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Surgical Management of Endobronchial Solitary Fibrous Tumors

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Abstract

Solitary fibrous tumors of the pleura are rare neoplasms with both benign and malignant behaviors that are not reliably predicted by histologic findings. We report the case of a 55-year-old woman with recurrent pneumonias whose left mainstem bronchus was occluded by a 2.1- × 1.4-cm endobronchial mass that extended extraluminally into the subcarina. The mass was tissue diagnosed to be a solitary fibrous tumor preoperatively, and was completely resected by sleeve resection of the left mainstem bronchus with lung preservation. This report describes the challenging surgical management of an infrequently encountered tumor in a unique location.

Solitary fibrous tumors of the pleura (SFTP) were first pathologically described by Klemperer and Rabin in 1931, and approximately 800 cases have been reported in the English literature [1]. A review by Briselli and colleagues [2] of 368 cases found that approximately 80% arise from the visceral pleura and 20% from the parietal pleura. The visceral pleural-based masses are typically pedunculated, encapsulated, peripheral masses with a well-vascularized stalk. Parietal-pleural based tumors are usually sessile and have a greater incidence of recurrence [3]. In case reports, there have been accounts of SFTP located entirely within the lung parenchyma, with no gross relationship to the pleura.

Only 15 cases of intraparenchymal lung tumors have been described in the English literature, [1, 4–6], less than 2% of the roughly 800 reported cases. Proposed mechanisms for these atypical lung masses include (1) direct continuity of subpleural mesenchyma with the intralobular septae connective tissue, (2) origination of tumors from fibroblasts of the lung parenchyma itself, and (3) invagination of the visceral pleura, with mechanical forces causing growth away from the chest wall [6]. The occurrence of SFTP in an endobronchial location is an even rarer finding; to our knowledge, there have only been two published instances [1, 5]. We report a case of an endobronchial SFTP that completely occluded the left mainstem bronchus and extended extraluminally into the subcarina.

A 55-year-old woman, who was a nonsmoker with a history of asthma, presented with recurrent pneumonias since January 2008. In February 2009, computed tomography scanning and bronchoscopy revealed a 2.1- × 1.4-cm mass within the left mainstem bronchus that extended into the subcarina, with postobstructive collapse of the lingula and left lower lobe (Fig 1A, Fig 2). Bronchoscopic biopsy revealed a solitary fibrous tumor.

Immunohistochemistry staining was positive for cluster of differentiation 34, and negative for cluster of differentiation 31, cytokeratin 5/6, calretinin, cluster of differentiation 45, epithelial membrane antigen, cytokeratin 7, cytokeratin 20, pankeratin, S100, synaptophysin, and thyroid transcription factor 1. Preoperative pulmonary function tests revealed forced expiratory volume in 1 second of 1.43 (57% of predicted) and diffusion capacity of the lung for carbon monoxide of 81% of predicted. Quantitative lung perfusion study showed 9.5% left and 90.5% right lung perfusion.

In March 2009, the patient underwent surgical resection, which entailed left thoracotomy and en bloc sleeve resection of the left mainstem bronchus and subcarinal tumor, with bronchoplasty repair and intercostal muscle flap coverage of the primary anastomosis (Fig 3). A left hilar pericardial release maneuver was performed to prevent tension on the bronchial anastomosis.

Pathology revealed an intramucosal, 3.3-cm solitary fibrous tumor with rare mitoses (<1/10 high-power field) and no evidence of tumor necrosis or lymphovascular invasion (Fig 4). The lymph nodes and margins were negative.

The patient had an uneventful postoperative course and was discharged on postoperative day 9 without complications.

Postoperative pulmonary function tests at 6 months showed marked improvement, with a forced expiratory volume in 1 second of 2.02 (85%) and diffusion capacity of the lung for carbon monoxide of 99%. A chest computed tomography scan in August 2009 revealed no evidence of recurrent disease and complete left lung aeration (Fig 1B). She has not required the use of inhalers and remains asymptomatic.

Comment

Although most SFTP arise in the pleura, an intraparenchymal or endobronchial location is a rare occurrence. To our knowledge, endobronchial SFTP has been documented in two prior reports. Histologic attributes have been described in an attempt to predict malignant potential. England and colleagues [7] described 223 patients, 141 (63%) benign and 82 (37%) malignant. They established the following features suggestive of malignancy: (1) mitotic counts exceeding 4/10 high-power fields, (2) necrosis, (3) hemorrhage, (4) pleomorphism, (4) stromal or vascular invasion, and (5) size exceeding 10 cm [7]. These pathologic features are helpful, but the absence of these characteristics is not completely predictive of benign behavior.

Perrot and colleagues [1] described a staging system based on pedunculated vs sessile attachment and malignant vs benign histology that predicted recurrence:

- stage 0: pedunculated tumor without signs of malignancy (<2% recurrence);
- stage I: sessile or “inverted” tumors without signs of malignancy (<8% recurrence);
- stage II: pedunculated tumor with histologic signs of malignancy (14% recurrence);
- stage III: sessile or “inverted” tumor with histologic signs of malignancy (63% recurrence); and
- stage IV: multiple synchronous metastatic tumors.

Other studies have described long-term survival to be 85% to 91% [2, 7, 8], with complete surgical resection being the most important prognostic factor [1, 3–5, 8]. In a recent report by Harrison-Phipps and colleagues [8], the average median survival for all types of SFTP in 84 patients was 24 years. In a previous study by England and colleagues [7], however,

malignant tumors metastasized or recurred in 55% of patients, eventually leading to death. Metastases have been reported in the liver, brain, spleen, peritoneum, adrenal glands, gastrointestinal tract, kidney, and bone [1].

Although there are anecdotal reports of good response to adjuvant radiotherapy in incomplete resections, use of induction and adjuvant therapies remain unclear [1, 5]. Adjuvant therapy in malignant sessile lesions should be considered, particularly in recurrent disease [1]. After resection, adjuvant therapy is generally not recommended in histologically benign tumors (sessile or pedunculated) or pedunculated lesions that appear malignant [1]. Given that these tumors can have high recurrence rates, regular follow-up with radiographic studies is advised.

Complete surgical resection with margin negativity is the mainstay of treatment. Intraoperative evaluation of margin status is recommended. In cases of endobronchial SFTP, sleeve resection of the airway with preservation of lung parenchyma is a feasible surgical option. Intrapericardial hilar release to reduce tension on the bronchial anastomosis and muscle flap coverage of the anastomosis to facilitate healing should be considered.

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Fig 1. Coronal reconstructed chest computed tomography image demonstrates (A) an enhancing 2.1- × 1.7-cm mass in the left main-stem bronchus (arrow) causing postobstructive atelectasis of the left lung preoperatively and (B) postoperative changes from endobronchial sleeve resection, with patent lumen and reexpansion of left lung without recurrence.

Fig 2.
Preoperative bronchoscopy reveals a completely occluding endobronchial mass in the left mainstem bronchus.

Fig 3. Gross specimen of the tumor demonstrates penetration through the bronchial wall with intraluminal (I) and extraluminal (E) protrusion. The arrow indicates bronchial wall cartilage.

Fig 4. (A) The tumor is shown penetrating between plates of cartilage (double arrow). The single arrow indicates bronchial epithelium (hematoxylin and eosin stain, whole mount). (B) The tumor is composed of dense spindle cells infiltrating between submucosal glands with characteristic thin-walled and ectatic vessels (hematoxylin and eosin stain at original magnification $\times 200$).