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(CASE REPORT)

Pleural malignant fibrosolitary tumor: A rare entity

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Abstract

Solitary fibrous tumors of the pleura represent a rare histological entity, belonging to the groups of pleural tumors of sub-mesothelial origin , therefore some authors evoke the hypothesis of its proliferation from submesothelial tissue of the pleura .The clinical manifestations are varied and the certain diagnostic can only be made by anatomopathological examination which evaluates precise malignancy criteria. Total surgical removal is the only way to ensure a definitive cure. We report the case of 65-year-old woman with a malignant solitary fibrous tumor operated by thoracotomy with complete resection. Through this presentation, we would like to underline the rarity of this entity

Keywords: Thorasic Oncology; Oncology; Thoracotomy; Pleural Tumors; Fibrosolitary Tumor; Thorasic Surgery

1. Introduction

Solitary fibrous tumors of the pleura represent a rare histological entity, belonging to the groups of pleural tumors of sub-mesothelial origin (2), therefore some authors evoke the hypothesis of its proliferation from submesothelial tissue of the pleura (1).The clinical manifestations are varied (2) and the certain diagnostic can only be made by anatomopathological examination which evaluates precise malignancy criteria.

Total surgical removal is the only way to ensure a definitive cure.

2. Case presentation

In March 2022, a 65-year-old schoolteacher Moroccan female with history of high blood pressure for 20 years, type 2 diabetes for 5 years, dysthyroidia, asthma for 30 years, was referred to our institution for chronic dyspnea and cough. She had no history of smoking or exposure to any chemical substances. The symptomatology goes back to 1 year by the progressive installation of a chronic dyspnea associated with an emetogenic cough with expectorations.

Thereafter, the dyspnea became more and more disabling (orthopnea, sleep disorders). The general state was moderately preserved, and she was apyretic.

Chest physical examination has noted at the left lung field less transmitted vocal vibrations and the patient was polypneic at 30 cycles per minute.

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As part of the initial investigation, a chest x-ray revealed a large mass located in the left hemithorax (Fig 1). Next, thoracic pre-operative CT scan with intravenous contrast in coronal (Fig.2. A,C) and sagittal plans (Fig.2. B,C) was performed. It demonstrated an heterogeneously enhancing mass with slightly lobulated margins that invades nearly the entire left hemi thorax sparing the left upper lung.

Also, we notice an ipsilateral pulmonary spiculated nodule (Fig 2.D) which is histologically similar to pulmonary aspergillosis, without any sign of malignancy.

Within the framework of the pre-operative assessment, the cardiac echography revealed a repression of the heart by the mass, plethysmography showed restrictive ventilatory disorder and a scan-guided biopsy was performed revealing the presence of localized spindle cell tumor proliferation.

Also, bronchial biopsies showed a bronchial mucosa with fibro-inflammatory remnants, without signs of malignancy.

As surgical treatment, the patient underwent a left posterolateral thoracotomy (Fig 3) that allowed complete surgical resection of a huge lung tumor of 20x15x7 cm long axis (Fig. 4) with mediastinal and left lower lobe invasion, taking a segment of the lower lobe.

The histopathological study of the mass showed a fairly dense tumor proliferation made of spindle cells with territories of fibrosis and necrosis estimated at 30%. The nuclei were elongated with moderate anisokaryosis in some places. The cells tended to form short bundles. The neoplasic cells presented a high mitotic activity (7-8 mitoses per high-power field).The stroma was fibro-inflammatory without visible emboli. This proliferation infiltrated the pleural tissue and adjacent lung parenchyma. Therefore, all these elements were in favor of a poorly differentiated sarcomatoid mesenchymal tumor.

Immunohistochemical staining was positive for CD34, Desmine and STAT6 but was negative for cytokeratin,EMA and PS100 and the Ki67 was estimated at 15%. The diagnosis of Solitary Fibrous Tumor (SFT) was made.

The patient stayed 3 days in intensive care and the thoracic drain was removed at day 3 post-op.

She was discharged in good condition on the 5th day after surgery. Postoperative complications were not detected. No recurrences have been reported so far.

3. Discussion

Solitary fibrous tumors are rare. There are no more than 1000 cases described in the literature (1). It represents 5% of all pleural tumors (1). In majority, the preferred seat of these tumors remains the pleural cavity. Their intra-pulmonary development from the visceral pleura is rare. Only about 20 cases have been described in the literature (1).

This pathology can occur at any age, but predominates around the age of fifty with a balanced sex ratio in the majority of studies (2).

Subcutaneous tissue is the most frequent location, however they can occur at any location(3).

Clinically, solitary fibrous tumors are often symptomatic, accompanied by chest pain, dyspnea or cough. The intensity of the symptoms is correlated to the size of the tumor by the compressive effect it exerts, not only on the respiratory system but also on the right heart chambers, responsible for dyspnea in decubitus as the case of our patient(4).

The chest X-ray is a diagnostic step, showing most often a rounded, homogeneous and well limited opacity, rarely associated with a pleural effusion.

However, for large tumors with pulmonary compression, the radiographic image is limited to a simple opacity of the entire pulmonary hemifield associated with a significant mediastinal deviation (5). The chest CT scan will complete the assessment by confirming the pleural origin and the tissue nature of the tumor, as well as by specifying its location and its relationship with the pleura and adjacent organs [2]. The tumor often appears well-limited and homogeneous, but a heterogeneous appearance may be seen when there are necrotic-hemorrhagic remodeling or myxoid degeneration. Calcifications may also be seen. This mass may enhance after contrast injection and may be mobile (5). Nuclear magnetic

resonance imaging allows to determine the fibrous character of the tumor especially in T1 sequence, which differentiates it from other mediastinopulmonary, parietal and diaphragmatic structures (6).

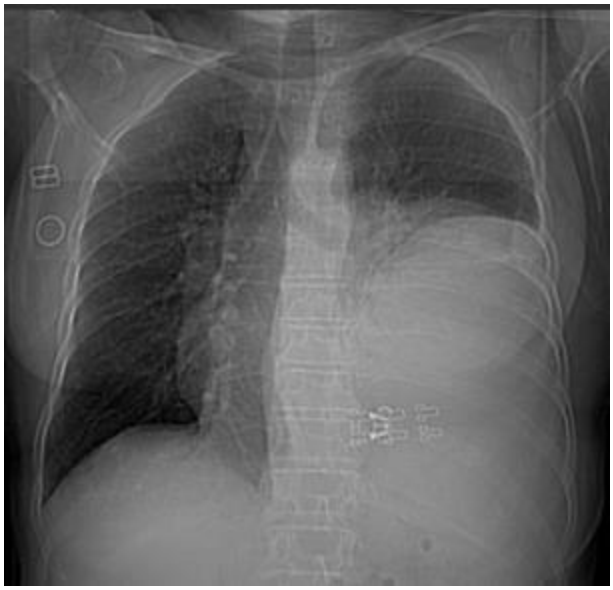

Figure 1 Chest X- ray of a large mass located in the left hemitorax

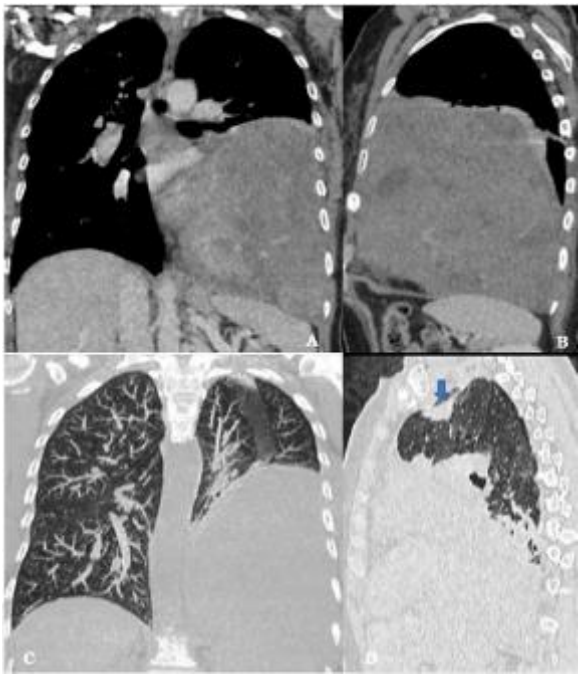

Figure 2 Thoracic pre-operative CT scan with intravenous contrast in coronal (A,C) and sagittal plans (B,C) demonstrated an heterogeneously enhancing mass with slightly lobulated margins that invades nearly the entire left hemi thorax sparing the left upper lung.

We notice an ipsilateral pulmonary spiculated nodule (Blue arrow).

However, certain diagnostic can only be provided by anatomopathological examination. Macroscopically, the tumor is encapsulated, variable in size, and may be sessile or pedunculated, which is not the case for our patient. Sessile tumors are associated with a higher risk of recurrence (63% versus 14%) (3). Histologically, the tumor is presented as ovoid to fusiform spindle cells accompanied by indistinct cell borders arranged haphazardly, the stroma is in general hyalinized

to collagenous and myxoid changes can be prominent (7). Many features are suggestive of malignant behavior, they include increased cellularity, increased mitotic activity, nuclear polymorphism, tumor necrosis and infiltrative borders.

The immunohistochemical study of fibrous tumors classically shows diffuse expression of CD34, variable intensity expression of CD99 and Bcl-2 protein and negativity of epithelial markers (cytokeratin, EMA) and S100 protein (8,9). Tumors lacking malignant histological features in primary resection specimens may acquire malignant features at time of recurrence and metastases.

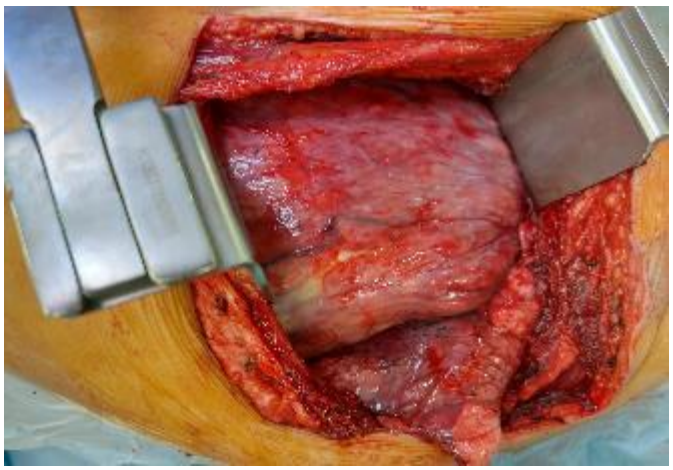

Figure 3 Intraoperative tumor image before the resection

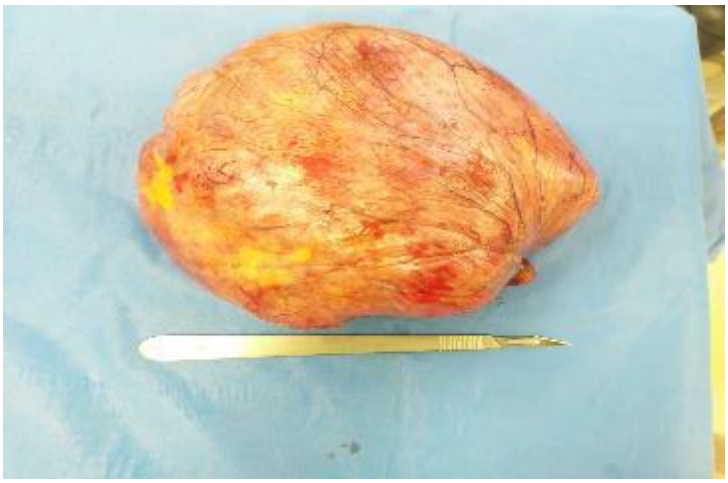

Figure 4 Resected surgical piece

It would be interesting to discuss in our case the discrepancy between the radiological image evoking a malignant nodule and the morphological aspect compatible with pulmonary aspergilloma.

Because of the difficult evolutionary profile, remote radioclinical monitoring is recommended by some authors (10). An annual check-up including essentially a complete clinical examination and a radiographic work-up, especially in patients at risk with giant tumors like the case of our patient, would be sufficient.

The complete resection of the tumor from its base of implantation remains the standard treatment (11) which can sometimes extend to the pulmonary parenchyma or to the wall in the event of invasion.

In a patient with a giant compressive solitary fibrous tumor, surgical management must be rapid in order to remove the tumor without compromising the hemodynamic and respiratory status of the patient. On the other hand, it must allow the complete carcinological removal of the tumor to hope for a recurrence-free evolution (12). the anterior thoracotomy

more or less associated with a partial sternotomy constitutes an excellent approach for patients who are generally severely dyspneic. This anterior approach is better supported than the large posterolateral thoracotomy in the presence of a large compressive pleural tumor(12).

Given the pleural origin of this tumor, its removal allows for immediate lung re-expansion. At this moment, the alveolar recruitment maneuvers are necessary because they condition the operative follow-up. In our patient's case, the immediate pulmonary re-expansion was satisfactory (10). In aggressive forms of solitary fibrous tumors of the pleura, surgical resection may be followed by chemotherapy associated or not with radiotherapy, as in the case of our patient.

4. Conclusion

These tumors remain very heterogeneous and the symptoms are various according to the location of the tumor. Gold standard of treatment is surgical resection and the anatomopathological examination is essential for the diagnosis but also to determine prognostic factors.

After surgery, a long terme surveillance is imperative to watch for a possible recurrence or complications.

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