stain) of an axillary lymph node reveals non-caseous granulomas, consistent with sarcoidosis. F, In comparison with the previous CT (C), areas of mosaic attenuation resolved in the follow-up chest CT 6 months after the administration of medication, and symptoms of fever and uveitis no longer recurred.

(Fig. 1C). Thoracoscopic wedge resection for a nodule (Fig. 1D) and excisional biopsy for a lymph node (Fig. 1E) were performed to exclude malignancy and confirmed non-caseating granulomas. The patient was diagnosed with sarcoidosis of multi-organ involvement based on the histopathologic detection of non-caseating granulomas in the lung and lymph node and the presence of systemic symptoms such as fever and uveitis. Fever subsided 5 days after the administration of prednisolone (30 mg), and levels of ESR and CRP were normalized. After tapering the steroid dosage, CRP level was increased again; therefore, azathioprine was added. After 6 months of medication, mosaic

attenuation disappeared in the follow-up chest CT (Fig. 1F), and symptoms of fever and uveitis no longer recurred.

3. Discussion

The diagnosis of sarcoidosis is established when clinical and imaging features are supported by histologic evidence.^[5] When a patient complains of recurrent uveitis as in our case, sarcoidosis should be considered as one of its underlying causes. Uveitis has been known to be the initial manifestation in 20% of patients with sarcoidosis.^[6] Our patient also showed elevated level of serum ACE. Chest CT was performed to identify the affected site that is amenable to biopsy, because diagnosis of sarcoidosis requires histopathologic evidence of non-caseating granulomas. Chest CT only demonstrated atypical imaging findings of sarcoidosis; therefore, it was not suspected based on the chest CT findings, but fortunately biopsy was performed to exclude malignancy and finally sarcoidosis was diagnosed. As such an unusual imaging finding may result in unlikely suspicion of sarcoidosis or delayed biopsy, eventually making diagnosis difficult.

Reported atypical features of pulmonary sarcoidosis include isolated lymphadenopathy, airspace consolidation, solitary pulmonary nodule, GGO, and mosaic attenuation.^[3] Among them, three atypical features, that is, isolated axillary lymphadenopathy, solitary pulmonary nodule, and mosaic attenuation, were interestingly observed at once and no other typical findings were seen in our case.

It is very rare but known to be possible that pulmonary sarcoidosis involves hilar lymph node as well as other unusual location of thoracic lymph nodes, such as the internal mammary, axilla, paravertebral, and retrocrural areas.^[3] Sometimes, it is misdiagnosed as lymphoma when lymphadenopathy is found in an unusual location.^[7]

Sarcoidosis can manifest as a solitary pulmonary nodule which is coalescence of individual granulomas.^[3] Michael et al^[8] reported a sarcoidosis that presented as a solitary pulmonary nodule showing similar interval growth as in our case. Although a malignant nodule cannot be excluded for an enlarging nodule, sarcoidosis should also be considered as a differential diagnosis, when clinically it was suspected.

Mosaic attenuation can occur in patients with pulmonary sarcoidosis from small airway involvement by granulomas which cause air-way luminal obstruction.^[3,9] When reviewing the imaging findings on serial chest CT scans, we found that mosaic attenuation appeared when fever occurred and it resolved when fever improved.

This case reminds clinicians that sarcoidosis should be considered as an uncommon cause of FUO, and awareness of its atypical CT manifestations may enable early diagnosis through histological examinations.

Author contributions

Conceptualization: Ran Song, Jung Im Kim.

Data curation: Ran Song.

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