

[CASE REPORT]

Pulmonary Multiloculated 'Lotus Torus-like' Sarcoidosis Mimicking Lung Adenocarcinoma

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Abstract:

A 34-year-old Japanese woman exhibited a 35×25-mm solitary multiloculated mass shadow in the left lower lobe mimicking lung adenocarcinoma. On computed tomography, the mass resembled a lotus torus. A transbronchial lung biopsy and mediastinal lymph node biopsy led to the diagnosis of sarcoidosis. This lotus torus-like mass regressed spontaneously. This is the second reported case of pulmonary cavitory sarcoidosis with a 'lotus torus-like' appearance. We propose several findings regarding the lotus torus-like appearance by comparing the findings to those of lung adenocarcinoma. Knowledge of this unique sign may be helpful for the differential diagnosis of pulmonary sarcoidosis from lung adenocarcinoma.

Key words: sarcoidosis, lotus, cavitory, nodular, multiloculated, adenocarcinoma

(Intern Med 58: 419-422, 2019)

(DOI: 10.2169/internalmedicine.1541-18)

Introduction

The lung is one of the most common sites of sarcoidosis involvement, and a wide spectrum of radiographic findings has been reported in patients with pulmonary sarcoidosis (1). Nodular sarcoidosis is an uncommon finding in patients with pulmonary sarcoidosis. Six of 150 patients (4.0%) with sarcoidosis were identified as having nodular sarcoidosis (2). Pulmonary cavitory lesions are also reported to be uncommon: the incidence was 2.2% (23 of 1,060 patients) in a large series of patients with pulmonary sarcoidosis (1). A case of pulmonary cavitory sarcoidosis with 'lotus seed-like' manifestations was reported (3); the cavitory lesions were multiloculated, and their appearance on computed tomography (CT) resembled that of a lotus torus. To our knowledge, there had been no prior detailed description of this manifestation.

We herein present the second case of pulmonary 'lotus torus-like' sarcoidosis, which was initially suspected to be lung adenocarcinoma. We describe the characteristics of the

lotus torus-like appearance in this unique sarcoidosis case to aid in the differential diagnosis from lung adenocarcinoma in future cases.

Case Report

An abnormal shadow was identified on a chest radiograph of a 34-year-old Japanese woman without any symptoms. She had no remarkable history. She was an office worker and a current smoker (5 cigarettes/day, 14 years). The chest radiograph showed a mass shadow in the left middle lung field (Fig. 1A), and chest CT showed a 35×25-mm mass with spiculation and pleural indentation in the left lower lobe (Fig. 1B). The mass was accompanied by multiple cystic changes and air bronchogram. The cysts were tiny, numerous and uniform in appearance. The wall of the mass was uniformly thick. These CT findings resembled a lotus torus (Fig. 1C).

No obvious small satellite nodules or granular shadows were observed around the mass. The mass was solitary, and no abnormal shadows were observed elsewhere in the lung.

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Received: May 15, 2018; Accepted: July 4, 2018; Advance Publication by J-STAGE: September 12, 2018

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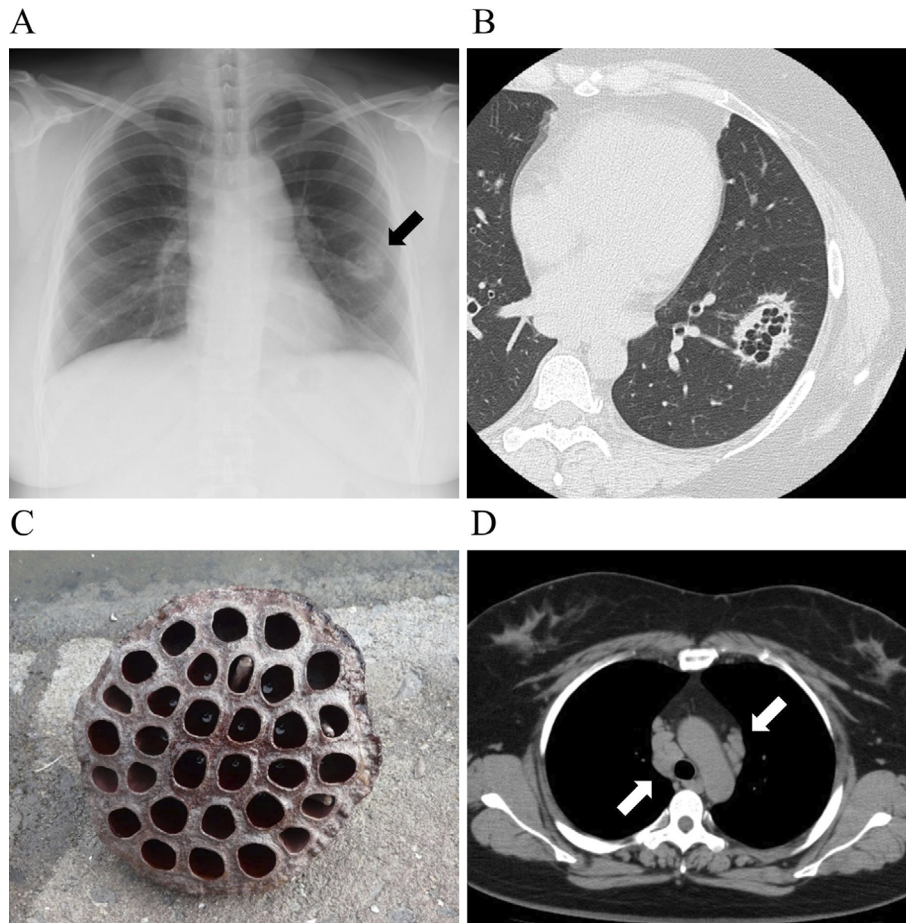


Figure 1. (A) Chest X-ray at the time of the diagnosis showing a mass shadow (arrow). (B) Chest CT showing the shadow of a 35×25-mm mass with a lotus torus-like appearance. (C) A lotus torus. (D) Swelling of the mediastinal lymph nodes (arrows).

Bilateral mediastinal lymphadenopathy was evident (Fig. 1D). No calcification was observed in the mass or the mediastinal lymph nodes. The general blood and urine data were normal, and the level of angiotensin-converting enzyme was within the normal range; 15.3 IU/L (7.7-29.4). In light of the possibility of lung adenocarcinoma, we performed a bronchoscopic examination. The biopsy specimens showed few multinucleated giant cells and no malignant cells (Fig. 2A). Mediastinal lymph nodes obtained by mediastinoscopy revealed non-caseous epithelioid cell granulomas without malignancy (Fig. 2B).

No acid-fast bacilli or fungi were detected. Based on these findings, we diagnosed the patient with sarcoidosis. Fluorine-18-fluorodeoxyglucose positron emission tomography revealed no involvement of organs other than the lung and lymph nodes (Fig. 2C). No treatment was administered as the patient had not experienced any symptoms. At six months after the diagnosis, CT images of the mass showed a decrease in the number of cysts and a thinner wall (Fig. 3A). At 1 year after the diagnosis, the mass had regressed, the cystic changes had disappeared, and the mediastinal lymph nodes had clearly decreased in size (Fig. 3B and C).

Discussion

The final diagnosis in this case was pulmonary sarcoidosis. However, the CT findings mimicked lung adenocarcinoma, and we initially considered the possibility of adenocarcinoma rather than sarcoidosis, because there was only a single mass with spiculation and with cystic changes inside. We next present some cases of lung adenocarcinoma that were encountered at our institute, which showed similar findings to the present case of sarcoidosis (Fig. 4). Adenocarcinoma cases with these findings are occasionally encountered, and a similar adenocarcinoma case has been reported (4). In comparison to these adenocarcinoma cases, the present case and the other case involving lotus torus-like sarcoidosis (3) have some distinct findings. We propose the characteristics of the lotus torus-like appearance: (I) an oval mass with spiculation; (II) multiloculation, the cysts are tiny, numerous and uniform in appearance, and (III) the wall of the mass is uniformly thick. The mass may be surrounded by tiny satellite nodules (3), although no obvious nodules were observed in the present case.

In contrast, the CT findings of adenocarcinoma are as follows: (i) an irregularly shaped mass; (ii) fewer irregular

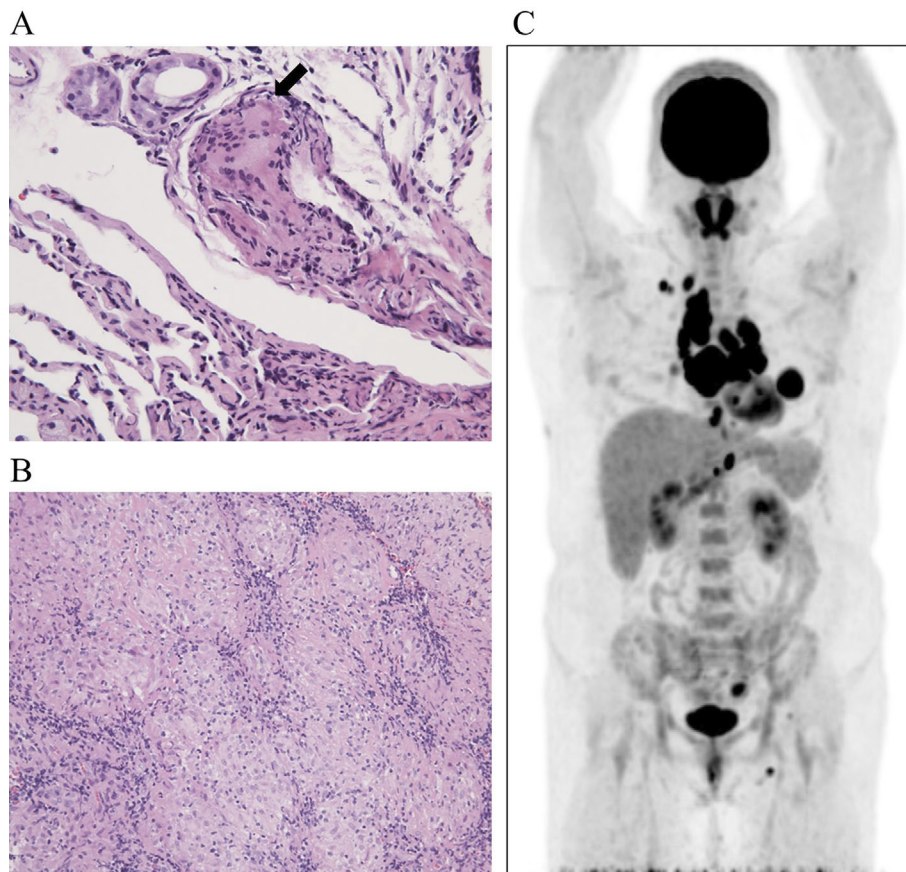


Figure 2. The histopathological findings of (A) the lotus torus-like mass obtained by bronchoscopic biopsy; a few multinucleated giant cells (arrow), and (B) a mediastinal lymph node obtained by mediastinoscopy; non-caseous epithelioid cell granulomas. (C) Positron emission tomography showing the accumulation of fluorine-18-fluorodeoxyglucose in the lung mass, and the bilateral hilar, mediastinal, right supraclavicular, and para-aortic lymph nodes.

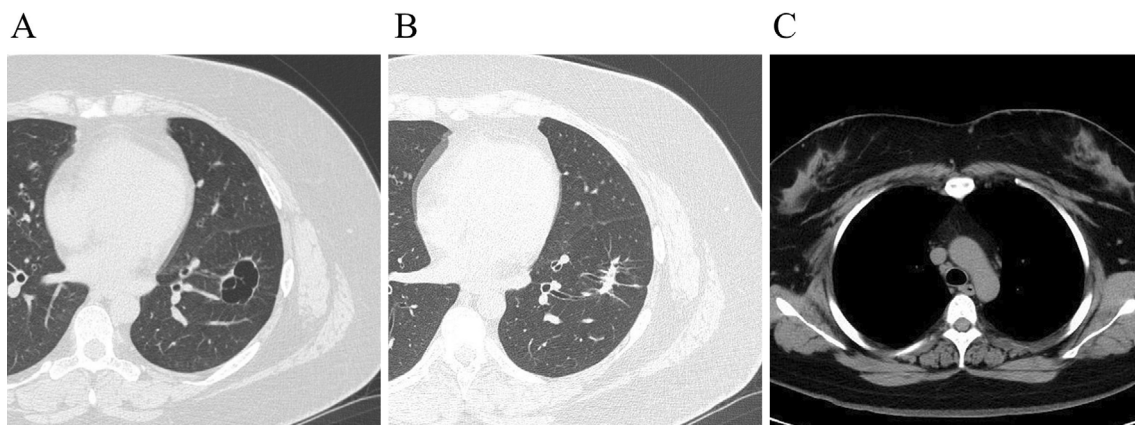


Figure 3. The natural course of the mass with a lotus torus-like appearance. (A) Six months and (B, C) twelve months after the diagnosis.

cysts; and (iii) laterality of the wall thickness (i.e., the coexistence of thick and thin parts of the wall) (Fig. 4). The knowledge of this unique lotus torus-like appearance may contribute to the differential diagnosis of sarcoidosis from adenocarcinoma. However, it should be noted that most cases with cavitory sarcoidosis do not present this lotus

torus-like appearance (1). The diagnostic accuracy of sarcoidosis will increase when these findings are used together with other CT findings of pulmonary sarcoidosis, such as the galaxy sign (5).

Some mechanisms of cavitation in pulmonary cavity sarcoidosis that have been proposed include alveolar disten-

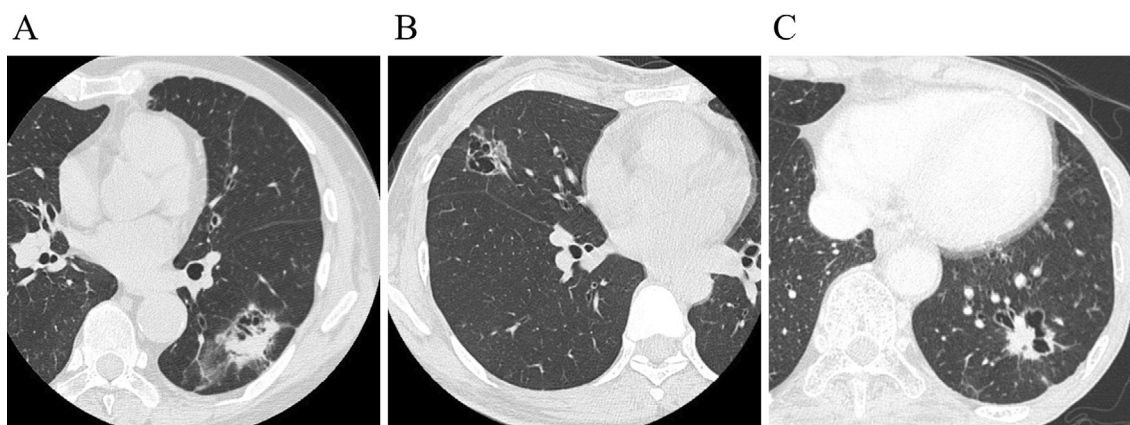


Figure 4. (A-C) Multiloculated mass shadows in three patients with lung adenocarcinoma. Each image is from a different patient.

sion with rupture by a check-valve obstruction due to endobronchial sarcoid granulomas or fibrosis, the accumulation of secretions into the airways (3), excretion of ischemic necrosis, complication with infections such as aspergillus, and cystic bronchiectasis (1, 6). We hypothesize that air trapping due to the above-mentioned check-valve mechanism occurred in the present case because the patient was asymptomatic and the cysts were uniform in appearance and disappeared during follow-up.

The natural course of these lesions is also interesting. In the present case, the multiple cysts decreased in number and became larger at 6 months after the diagnosis, which might have been the result of cysts fusing with each other due to the check-valve mechanism. At 12 months after the diagnosis, the mass had clearly regressed. The mass of the previous case also regressed within 14 months (3). The lotus torus-like appearance may be associated with spontaneous regression. Because the lotus torus-like appearance would only be observed for a short period, the actual frequency of this sign in pulmonary sarcoidosis may be higher than expected. In addition, some frequently observed CT findings (e.g., post-inflammatory nodules or post-infectious bleb) may include post-lotus torus-like sarcoidosis lesions.

In conclusion, knowledge of the features of the lotus torus-like appearance in pulmonary sarcoidosis may be useful for differentiating sarcoidosis from lung adenocarcinoma.

This unique sign may regress spontaneously.

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

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