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Intrapulmonary Solitary Fibrous Tumor of the Lung: A Rare Case Report and Comprehensive Literature Review

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ABSTRACT

We report a case of a neoplasm in the upper lobe of the left lung in a 54-year-old man. The morphological picture and the immunohistochemical profile are consistent with the diagnosis of intrapulmonary solitary fibrous tumor (SFT). The study of the literature and the data emerging from the present observation suggest that intrapulmonary localization of SFT is not necessarily of pleural origin and may be associated with origin from mesenchymal structures specific to the lung.

Keywords: *Solitary fibrous tumor; Intrapulmonary solitary fibrous tumor; Lung neoplasm; STAT6; Immunohistochemistry; Spindle cell tumor; Case report.*

Introduction

Solitary fibrous tumor (SFT) is a fibroblastic tumor characterized by a prominent, branching, thin-walled, dilated (staghorn) vasculature and NAB2-STAT6 gene rearrangement (WHO 2020)[1]. Solitary Fibrous Tumor (SFT) is the term used to group together several morphological features previously considered distinct entities, which have subsequently been demonstrated to share a common immunophenotypic pattern (STAT6, CD34, BCL2, CD99) and the possibility of mixed morphological patterns occurring in the same case. The Stout's hemangiopericytoma has essentially been absorbed into the morphological spectrum of SFT, in which it represents a variant or a component mixed with other morphological expressions. Lesions arising at the pleural level, one of the most frequent sites, were previously classified as Fibrous Mesothelioma. Over the years SFT has been reported in almost all anatomical locations. SFT represents a diagnostic problem because it can mimic many unrelated tumor entities. The availability of STAT6 immunostaining has certainly contributed to improving the diagnosis. Thoracic locations, as already said, represent the most frequent site of the tumor initially indicated as a pleural neoplasm. The pleural attribution to neoplasms developing intrapulmonary is more problematic. A case of this type is the subject of this study.

CASE. A 54-year-old man has been experiencing a persistent cough and mild dyspnea for some time. He underwent a CT scan with the following results: in the most cranial sectors of the apico-dorsal segment of the left upper lobe, a solid, expansive lesion was found, with finely irregular margins, measuring 48 × 37 mm in the axial plane and extending 25 mm craniocaudally. The lesion makes extensive contact with the pleural surface along the costovertebral groove and with the mediastinal pleura and shows homogeneous, discrete enhancement after contrast medium. He subsequently underwent surgery in which a large neoplasm of the upper lobe of the left lung was found. Wedge resection was performed.

Materials and Methods

The surgical specimen consisted of a fragment of pulmonary parenchyma measuring 6 × 3 cm, almost entirely occupied by a whitish neoplasm that extends to the visceral pleura. Numerous fragments fixed in buffered formalin and embedded in paraffin were taken from it. Sections were prepared from each fragment, some of which were stained with Hematoxylin and Eosin and Gomori's Silver stain, while others were subjected to immunohistochemical investigation with a panel of antibodies indicated in the following Table 1.

Table 1. Panel of antibodies used for immunohistochemical investigation.

CD34	CD99	BCL2	VIM	CALR	SMACT	DESM	S100	CKAE1-AE3	EMA	CD56

Histology

The tumor presents a pushing expansion, and appears very well demarcated from the surrounding structures and allows a clear cleavage plane to be recognised from the pleural serosa (Figure 1a). The tumor's structure is highly varied. Hypercellular areas alternate with hypocellular areas..

The former are composed of spindle-shaped, ovoid-shaped elements, usually arranged in a disorderly fashion (Figs. 1b–d), in hemangiopericytomatous aggregation (Figs. 2a, b, c), more rarely storiform (Fig. 2d). The cells have a hyperchromatic nucleus, rather voluminous, and little amphophilic cytoplasm. Mitotic activity is not evident.

The latter are made up of dense, keloid-like connective tissue, sometimes sparsely cellular and compact (Figure 3a), sometimes composed of ropy bundles (Figure 3b) delimited by small cellular elements (Stout's patternless) (Figure 3c). There are also some small areas of lipomatous differentiation (Figure 3d). Scattered throughout the tissue are recognizable vessels with hyaline walls (Figure 4a) and variously branched thin-walled vessels (staghorn) (Figures 3a [arrows], 4b-d).

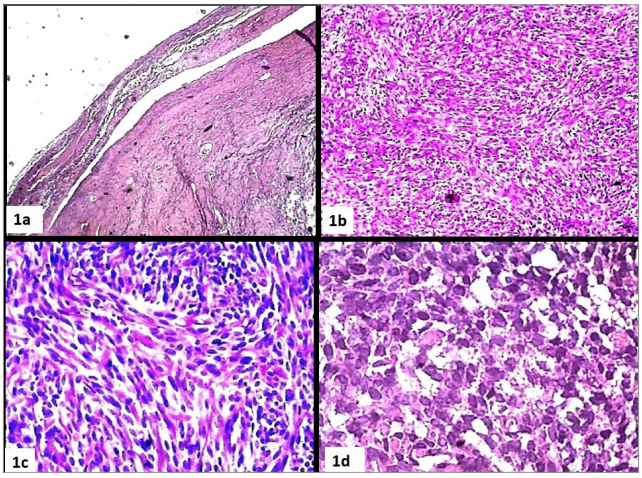

Figure 1. ***(a)*** *Pushing expansion of the tumor. Pleural detachment is evident through a cleavage line (75×);* ***(b)*** *Cellular pattern with spindle elements (HE 125×);* ***(c)*** *Cellular pattern with spindle cells arranged in bundles (HE 125×);* ***(d)*** *Cellular pattern with globose elements (HE 125×).*

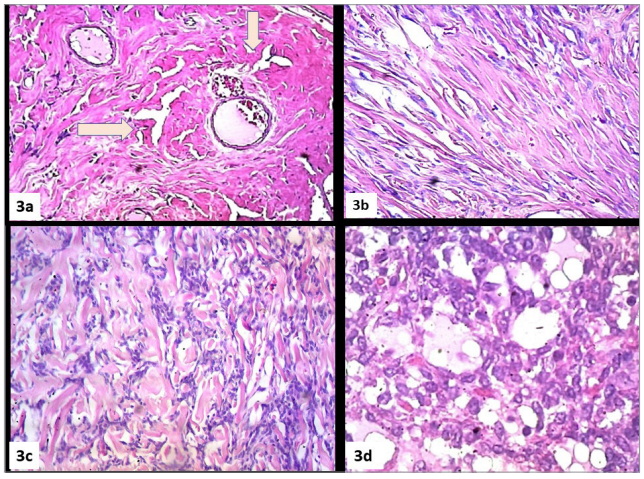

Figure 3. *Stout's patternless pattern:* ***(a)*** *Area of massive fibrosis with staghorn vessels (arrows);* ***(b)*** *Bands of thick ropey collagen;* ***(c)*** *Thick collagen bands bordered by small neoplastic cells;* ***(d)*** *Small area of lipomatous differentiation (HE 125×).*

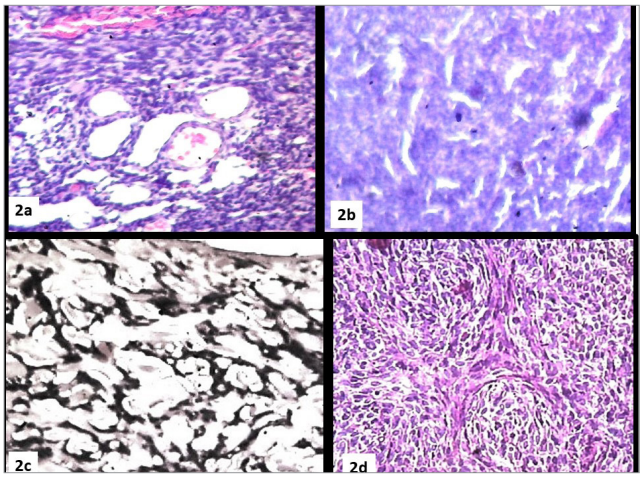

Figure 2. ***(a)*** *Hemangiopericytomatous pattern with microcystic aspects and visible thin vascular slits (HE 125×);* ***(b)*** *Hemangiopericytomatous pattern with thin vascular spaces (HE 125×);* ***(c)*** *Argyrophilic reticulum of hemangiopericytoma pattern (Gomori's Silver stain 175×);* ***(d)*** *Cellular pattern with spindle cells arranged around vascular spaces (HE 125×).*

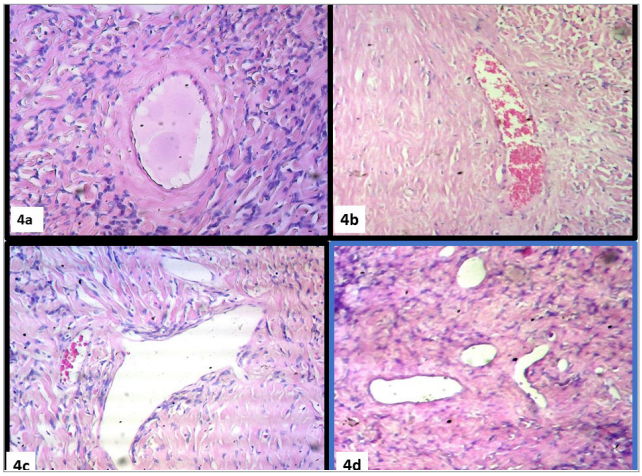

Figure 4. *Various aspects of the vascular component:* ***(a)*** *Hyalinosis of the vascular wall;* ***(b-d)*** *Vascular slits variably branched (HE 175×).*

Immunohistochemistry

The results of the immunohistochemical investigations are summarized in the following Table 2.

Table 2. Immunohistochemical findings.

STAT6	CD34	BCL2	CD99	VIM	CALR	SMACT	DESM	S100	CKAE1-AE3	EMA	CD56
+++	+++	+++	++	+++	-	-	-	-	-	-	-
Figure 5a	Figure 5b	Figure 5c	Figure 5d	Figure 6a	Figure 6b						

The results of the morphological and immunohistochemical investigations are consistent with the diagnosis of intrapulmonary Solitary Fibrous Tumor.

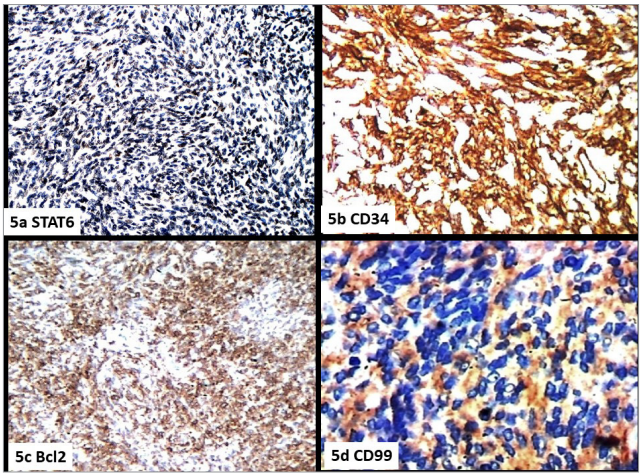

Figure 5. (a) *STAT6,* ***(b)*** *CD34,* ***(c)*** *BCL2,* ***(d)*** *CD99 (125×).*

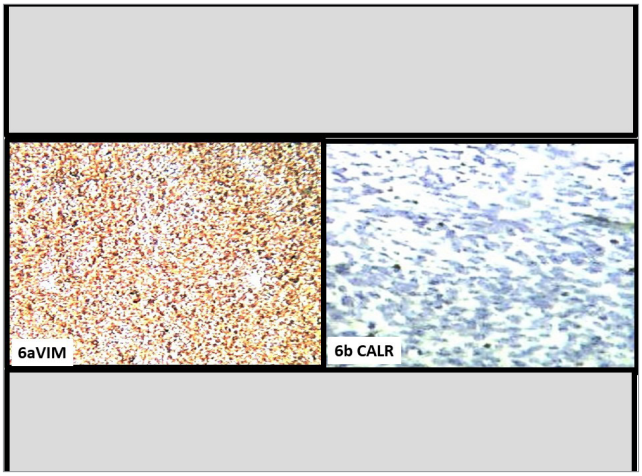

Figure 6. (a) *Vimentin,* ***(b)*** *Calretinin (125×).*

Discussion

In a review of 79 SFTs, observed in a single institution (SKCC) over eighteen years, 54 were located in the thorax (68%); of these, 29 in the pleura (37% of the total) and 19 in the lung (24% of the total). The same study reports a median age (range) of 58 (26–82), with male 40% (51) and female 39 (49). In the 54 cases with thoracic location, the median age was around 60 years and the female gender represented 52% [2].

Scrolling through PubMed under the heading "Intrapulmonary Fibrous Tumor" between 1996 and 2025, 35 items are reported. Of these, thirty-th ree were single case reports, one reported twenty-four cases retrieved from numerous institutions [3], and one reported four cases [4]. In the twenty cases where it was possible to determine the gender, the MF ratio was 9/11. The age ranged from 0–83 years. The age of the three pediatric cases was 0, 7, 8 years. The reports come from seventeen countries, fifty percent of them were reported from the Far East: 13 from Japan, 3 from China, 1 from Indonesia and 1 from Korea.

SFT presents three classic morphological patterns: 1) the so-called Stout's patternless, consisting of bands of thick ropey collagen with fissure-like spaces delimited by small neoplastic cells; 2) hemangiopericytomatous pattern comprising thin branched vessels (staghorn); 3) cellular pattern with spindle cells of varying density, often with storiform aspects. These patterns can occur in pure form, but more often they are present simultaneously in the same tumor.

The existence of some variants adds to the already complicated morphology of the classic forms: CELLULAR SFT: highly cellular with minimal collagen, often mimicking other spindle cell tumors; FAT-FORMING SFT: contains adipocytes, mimicking lipomatous tumors; GIANT CELL-RICH SFT: features numerous multinucleated giant cells; EPITHELIOID SFT: exceptionally rare, featuring epithelioid morphology with less collagen, high cellularity, and sometimes a focal papillary pattern; DEDIFFERENTIATED SFT: a highly aggressive, malignant transformation.

All this makes a differential diagnosis very difficult as it must deal with a series of innumerable entities. As many as thirty-four entities come into the differential diagnosis among the various patterns and variants of SFT [5].

The advent of immunohistochemistry has certainly made it easier to diagnose SFT with an immunophenotypic profile consisting of high expression of CD34, BCL2 and CD99. Unfortunately, these antibodies are expressed in a wide range of neoplasms, so the diagnosis must always be supported by morphological congruence. In 2014, a series of publications reported the specificity of the STAT6 antibody for SFT [6,7], making SFT diagnosis much easier.

In a series of 110 cases (all locations), local recurrence was recorded in 10% and metastases were present in 26%. In this series, the strongest predictors of time to tumor metastasis and disease-specific death were patient age at presentation, tumor size, and mitotic index [8]. A study on 147 patients shows that intrapulmonary SFTs have a benign clinical course, but an estimated subset of around 8.5% will recur or metastasize [9]. A recent report illustrates even better the course of these malignant forms [10].

A study dedicated to the so-called dedifferentiated SFT deserves special mention. These are lesions in which, in the same tumor, the patterns of classic SFT coexist with clearly sarcomatous aspects with spindle cells or epithelioid cells in which P53 is expressed (negative in classic SFT) and, instead, CD34 is not expressed (similarly to what is found in Dermatofibrosarcoma Protuberans in malignant transformation). The study reports 7 cases with various localizations, including one intrapulmonary with local recurrence and metastasis to the contralateral lung and bone [11].

The development of intrapulmonary SFT may be attributed to the direct continuity between the subpleural mesenchyme and interlobular septa or the presence of lung fibroblasts in the submesothelial areas of normal pulmonary parenchyma [12].

Conclusion

The case we studied represents a classic example of an intrapulmonary solitary fibrous tumor, both morphologically and immunophenotypically. The lack of abnormal mitotic activity and cellular atypia, with preservation of immunophenotypic expression, suggests a favorable prognosis. The presence of an evident cleavage plane from the pleura would suggest that the neoplasm is not of pleural origin. The extreme morphological variability of this neoplasm makes the statement made when it was first reported still relevant: “When one first undertakes to unravel the complex histological pictures exhibited by this tumor, the task seems hopeless, for the variations appear endless” (A.P. Stout, 1949) [13].

Acknowledgements

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Conflicts of Interest

The authors declare no conflict of interest and received no specific funding for this work.

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