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Case Report

# Primary Pulmonary Synovial Sarcoma: A Rare Lung Tumor

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## Abstract

Pulmonary synovial sarcoma represents a rare form of lung malignancy when juxtaposed with others. The diagnosis of primary pulmonary sarcoma is usually made after ruling out the common primary lung malignancies and metastatic sarcomas. Metastatic synovial sarcoma of the lung occurs more frequently than primary pulmonary synovial sarcoma. The usual presentation will be cough, chest pain, breathlessness and radiologically a mass in the lung. Histopathological examination and immunohistochemical staining of the biopsy sample are needed to confirm the diagnosis; if possible genetic study should also be done. Since the tumor is very rare, we describe the case of a 66-year-old woman presenting with a large right lung mass and ipsilateral pleural effusion, diagnosed as pulmonary synovial sarcoma via ultrasound-guided lung biopsy.

**Keywords:** Pulmonary synovial sarcoma, TLE1, Surgical excision.

## Introduction

Pulmonary synovial sarcoma is a rare lung tumor, making up about 0.5% of all primary lung cancers.[1] It is an aggressive tumor originating from mesenchymal tissues. It commonly affects the periarticular sites but very rarely involves the lung.[2] It is more common in males.[2] "The non-specific clinical and radiological features of this tumor make its diagnosis challenging. Therefore, histopathological examination along with immunohistochemistry is essential for definitive diagnosis. TLE1 is a specific and sensitive IHC marker for synovial sarcoma, which aids in differentiating it from its histologic mimics. To establish the diagnosis, other common primary lung tumors must be excluded. As of yet, only a limited number of cases have been documented in the literature.

## Case Report

A 66-year-old female who is a daily wager, presented with breathlessness and chest pain for 3 months. Breathlessness was progressive, grade IV at the time of admission, and was associated with orthopnea. She had a myocardial infarction 10 years ago and started on antiplatelet drugs. No other significant past history was there. No history of any substance addiction was disclosed. Routine blood investigations were normal. ECG was done, showed old ischemic changes (Q waves seen in II, III, and aVF), and Trop T was negative. A chest X-ray was done, which showed homogenous opacity in the right middle and lower zones with some lucency in the lower zone, and the costophrenic angle was obliterated (Figure 1A). The upper border of the lesion was lobulated, suggesting suspicion of a mass lesion and loculated effusion. The CECT chest showed a right-sided well-defined heterogeneous mass lesion occupying 2/3rd of the hemithorax and abutting the mediastinal structures (Figure 1B). There was also a collection in the right pleural space, which was aspirated under USG guidance and sent for analysis. The analysis of the pleural fluid revealed an exudative lymphocytic effusion, characterized by low ADA levels (22 U/L) and the absence of malignant cells. Whole-body CT was done to rule out metastasis in other sites. USG guided lung FNAC, and a biopsy was done. Lung FNAC was inconclusive, and the biopsy specimen revealed a biphasic tumor with predominantly monomorphic spindle cells arranged in fascicles and a palisading pattern (Figure 1C). The spindle cells were seen

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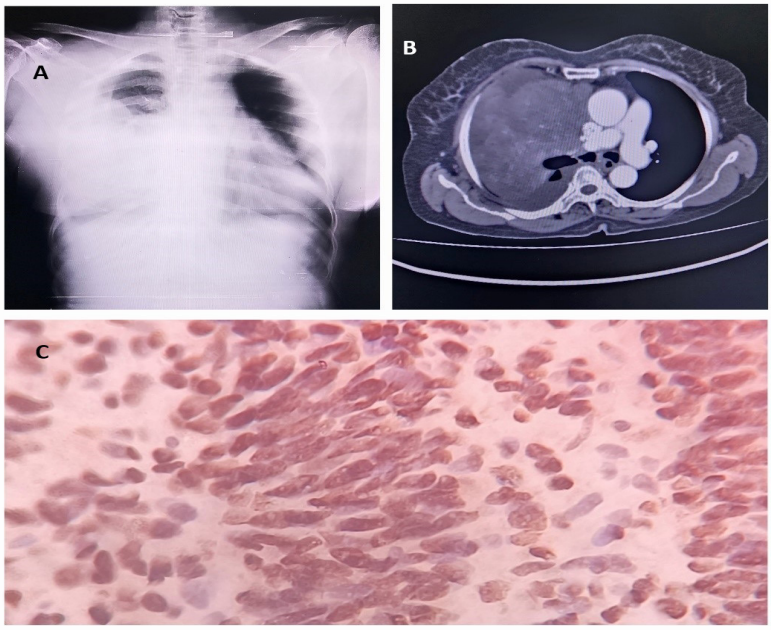


**Figure 1:** (A) - Chest X-ray shows Homogeneous opacity in the right middle and lower zones with focal lucency inferiorly and obliteration of the costophrenic angle. (B) - CECT chest shows a right-sided heterogeneous mass lesion occupying 2/3rd of the hemithorax and abutting the mediastinal structures with pleural effusion. (C) - HPE shows a biphasic tumor with predominantly monomorphic spindle cells arranged in fascicles and a palisading pattern on TLE1 staining

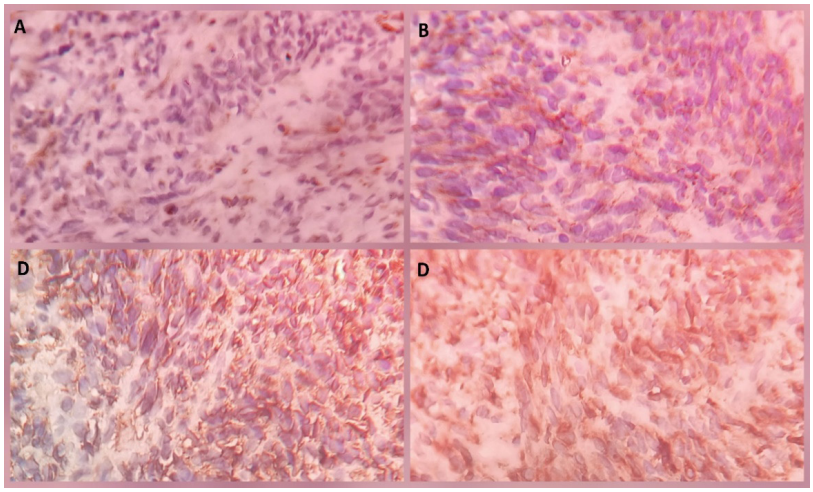


**Figure 2:** Immunohistochemical staining of lung mass biopsy showing negative staining for Desmin (A), positive for S100 (B), Cd99 (C) and Bcl2 (D)

with hyperchromatic nuclei and scanty cytoplasm (stubby nuclei). For further confirmation, immunohistochemical staining was done. TLE1, CD99, Bcl2 and S100 were positive (Figure 2A). Desmin (Figure 2B-D) and SMA were negative. There was no evidence of metastasis in any other sites. The final impression was primarily biphasic synovial sarcoma. Since the treatment of choice for this condition is surgery followed by chemotherapy, the patient was referred to cardio thoracic surgeon for surgical intervention.

## Discussion

Synovial sarcoma is a misnomer, as the tumor originates from mesenchymal cells rather than the synovium. However, the tumor histologically resembles the synovial tissue, it was named synovial sarcoma. It is more common in males. Breathlessness, cough, and chest pain are the commonest symptoms.[3] Usually presents as a central tumor and is rare to present as a peripheral lesion. On radiological examination, the mass lesion appears well-defined and heterogeneous, featuring necrotic areas and an accompanying pleural effusion. Lymphadenopathy is usually rare in this tumor, which is evident in our case.[4]

Synovial sarcoma comprises four histological variants: monophasic fibrous, monophasic epithelial, biphasic, and poorly differentiated.[5] Biphasic sarcoma is the most common, as observed in our case.[6] In the biphasic type, both epithelial and

sarcoma components will be present. Immunohistochemical markers like CD99, S100, Bcl2, vimentin, desmin, and TLE1 help to confirm the diagnosis.[7,8] TLE1 serves as a highly sensitive and specific immunohistochemical (IHC) marker for the diagnosis of synovial sarcoma that aids in differentiating it from synovial sarcoma from other that histologic mimics of synovial sarcoma.[9] The gold standard investigation of this tumor is the demonstration of chromosomal abnormality (translocation X:18), which is present in almost 90% of the patients. This translocation fuses the SYT gene on chromosome 18 with the SSX1 or SSX2 genes at Xp11. The resulting SYT–SSX1 or SYT–SSX2 fusion transcripts alter cellular transcription. Biphasic tumors generally express the SYT–SSX1 fusion, but monophasic tumors can express either type. In our case, the diagnosis was confirmed with histopathological examination and immunohistochemical staining. We could not do the genetic study due to non-availability of the test in our setup, which is a limitation in our report.[10]

The mainstay of treatment is surgery, chemotherapy, and radiotherapy.[11] Patients with locally invasive disease or metastasis, surgery is not recommended. This tumor is relatively chemo sensitive. A treatment regimen using Adriamycin, either alone or in combination with ifosfamide, is used particularly in cases with metastasis.[12] The 5-year survival rate of pulmonary synovial sarcoma is less compared to synovial sarcoma in other sites (30 *vs* 50%). Poor prognostic factors of this tumor are young age < 20 years, male gender, size of the tumor>5 cm, extensive necrosis, presence of high mitotic figures, and neurovascular involvement.[13] Since our patient had a huge tumor occupying almost 2/3 of the hemithorax and associated with pleural effusion, we referred our case to the CTVS department for further management.

## Conclusion

Rare tumors may present with non-specific clinical and radiological features, which need an extensive workup to make a final diagnosis. In our case, histopathology along with immunohistochemical staining played a very important role in the diagnosis, to the level where no need to go for gold standard investigation (genetic study).

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