

Case report of medical thoracoscopy and endobronchial ultrasound bronchoscopy in the workup of giant solitary fibrous tumor of the pleura

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Abstract

Solitary fibrous tumor of the pleura (SFTP) is a rare tumor of fibroblastic origin. It can be quite vascular, and its surgical management carries the risk of a major intra-operative bleed. The pre-operative use of endobronchial ultrasound (EBUS) to visualize the vascular supply of the tumor has not been reported.

We report a case of a patient presenting with progressive shortness of breath and cough who was found to have a very large pleural-based tumor. We describe the use of medical thoracoscopy and EBUS to establish the diagnosis of SFTP and to characterize the blood supply of the tumor.

In the future, EBUS may provide an alternative to conventional angiography for both mapping and embolizing tumor blood supply.

Abbreviations: CT scan = computed tomography scan, EBUS = endobronchial ultrasound, EUS = endoscopic ultrasound, IR = interventional radiology, SFTP = solitary fibrous tumor of the Pleura.

Keywords: case report, endobronchial ultrasound, medical thoracoscopy, solitary fibrous tumor of the pleura

1. Introduction

Solitary fibrous tumor of the pleura (SFTP) is a rare mesenchymal tumor of fibroblastic origin. Approximately 80% of SFTP arise from the visceral pleura.^[1] SFTP can be highly vascular and involves major systemic arteries, such as the subclavian.^[2] The treatment of SFTP is surgical resection; however, no standardized approach to the pre-operative evaluation of SFTP has been described. Herein, we report the case of a large SFTP that was evaluated pre-operatively using endobronchial ultrasound (EBUS) to assess tumor vascularization and thoracoscopy to obtain tissue samples for diagnosis. Given the exceptional vascularization noted during this pre-operative assessment, interventional radiology-guided embolization of a branch of the phrenic artery was performed before surgical resection. Institutional review board approval and informed consent were waived, as this is a retrospective case report with no identifying patient information presented.

Editor: Levent Dalar.

The authors have no conflicts of interest to disclose.

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Medicine (2016) 95:27(e4100)

Received: 18 January 2016 / Received in final form: 4 June 2016 / Accepted: 9 June 2016

<http://dx.doi.org/10.1097/MD.0000000000004100>

2. Case report

A 65-year-old healthy male with no prior past medical history presented to his primary care provider with complaints of worsening shortness of breath and cough for several months. His physical examination was significant for dullness to percussion over the right hemithorax and absent airflow on auscultation of the same area. Chest x-ray revealed a dense opacification of the right lung base with marked hypoinflation of the right lung. A subsequent CT scan of the chest confirmed the presence of a large mass filling the lower right hemithorax, compressing the right lung, and causing mild mediastinal shift (Fig. 1). The patient was referred for a transthoracic CT-guided needle biopsy. The resultant histopathology revealed fibrous tissue, but was inadequate to make a definitive diagnosis.

The case was discussed with a multidisciplinary team, including members of the interventional pulmonary and thoracic surgery services. Because of the tumor's very large size and concern about proceeding with en bloc resection without definitive pathology or characterization of its blood supply, the decision was made to do further testing. Medical thoracoscopy, bronchoscopy, and EBUS were performed in a single diagnostic session by the interventional pulmonologist. The patient received intravenous anesthesia. An endotracheal tube was placed by an anesthesiologist. Flexible bronchoscopy was performed to exclude endobronchial involvement of the lesion.

Because the vascular supply of SFTP often originates from the subclavian or central arteries, we used curvilinear EBUS to evaluate the tumor vasculature by starting from the upper trachea. The tumor was visualized beginning at the level of the main carina. Scanning, at the distal end of the bronchus intermedius, revealed a large vessel supplying the mass originating from the direction of the diaphragm. Based on the anatomical location, we felt that this likely corresponded to a branch of the phrenic artery (Fig. 2). No medial or superior feeding vessels were identified by EBUS. Medical thoracoscopy was then performed

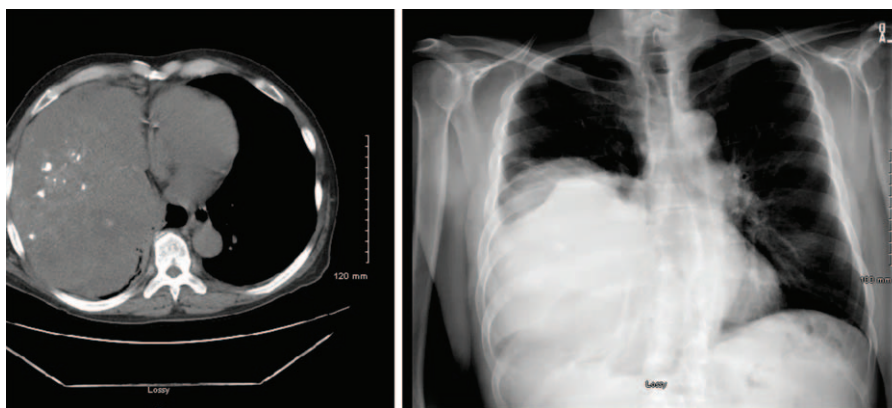


Figure 1. Computed tomography scan and chest radiograph showing a large mass occupying the right lower hemithorax.

with a semi-rigid thoracoscope in order to obtain biopsies and inspect the surface of the tumor (Fig. 2). Very brief, single-lung ventilation was performed by placing a balloon blocker to the right main stem bronchus. Then, careful blunt dissection at the 5th intercostal space (midline axillary) was performed. The blunt 8mm trocar then passed into the pleural space successfully. The bronchial blocker was then removed and the right lung was allowed to re-expand. The tumor’s lateral surface was examined and biopsies were obtained with flexible forceps. To obtain immediate hemostasis in a possible event of

bleeding during thoracoscopic biopsy, argon plasma coagulator and a thoracic surgeon were present in thoracoscopy room. However, there was no significant bleeding during thoracoscopic biopsy of tumor mass. A total of 6 biopsies ranging in size from 0.3 to 1.0cm were obtained. Pathology of SFTP was confirmed.

Because of both the observed vascularity of the tumor and its large size, the patient was referred for IR-guided angiography and embolization of the phrenic artery (Fig. 3). The following day, a right thoracotomy was performed and the lesion was removed en

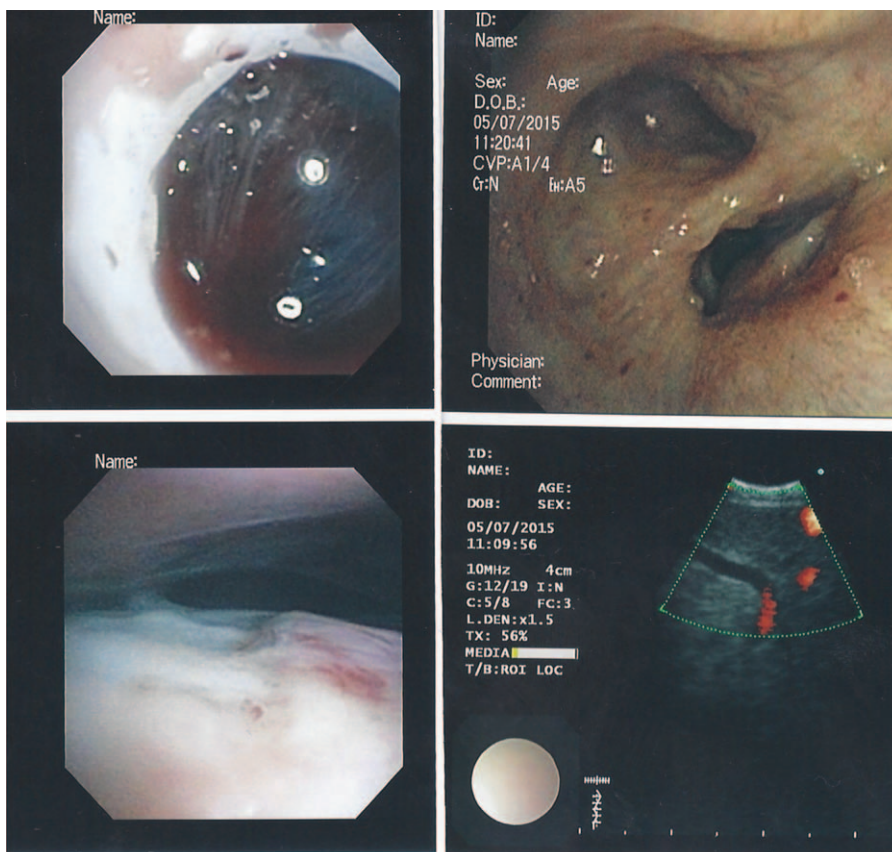


Figure 2. Thoracoscopy revealed a large tumor with both vascular (upper left) and encapsulated (lower left) components. EBUS (right) showed no endobronchial invasion but evidence of a large vessel feeding the tumor. EBUS, endobronchial ultrasound.

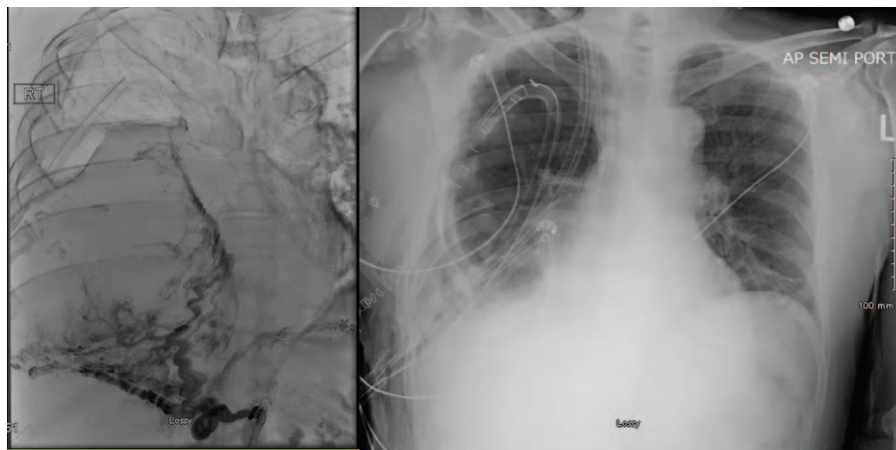


Figure 3. Angiography identified the phrenic artery as the main blood supply of the tumor (left). Chest radiograph on post-operative day 1 showed adequate lung re-expansion (right).

bloc. The excised tumor measured 25 cm in diameter and weighed 2606 g, one of the largest reported in the literature (Fig. 4). Three chest tubes were left in place. On post-operative day 1, a chest x-ray showed significant re-expansion of the right lung (Fig. 3). The patient's hospital course was complicated by brief new-onset atrial fibrillation with rapid ventricular response and development of a large pleural effusion. He remained afebrile and his incisions were without signs or symptoms of infection throughout his stay. The chest tubes were removed on post-operative day 8 and the patient was discharged home on

post-operative day 9. At the time of discharge, he reported marked improvement of shortness of breath and cough.

3. Discussion

Primary tumors of the pleura are classified into two major categories: diffuse and solitary. Solitary tumors are typically fibrous and not asbestos related, whereas diffuse pleural neoplasms are typically asbestos related. SFTP was first described by Wagner in 1870, and defined pathologically by Klemperer and Rabin in 1931.^[3] SFTP occurs equally in men and women, affecting most patients in the 6th and 7th decades of life.^[4] The incidence of SFTP is low in comparison to that of malignant pleural mesothelioma. Approximately 80% of SFTP are benign, and treatment for both benign and malignant tumors consists of surgical resection.^[1]

Whereas surgical resection is the standard treatment for SFTP, the pre-operative evaluation of such tumors varies. Tissue diagnosis can be difficult to obtain before surgical resection. As with our patient, percutaneous transthoracic needle biopsy of the mass often fails to yield enough tissue for diagnosis.^[1] We believe that medical thoracoscopy may provide an alternative means of obtaining adequate tissue samples. It has also been suggested that pre-operative bronchoscopy is of no utility beyond ruling out endobronchial involvement of the tumor.^[1] However, prior case reports note that SFTP can be highly vascular^[5] and involve major systemic arteries, such as the subclavian, predisposing to a high risk of intra-operative complications.^[2] This has led some surgeons to recommend pre-operative angiography and potential embolization of massive SFTP to reduce the risk of intra-operative bleeding and improve overall outcomes.^[6]

We believe that the present EBUS technology provides limited, but important information in evaluating the vasculature of large SFTPs. Endoscopic ultrasound endoscopy (EUS), on the contrary, has been used in other settings to control bleeding and embolize vessels.^[7,8]

As higher depth EBUS becomes available, this technology may mitigate the need for contrast exposure via angiogram, particularly in high-risk patients such as those with renal failure or contrast allergies.

Although limited to a single case, the present report is the first, up to our knowledge, to suggest that future advances in EBUS technology may provide an alternative option to angiography,

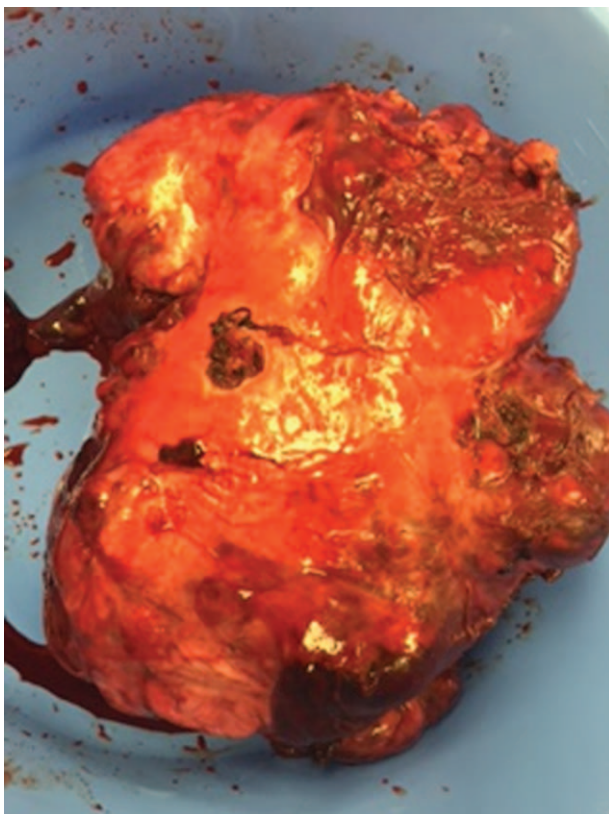


Figure 4. The excised solitary fibrous tumor of the pleural measured 25 cm in diameter and weighed 2606g.

both diagnostically and therapeutically in the pre-operative evaluation of SFTP. Further investigation is warranted to validate this approach.

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