FIGURE 2. Interventional closure of paravalvular leak in patient 2. Two occluder devices are correctly placed (arrows). AP, Aortic prosthesis.

event-free survival (freedom from reoperation, reintervention, and significant residual shunt) was as low as 48% in a series of 24 patients.⁵ Common reasons for failure of this demanding intervention are poor imaging and unsuitable anatomy, such as crescent-shaped or multiple leaks and severe calcification. Additional limitations result from inadequate device configuration, because the occluders in use were originally designed for atrial and ventricular septal defects or patent ducts. Consequently, surgery still remains the criterion standard of treatment for paravalvular leaks while the technology of interventional closure is being advanced.

Our program comprised 1842 heart valve surgeries in 2006, including 26 patients (0.14%) operated on for para-

valvular leaks. Even in this large series, the 3 cases presented here are unusual because of the multiple previous surgeries. These cases were technically challenging and of high risk, and avoiding fourth-time cardiac surgery for reoperative valve exchange by use of catheter-based intervention would have been preferable. We therefore advocate a multidisciplinary approach that includes interventional cardiologists and surgeons. Depending on level of hemolysis, severity of regurgitation, and anatomy of the paravalvular leak, the risk of surgery must be weighed against the likelihood of success of intervention. Both treatment options are clearly explained to the patient so that an informed decision can be made. Surgical backup in case of interventional complications is provided for every intervention as well. Future studies, perhaps with more suitable device technology, may address a hybrid approach, so that outcomes can be improved even for most complex cases of paravalvular leakage.

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Resection of a giant thoracic solitary fibrous tumor through two separate thoracotomies

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Solitary fibrous tumor (SFT) of the pleura is an uncommon intrathoracic neoplasm. A middle-aged woman had progressive exertional dyspnea for 6 months, and eventually respiratory failure developed which necessitated ventilatory

support. After examination, a giant tumor was found to occupy the whole left pleural cavity with chest wall distention and severe mediastinal shift to the right side. We completely resected this giant tumor by two separate thoracotomies. The pathologic examination revealed an SFT weighing 5.1 kg. Her respiratory condition improved markedly after the operation and she was discharged 23 days postoperatively.

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CLINICAL SUMMARY

A 51-year-old woman, a nonsmoker, was referred to our hospital with endotracheal intubation and mechanical ventilation owing to respiratory distress caused by a huge lesion occupying the left chest cavity. A large tumor had been

FIGURE 1. A, Initial chest radiography showed complete whiteout of the left thoracic cavity. B, Initial computed tomogram with contrast revealed one mild heterogeneous, well-defined mass that compressed the mediastinum to right side.

discovered in the left side of the chest 3 years earlier during a physical examination, but she had ignored it because it was asymptomatic. Six months before admission, she had exertional dyspnea that she had treated with oxygen therapy without seeking medical intervention. Seventeen days before referral, the sudden onset of progressive dyspnea, drowsy consciousness, generalized edema, and eventually a faint occurred at home. She was sent to a district general hospital immediately, where emergency intubation was performed.

On physical examination, bilateral decreased breath sounds were noted without finger clubbing. Laboratory examination data were all within normal ranges, including a fasting sugar level of 107 mg/dL. Chest radiography (Figure 1, A) and computed tomography (CT) (Figure 1, B) both revealed one huge tumor that occupied the whole left chest cavity with chest wall distention and shifting of the mediastinum to the right side. Echo-guided biopsy favored an SFT.

After referral to our hospital, an entirely en bloc tumor excision was done by two thoracotomies through the fifth and ninth intercostal spaces. First, we made an incision through the fifth intercostal space for the upper and middle parts of

tumor dissection. Because the huge tumor measuring $32 \times 21 \times 8$ cm extended from the left apex downward to the level of left kidney with downward compression of the diaphragm and complete atelectasis of the left lung, we added an incision through the ninth intercostal space to dissect the lower part of the tumor. Finally the tumor was taken as a whole through the fifth intercostal space. The diaphragm was preserved but became rather redundant. The lung could be re-expanded but was diffusely congested owing to re-expansion lung edema. After complete resection, the tumor weighed 5.1 kg with focal necrosis (Figure 2, A). Microscopic examination disclosed a typical patternless pattern with occasional mitosis (4/30 high-power fields) (Figure 2, B). With the positive immunoreactivity of CD-34 and vimentin, it was confirmed as an SFT with malignant potential. On the 23rd day after surgery, she was discharged in excellent condition without sequel. During follow-up, thalidomide was prescribed for prevention of recurrence. Seventeen months after the operation, there was no evidence of local recurrence on chest radiograph (Figure 3). She was well without any respiratory symptoms.

FIGURE 2. A, The giant SFT was well circumscribed, white, and firm, with a whorled appearance and focal necrosis on cut surface. B, The tumor typically exhibited a patternless architecture characterized by the coexistence of hypocellular and hypercellular areas. (Hematoxylin and eosin; original magnification $\times 33$.)

FIGURE 3. Follow-up chest radiograph 17 months after the operation showed no evidence of recurrence or lung metastasis.

DISCUSSION

SFTs were first described by Klemperer and Rabin¹ in 1931. The growths were initially termed localized mesothelioma because the tumor was believed to arise from mesothelial cells.² However, according to immunohistochemical analysis, SFTs of the pleura express vimentin and CD-34, which are markers of mesenchymal cells, and do not express cytoplasmic keratins, which are found in mesotheliomas, indicating that these fibromas originate from the mesenchyme rather than from mesothelial cells.³

SFTs represent less than 5% of all neoplasms involving the pleura. Two thirds of these tumors arise from the visceral pleura, and one third arises from the parietal pleura.⁴ The etiology of SFTs is still unknown. The most frequent symptoms are dyspnea, cough, and chest pain. Paraneoplastic manifestations include arthritic pain, clubbed fingers, and hypoglycemia, but about 54% of the patients are asymptomatic. The tumors are noted by chest x-ray screening. By the time the tumors are found, their size can vary from centimeters to occupying the whole lung field. The SFTs on chest radiograph vary from a sharply delineated round or lobulated mass, with or without pleural effusion, to opacification of the complete hemithorax. Computed tomographic scanning and magnetic resonance imaging are important to evaluate the relationship of the tumor to neighboring structures

and to evaluate the resectability of the tumor. Displacement rather than invasion of adjacent structures is a characteristic finding of SFTs on computed tomography or magnetic resonance imaging.⁵

Histologic and immunohistochemical analysis are the most important diagnostic measures. Surgical resection is needed. Fine needle aspiration is mostly inconclusive because of the combination of acellular and hypercellular portions. The microscopic picture is dominated by a “patternless pattern,” with fibroblasts, collagen, and reticular fibers haphazardly arranged. Immunohistochemical study shows immunoreactivity for vimentin and CD-34. England, Hochholzer, and McCarthy⁶ listed criteria of malignant SFTs as followed: 4 mitotic figures in 10 high-power fields, hemorrhage, high cellularity, pleomorphism, and necrosis.

Most of these tumors have a benign clinical course if resected completely. However, according to the literature, even after complete resection, benign SFT still can result in local recurrence, malignant transformation, distant metastasis, and even death. On the contrary, even if the pathologic diagnosis is malignant SFT, cure may be effected by wide local resection. Because SFTs have the potential for adverse biological behavior, such as repeated recurrences or malignant transformation, the principle of treating this tumor is complete resection and followed by long-term follow-up.³

Oral thalidomide was prescribed because of our previous successful experience with thalidomide as an adjuvant therapy for patients with malignant SFT, margin-involved desmoids, and unresectable metastatic osteosarcoma. Thalidomide is postulated to have antiangiogenic activity through its ability to interfere with the vascular endothelial growth factor and basic fibroblast growth factor pathway.⁷

In summary, our patient had an exceptionally huge SFT weighing 5.1 kg that occupied the whole left thoracic cavity. To our knowledge, it is the biggest one ever reported in the literature. Thalidomide was prescribed for 9 months after the operation. There has been no recurrence or any sequel during 17 months’ follow-up.

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