

CASE REPORT

Complete Surgical Excision of A Challenging Giant Intrathoracic Mass - Solitary Fibrous Tumor

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ABSTRACT

Background: Solitary fibrous tumors are rare mesenchymal tumors that remain asymptomatic and are diagnosed incidentally. They grow at a slow rate and expand to giant size by the time of their presentation.

Introduction: This is a case of a female who presented with exertional dyspnea, orthopnea and dull chest pain for 4-5 years. Clinical examination showed reduced movement over the left chest along with dull percussion note and absent breath sounds. Computed tomography showed a large 20*11*12cm heterogeneous opacity occupying the left hemi thorax. Biopsy was inconclusive. A complete excision of the mass was achieved. Intraoperatively, mass was consistent with radiological findings, however, was adherent to surrounding structure with atelectasis. Patient had an uneventful recovery period with histopathological evidence of solitary fibrous tumor.

Conclusion: Complete surgical excision remains the mainstay of treatment for solitary fibrous tumors and can achieve favorable outcomes.

Keywords: Dyspnea, intrathoracic mass, solitary fibrous tumor

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INTRODUCTION

Solitary fibrous tumors (SFT) are rare mesenchymal tumors accounting for less than 5% of all pleural tumors¹. It can develop in any body cavity with intrathoracic being the most common site^{2,3}. Majority of the thoracic SFT are usually benign but they exhibit aggressive behaviour in terms of local invasion and recurrence as reported in 12 % of all cases^{4,5}. SFT are often asymptomatic or have nonspecific symptoms like dyspnea, cough, and chest pain because of which they remain undiagnosed and are incidentally found on chest imaging⁶. Here, we present a rare case of large intrathoracic mass presenting as pleural effusion.

Case Presentation:

An elderly female, known hypertensive, presented with exertional dyspnea, orthopnea, and dull chest pain for

the last 4 to 5 years. Her symptoms worsened over a year. She was initially managed by a medical team with needle thoracocentesis followed by a pigtail catheter placement as a case of loculated pleural effusion. On examination, reduced movement over the left chest was observed along with dull percussion note and absent breath sounds. A chest radiograph revealed a homogenous opacity in the middle and lower zone along with blunting of both angles and obscuration of cardiac silhouette (figure 1).

A computed tomography was done which was suggestive of a 20*11*12cm size heterogeneous opacity occupying the left hemi thorax. Additionally, the mass was abutting trachea, left main bronchus and a collapsed left lower lobe was seen along with a mediastinal shift towards the right side. Multiple radio-guided biopsies were inconclusive, likely due to necrosis or inadequate tissue sampling. It is worth mentioning, that surgery was already suggested by another institute which was refused by the patient. Eventually, because of the large tumor size and inadequate diagnosis, patient was referred to our institution for surgical intervention. Preoperative embolization was not performed as the tumor had multiple tortuous feeding vessels arising from both the intercostal arteries and pulmonary circulation, making selective embolization technically

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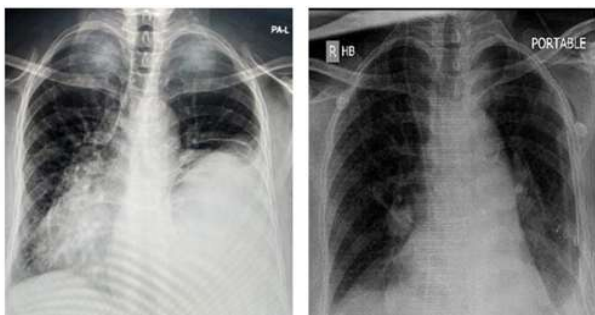


Figure 1: Preoperative Chest Radiograph Showing a Large Homogenous Opacity, Postoperative Chest Radiograph Demonstrating Complete Expansion of Lung



Figure 2: Intraoperative Image Showing a Giant Mass with Dilated Torturous Veins

difficult with uncertain benefit. In addition, the patient was hemodynamically stable, and complete surgical excision was considered achievable without prior embolization.

After multidisciplinary team meeting, general consensus was made among oncology, radiology, and thoracic surgery regarding complete excision of the tumor. Preoperative preparation was done including medical optimization of hypertension, breathing exercises, and obtaining high risk consent.

A standard left posterolateral thoracotomy was done with the excision of the 7th rib after advancing 32 FR left sided double lumen tube. Most importantly, graded posterolateral position was made to avoid sudden mediastinal shift to prevent hypotension. Intraoperatively, a tanned white encapsulated, lobulated, and highly vascular tumor was identified receiving dilated torturous supply from the upper lobe, chest wall, and intercostal arteries (aorta). Mass was adherent to diaphragm with neovascularization and a consolidated lower lobe due to prolonged compression was seen (figure 2).

A complete excision of the mass was achieved, resulting in expansion of lower lobe. Operative time was approximately 2.5 hours, with an estimated blood loss of 1200-1500ml requiring transfusion of 2 units of red blood cells intraoperatively.

Complete expansion of lung was noticed postoperatively. Patient had an uneventful recovery and was discharged later on 7th postoperative day. Histopathology confirmed the diagnosis of solitary fibrous tumor with immunohistochemistry positive for Cd34, Stat 6, BCL2, and CD99. During 6th months follow up, patient remained asymptomatic with no evidence of recurrence on clinical and radiological examination.

DISCUSSION

Solitary fibrous tumors are rare tumors that originate from mesenchymal cells within sub mesothelial tissue of pleura, distinguishing them from mesothelioma, which derives from mesothelial cells⁴. SFTs typically occurs in 4th to 6th decade of life and affects men and women equally¹. Diagnosis and management of SFT are challenging as they are rare and are often discovered incidentally as small asymptomatic tumors but can grow to massive sizes³. The definitive diagnosis will, however, be made histopathologically⁷.

Benign SFTs generally develop from the visceral pleura, are pedunculated, and extend into the pleural space, while malignant tumors more often originate from the parietal or diaphragmatic pleura and can invade the lung. The presence of symptoms like pleural effusion, and lack of pedicle are key indicators of potential malignancy⁵.

Computed tomography usually reveals well defined lesions, but these findings are not definitive of diagnosis. CD34 and NAB2-STAT6 is molecular hallmark found in most of SFT, hence immunohistochemistry can be used for diagnostic purpose^{2,8}. Rarely, SFTs is linked to paraneoplastic syndromes such as digital clubbing, and hypertrophic pulmonary osteoarthropathy (HPO), possibly due to tumor secreted cytokines, hyaluronic acid, or chronic hypoxia. Doege-Potter syndrome, characterized by refractory hypoglycemia, occurs in less than 5% of SFT, is associated with insulin like growth factor 2 secretion⁹.

Radical surgical resection with wide clear margins is currently the cornerstone of treatment¹⁰. Depending on the vascularity of the tumor, surgical removal can be complemented by the endovascular embolization to prevent blood loss⁵. In some cases, minimal invasive techniques may be considered according to dimension,

position, and infiltration of adjacent structures, although there is risk of tumor seeding at the incision site⁹. Long term outcomes are generally favourable, but there is always a risk of local or distant recurrence⁵. Role of neoadjuvant/adjuvant chemo radiotherapy is still controversial in irresectable cases⁷.

CONCLUSION

Fibrous tumors are often asymptomatic and may grow to be giant in size by the time of diagnosis. The gold standard treatment of thoracic solitary fibrous tumor is complete surgical resection. Due to rarity of tumor, longer follow up is required.

Patient Consent: Informed and written consent was acquired from the patient before the initiation of write-up of case report.

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Authors' Contribution: SN conducted the literature search, prepared the original draft, and contributed to manuscript writing. TA conceptualized the study and provided financial guidance. MM supervised the study process and contributed to review and editing of the manuscript. NS contributed to the literature search and manuscript editing.

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