CASE REPORT

A Case of Solitary Fibrous Tumor of the Pleura That Increased in Size After Resection of Renal Cell Carcinoma

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ABSTRACT — Background. Solitary fibrous tumor (SFT) is a rare tumor of mesenchymal origin that occurs preferentially in the pleura. When a solitary thoracic nodule is detected in a patient suffering from any type of cancer, it is difficult to determine whether the nodule is a metastatic lesion or a de novo primary thoracic tumor. Case. A 62-year-old man, previously treated for stage IB renal cell carcinoma by nephrectomy, was admitted to our hospital for further examination following detection of a solitary thoracic nodule, which was suspected to be thoracic metastasis. The patient underwent computed tomography (CT)-guided lung biopsy and SFT was subsequently diagnosed, although hemothorax occurred due to intercostal artery laceration. Video-assisted thoracoscopic surgery (VATS) was performed and no recurrence has occurred thus far. Conclusion. When a solitary thoracic nodule is detected in a patient suffering from any type of cancer, SFT should be considered in the differential diagnosis, and surgical resection may be considered as initial treatment if the primary tumor is controlled, no extrathoracic metastases are present, and the patient is in good enough condition to undergo the operation.

KEY WORDS — Solitary fibrous tumor, Renal cell carcinoma, Differential diagnosis, CT-guided lung biopsy, Intercostal artery laceration

INTRODUCTION

Solitary fibrous tumor (SFT) is a rare tumor of mesenchymal origin that occurs preferentially in the pleura. When a solitary thoracic nodule is detected in a patient suffering from any type of cancer, it is difficult to determine whether the nodule is a metastatic lesion or a primary thoracic tumor. The necessity of preoperative puncture biopsy for diagnosis of a solitary thoracic nodule remains controversial and surgical resection may be considered as initial treatment if the primary tumor is controlled, no extrathoracic metastases are present, and the patient is in good enough condition to undergo the operation. Here, we present a case of SFT of the pleura that had increased in size after resection of renal cell carcinoma (RCC) with review of the relevant literature.

CASE REPORT

A 62-year-old man was admitted to our hospital for further examination following abnormal chest radiograph and computed tomography (CT) findings in 2012. He had no specific thoracic symptoms. He was an ex-smoker of 75 pack years and denied any history of asbestos exposure. He had received nifedipine, bisoprolol fumarate and rosuvastatin calcium as treatment for hypertension, hyperlipidaemia, and paroxysmal atrial fibrillation since age 56.

One year previously (2011), left RCC was suspected by the Department of Urology. A CT showed a left re-
nal tumor (5.0 cm in diameter) and a small nodule (2.8 cm in diameter) in the lower left lung field. Since the small thoracic nodule was not suspected to be thoracic metastasis of RCC at that time, he underwent left nephrectomy alone. Pathological diagnosis of the left renal tumor was stage IB RCC and the cell type was clear cell carcinoma.

He was monitored and did not show any symptoms after surgery. However, the size of the thoracic nodule was found to be increased one year later (in 2012), and thoracic metastasis of RCC was suspected. A physical examination upon admission revealed body temperature of 36.6°C, blood pressure of 119/72 mmHg, and regular pulse of 89 beats/min. The cervical lymph nodes were not palpable. Lung and heart auscultation was normal, and abdominal examination revealed an operation scar from the radical nephrectomy on the left upper abdomen. Laboratory examinations on admission were as follows: complete blood count as well as liver and renal function tests were close to normal. The prothrombin and bleeding times were within normal limits. Tumor marker tests found that prostate specific antigen (PSA) was slightly elevated (4.59 ng/ml, normal range < 4.00 ng/ml), but carcinoembryonic antigen (CEA) (2.6 ng/ml), soluble cytokeratin fragment 19 (CYFRA 21-1) (28 ng/ml), and pro-gastrin-releasing peptide (ProGRP) (40.0 pg/ml) were within normal limits. A chest radiograph in 2012 showed a well-defined, left mediastinal mass (Figure 1). In addition, a contrast-enhanced CT scan of the chest in 2012 showed a well-defined, circumscribed, heterogeneous mass that was 5.0 × 3.4 cm in size in the left paraspinal area without enlargement of the hilar or mediastinal lymph nodes (Figure 2A). Magnetic resonance imaging (MRI) features of the chest in 2012 showed a well-defined mass with a low signal intensity on T1-weighted images, mixed low and intermediate signal intensities on T2-weighted images, and relatively strong enhancement on T1-weighted contrast-enhanced image (Figure 2B). By contrast, 18-fluorodeoxyglucose-positron emission tomography (FDG-PET) in 2011 showed a well-defined, circumscribed nodule that was 2.8 × 2.4 cm in size with no uptake (the maximum standardized uptake value was 1.65) (Figure 2C). From these findings, the mass was suspected to be a lung or pleural tumor, thoracic metastasis of a malignant tumor (notably, the resected RCC), or posterior mediastinal tumor such as neurogenic tumor. Since he rejected the option of diagnosis by video-assisted thoroscopic surgery (VATS), CT-guided lung biopsy was performed, and bleeding and hemothorax occurred just after biopsy due to intercostal artery laceration. He underwent chest tube insertion, which immediately drained about 700 ml of blood and the bleeding stopped spontaneously. Subsequently, his blood pressure and heart rate became stable on receiving fluid replacement. The biopsy specimen demonstrated proliferating oval or spindle-shaped cells with various densities of collagenous stroma and positive immunohistochemical staining for CD34, which was suspected to be SFT (Figure 3A). In addition, the biopsy specimen included a piece of arterial wall, which was thought to be part of the intercostal artery (Figure 3B). VATS was performed one month later, and the tumor that had a pedicle projecting from the visceral pleura into the left lung S5 area was resected with a histologically negative margin (Figure 4A). Histopathological findings showed patternless distributions of closely packed oval or spindle-shaped cells with varying degrees of fibrosis and interspersed branching staghorn-shaped vessels as well as positive immunohistochemical staining for CD34. Myxoid and hyaline degeneration were found, but no necrosis or hemorrhage was observed and the Ki-67/MIB1 labeling index was less than 5%. No mitotic figures, nuclear pleomorphisms, or multinucleated giant cells were present (Figure 4B). Therefore, we ultimately diagnosed benign SFT and monitored him without postoperative adjuvant therapy.
Figure 2. (A) A contrast-enhanced CT scan of the chest performed in 2012 shows a well-defined, circumscribed, heterogeneous mass, 5.0×3.4 cm in size, in the left paraspinal area. (B) A sagittal T2-weighted MRI of the chest conducted in 2012 shows a well-defined mass with mixed low and intermediate signal intensities. (C) FDG-PET performed in 2011 showed a well-defined, circumscribed nodule, 28×24 cm in size, with no uptake (the maximum standardized uptake value was 1.65).

Figure 3. The biopsy specimens were obtained by CT-guided lung biopsy and examined by histopathological analysis. (A) Proliferating oval or spindle-shaped cells with various densities of collagenous stroma (hematoxylin and eosin staining, ×200) and positive immunohistochemical staining for CD34 (inset) were found. (B) A piece of arterial wall, which was thought to be part of the intercostal artery, was found (hematoxylin and eosin staining, ×100).
because complete resection seemed to have been achieved. No recurrence has occurred at 12 months.

**DISCUSSION**

SFT is a rare tumor of mesenchymal origin that occurs preferentially in the pleura.\(^1\) SFT of the pleura accounts for less than 5\% of all pleural tumors, and its incidence is 2.8 per 100000 according to the literature.\(^4\) SFT of the pleura occurs in a wide age range, but it is much more common in the fifth and sixth decades of life. Although some authors have reported a slight female predominance, it seems to affect both genders equally.\(^5\) SFT of the pleura often has a silent clinical course for several years, when it is discovered incidentally during chest X-ray or CT examination.\(^1,5\) SFT of the pleura arises more frequently from the visceral pleura than from the parietal pleura. Pedunculated tumors may change in location and shape of the mass during respiration and with the patient’s position.\(^1,5\) Histologically, SFT is composed of patternless distribution of closely packed oval or spindle-shaped cells with varying degrees of fibrosis and interspersed branching staghorn-shaped vessels as well as positive immunohistochemical staining for CD34.\(^1,4,5\) CT scan usually demonstrates a well-defined and occasionally lobulated mass with soft tissue attenuation, appearing typically in contact with the pleural surface.\(^5\) On contrast-enhanced CT, most cases show mild or greater enhancement, which is consistent with the histological features of a hypervascular tumor such as SFT.\(^5\) As in the present case, heterogeneous attenuation on contrast-enhanced CT is also observed and seems to be correlated with gross descriptions of myxoid degeneration and various cellularity.\(^5\) The necessity of preoperative puncture biopsy for diagnosis of SFT remains controversial. A definitive diagnosis of SFT is achieved by obtaining a sufficient amount of biopsy tissue and immunohistochemical staining.\(^5,7\) However, when puncture biopsy cannot obtain adequate tissue, it is difficult to reach a histological diagnosis.

In the present case, RCC was initially diagnosed. It was necessary to differentiate between solitary thoracic nodule and thoracic metastasis of RCC because the size of the lesion had increased within one year. RCC accounts for approximately 3\% of all adult cancers and 20-30\% of patients with RCC have distant metastases at presentation. Clear cell carcinoma is the most common histologic subtype representing 70-75\% of all RCCs and is known to be hypervascular.\(^5\) The lungs are the most frequent metastatic site and are affected in 50-60\% of patients with metastases.\(^9\) In general, metastatic pulmonary tumors commonly have multiple nodules, but occasionally, they have solitary nodules. When a solitary thoracic nodule is detected in a patient suffering from any type of cancer, it is difficult to determine whether the nodule is metastatic or a de novo primary thoracic tumor. According to the study of
solitary pulmonary metastases from clear cell carcinoma by Yanagawa et al., the nodules are solid lesions, round or oval in shape with relatively well-defined borders, have homogeneous soft tissue density, and show greater enhancement on contrast-enhanced CT. These features are similar to SFT of the pleura, therefore, it is difficult to differentiate from thoracic metastasis of RCC. The resection of pulmonary metastases can prolong the survival of carefully selected patients and its therapeutic value is now accepted. In the present case, the patient underwent CT-guided lung biopsy and SFT was diagnosed although hemothorax occurred due to intercostal artery laceration. The rate of hemothorax as a complication of CT-guided lung biopsy is generally very low (<0.10%). The risk of intercostal laceration by thoracentesis increases in elderly patients because the arteries become increasingly tortuous with age, and the amount of space available for safe insertion of needle decreases. In addition, the safety rate of puncturing the paraspinal area in cases such as the present, tends to be low.

In summary, we present a case of SFT of the pleura which increased in size after resection of RCC. When a solitary thoracic nodule is detected in a patient suffering from any type of cancer, SFT should be considered in the differential diagnosis, and imaging studies such as X-ray and CT should be performed while changing the patient’s position. Furthermore, surgical resection may be considered as initial treatment if the primary tumor is controlled, no extrathoracic metastases are present, and the patient is in good enough condition to undergo the operation.

REFERENCES