A pulmonary chondromatous hamartoma resembling multiple metastases in the ^{18}F-FDG PET/CT scan

Li Li[1*] **MD,**
Chong Jiang[1*] **MD,**
Rong Tian[1] **MD**

**Li Li and Chong Jiang are co-first authors due to their equal contribution to this work.*

1. Department of Nuclear Medicine, West China Hospital, Sichuan University, 37 Guoxue Lane, Wuhou District, Chengdu 610000, P.R. China

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Corresponding author:
Rong Tian MD
Department of Nuclear Medicine, West China Hospital, Sichuan University,
37 Guoxue Lane, Wuhou District, Chengdu, Sichuan 610000, P.R. China
Tel:+86-18980601586
Fax:+86-028-85422187
rongtiannuclear@126.com

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Abstract
Multiple pulmonary hamartomas (PH) occur rarely, are mostly seen in females, and are usually leiomyomatous hamartomas. Here, we report an extremely rare case of a 30 years old male patient diagnosed as multiple pulmonary chondromatous hamartomas. He was admitted on May 2015 to our hospital for a 3 months history of cough. Multiple nodules in the right lung were detected on chest X-rays during a routine checkup 9 months ago and in a subsequent chest computed tomography (CT). However, he abandoned medical follow-up because he was asymptomatic. Nine months later, rare and atypical CT findings with progression were observed during this visit so that pulmonary metastases from an unknown primary tumor was suspected. Positron emission tomography/computed tomography (PET/CT) scan showed mild fluorine-18 fluorodeoxyglucose (^{18}F-FDG) uptake in the lesions and no abnormal foci in any other part of his body. A posterolateral thoracotomy was performed. Pathologic features were consistent with those of pulmonary chondromatous hamartomas.

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Introduction

Hamartomas are tumor-like malformations resulting from abnormal mixing or abnormal development of normal tissue components in the organ where they occur [1]. Pulmonary hamartomas (PH) constitute 5%-8% of all solitary lung tumors, account for 75% of all benign lung tumors [2], and are primarily (90%) solitary [3]. Multiple PH are rare, appear mostly in females, and are usually leiomyomatous [4, 5]. Here, we report an extremely rare case of pathologically verified multiple pulmonary chondromatous hamartoma in a male patient, with mild ^{18}F-FDG uptake on PET/CT scan.

Case Report

A 30 years old male smoker was admitted to our hospital for a 3 months history of cough in May 2015. He was informed of abnormal dense nodules of the right lung when he received a routine chest X-rays checkup 9 months ago, but that was unavailable for us to comparison. He was seen for a second opinion on these lesions in another institution and underwent a chest CT scan which revealed multiple nodules in the right lung, the upper and middle lobes affected predominantly. Some of the nodules were atypically calcified (Figure 1A-B). Routine laboratory tests were normal and transthoracic fine needle aspiration biopsy did not establish diagnosis. At that time, he was asymptomatic and abandoned medical follow-up. A CT scan performed by us demonstrated more multiple pulmonary nodules. Some of the nodules were enlarged compared to the previous CT images. Pulmonary metastases from an unknown primary tumor were suspected. The patient subsequently underwent a ^{18}F-FDG PET/CT scan. The pulmonary nodules were mildly ^{18}F-FDG avid (maximal standardized uptake value, SUVmax=2.6), and no abnormal foci were observed in any other part of his body (Figure 2A-D). Because of obvious symptoms ascribed to progression, thoracotomy was proposed. Based on the histopathological findings, the patient was diagnosed as multiple pulmonary chondromatous hamartoma (Figure 3A-B).

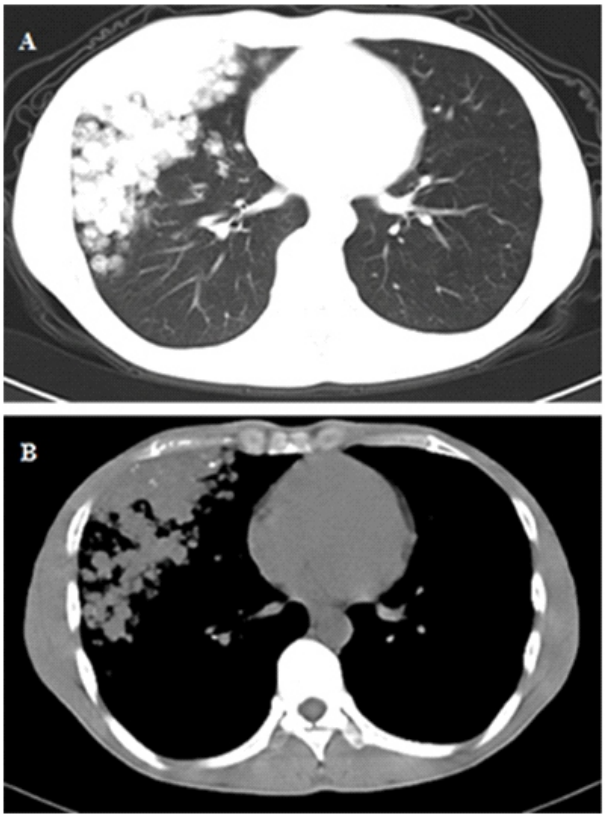

Figure 1. A CT scan (A-B, lung and mediastinal windows, respectively) subsequent to the routine cheat X-ray checkup showed multiple nodules in the right lung, some of which were calcified.

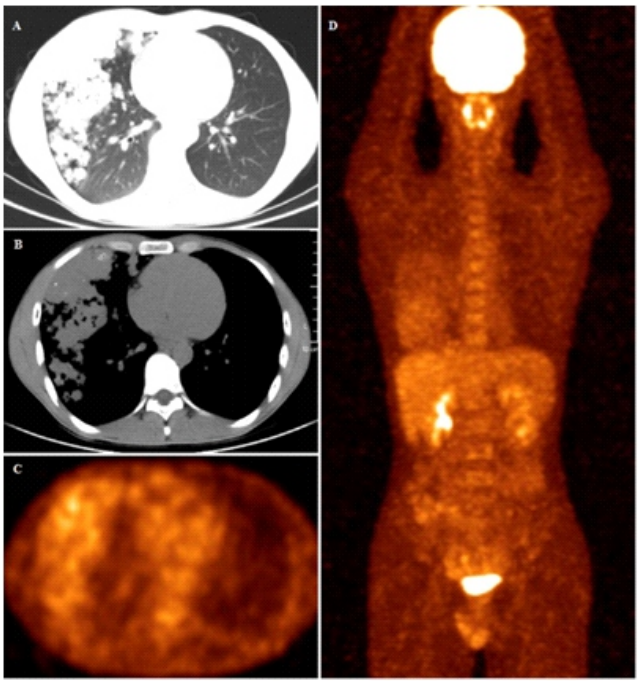

Figure 2. CT (A-B, lung and mediastinal windows, respectively) and PET scans (C-D) 9 months after abandoning medical follow-up showed progression of the lesions and slight ^{18}F-FDG accumulation (SUVmax=2.6). No ^{18}F-FDG-avid primary tumor was demonstrated.

Discussion

Hamartomas occur frequently and account for 75% of all benign lung tumors [2, 6]. They arise most frequently when patients are in their sixties [7]. Patients with PH are usually asymptomatic, whereas some patients may have respiratory symptoms such as cough, phlegm, hemoptysis, pyrexia or chest tightness [8]. Pulmonary hamartomas are frequently discovered on chest radiographic examination, in which they typically appear as single, well-circumscribed round nodules [8, 9]. Multiple PH rarely occur, appear mostly in females, and they are usually leiomyomatous [4, 5]. In Nili M et al. study (1979), all reported cases (12/12) of multiple PH occured in women and 6 of the 12 women (50%) had a history of leiomyomatous disease of the uterus [4]. These authors found that the histological appearance of multiple hamartomas differs from the solitary hamartoma in that no cartilage is usually seen in the former. The tumor is consistent of fibrous ends interlaced with bundles of smooth muscle. Here we report an extremely rare case of a multiple pulmonary chondromatous hamartoma. Only 22 of such cases have so far been reported [3, 5, 9-11]. This type of hamartoma usually represents a manifestation of Cowden syndrome (mucocu-taneous lesions, multiple benign tumors in internal organs, and an increased risk for breast, thyroid, urogenital,

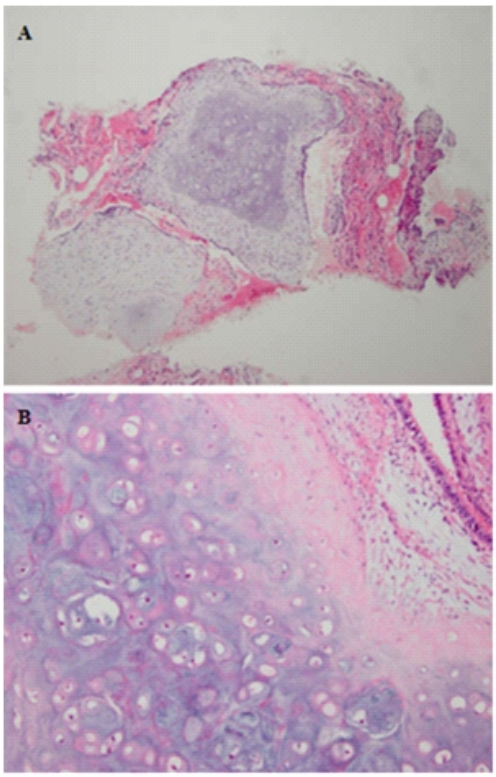

Figure 3. (A) Islands of mature cartilage were detected with intervening fragments of lobules and hyalinized stroma (H&E stain, original magnification ×10). (B) Islands of mature cartilage and immature mesenchymal tissue containing spindle cells were present within the intervening stroma (H&E stain, original magnification ×20).

and digestive tract cancer) or Carney triad (gastric leiomyoblastoma, pulmonary hamartoma, and extra-adrenal paraganglioma) [5, 9]. The current case presented in a young male without any of the concomitant features of these diseases.

Intranodular fat (attenuation, -40 to -120HU) and popcorn-like chondroid calcifications are reliable indicators of hamartomas. However, some hamartomas contain neither fat nor calcification. Siegelman et al. (1986) reported that fat is only observed in approximately 34% of hamar-tomas and fat with calcification is present in approximately 21% of hamartomas [12]. In addition, some cases showing rapid growth have been reported, although PH typically show slow annual growth [1, 13]. These can cause a dilemma when making a differential diagnosis from malignant tumor in daily practice. In the present case, a suspicion of metastatic malignancy was raised in the CT report in view of rare and atypical CT findings with progression of the scanning image. Therefore, ^{18}F-FDG PET/CT was performed to further evaluate the lesions and to search for metastatic disease. Mild ^{18}F-FDG uptake in the lesions was observed and no ^{18}F-FDG avid foci in any other part of his body was demonstrated, which was highly suggestive of begin nature of the lesions. Finally, postoperative pathological examination confirmed the diagnosis of pulmonary chondromatous hamartomas.

Fluorine-18-FDG PET/CT imaging features of pulmonary chondromatous hamartomas are less well known [14]. There is no satisfying explanation for the mild tracer uptake [6, 15], as was observed in the present case. However, ^{18}F-FDG PET/CT is a useful noninvasive method in the differential diagnosis of indeterminate lung lesions. A study conducted by De Cicco et al. (2008) revealed that ^{18}F-FDG PET/CT provided better performance than CT alone when diagnosing hamartoma lesions as benign [6]. Our case offers an example of successful application of ^{18}F-FDG PET/CT imaging in excluding malignancy for a patient with multiple pulmonary chondromatous hamartomas.

In conclusion, pulmonary chondromatous hamartoma presenting as multiple nodules in a male as in the case we present is extremely rare in the literature. When occurs, its multiple types can complicate the differential diagnosis from metastatic malignancy, especially in cases with no evidence of intranodular fat and popcorn-like chondroid calcifications. Fluorine-18-FDG PET/CT as a noninvasive assessment is useful for differentiating multiple pulmonary hamartomas from metastatic lung lesions.

The authors of this study declare no conflict of interest

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