Case Series: Benefit of Angioembolization Prior to Surgery for Giant Fibrous Pleural Tumors

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Abstract

Only 5% of malignant pleural neoplasms are fibrous tumors of the pleura that typically develop from sub-mesothelial mesenchymal tissue of the visceral pleura. When a tumor measures more than 15 cm, it is referred to as a "giant."

The best option for treating both benign and malignant neoplasms is surgery. We conducted a retrospective analysis of the three case series of pleural large fibrous tumors. Additionally, we will discuss our experience with pre-surgical Angioembolization, involving implanting coils and plugs, to treat spindle cell neoplasm. Surgery was completed without any issues.

Keywords: *Pleural tumor, angioembolization, solitary fibrous tumor, giant fibrous pleural tumor.*

INTRODUCTION

SFTs exhibit a range of behavior from benign to malignant, with evidence linking metastasis risk to age at diagnosis, the extent of necrosis, mitotic rate, and tumor size. Surgical excision with or without radiotherapy is the standard treatment for localized disease [1].

Tumor resectability is an important determinant of clinical prognosis in SFT. It is affected by the type of mass attachment, angiogenesis, and accessibility. SFTs are more commonly attached to the visceral pleura and are mostly pedunculated. The pedicle attached to these tumors contains abundant feeding vessels. Therefore, angiography should be performed to identify the pedicle. Preoperative embolization is required here. Other options are mini-thoracotomy or thoracoscopic ligation of the vessel. In contrast, tumors originating from the parietal pleura tend to adhere [2].

CASE NO. 1

A 50-year-old male with no known co-morbid was presented in the outpatient department with complaints of shortness of breath and cough. His history was unremarkable. On examination, there was a decreased breath sound in the right upper zone of the lung.

Chest X-ray showed a right-sided large lobulated mass covering 50% of the chest.

CT scan was performed that showed a large tumor with heterogeneous enhancement with adhesions to adjacent structures without invasion with intact fat planes measuring 20cmx19cmx14xm (**Fig. 1**).

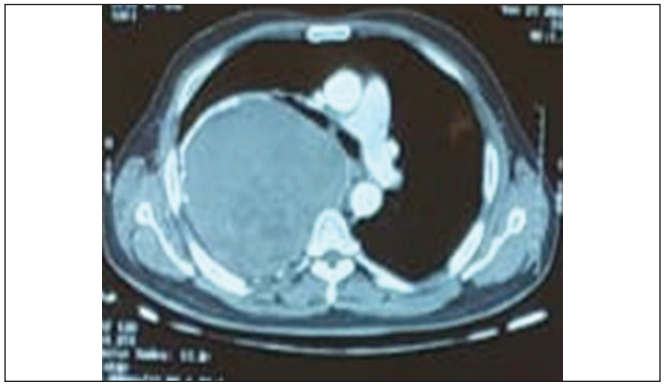

Fig. (1): CT-scan Chest.

Surgery was planned, a Right thoracotomy was performed and a biopsy was taken. The sample was sent for the frozen section which showed benign spindle cell neoplasm. So we continued the procedure but it was a large highly vascular tumor with dense adhesions between the lung and tumor, and between the lung and chest wall. The pedicle of the tumor was short and arising from intercostal vessels.

Therefore, it was difficult to approach maneuver and ligate the pedicle. In the pedicle, after the removal of the

Fig. (2): Rt. Pleural Mass.

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tumor the vessel was ligated separately and transfixed (**Fig. 2**).

Due to the lack of length of feeding vessels, the pedicle was over-sewn as well. His total blood loss was 3.5 liters and he required a massive transfusion.

Postoperatively patient was extubated but shifted to the ICU setup for 24 hours observation. The recovery was otherwise uneventful and the patient was discharged from home after 7 days. Postoperative chest X-ray was improved (**Fig. 3**).

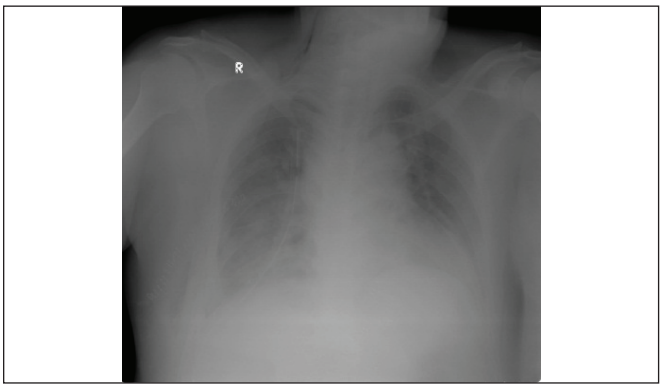

Fig. (3): Post-op chest x-ray.

CASE NO. 2

A 35-year-old male with no known co-morbid came with complaints of left-sided chest pain and chest congestion with no relevant past medical and family history. Examination of the chest was unremarkable. There was no lymphadenopathy. Chest X-ray showed a lobulated left-sided mass (**Fig. 4**). CT-scan chest angiogram showed a lobulated faintly enhancing lesion measuring 6.5x 6.3cm on axial dimension in the left upper lobe. A small feeding artery is seen arising from the left main pulmonary artery supplying to the core of the lesion (**Fig. 5**).

CT-guided biopsy showed spindle cell neoplasm with no atypia and mitosis. Preoperative angioembolization of the vessel supplying the lesion was performed then the patient was prepared for surgery after 48 hours of angioembolization.

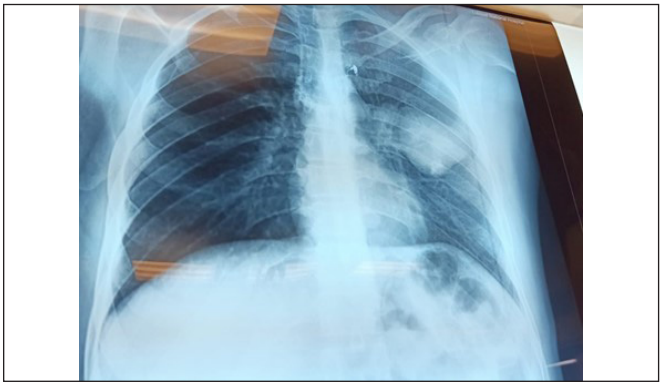

Fig. (4): pre-operative chest x-ray.

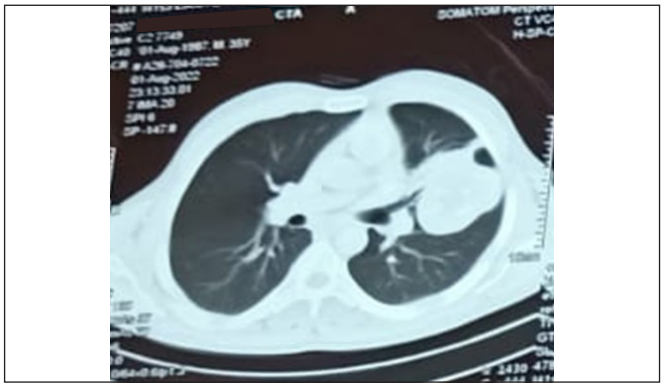

Fig. (5): CT-scan image.

Left-sided thoracotomy and left upper lobectomy were performed There was no obvious blood loss (100ml) and the surgery went uneventful. The postoperative course was uneventful. Chest X-ray postoperatively was improved (**Fig. 6**).

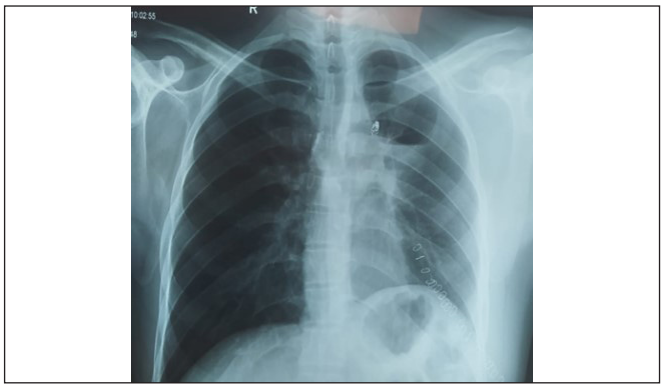

Fig. (6): Post-op chest x-ray.

CASE NO. 3

A 58-year-old male with a known case of diabetes mellitus came with complaints of chest pain and shortness of breath for 1.5 months. On examination prominently visible neck veins were present and breath sounds were absent on the right side. Pedal edema was also present.

Chest X-ray showed complete whiteout on the right side with the shifting of the trachea on the opposite side (**Fig. 7**).

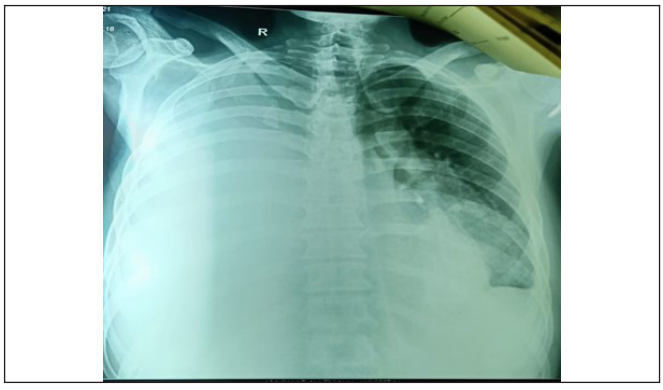

Fig. (7): Pre-op chest x-ray.

The CT-scan chest was advised which showed: there was a large hypodense mass lesion involving the right hemothorax. The mass measured about 21.6 x 20.0cm and extended for a length of about 21.9cm in the craniocaudal direction. It showed a few areas of necrosis. Anterolaterally it was abutting the chest wall with no bony erosion or chest wall extension. Posteriorly it was abutting the posterior ribs and vertebral bodies with no bony erosion or intraspinal extension. Medially it was infiltrating the mediastinum and compresses the right pulmonary artery, veins, and superior vena cava. It was abutting the pericardium, heart, and esophagus displacing them towards the left side. Inferiorly it was reaching up to the diaphragmatic region displacing the right hemidiaphragm inferiorly (**Fig. 8**).

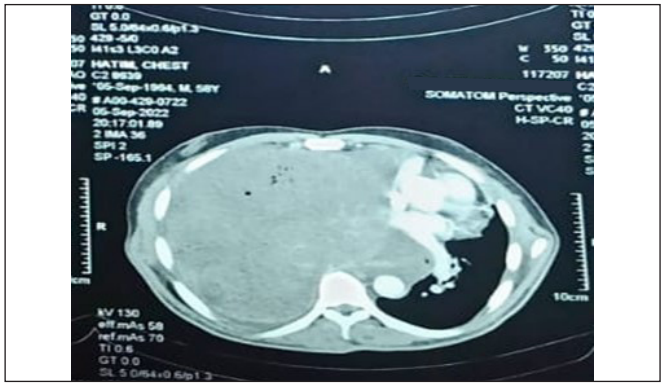

Fig. (8): Pre-op CT-scan chest.

CT-guided biopsy was performed and showed spindle cell neoplasm without atypia and mitosis with CD-34 + IHC staining. The case was discussed in a multidisciplinary meeting and preoperative Angio-embolization was planned before surgical intervention and successful embolization of feeding vessels Right 10th, 11th, 12th intercostal and artery arising from the abdominal aorta at L2 level (predominant) was performed with PVA and glue lipoidal. Surgery was planned after 2 weeks of Angio-embolization.

Right thoracotomy and resection of the mass were performed intra-operatively mass was adherent to the diaphragm as well and the pedicle was arising from the diaphragm (**Fig. 9**). The surgery went uneventful and there is less blood loss (500ml) than expected and need less blood transfusion than expected.

Fig. (9): Intra-operative image.

Post-operative recovery was uneventful. The patient's condition improved as shown in the chest x-ray (**Fig. 10**).

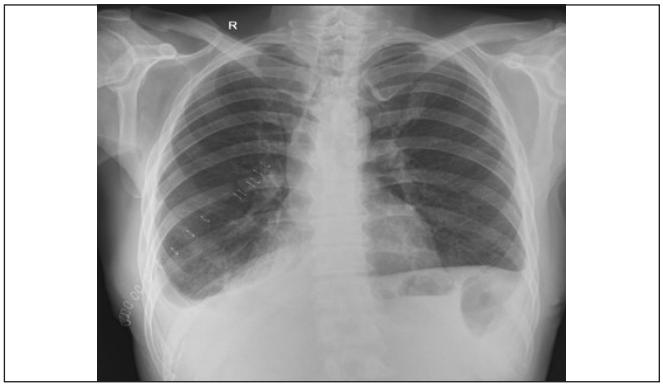

Fig. (10): Post-op chest x-ray.

DISCUSSION

Giant benign fibrous tumors are described as having a diameter of more than 15 cm or occupying more than 40% of the hemithorax. These tumors can develop from sub-mesothelial stromal cells with fibroblastic or myofibroblastic phenotypes, whose development is aided by an abnormal inflammatory response as well as hormonal stimulation. The surface of a solitary fibrous tumor is greyish-white, with areas of soft tissue, necrosis, and hemorrhage. Sections are made up of thick fibrous tissue nodules that frequently contain cystic features, as well as a tiny vascularized peduncle [3].

Fibrous tumors of the pleura are slow-growing tumor that persists without pain. There are usually compressive symptoms in line with the tumor's growth. It is uncommon to have associated pleural effusion, and it typically has a negative cytology. It is possible for fibrous tumors to occasionally cause distant metastases, Para neoplastic syndrome, and malignant transformation. Moderate to mild restrictive ventilation dysfunction may exist. For the clinical diagnosis of a Fibrous Tumor of Pleura, a CT scan chest is an essential tool [4].

Rarely, SFPT manifests as Doege-Potter syndrome, which is characterized by hypoglycemia caused by incorrect insulin-like factor II production.

For lesions with a diameter of less than 5 cm, video-assisted thoracoscopic surgery (VATS) is recommended. Surgical excision may be difficult due to the tumor's size, proximity to nearby tissues, and identification of the vascular peduncle. The internal mammary and bronchial arteries, the intercostal arteries, and collateral branches from the phrenic artery most frequently provide the tumor's blood supply [5].

Heterogeneous contrast enhancement can also occur, typically seen in malignant changes and 60% of benign tumors in the case of pleural SFT. The pattern of contrast enhancement depends on the cellularity of the tumor,

Table 1: Comparison of Research articles with our case series.

Papers	Case	Gender	Age (yr.)	Pre-operative Angioembolization	Blood loss	Site	Size of Tumor
Our case series	No. 1	Male	50	No	3.5 lit	Right upper pleural cavity	20cm x 19cm x 14cm
	No. 2	Male	35	Yes	100ml	Left upper lobe	6.5cm x 6.3cm
	No. 3	Male	58	Yes	500ml	Right lower pleural cavity	21.6cm x 20cm x 21.9cm
[1]	-	Female	67	No	100ml	Left lower pleural cavity	13cm x 12cm x 19cm
[3]	-	Female	44	Yes	500ml	Left pleural cavity	14cm x 12cm x 8cm
[4]	-	Female	64	No	-	Within left lung	-
[5]	-	Female	76	Yes	Nil	Left pleural cavity	15cm
[7]	-	Male	68	No	-	Right pleural cavity	30cm x 21cm x 15cm
[8]	-	Female	66	Yes	-	Right pleural cavity	23cm x 16cm x 15cm
[9]	-	Female	39	Yes	100ml	Right lower pleural cavity	14.7cm x 15.4cm x 13.5cm
[10]	5 Patients	Male >female 4:1	44-78	Yes (in all cases)	Average: 800ml	Right >left pleural cavity 3:2	Mean: 25cm
[11]		Male	28	Yes	Nil	Retro auricular	2cm
[12]		Female	52	Yes	Nil	Bilateral pleural cavity	>15cm
[13]		Female	57	Yes	1000ml	Right pleural cavity	33cmx20cmx15cm

the number of connective tissue elements, and the presence of degenerative changes.

NAB2-STAT6 fusion genes define SFTs and can be visualized by STAT6 Immuno-histochemical staining. This is very reliable and therefore helps him in diagnosing SFT. CD34 is a characteristic antigen overexpressed in SFTs but may be lost in some malignant SFTs [6].

The likelihood of the tumor returning is dependent primarily on the histological features of SFTP and the radically of the surgical operation. Around 2% of benign pedunculated tumors, 8% of benign sessile tumors, 14% of malignant pedunculated tumors, and 63% of malignant sessile tumors have a recurrence risk [7].

Rarely, both benign and malignant tumors of a large size exhibit calcification. Additionally, MRI can help differentiate between benign and malignant tumors, with variable signal intensity and contrast uptake having been linked to malignancy [8].

A highly vascular pedicle connects the tumor to the pleura in 38%–50% of patients. They grow in the submesothelial connective tissue. They may arise from both the visceral and parietal pleura, with the visceral pleura accounting for two-thirds of them and the parietal pleura for the remaining one-third. The visceral pleura is the site of origin for 80% of cancers, yet they rarely spread there. Tumors that are sessile or pedunculated are divided in half equally. The blood supply to the tumor is typically ensured by collateral branches from the phrenic artery, the intercostal arteries, internal mammary arteries, and bronchial arteries [9].

When the thoracic cavity is open, a giant sessile tumor with good movement may compress the heart and lower blood pressure. During operation, the 45° lateral decubitus position is a simple and reasonable way to avoid compression. Extensive peri-tumor adhesions make ligating feeding vessels and handling hilar vessels more difficult, especially in a tumor with a broadbased pedicle. Because it allows for better exposure and avoids impairing adjacent structures, piecemeal resection proved to be a practical method for removing giant fibrous tumors. In cases of “inverted fibroma,” an invasive tumor, extended excision is required [10].

Depending on the vascularity of the tumor, surgical removal can be complemented by endovascular embolization to reduce bleeding during surgery, allowing for better visualization of anatomical relationships and clear surgical margins can be secured. Furthermore, the resulting necrotic softening of the tumor can simplify resection. The use of spherical particles, polyvinyl alcohol (PVA), and onyx (ethylene vinyl alcohol copolymer dissolved in dimethyl sulfoxide and micronized tantalum powder; EV3, Irvine-CA) is described in transarterial embolization of SFT.

Similar to PVA, microspheres (including Embozene that was used in our case) block the vessel inducing inflammatory reaction, angionecrosis, and fibrosis [11].

Benefits of Angioembolization in the preoperative setting include reduced perioperative blood loss, creation of an edematous tissue plane to facilitate dissection, and reduced tumor burden, including the extent of vascular thrombosis, if present. Significant differences in reporting markers such as reduction in intraoperative blood loss need for blood transfusion, duration of surgery, surgical complications, and survival outcomes have limited their use to local practice patterns [12].

Table **1** showed a comparison of our cases with other research papers.

Preoperative embolization of arteries supplying the tumor was effective in reducing the risk of massive bleeding. A confirmed diagnosis depended on postoperative histological and Immuno-histochemical investigations.

Surgical resection with wide margins is currently the main treatment [13].

As we have seen in our cases, a patient with a large fibrous tumor in which pre-operative Angio-embolization was not performed, led to massive blood loss of 3.5 liters which required multiple blood transfusions (case no .1). As compared to that patient (Case No. 3), in whom Angio-embolization was performed pre-operatively, this patient had a better outcome in terms of less blood loss.

CONCLUSION

In one of our cases, which was not angioembolized preoperatively, there was considerable blood loss from the pedicle as the pedicle was short and behind the tumor, and it was technically difficult to control the pedicle before dividing the various adhesions between the lung and tumor.

Due to Angioembolization of the pedicle, we found that all the feeding vessels had coagulated and there was no bleeding from any of them. This reduces overall blood loss considerably.

The only blood loss was encountered when adhesion between the lung and the tumor was divided.

In large fibrous tumors, it is often the case that the pedicle is inaccessible and behind the tumors. Furthermore, the pedicle can be short and arise from the chest wall, therefore we recommend that Angioembolization should be carried out before surgery for these tumors.

DECLARATION OF PATIENT CONSENT

We received informed written consent from the patient for the publication of this case report and accompanying images.

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Nil.

CONFLICTS OF INTEREST

The authors declare no conflict of interest.

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Declared none.

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