Slight uptake of ^{18}F-FDG on positron emission tomography in pulmonary hamartoma: A case report

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Abstract. The present study reports the case of a 77-year-old female that was asymptomatic at presentation and was found to possess a lesion that was incidentally identified on a computed tomography (CT) scan. The CT scan revealed a non-homogeneous, hypodense, non-lobulated solid mass, ~1.2 cm in diameter, in the left upper lobe of the lung that demonstrated minimal contrast enhancement. The following CT scan was performed only two years later. This scan revealed that the non-homogeneous round mass had increased in size to ~1.7 cm in diameter, and possessed an irregular margin, in addition to being slightly lobulated with no calcification or fat. Combined positron emission tomography and CT revealed a lobulated mass that was ~1.9 cm in diameter, demonstrating an irregular margin with involvement of the mediastinal pleura. Slight uptake of ^{18}F-fluorodeoxyglucose was also detected. The final histological diagnosis was pulmonary hamartoma.

Introduction

Pulmonary hamartoma is the most common benign tumor of the lung, accounting for 6% of all solitary pulmonary nodules (1). The majority of hamartomas are identified incidentally, with the peak incidence occurring in patients in their sixth decade (2). Hamartomas are rarely symptomatic, but in symptomatic cases the hamartoma is associated with hemoptysis or cough. Originally, hamartomas were considered to be congenital lesions, but pulmonary hamartomas are currently considered to be true neoplasms by the majority of investigators. Consistent with this concept are the observations that hamartomas are often identified in the elderly and that the lesions grow slowly. The typical radiographic appearance of a hamartoma is that of a smooth or slightly lobulated peripheral solitary pulmonary nodule. The presence of fat or popcorn-like calcifications may enable the confident diagnosis of a hamartoma, but these findings are not usually identified.

The radiological diagnosis of hamartoma is usually based on computed tomography (CT) findings, particularly the detection of popcorn-like calcifications and fat (3). ^{18}F-fluorodeoxyglucose (^{18}F-FDG) positron emission tomography (PET)/CT investigations may be of use when neither calcification nor fat is identified on CT scans (4). The present study reports the case of a 77-year-old female patient that was diagnosed with a pulmonary hamartoma that demonstrated atypical imaging findings on two imaging modalities.

Case report

An asymptomatic 77-year-old female was admitted to The 117th hospital of PLA (Hangzhou, China) for the investigation of a lesion in the left upper lobe of the lung that had been present for 3 years.

The patient had undergone a routine health examination in The 117th hospital of PLA on January 29, 2007. The patient was asymptomatic and no abnormal findings were detected in the examination, with the exception of a 1.2-cm solitary pulmonary nodule that was identified on a CT scan. The scan revealed a round non-homogeneous parenchymal neoformation in the anterior segment of the left upper lobe of the lung, with a clear border (Fig. 1A). The contrast-enhanced CT that was performed four days later revealed that the neoformation contained hypodense regions with possible involvement of the vessel. In addition, non-homogeneous enhancement during the arterial phase was demonstrated. There was no indication of lobulation and spiculation. Neither the mediastinal nor hilar lymph nodes were enlarged (Fig. 1B and C).

Since there was no evidence of a malignant neoplasm, it was decided continue to follow-up the present patient. The following CT scan was performed two years subsequent to the first presentation (March 9, 2009). The CT scan revealed that the non-homogeneous oval mass had increased in size, to

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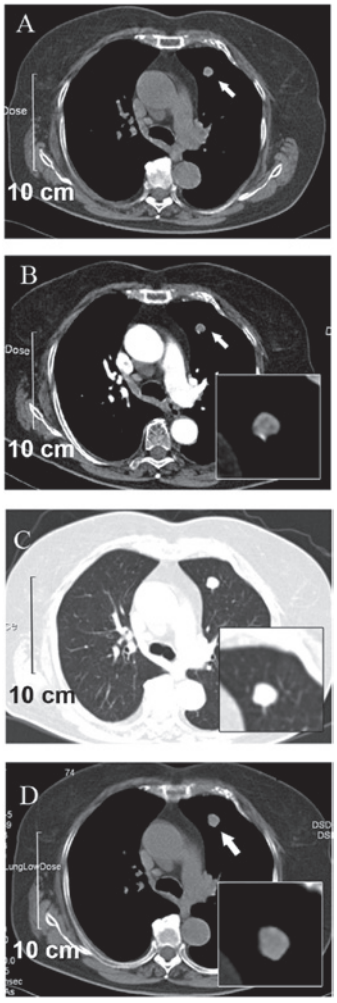

Figure 1. (A) CT of the chest revealed a round non-homogeneous parenchymal neoformation (arrow) that was ~1.2 cm in diameter and located in the anterius segment of the left upper lobe of the lung. The lesion was sharply demarcated. (B) The contrast-enhanced CT revealed minimal contrast-enhanced areas, and the lesion did not include any calcifications or fat. There was no sign of lobation and speculation. Neither the mediastinal nor hilar lymph nodes were observed to be enlarged in the (B) mediastinal and (C) pulmonary windows. CT, computed tomography.

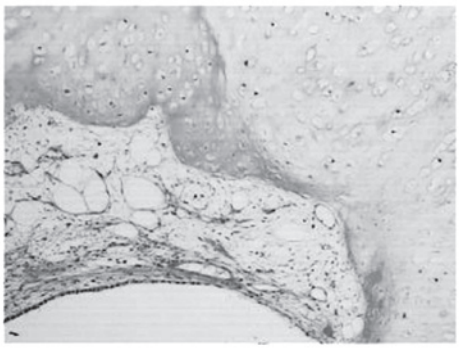

Figure 2. The lesion was composed of fat, cartilage and smooth muscle, covered by normal bronchial epithelial cells.

~1.7 cm in diameter, with a regular margin and no calcification (Fig. 2). The results of physical examination and laboratory tests (serum tumor markers detection, including AFP, CEA and CA125, performed on November 6, 2010) were negative and the

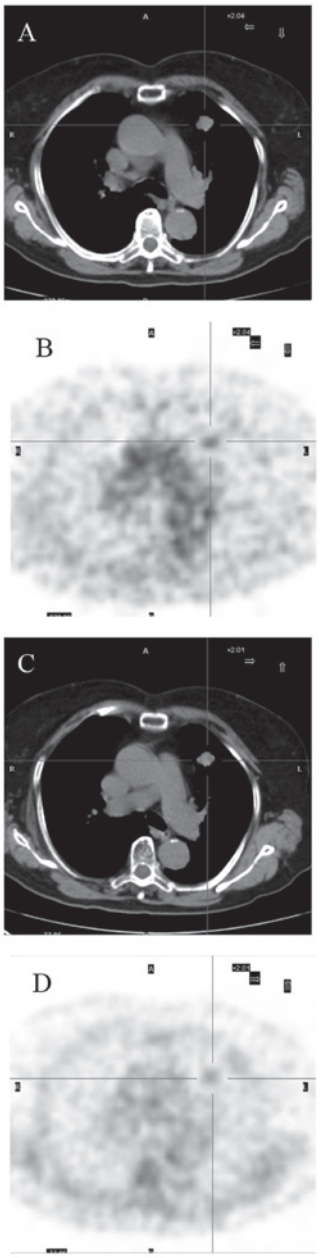

Figure 3. (A) Positron emission tomography/computed tomography revealed a slightly lobulated mass in the left upper lobe of the lung that was ~1.9 cm in diameter and well-defined, with involvement of the mediastinal pleura. (B) Slight uptake of ^{18}F-fluorodeoxyglucose was also detected. (C and D) Delayed scanning demonstrated that the lesion possessed diminishing metabolism.

appearance of the radiology results were non-specific. Therefore, PET/CT was performed on November 8, 2010. PET/CT revealed a slightly lobulated mass in the left upper lobe of the lung that was ~1.9 cm in diameter and well defined, with involvement of the mediastinal pleura. Slight uptake of ^{18}F-FDG was also detected. The maximum standardized uptake value (SUV_{max}) was found to be 2.44. Delayed scanning demonstrated that the lesion possessed diminishing metabolism, with an SUV_{max} of 1.64 (Fig. 3). Review of the laboratory tests, including those for serum tumor markers, remained within the normal range.

The lesion was suspected to be a pulmonary benign tumor or primary lung cancer, such as well-differentiated adenocarcinoma, but the diagnosis was unable to be confirmed

pre-operatively. Since the patient requested removal of the lesion, it was resected by video-assisted thoracic surgery. Gross examination of the resected specimen revealed that the tumor measured 2.5x2.0 cm, and was a white, multilobular, flexible mass without an envelope. The margin of the lesion and peripheral lung tissue was clear and there was no area demonstrating infiltrative growth. Histologically, the lesion was composed of fat, cartilage and smooth muscle, covered by normal bronchial epithelial cells (Fig. 3). On the basis of these findings, the tumor was diagnosed as a hamartoma. Six months subsequent to the surgical procedure, the patient is completely well, with no evidence of recurrence.

Discussion

Hamartomas are the third most common solitary pulmonary nodule, following granuloma and carcinoma, and usually account for 6-8% of localized parenchymal masses treated by thoracotomy (1). Individuals aged 60-79 years old develop hamartomas with the highest frequency (2). The majority of the patients with pulmonary hamartoma are free of symptoms, and the tumor is found incidentally on chest X-ray examination. On X-ray examination, a pulmonary hamartoma usually presents as a sharply demarcated coin lesion with slow growth, occasionally with calcification, but this is not diagnostic since calcifications may appear in carcinomas.

The radiological diagnosis of a pulmonary hamartoma has frequently been made using CT. In particular, CT densitometry with an advanced narrow collimated technique has been accepted as one of the best sensitive methods from eliminated partial volume averaging in detecting intranodular fat (-40 to -120 Hounsfield units), providing a highly predictive diagnosis of a pulmonary hamartoma or characteristic popcorn calcifications (1,2). However, hamartomas are extremely challenging to diagnose when the lesions possess neither fat nor calcification in 50% of pulmonary hamartomas, at most (1). This results in a diagnostic challenge, as the hamartomas cannot be differentiated from a primary or secondary lung cancer or other benign nodules. In the present study, the patient was asymptomatic and the tumor was identified incidentally on a CT scan. The mean size of hamartomas increased between 1.2 and 1.9 cm in three years, with an irregular margin and no calcification or fat identified on CT. The lesion was suspected to be a benign or primary lung cancer, such as well-differentiated adenocarcinoma, but the diagnosis was unable to be confirmed pre-operatively. ^{18}FDG PET/CT studies may aid diagnosis when neither calcification nor fat is demonstrated on CT studies.

The use of an ^{18}F-FDG-PET scan is based on the principle that cancer cells demonstrate an increased glucose uptake and higher rate of glycolysis compared with non-cancerous cells. In a meta-analysis reported by Gould *et al*, a sensitivity of 97% and a specificity of 78% were determined for the differentiation of malignant from benign pulmonary lesions using an ^{18}F-FDG-PET scan (4). However, ^{18}F-FDG-PET possesses a limited capacity for the detection of tumors with low glycolytic activity, including small tumors, bronchioloalveolar cell carcinoma and carcinoid tumors (5). As a result, the use of ^{18}F-FDG-PET scans for the identification of hamartomas is limited.

To the best of our knowledge, there are only two published studies that investigated the ^{18}F-FDG-PET scan findings in hamartomas. Teramoto *et al* reported that the uptake of ^{18}F-FDG did not increase in pulmonary hamartomas (6). By contrast, in the case reported by Himpe *et al*, only slightly elevated uptake with an SUV_{max} of 3.3 and SUV_{mean} of 1.7 was identified (7). Consistent with the findings of Himpe *et al*, the present patient possessed a lesion with an SUV_{max} of 2.44. Delayed scanning demonstrated that the lesion possessed diminishing metabolism, with an SUV_{max} of 1.64. The mechanism of slightly enhanced uptake of ^{18}F-FDG in hamartomas remains unknown. Benign and slow-growing tumors usually demonstrate low glucose metabolism. Therefore, familiarity with this false positive finding would aid the interpretation of ^{18}F-FDG-PET scan results on solitary pulmonary nodules.

The clinical features are usually benign and the prognosis subsequent to surgical excision is usually excellent. However, certain studies consider hamartomas to be a potentially low-grade malignancy, since a small number of cases of squamous cell carcinoma, adenocarcinoma or sarcoma arising from pulmonary hamartoma have been reported (8). In addition, a previous study reported that synchronous or metachronous lung cancer with hamartoma in an adjacent region (9). If a benign tumor is strongly suspected, observation of the patient is reasonable. However, if the lesion has increased in size more rapidly than usual, in addition to being large and symptomatic, patients should undergo resection, unless surgery is contraindicated due to poor pulmonary function, comorbidities, or withholding of consent. Furthermore, if malignant causes cannot be excluded and patients agree to undergo resection, the mass may be excised for diagnosis and intent to cure. In the present patient, although the nodule was strongly suspected to be benign, a malignant cause could not be completely excluded and the patient agreed to undergo resection to obtain a diagnosis. Therefore, the nodule was removed using thoracoscopic enucleation. The findings of the present case add further support to the continued use of 18F-FDG-PET scan in hamartomas

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