Comment

Liebow and Hubbell [3], the first to describe SH, suggested that this benign tumor was an endothelial proliferation; however, electron microscopic and immunohistochemical studies suggest that SH originates from epithelial cells [4, 5]. The exact histogenesis of SH thus remains uncertain [1, 5].

This is the first report to describe SH isolated to the mediastinum. One case of mediastinal SH was found among a series of 100 patients with pulmonary SH, but was considered to have arisen in the periphery of the lung and protrude into the mediastinum, mimicking mediastinal SH [6]. Many investigators consider that SH develops from type II alveolar epithelial cells, although this remains controversial. Positive TTF-1 staining supports the hypothesis that the tumor originated from respiratory epithelial cells [6]. In our patient, however, the tumor developed outside the lung.

Three mechanisms may account for the development of mediastinal SH. The first is metastasis from the lung to the mediastinum, although there was no evidence of tumor in the lung. All previously reported cases of lymph node metastasis from SH had evidence of primary tumor measuring at least 3.5-cm diameter in the lung [2, 6, 7]. The second mechanism is that the tumor could have originated from ectopic lung tissue, such as bronchogenic cysts. However, no ectopic lung tissue was found in the surgical specimen. The final mechanism is that the SH developed from the lung as a pedunculated pleural mass that slowly pulled away from the lung surface.

All three of these potential mechanisms are plausible; however, none can satisfactorily explain the apparent extrapulmonary location of the SH in our patient. Although we consider this to be the first documented case of SH isolated to the mediastinum, immunohistochemical studies suggest that the tumor arose from lung tissue. Our report demonstrates that, although rare, SH should be considered in the differential diagnosis of mediastinal masses.

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Postpneumonectomy Syndrome in Single Lung Transplantation Recipient Following Previous Pneumonectomy

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Successful single lung transplantation following previous pneumonectomy has not been reported in the literature. We report a patient with cystic fibrosis who underwent left single lung transplantation following right pneumonectomy 13 years previously. The outcome was adversely affected by postpneumonectomy syndrome.

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Respiratory failure is the common cause of death in patients with cystic fibrosis. Lung transplantation is an accepted treatment when these patients present with end-stage lung disease. By the nature of the suppurative lung disease, bilateral lung transplantation is generally required to remove the infection, and this is usually achieved by performing a bilateral single lung transplant or a heart and lungs transplantation [1–3]. Exacerbation of respiratory infection and hemoptysis are not uncommon during the course of the disease and when there is massive hemoptysis pulmonary resection could be lifesaving. We report a patient with cystic fibrosis who previously had a right pneumonectomy for recurrent hemoptysis. Subsequently, 13-years later, she underwent a left single lung transplantation. Postpneumonectomy syndrome was a significant and challenging problem following the lung transplantation. She remains alive at 30 months post-transplant.

The patient is a 40-year-old white female with cystic fibrosis diagnosed at birth, who received no treatment until 21 years old. Patient underwent right upper and middle lobectomy when she was 24 years old for recurrent infection and pneumothorax. She underwent a completion pnuemonectomy for recurrent hemoptysis when she was 27 years old. Her respiratory symptoms progressed and, therefore, she was evaluated for lung trans-

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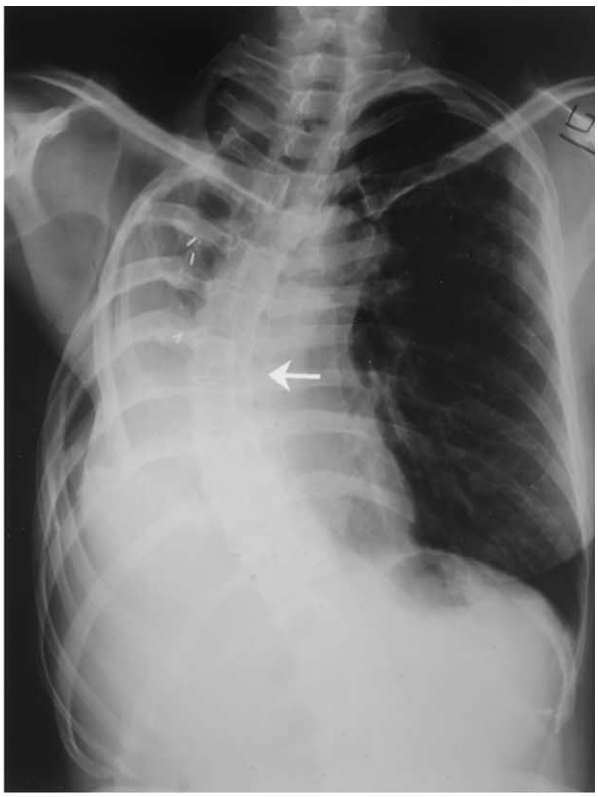
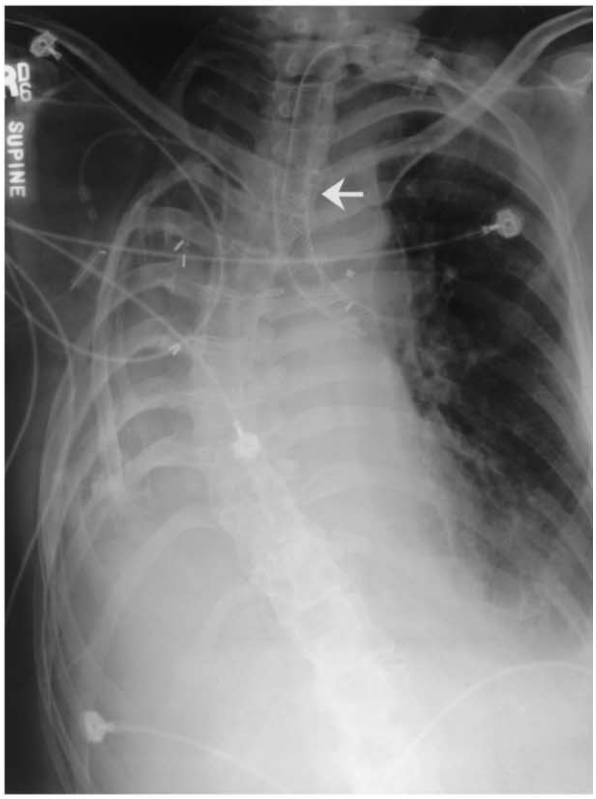


A

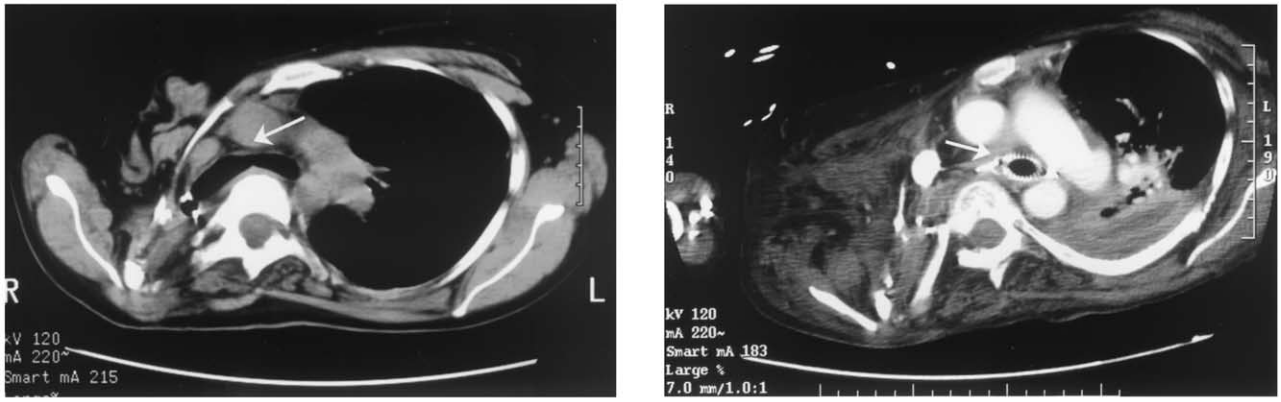


B

Fig 1. (A) Preoperative chest roentgenogram revealed a marked scoliosis with convexity toward the left (arrow) *and prominent interstitial markings in the remaining left lung field. (B) Postoperative chest roentgenogram revealed multiple wall stents in distal trachea and bronchus* (arrow).

plantation. She weighed 45 kg (85% IBW). Her pulmonary function testing revealed a forced expiratory volume (FEV_1) of 0.78 L/second, DLCO 11.76% of predicted value, $PaCO_2$ 67 mm Hg, PaO_2 103 mm Hg, and resting saturation 97%, all measurements were conducted at room air. Her 6-minute walk test was 1200 feet. Her chest roentgenogram revealed marked scoliosis with convexity towards the left and prominent interstitial markings in the remaining left lung field (Fig 1A). Her computed tomographic (CT) scan illustrated bronchiectasis in left upper lobe and lingula. She was listed for a left lung transplantation. Two weeks before transplantation, the patient developed respiratory failure requiring mechanical ventilation until transplantation.

In the view of the large pleural space and significant shifting of the mediastinum, the donor lung was oversized to the patient's predicted tidal volume by 37%. She was placed in a right semilateral position and her chest abdomen and groins were prepared and draped. A left anterolateral thoracotomy was performed entering the pleural cavity. In view of severe adhesions of the lungs, the femoral vessels were cannulated for cardiopulmonary bypass. The recipient lung was removed uneventfully. The lung transplantation was performed in a standard fashion, a 40 absorbable monofilament suture was used to anastomose the bronchus end-to-end using continuous suture to the membranous portion and interrupted figure-of-eight sutures to the cartilaginous portion. Pulmonary vessels were anastomosed with 40 nonabsorbable monofilament continuous sutures. Following the transplantation the cardiopulmonary bypass was easily weaned off and the patient was transferred to ICU in a very stable condition. Immediately in the post-transplant bronchoscopy it was noted her left main stem bronchus and trachea were compressed posteriorly. Despite good gas exchange and clear lung field she could not be weaned off the ventilator. On postoperative day 10, she developed sepsis secondary to perforated duodenal ulcer. Exploratory laparotomy and gram patch of perforated ulcer were done without further complications. After this operation, in view of the tracheobronchial narrowing, wall stents were placed in distal trachea and left main bronchus. Tracheostomy was also done to facilitate the weaning process. She was able to be weaned off positive pressure ventilation for transient periods but supported most of the times with portable ventilatory support. Surgical correction for postpneumonectomy syndrome was discussed with the patient and family but they declined. The patient remains alive at 30 months following single lung transplant.



A B

Fig 2. (A) Preoperative computed tomographic (CT) scan demonstrating stretching of the distal trachea, the main bronchus, and the pulmonary artery over the vertebral column (arrow). *(B) Postoperative CT scan, in addition to the structures mentioned in Fig 2 (A), reveals the stent in the distal trachea* (arrow).

Comment

Anatomic changes after pneumonectomy had been well described. Significant shifting of the mediastinum and profound scoliosis can occur in some, particularly in the young following a long period of a pneumonectomy. Postpneumonectomy syndrome is a rare complication due to severe anatomic changes in the postoperative chest causing extrinsic compression of the distal trachea or main stem bronchus by the vertebral column [4, 5]. This complication is usually seen in patients who underwent right pneumonectomy, especially in childhood or early adulthood. The syndrome occurs when the heart and remaining mediastinal structures shift rightward and posteriorly into vacated pleural space coupled with severe hyperinflation of the remaining lung. Significant scoliosis also develop after pneumonectomy and the convexity of the spine is toward the contra-lateral side. The result is stretching of the distal trachea, the remaining main bronchus, and the pulmonary artery over the vertebral column (Fig 2A). When this occurs symptomatic surgical treatment aimed at correcting the anatomic derangement is recommended. The current procedure of choice is repeat thoracotomy on the pneumonectomy side for freeing the mediastinum of adhesion and filling the pleural space with an expandable prosthesis.

Successful single lung transplantation following a long period after previous pneumonectomy has not been reported in the literature. In this case, we demonstrated that this procedure could be done safely. However, postpneumonectomy syndrome causing compression of the bronchus and trachea can be a significant problem if the interval between pneumonectomy and transplantation is long. In retrospect, the preoperative chest roentgenogram and CT scan was suggestive of postpneumonectomy syndrome including severe scoliosis, with compression of the tracheobronchi. Before transplantation the patient was supported by positive pressure ventilation, preoperative bronchoscopy was not performed, and, therefore, the tracheal compression before transplantation was not verified.

Stent placement for extrinsic bronchial compression including in postpneumonectomy syndrome has been described in the literature. We were able to partially correct the tracheobronchial narrowing with multiple wall stents (Figs 1B and 2B). Our follow-up to date demonstrated patency of the stents at 30 months. Surgical correction of postpneumonectomy syndrome is possible even in the presence of immunosuppression regimen. We recommend a bronchoscopic examination in all patients with pneumonectomy before transplantation. If tracheobronchial compression is observed this should be corrected at the time of transplantation.

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