Video-Assisted Thoracoscopic Surgery (VATS) for Patients with Solitary Fibrous Tumors of the Pleura

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Objectives: To present our experience of video-assisted thoracoscopic surgery (VATS) for patients with solitary fibrous tumors of the pleura (SFTPs) and to discuss the treatment of choice of such neoplasms.

Methods: Between June 2000 and September 2008, 21 patients with SFTPs (9 men and 12 women) underwent VATS at our department. The mean age was 52.5 years (range, 33–76 years).

Results: Surgical excision was performed in all patients. Surgical excision was performed by VATS in 15 patients (71.4%), by VATS plus a small thoracotomy (<5 cm) in 4 patients (19.0%), and by posterolateral thoracotomy accompanied by VATS in 2 patients (9.5%). Mean chest drain duration was 2.3 days (range, 1–4 days), and the mean hospital stay was 7.2 days (range, 4–15 days). There were 18 pathologically benign SFTP cases (85.7%) and 3 malignant SFTP cases (14.3%). There was no operative morbidity or mortality. No recurrence or metastasis of SFTPs developed during postoperative median follow-up period of 43 months.

Conclusions: Complete resection and close follow-up for years after operation is recommended for SFTPs. VATS may play an important role in reducing the size of the thoracotomy incision in the treatment of SFTPs, which results in less invasive surgery.

Key Words: Solitary fibrous tumors of the pleura, Video-assisted thoracoscopic surgery, Immunohistochemical analysis.

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Solitary fibrous tumors of the pleura (SFTPs) are rare neoplasms that usually originate from the visceral pleura. Because its pathologic characteristics was first described by Klemperer and Rabin1 in 1931, the nomenclature has become confused, and the disease has also been referred to as a localized mesothelioma, localized fibrous tumor, fibrous mesothelioma, or a pleural fibroma.2 The further development of electron microscopy and immunohistochemistry has clarified that the tumor does not originate from the mesothelial layer but from the submesothelial, noncommitted mesenchymal layer.3,4 Thus, the various names used for this disease have become unified, and the disease is now referred to as solitary or localized fibrous tumors of the pleura. Mesenchyme is pluripotent tissue and possesses diverse differentiation potential to bone, cartilage, or blood vessels. Because of this diversity of the mesenchyme, the pathologic morphology of SFTP seems variable.5 Recent studies on SFTP have mainly involved immunologic markers for pathologic diagnosis6,7 and some clinical reports that include postsurgical resection results and description of its clinical behavior8–10. Complete surgical resection is the treatment of choice for SFTP.11 Pedunculate tumors can be safely treated by wedge resection. For these pedunculate tumors, video-assisted thoracoscopic surgery (VATS) provides a powerful and useful approach. In addition, some authors have reported the assistance of thoracoscopy to obtain a more precise view of the resection margins, even in parietal tumors.12,13 Although a small thoracotomy can be added to the VATS for the removal of large tumors, VATS plays an important role in reducing the size of the thoracotomy incision, which results in less invasive surgery.14 Initially, we selected a thoracoscopic approach for the resection of SFTPs in all cases. Even with large tumors, if a VATS resection is technically feasible, it can be carried out. If necessary, we add a small thoracotomy incision to remove the tumor safely and completely, with free surgical margins. The aim of this study was to present our experience of VATS for 21 consecutive cases with SFTPs and to discuss the treatment of choice of such neoplasms.

PATIENTS AND METHODS

Between June 2000 and September 2008, 21 patients with SFTPs (9 men and 12 women) underwent VATS at our department. The mean age was 52.5 years (range, 33–76 years). Chest pain (in one patient), dyspnea (in five patients), cough (in six patients), and hemoptysis (in one patient) were the symptoms. Fifteen patients were symptom-free. On physical examination, dullness to percussion and absence of...
Surgical excision was performed in all patients. Surgical excision was performed by VATS in 15 patients (71.4%), by VATS plus a small thoracotomy (<5 cm) in 4 patients (19.0%), and by posterolateral thoracotomy accompanied by VATS in 2 patients (9.5%). Mean chest drain duration was 2.3 days (range, 1–4 days), and the mean hospital stay was 7.2 days (range, 4–15 days). There was no operative morbidity or mortality. At surgery, 14 tumors (66.7%) arising from the visceral pleura were pedunculated and 7 tumors (33.3%) from the parietal pleura presented with a broad base of attachment. All tumors were resected with free surgical margins. The largest mass excised in our patients was 18.5 cm \( \times \) 17 cm \( \times \) 13.5 cm. The smallest one was 0.5 cm \( \times \) 0.6 cm \( \times \) 4.0 cm. The median size of the tumors resected was 6.5 cm (range, 4.0–18.5 cm). Pathologic margins of all cases were confirmed by frozen sectioning free of cancer. On sectioning, the tumors showed a whorled, fleshy appearance, sometimes alternating with some myxoid areas. Focal necrosis and hemorrhagic zones were found in three cases (14.3%), all relatively large in diameter (>5 cm).

Microscopically, all the tumors consisted of spindle-shaped cells. The tumor cells and intercellular collagen fibers proliferated without obvious direction or structure, which corresponded to the so-called “patternless pattern” (Figure 1A). There were 18 pathologically benign SFTP cases (85.7%) and 3 malignant SFTP cases (14.3%) who were focally characterized by a mitotic count in excess of 4 mitoses per 10 high-power fields and by cellular pleomorphism. Chest CT scan of all three histologically malignant SFTP showed the supply of blood vessels within the tumor and/or liquefaction necrosis (Figure 1D). Immunohistologic analysis revealed positivity for CD34 and vimentin in all tumors (Figures 1B, C) and no expression of cytokeratin, desmin, S-100, Ki-67, or actin in any of the tumors. No adjuvant therapy was used. All patients were included in a follow-up program that included clinical examination and chest roentgenogram after 1, 3, and 6 months postoperatively, and annually thereafter. The median follow-up time was 43 months (range, 3–96 months). All of the patients have remained well, with no recurrence or metastasis of the tumors.

DISCUSSION

SFTPs are uncommon, representing less than 5% of all neoplasms involving the pleura. Diagnostic tools, such as chest radiography, CT scanning, and magnetic resonance imaging, are helpful but not decisive in establishing the diagnosis. In most cases, thoracic CT scan shows a well-circumscribed round tumor with a homogenous density. However, these findings lack specificity and other imaging findings are possible. Thus, CT scan cannot differentiate benign from malignant SFTPs. Difficulties in differentiating these tumors from others originating from mediastinum or chest wall are possible. Furthermore, if the lesion is not homogeneous, the differential diagnosis with a bronchogenic carcinoma may be also more difficult, especially in the presence of a smoking history (as in 57.1% of our patients). Even fine-needle aspiration is often inconclusive, in fact, only four of nine cases (45%) were identified by CT guided aspiration biopsy. Only surgical excision, with subsequent immunohistologic examination, can be diagnostic. Immunohistochemically positive CD34 staining is decisive in establishing the diagnosis differentiating SFTPs from mesotheliomas, synovial sarcomas, fibrosarcomas, and neurogenic tumors. In addition, the bel-2 essay can confirm the diagnosis of SFTPs in case of CD34 negativity. In our experience, malignant forms of SFTPs accounted for 14.3% of all cases, whereas it was observed 7% in the experience of Cardillo et al.\(^9\) 30% in the experience of de Perrot et al.\(^8\) 36% in the experience of England et al.,\(^21\) 38% according to Rana et al.,\(^9\) and 60% in the experience of Suter et al.\(^22\) This variability could be probably justified either by the heterogeneity in studied populations or by relative subjectivity in the recognition of some pathology criteria, especially hypercellularity and pleomorphism.

Complete surgical excision with underlying tissue removal lends itself to complete cure and minimizes tumor...
recurrence. For the pedunculate tumors, therefore, VATS is a useful approach. Even when it is necessary to perform a small thoracotomy in addition to VATS for the removal of a large tumor, VATS may play an important role in reducing the size of the thoracotomy incision, which results in less invasive surgery. To minimize postoperative morbidity, VATS may be the most promising surgical approach for the resection of SFTPs. Six of 60 patients (10.0%) and 22 of 63 patients (34.9%) with SFTPs successfully underwent VATS in the experience of Magdeleinat et al.\textsuperscript{10} and Sung et al.,\textsuperscript{23} respectively. Takahama et al.\textsuperscript{14} did the first study to evaluate the advantage of VATS for patients with SFTPs. In that study, surgical excision was performed with VATS only in nine patients, with VATS plus a small thoracotomy in three patients, and by a posterolateral thoracotomy without VATS in one patient. The mean chest-drain duration was 1.3 days (range, 1–3 days), and the mean duration of hospital stay was 8.6 days (range, 3–30 days). With a median of 42-month follow-up (range, 6–120 months), all patients have remained well with no recurrence or metastasis. Nomori et al.\textsuperscript{12} reported that contact metastasis and local recurrence had occurred at the port site, which was used during the VATS procedure. To avoid contact metastasis and local recurrence at the port site, we extract the surgical specimen by means of a retrieval bag. In our series of 21 cases, no recurrence or metastasis of SFTPs developed during postoperative median follow-up period of 43 months. However, three cases showed a mitotic count in excess of 4 mitoses per 10 high-power fields or cellular pleomorphism, all of which are considered to indicate potentially malignant SFTPs.

Because of the rarity of these tumors, there is no systematic assessment of the role of adjuvant therapy in SFTPs.\textsuperscript{2,11,24} Anecdotal reports describe long-term survivals with postoperative radiotherapy in patients with incomplete resection of the tumor. Responses to ifosfamide and doxorubicin have been reported for recurrent, inoperable SFTP. Nevertheless, recurrent benign or malignant tumors should be strongly considered first for repeat surgical resection. After resection, adjuvant therapy should be considered for recurrent tumors, particularly the sessile, malignant variety, although little experience is described in the literature with postoperative treatment.\textsuperscript{2}

In conclusion, complete resection and close follow-up for years after operation is recommended for SFTPs. VATS may play an important role in reducing the size of the thoracotomy incision in the treatment of SFTPs, which results
in less invasive surgery. Further studies are required to identify reliable prognostic factors and multicenter trials to evaluate the effectiveness of preoperative or postoperative systemic therapy.

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REFERENCES