

Case Report

A rare case of solitary fibrous tumor of the pleura: An unusual presentation

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ABSTRACT

A 69-year-old male presented with rapidly worsening symptoms, including breathlessness, productive cough, and weight loss. Physical examination revealed a deviated trachea and absent breath sounds on auscultation of the left side of the chest. Radiographic imaging revealed a large pleural mass associated with a significant pleural effusion, subtotal lung collapse, and mediastinal shift. Pulmonary function tests showed severe impairment, with a 39% forced expiratory volume in 1 second (FEV1) and 62% diffusing capacity of the lungs for carbon monoxide (DLCO). A positron emission tomography-computed tomography (PET-CT) scan showed mild avidity in the lesion. Definitive management involved a staged approach with initial decompression followed by surgical resection via a left double-space open thoracotomy. Intraoperatively, the tumor was found to occupy three-quarters of the chest cavity. The entire lung was adherent to the chest wall. Histological analysis showed pattern less architecture with high vascularity, hypercellularity, necrosis, elongated nuclei, pale cytoplasm, and high mitotic activity. Immunohistochemical staining was positive for CD34, BCL2, CD99, Ki-67, and STAT6, leading to a diagnosis of solitary fibrous tumor (SFT). The malignant potential could not be determined. The patient was monitored every six months for five years, with an estimated recurrence risk of 20%.

Keywords: Case report, Rare tumor, SFTP, Solitary fibrous tumor of the pleura, Thoracic surgery

INTRODUCTION

Solitary fibrous tumors of the pleura (SFTP) are rare mesenchymal neoplasms with a poorly recognized etiology and highly variable presentations.

The most recognized signs and symptoms reported in the literature are chest pain, cough, and dyspnea.^[1] However, presentation with cardiac compression and subsequent hemodynamic instability due to mass effect is a rare occurrence. To date, only one such case has been reported in the literature.^[2]

This case report aims to describe an unusual presentation of SFTP, where the tumor mass effect resulted in significant lung collapse, mediastinal shift, and a deviated trachea, a presentation not previously reported in the literature.

CASE REPORT

A 69-year-old male presented with rapidly worsening symptoms, including breathlessness, productive cough, and weight loss. These symptoms significantly impacted

the patient's daily activities. His medical history included idiopathic thrombocytopenic purpura (ITP), which had been managed conservatively for the past 15 years. He had no history of smoking or exposure to asbestos.

Initial physical examination revealed a deviated trachea, absent breath sounds on the left side of the chest, and signs of respiratory distress. Pulmonary function tests revealed severe impairment, with an FEV1 of 39% predicted and DLCO of 62% predicted, indicating significant respiratory compromise.

Investigation

Radiographic imaging revealed a giant pleural mass with a large pleural effusion, subtotal left lung collapse, and mediastinal shift. Figure 1 shows the initial radiograph obtained at presentation.

A subsequent CT scan confirmed a large lesion with pleural effusion occupying most of the left pleural cavity, causing subtotal left lung collapse and mass effect on the heart [Figures 2 and 3].

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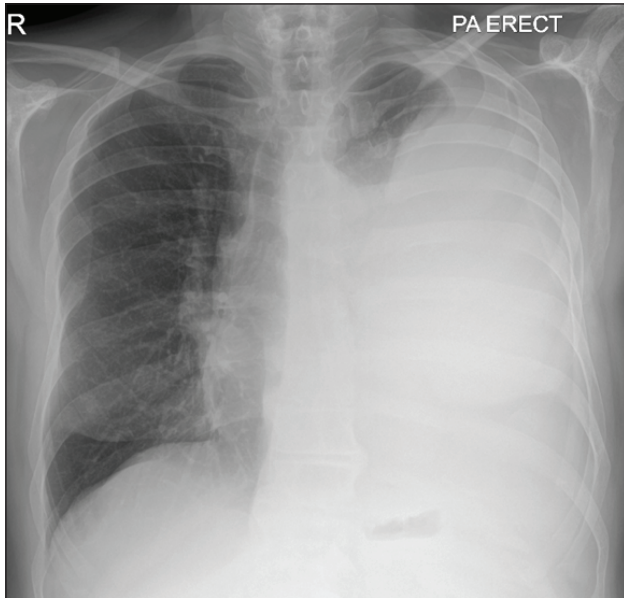


Figure 1: Radiographic imaging of a lung mass on presentation.

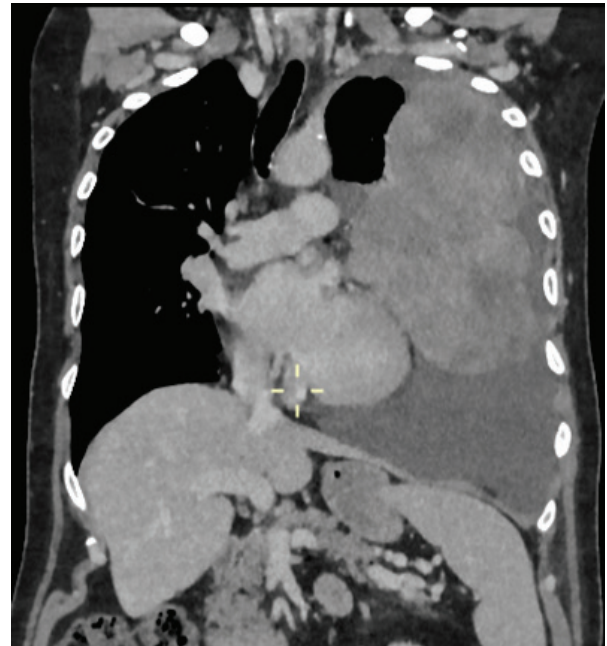


Figure 3: Coronal cross-section CT scan of massive left-sided SFTP and pleural effusion. CT: Computed tomography, SFTP: Solitary fibrous tumours of the pleura.



Figure 2: Transverse cross-section CT scan of massive left-sided SFTP. CT: Computed tomography, SFTP: Solitary fibrous tumours of the pleura.

The PET-CT showed mild avidity of the lesion, suggesting local disease activity but no distant metastasis [Figure 4].

A CT-guided biopsy was performed to aid diagnosis, and histology showed spindle cells with minimal cytological atypia, suggesting a solitary fibrous tumor.

The pleural fluid aspirate was blood-stained; however, cytology revealed no malignant cells. Cytology of the pleural fluid aspirate was negative for malignant cells.

Management

Due to the patient's poor functional status, resulting from complete left lung collapse and mediastinal shift with associated hemodynamic compromise, major surgical

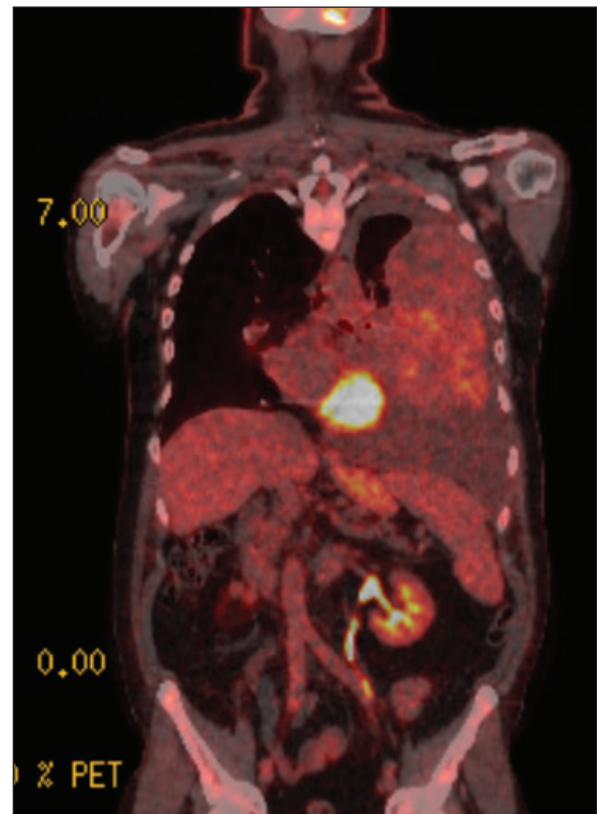


Figure 4: Coronal cross-section PET-CT scan of left-sided SFTP with mild avidity. PET-CT: Positron emission tomography and computed tomography, SFTP: Scan and solitary fibrous tumours of the pleura.

resection was not initially feasible. As a result, a staged management approach was adopted.

First Stage: Decompression and Inspection

The patient underwent pleural decompression with an evacuation of 1000 ml of pleural effusion. This allowed for gross tumor inspection, which revealed a highly vascular mass attached to the upper lobe, the anterior mediastinum, and the parietal pleura. The pleura appeared thickened but not overtly malignant.

Pleural biopsies confirmed the gross findings. An indwelling catheter was placed to prevent further fluid accumulation and allow the lung to re-expand. The postoperative X-ray confirmed the re-expansion of the lung [Figure 5].

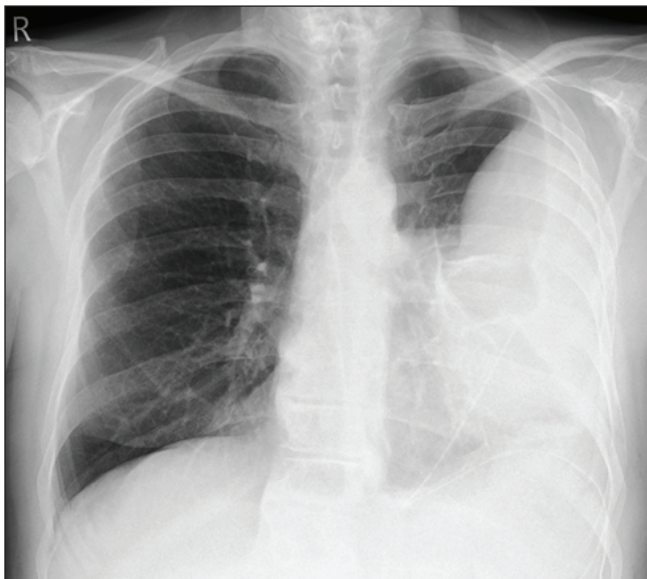


Figure 5: Post-operative X-ray following thoracoscopic evacuation of pleural effusion and insertion of indwelling pleural catheter.

Second Stage: Surgical Resection

After 10 days, the patient showed significant clinical improvement and underwent second-stage resection. Due to the tumor's massive size and its multiple pedicles of vascular supply, access to the chest cavity was challenging. The tumor's massive size and multiple vascular pedicles made access to the chest cavity technically challenging.

- Lateral parietal pleura
- Thick pedicle from an internal thoracic arterial branch at the superior mediastinum
- Thin pedicle next to the left phrenic nerve
- Thin pedicle from the aortopulmonary window
- Pericardial fat near the left ventricle of the heart
- Anterior segment and lingual of the upper lobe

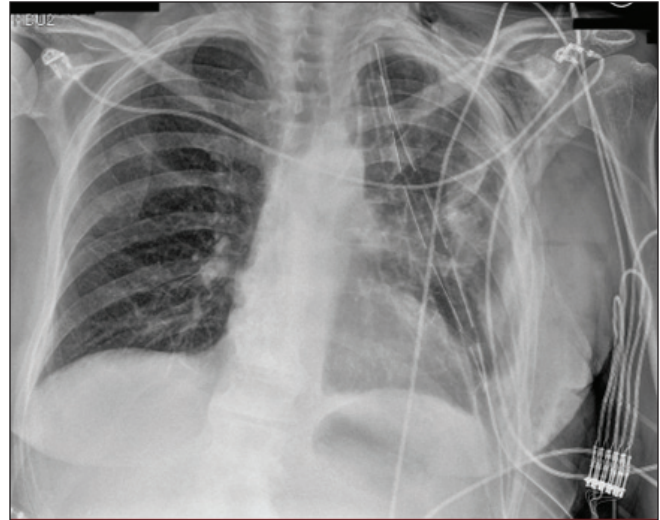


Figure 6: Chest X-ray post-resection of tumor mass.

A double-space thoracotomy was performed through the 4th and 7th intercostal spaces, providing adequate exposure for tumor resection. The tumor was dissected from the chest wall using extra-pleural dissection and from the lung via wedge resection of the upper lobe. All mediastinal pedicles were ligated, and the tumor was completely excised with adequate macroscopic margins (Postoperative radiograph is shown in Figure 6).

Macroscopy, Microscopy and Immunohistopathology

Macroscopically, the tumor was irregular, lobulated, and measured 21×14×8 cm. The cut section revealed a mixed solid hemorrhagic tumor with focal areas of necrosis. Microscopy revealed a spindle cell tumour with patternless architecture, high vascularity, hypercellularity, necrosis, elongated nuclei, pale cytoplasm, and mitotic activity of 2-3 mitotic figures per 10-high-power-fields. Immunohistochemistry stained positive for CD34, BCL2, CD99, Ki-67, and STAT6.

A diagnosis of SFTP was confirmed; however, malignant potential could not be determined.

Recovery and Follow-up

The patient had an uncomplicated recovery following the procedure and was discharged on postoperative day 7. His clinical condition improved rapidly. Within three months, the patient was able to resume day to day activities without pain or respiratory compromise.

Given the high recurrence rate of SFTP, the patient was scheduled for follow-up every six months for five years, with repeat CT scans. At the six-month follow-up, the CT scan

revealed mild pleural thickening and some fluid anteriorly in the upper left hemithorax, but no signs of recurrence were detected [Figures 7-11].

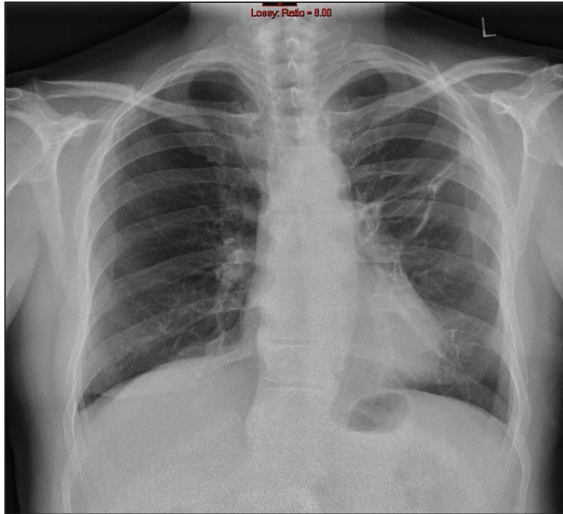


Figure 7: Chest X-ray at 6-month follow-up.



Figure 8: Transverse cross-section CT scan at the 6-month follow-up. CT: Computed tomography.



Figure 9: Transverse cross-section CT scan at the 6-month follow-up. CT: Computed tomography.



Figure 10: Transverse cross-section CT scan at the 6-month follow-up. CT: Computed tomography.

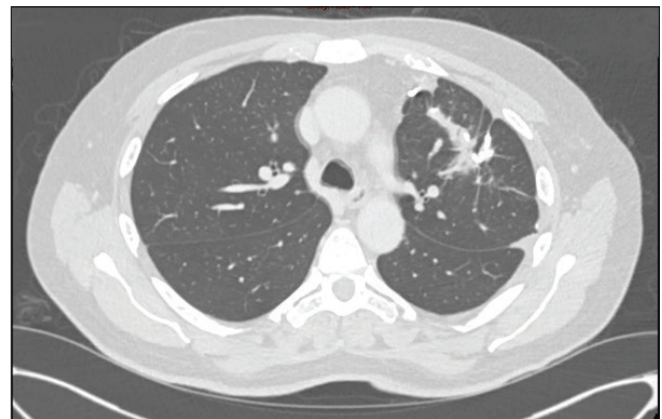


Figure 11: Transverse cross-section CT scan at the 6-month follow-up. CT: Computed tomography.

DISCUSSION

This case is unique in that it presents a SFTP causing bilateral lung collapse and tracheal deviation due to mass effect, which has not been reported in the literature to date. The only other case with a similar mass effect described an SFTP originating from the diaphragm, leading to compression of the pericardium and other mediastinal structures.^[2]

The size of the tumor is often correlated with its malignant potential, with larger tumors generally considered at higher risk of malignancy^[1]. In our case, despite the large tumor size and associated mass effect, neither the gross appearance nor fluid cytology suggested malignancy. Histopathology and immunohistochemistry were also inconclusive regarding malignant potential.

Complete resection of SFTPs is considered the best management strategy, with an excellent prognosis for benign tumour variants. Surgical resection has also been shown to prolong survival in malignant cases^[3,4]. In this case, due to the massive size of the tumor, a two-stage surgical approach

was required. Video-assisted thoracoscopic surgery (VATS) has been successfully used for tumors of up to 15 cm in diameter^[3], but in this case, the tumor's size necessitated an open surgical approach.

CONCLUSION

SFTP is a rare and usually benign tumor, but it can present in ways that are potentially life-threatening if not diagnosed early. While tumor size is commonly used as a predictor of malignancy, clinical and radiological features alone are unreliable for distinguishing between benign and malignant variants. In cases of advanced disease with significant mass effect, open surgical resection may be required, as opposed to less invasive techniques such as VATS. Given the potential for recurrence, long-term follow-up is crucial for monitoring tumor regrowth.

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