

A Case of Recurrence and Rapid Growth of Pleural Solitary Fibrous Tumor 8 Years after Initial Surgery

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A 69-year-old woman underwent resection of a solitary fibrous tumor (SFT) of the left pleura in April 1997 and of locally recurrent SFT in the left thoracic cavity in September 2003. A postoperative follow-up chest CT scan in March 2005 revealed pleural thickening at two sites of the left thoracic cavity. A further chest CT scan performed in September of the same year showed the enlarging tendency of the lesions, suggesting SFT recurrence, for which surgery was performed in January 2006. The two resected tumors were benign SFT, and were diagnosed as locally recurrent SFT in the left thoracic cavity. It has been reported that despite its benign histopathology, pleural SFT recurs more than once after surgery, and the interval between recurrences tends to shorten from the second recurrence. In this patient, the tumor recurred twice and showed a rapidly enlarging tendency at the time of the second recurrence, suggesting the need for careful follow-up at short intervals. (Ann Thorac Cardiovasc Surg 2009; 15: 178–181)

Key words: solitary fibrous tumor, recurrence, rapid growth

Introduction

It has been reported that despite its benign histopathology, solitary fibrous tumor (SFT) recurs more than once after resection. We report a case of SFT that recurred in the 6th and 8th years after resection and rapidly enlarged at the time of the second recurrence, along with a review of the literature.

Case Report

A 69-year-old woman with no contributory family his-

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Received February 5, 2008; accepted for publication May 27, 2008
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tory and no history of smoking underwent resection of SFT of the left pleura in April 1997 and of locally recurrent SFT in the left thoracic cavity in September 2003. A postoperative follow-up chest CT scan in September 2005 revealed abnormal shadows in the left thoracic cavity, suggesting recurrent SFT. Thus she was admitted in January 2006 for further evaluation and treatment.

On admission, she measured 153.6 cm (5 ft) in height and weighed 53.0 kg (116 lb). Her body temperature was 36.6°C, blood pressure 132/60 mmHg, and pulse 82/min. The bulbar conjunctivas were not stained yellow, and the palpebral conjunctivas were not anemic. No superficial lymph nodes were palpable. A surgical scar was noted on the left chest. Chest auscultation revealed no abnormal breathing sounds or heart murmurs. There were no abnormalities of the abdomen, no neurological abnormalities, and leg edema was not observed. Blood chemistry and respiratory function tests on admission also revealed no abnormalities. Chest CT scans performed in March 2005 (Fig. 1) showed pleural thickenings, 3 × 1 cm (Fig. 1A) and 5 × 1 cm (Fig. 1B), at the sites of surgical scars in the left pleural cavity. Since these

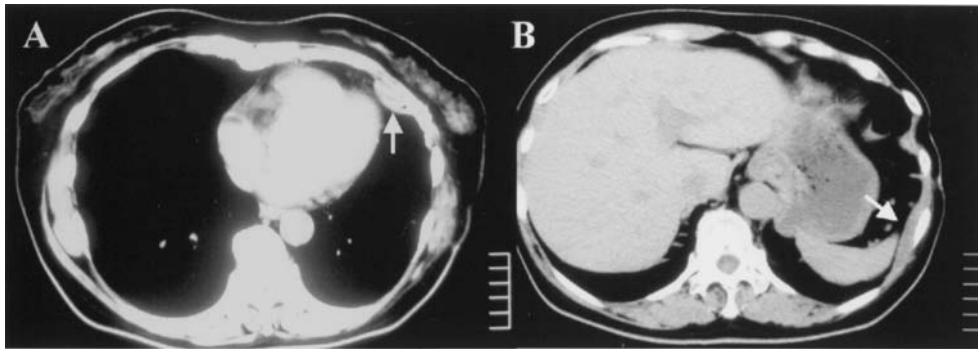


Fig. 1. Chest CT scans performed in March 2005 showed pleural thickenings, 3 × 1 cm (A, arrow) and 5 × 1 cm (B, arrow), at the sites of surgical scars in the left pleural cavity.

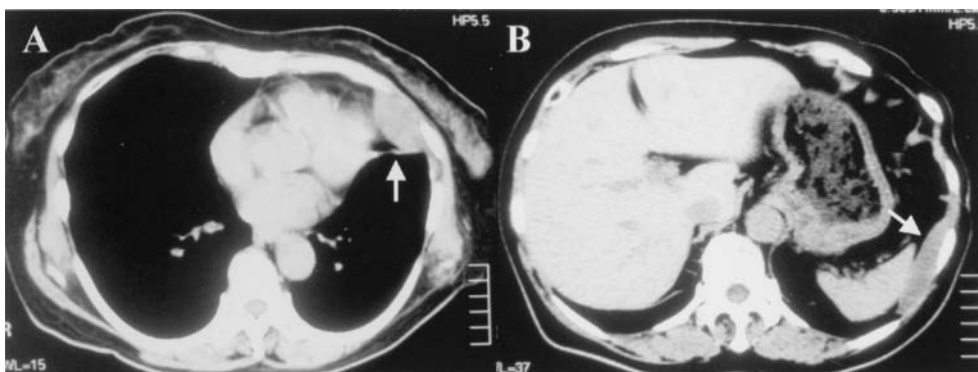


Fig. 2. Chest CT scans performed in September 2005 revealed that the previously observed pleural thickenings had enlarged to 5 × 4 cm (A, arrow) and 7 × 3 cm (B, arrow), respectively.

findings may have represented postoperative changes, the patient was followed up. Chest CT scans performed in September 2005 (Fig. 2) revealed that the previously observed pleural thickenings had enlarged to 5 × 4 cm (Fig. 2A) and 7 × 3 cm (Fig. 2B), respectively. From these findings, we suspected locally recurrent SFT in the left pleural cavity and performed surgery in January 2006. (Fig. 2A) An incision was made lateral and inferior to the left breast, followed by a left anterior thoracotomy at the level of the 5th intercostal space. A tumor was present on the inside of the chest wall, separable from the pericardium, osseous chest wall, and lung, and it was removed. (Fig. 2B) A left posterior thoracotomy was performed at the 9th intercostal space via a posterior oblique incision. A second tumor was also present on the inside of the chest wall. Since the tumor was expected to invade to the previous combined resection site of the 9th rib and diaphragm, it was resected together with part of the 9th rib and diaphragm. The resected tumors (Fig. 3) measured 4.5 × 4 cm (Fig. 3A) and 7 × 4 cm (Fig. 3B) and were solid, relatively soft, encapsulated, and smooth surfaced.

Histopathological examination of the tumors showed

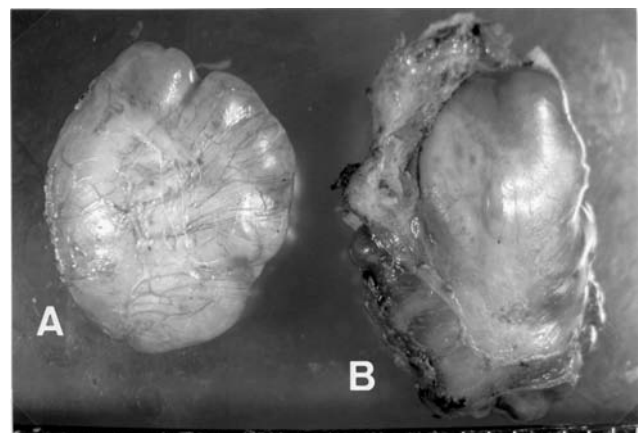


Fig. 3. The resected tumors were 4.5 × 4 cm (A) and 7 × 4 cm (B) in size, respectively, and both were solid, relatively soft, encapsulated, and smooth surfaced.

fibrous encapsulation and the dense proliferation of spindle-shaped cells with ovoid nuclei, along with hyperplasia of collagen fibers, with scattered areas of cyst formation and dilated lymph vessels (Fig. 4). A few mitotic figures were seen. The density of cells was

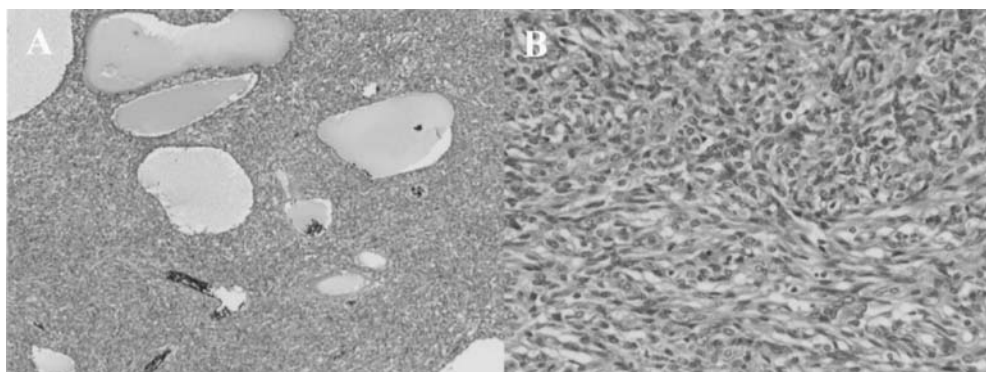


Fig. 4. Histopathological examinations of the tumors showed fibrous encapsulation and a dense proliferation of spindle-shaped cells with ovoid nuclei, along with hyperplasia of collagen fibers, with scattered areas of cyst formation and dilated lymph vessels. The density of cells was moderate, and multinucleated giant cells were present in some areas. There was no evidence of malignancy. (HE stain: A, ×20; B, ×100)

moderate, and multinucleated giant cells were present in some areas. There was neither cell invasion in the vicinity of the rib nor evidence of malignancy. Based on these findings, locally recurrent pleural SFT was diagnosed. The patient's postoperative course was uneventful, and she was discharged on the 12th postoperative day.

Discussion

The concept of SFT dates back to 1931, when Klemperer and Rabin¹⁾ proposed the classification of primary pleural tumors into localized and diffuse mesotheliomas. Currently, SFT is immunohistochemically characterized by negativity for cytokeratin, suggestive of a mesenchymal origin, and by positivity for CD34, suggestive of an epithelial origin, and it is considered to arise from undifferentiated mesenchymal cells in the subpleural connective tissue.^{2,3)} It usually develops in the intrathoracic pleura and has also been reported to arise in extrathoracic locations such as the peritoneum, thyroid gland, parotid gland, and soft tissue.^{4,5)}

Histopathologically, SFTs are classified into benign and malignant forms. England et al.⁵⁾ listed, as indicators of histological malignancy, (1) cell density, (2) the number of mitotic figures, (3) cell atypia, and (4) the presence or absence of bleeding and necrosis. Although surgical resection is the treatment rule for pleural SFT, many SFTs were reported to recur even if they had been diagnosed as benign according to these criteria.^{4,6,7)} Studies have reported that 15.7% of SFTs recur even after

complete resection,⁸⁾ and some recur more than once after surgery.^{4,6,7,9)} One of these tumors recurred after a postoperative period of 22 years.⁴⁾ In the present patient, the tumor recurred in the 6th and 8th years after surgery, indicating that SFT is histopathologically benign, but requires long-term postoperative follow-up even after complete resection.

Reported patterns of recurrence include local recurrence, pleural dissemination, metastasis to other lobes of the lung, and extrathoracic metastasis, with local recurrence being the most common pattern.^{4,7,9)} Although this patient twice exhibited local recurrence, SFT may metastasize to extrathoracic locations; therefore distant metastasis should be considered in the postoperative follow-up of this patient.

Kobayashi et al.¹⁰⁾ accumulated reported cases of recurrent SFT and noted that the time to the first recurrence was 24–120 months, but to the second and third recurrences it had shortened to 16–36 months. He suggested that patients showing more than one recurrence should be carefully followed up after surgery. Studies have also reported that benign primary SFTs showed histologically malignant transformation at the time of recurrence.^{11,12)} In this patient, the tumor was histopathologically benign at the time of the first resection and recurrence, but it had soon rapidly grown before the second recurrence, raising a suspicion of malignancy. However, the resected tumor was histopathologically benign and showed no invasion into the rib. Since even histopathologically benign SFTs may show a rapid growth tendency after more than one recurrence, we consider

that careful postoperative follow-up at short intervals is necessary.

The prognosis of SFT is not considered to reflect the degree of histological malignancy, presumably because even if the tumor is histopathologically benign at the time of resection, it may recur more than once and become malignant. Briselli et al.¹³⁾ emphasized the invasiveness and growth speed of SFT as its prognostic factors. At the time of the second recurrence, the tumor in this patient was histopathologically benign and apparently not invading the surrounding tissue, but it was rapidly growing, strongly suggesting a poor prognosis.

In summary, we encountered a case of SFT that recurred in the 6th and 8th years after initial surgery. Although the resected tumors were histopathologically benign, they showed a rapidly enlarging tendency at the time of the second recurrence. We consider that SFT requires careful follow-up at short intervals even if it is benign.

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