

Solitary Fibrous Tumor of the Pleura Presenting Dry Cough Induced by Postural Position

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Solitary fibrous tumor of the pleura is one of the uncommon diseases. About 40% of solitary fibrous tumors of the pleura are of a visceral and peduncled type. Several visceral pleural tumors have motility. A proof of motility of the intrathoracic tumor is useful for preoperative diagnosis. We report a resected case of solitary fibrous tumor of the pleura presenting dry cough induced by postural position. (Ann Thorac Cardiovasc Surg 2009; 15: 401–403)

Key words: solitary fibrous tumor, visceral pleura

Introduction

The solitary fibrous tumor of the pleura (SFTP) is one of the rare diseases. The exact origin of SFTPs is still unknown; however, immunohistological studies have revealed its association with positive staining for the CD34 antibody and bcl-2 oncoprotein.^{1,2)} CD34 and bcl-2 positive staining is found in stromal tumors with exception of mesothelioma, suggesting that SFTP is a tumor arising not from the mesothelial layer, but from the submesothelial mesenchymal layer.¹⁾ In the earlier years, this uncommon tumor was known by various names, such as localized pleural mesothelioma, pleural fibroma, localized fibrous mesothelioma, submesothelial fibroma, and localized fibrous tumor.^{1–3)} Recently, the SFTP was defined as an uncommon spindle-cell mesenchymal tumor of probable fibroblastic derivation that often presents a prominent hemangiopericytoma-like vascular pattern, but it may exhibit other histological patterns.⁴⁾ The SFTP is grossly a firm, encapsulated, occasionally lobular mass with a characteristic whorled appearance.³⁾ SFTPs should be

treated because they have malignant potential. The most recommended treatment is complete surgical resection. We report a resected case of the SFTP presenting dry cough induced by postural change.

Case Report

A 66-year-old woman who complained of a dry cough induced by postural position was admitted to a nearby hospital where the radiographic findings revealed an abnormal mass shadow of the chest. She was admitted to our hospital for further examinations. Her medical history included sarcoidosis, large thyroid adenomatous goiter, and meningioma of the cerebellopontine angle. Her vital signs were within normal limits. A dry cough was induced when she lifted herself up from a lying position, or when she assumed a lying position from a sitting-up position. A plain chest radiograph showed a mass shadow on the left chest wall (Fig. 1). The mass shadow became indistinct at the right decubitus chest radiograph (Fig. 2). Contrast-enhanced CT images revealed a heterogeneous mass shadow with a clear defined border and a slightly enhanced region (Fig. 3). Magnetic resonance (MR) images revealed that the tumor was high intensity under T2 weighted conditions, which was further enhanced under gadolinium-contrasted T1 conditions. ¹⁸F-fluoro-2-deoxy-D-glucose positron emission tomography (FDG-PET) revealed that the tumor was not cumulated, and there were no abnormal accumulations. Ultrasonography showed the tumor as a heteroechoic mass (i.e., most of it

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Fig. 1. The anterior-posterior chest radiograph. The mass shadow is observed on the left chest wall.



Fig. 2. The chest radiograph in the right lower decubitus position. The mass shadow on the left lower lung field at Fig. 1 is indistinct, and the left chest wall is observed clearly.

was low echoic) and motile with respiration and following repositioning. We performed a complete surgical resection via the video-assisted thoracic surgery (VATS) approach. The gross findings showed no connection between the mass and the chest wall, with a pedicle from the visceral pleura, a form similar to ganodermataceae (*Ganoderma lucidum*), and many feeding vessels from it (Fig. 4A). Although we tried to resect the tumor using the VATS approach, its size and solidity required a small thoracotomy of at least 10 cm. The tumor was resected with adequate margins, using a simultaneous stapler/cutter, and was removed smoothly from the minithoracotomy. The resected specimen measured 85 × 80 × 35 mm and weighed 112 g. Immunopathological findings revealed that the short spindle cells were patternless (so-called patternless pattern, Fig. 4B) without necrosis and without severe nuclear atypia, and they were rarely mitotic (less than 4 mitoses per high-powered field); they stained positive for CD34 and bcl-2 (Figs. 4C and 4D), and negative for s-100 protein, α-SMA, AE1/AE3, and calretinin. The final diagnosis was a benign SFTP. There were no residuals at the edge of the resection either macro- or microscopically. Our patient was discharged soon after the operation and was well at the 6-month follow-up.

Discussion

Two-thirds of SFTPs arise from the visceral pleura; the remaining third arises from the parietal pleura. About 60% of solitary fibrous tumors of the visceral pleura are



Fig. 3. The contrast-enhanced chest CT image indicates the mass shadow of the left hemithorax with the extrapleural sign.

of the pedicled type, and about 40% the sessile type. As for solitary fibrous tumors of the parietal pleura, one-third is the pedicled type and two-thirds the sessile type.³ SFTPs have malignant potential. Some papers reported that 20%–30% of resected SFTP had malignant components, and the size of the tumor was associated with malignancy.^{2,3,5–7} The average size of SFTPs is about 6 to 8 cm, ranging from 1 cm to the size of the hemithorax. SFTPs of more than 10 cm in diameter are quite likely to have malignant components, but all SFTPs should be treated as soon as possible, with complete resection being the recommended treatment.

In our case, the physical finding of dry cough induced by postural position may imply that the visceral pleura is extended by the transposition of a pedicled pleural tumor.

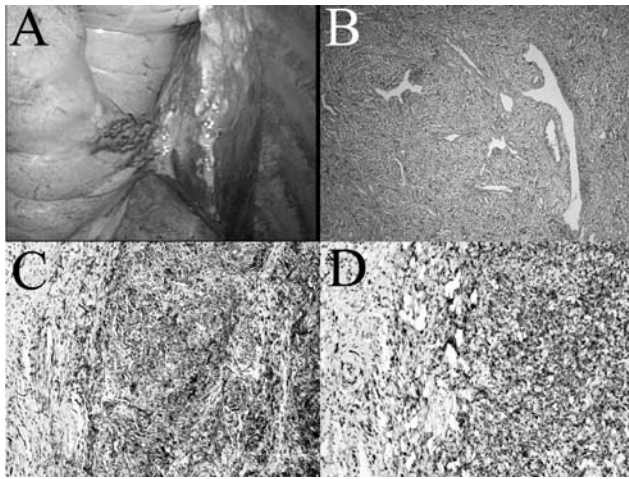


Fig. 4.

- A:** Gross findings of the tumor. A richly vascularized pedicle connected the tumor to the pleura of the left lower lobe.
- B:** Microscopic view of the central tumor showing a patternless arrangement of cells.
- C:** Positive staining for CD34 in tumor cells.
- D:** Positive staining for bcl-2 in tumor cells.

Extension of the visceral pleura induces a cough as a reflex (Hering Breuer's reflex). However, this on its own does not confirm that the pleural stimulation is due to a visceral pleural tumor or adhesion of a parietal pleural tumor if the cause of the dry cough is only a pleural extension. The plain chest radiograph revealed a mass shadow like a tumor at the chest wall (Fig. 1), and the contrast-enhanced chest CT showed a heterogenous mass shadow with the extrapleural sign (Fig. 3). On the other hand, the chest radiograph in the right decubitus position (Fig. 2) revealed disappearance of the tangent between the mass shadow and the chest wall, leading us to surmise that we considered there was no connection between the mass and the chest wall. These radiographic findings were reported by some papers.^{8,9)}

Although the minimally invasive approach has recently been recommended for SFTPs,¹⁰⁾ thoracic surgeons should not hesitate to convert a VATS procedure to a thoracotomy and should therefore decide on the thoracotomy approach before starting the VATS. This is because we must avoid tissue residuals, dissemination of the SFTP through destruction of or damage to the tumor as it is forcibly removed through a narrow gap such as the VATS port, and implantation via the VATS port. Malignant or benign SFTPs, however, can recur or metastasize after complete resection. A benign SFTP has an excellent prognosis, though malignant SFTPs have a higher tumor-related

mortality. Most patients with recurrence die of the tumor within 2 years. The SFTP patient should be followed over a long term because Yokoi and colleagues have reported that recurrence or metastasis can occur after more than 10 years following complete resection in benign SFTP.¹¹⁾

Conclusion

We reported a resected case of SFTP, which is a rare disease with malignant potential. The motility of intrathoracic tumor can be proven by chest radiograph with different plural positions. Complete resection is required for the radical treatment, but SFTP can recur or metastasize long after complete resection. Despite an SFTP being benign, a minimally invasive approach is apt to be difficult.

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