A 68-Year-Old Man With Intractable Dyspnea and Wheezing 45 Years After a Pneumonectomy*

Septimiu D. Murgu, MD; and Henri G. Colt, MD, FCCP

A 68-year-old man with a 20-year history of wheezing presented with a recent increase in dyspnea, productive cough, and inability to clear secretions. He denied fevers, rigors, chest pain, palpitations, lightheadedness, syncope, headaches, nasal symptoms, or gastroesophageal reflux. During the last 15 years, he received treatment with albuterol and ipratropium inhalers in addition to daily nebulizers, prednisone, salmeterol, montelukast, and fluticasone for presumed asthma. His symptoms had worsened despite being compliant with medication. In fact, during the 5 years prior to our evaluation, he had been hospitalized 12 times for asthma exacerbations, bronchitis, and recurrent respiratory infections. The patient was a nonsmoker, had no second-hand smoke exposure, and had no occupational exposures to toxic substances. Family history was not significant. The patient had no known drug allergies. At the age of 5 years, however, he had aspirated a foreign body that was incompletely removed, resulting in recurrent respiratory infections and a left pneumonectomy at age 23.

Physical examination revealed rhonchi and wheezing over the trachea and right hemithorax. Transmitted bronchial sounds were audible on the left. The trachea was slightly deviated to the left, but the rest of the examination was normal. The results of blood tests consisted of normal CBC count and normal basic metabolic panel. Dobutamine stress echocardiography was normal. Pulmonary function testing showed severe reduction in expiratory volumes (FVC, 43% of predicted; FEV₁, 39% of predicted; FEV₁/FVC ratio, 0.63; peak expiratory flow, 29% of predicted). There was worsening in spirometry values after administration of bronchodilators. The chest radiograph is shown in Figure 1.

What is the cause of this patient’s symptoms?
What additional studies are needed to establish the diagnosis?

*From Pulmonary and Critical Care Medicine, Department of Medicine, University of California School of Medicine, Irvine, CA.

Manuscript received August 29, 2005; revision accepted December 10, 2005.

Reproduction of this article is prohibited without written permission from the American College of Chest Physicians (www.chestjournal.org/misc/reprints.shtml).

Correspondence to: Septimiu Murgu, MD, UCI Medical Center, 101 the City Drive South, Building 53, Room 119, Rt 81, Orange, CA 92868; e-mail: smurgu@uci.edu

Figure 1. Chest radiograph (posteroanterior view).
Diagnosis: Tracheobronchomalacia caused by postpneumonectomy syndrome and diagnosed by dynamic CT, cine MRI, or dynamic bronchoscopy.

The differential diagnosis of dyspnea in patients with a previous pneumonectomy includes recurrence of primary disease (ie, malignancy), pulmonary hypertension, progression of underlying lung dysfunction, thromboembolism, congestive heart failure, and postpneumonectomy syndrome. Postpneumonectomy syndrome is characterized by dyspnea and recurrent infections in the remaining lung, and is caused by marked mediastinal shift (counterclockwise after right pneumonectomy or clockwise after left pneumonectomy), rotation of the heart and great vessels, and herniation of the remaining lung into the contralateral hemithorax.

Postpneumonectomy syndrome is usually a delayed finding after pneumonectomy, with an estimated incidence of 1 in 640 pneumonectomies. It is more frequent when pneumonectomy is performed at an early age, probably because of increased elasticity and compliance of the lung and mediastinum during childhood and early youth. Originally described after right pneumonectomy, postpneumonectomy syndrome is also documented after left pneumonectomy, especially in patients with a right-sided aortic arch. Postpneumonectomy syndrome is relatively rare after left pneumonectomy in patients with left-sided aortic arches because the descending aorta acts as a barrier to the marked leftward shift of cardiomedialstinal structures. Regardless of the side of pneumonectomy, however, airway compression by the thoracic spine, descending thoracic aorta, ligamentum arteriosum, or pulmonary artery can cause tortuosity and stretching of the trachea and bronchial compression. Secondary tracheobronchomalacia may thus develop over time.

Tracheobronchomalacia is defined as weakness of cartilaginous structures of the tracheobronchial wall. The trachea and main bronchi lose their usual degree of rigidity, and the airway walls come closer together, reducing the caliber of the airway lumen, particularly during expiration. Depending on whether the anterior, lateral, or both airway walls are weakened, tracheobronchomalacia can be classified morphologically as being of the crescent, saber sheath, or circumferential type after bronchoscopic or radiographic examinations. The adult forms of tracheobronchomalacia are classified as either idiopathic or acquired during the course of other illnesses, and are usually disorders of middle-aged and older persons. Malacia may occur alone or may be accompanied by excessive dynamic airway collapse caused by increased invagination of the membranous posterior airway wall during expiration. Etiologies include chronic inflammation; systemic diseases such as relapsing polychondritis; recurrent infections; sequelae from infections such as tuberculosis; pressure necrosis from overinflated tracheostomy or endotracheal tube cuffs; unrecognized tracheobronchomalacia caused by increased invagination of the membranous posterior airway wall during expiration. Etiologies...
degree of collapse, and correlates well with bronchoscopy findings. Cine MRI can document the effect of pulsating vessels on adjacent bronchi and may also demonstrate airway collapsibility without exposing patients to ionizing radiation or iodinated contrast media. In most cases of postpneumonectomy syndrome, there is a marked shift of the mediastinum and the postpneumonectomy space is severely reduced.

Bronchoscopy confirms narrowing from extrinsic compression and may reveal tracheobronchomalacia. Flexible bronchscopy is preferable to rigid bronchoscopy for diagnosis because the patient is able to breathe spontaneously and follow commands to perform deep breathing, forced exhalation, and cough in order to elicit airway collapsibility (Fig 3). During these dynamic bronchoscopic examinations, changes in bronchial and tracheal caliber can be measured, the extent of collapse noted, and the morphology of airway narrowing can be classified as being of the crescent, saber-sheath, or circumferential type.

Therapeutic alternatives for patients with tracheobronchomalacia include disease-specific medical treatment; conservative management using bronchodilators, noninvasive ventilatory support, and methods of secretion clearance; minimally invasive modalities such as airway stent insertion; and open surgical techniques that remove the diseased segment or reinforce the airway structure. Choice of therapy depends on the etiology, extent, type, and severity of airway abnormalities noted. In some patients, tracheobronchomalacia is an incidental finding that does not require treatment. In symptomatic patients, therapy should be individualized. Underlying diseases such as COPD, asthma, or relapsing polychondritis should be treated. Additional therapy might depend on extent of the disease. For focal malacia, surgical resection of the involved segment is often proposed in experienced centers. Stent insertion can be offered to nonsurgical candidates or as a therapeutic trial in patients being considered for surgery. In patients with multifocal or diffuse disease, a stent trial might also be warranted. Noninvasive positive pressure ventilation can be used as adjunctive therapy for patients with residual symptoms because the positive airway pressure acts as a pneumatic stent. Open surgical techniques such as membranous wall tracheoplasty should be reserved for good surgical candidates and offered only in experienced centers.
The treatment of tracheobronchomalacia in the setting of postpneumonectomy syndrome deserves special attention because published experience is very limited. Surgical repositioning of the mediastinal structures may be curative if malacia is minimal or has not occurred in the obstructed airway. In patients with significant malacia, however, surgical treatment prevents recurrent mediastinal shift but may not reverse dynamic central airway obstruction. Tracheobronchial resections of malacic segments and aortic division with bypass have been attempted, but malacia is usually an indicator of poor outcome. The use of airway stents for postpneumonectomy syndrome is rarely reported and has been used mainly in cases of respiratory failure or when surgery was declined.

In the case described herein, tracheobronchomalacia was diagnosed 45 years after left pneumonectomy in a patient with a left-sided aortic arch. Prolonged stretching and compression of the trachea and the right main bronchus probably caused weakness of the cartilage, resulting in secondary tracheobronchomalacia. The degree of softening of the cartilaginous rings may be related to the length of time the airway was compressed. This may explain why our patient had symptoms many years after left pneumonectomy. Paired inspiratory/expiratory dynamic CT showed severe, diffuse crescent type of tracheobronchomalacia as well as postpneumonectomy syndrome. The marked delay in onset of symptoms may also be explained by the lesser degree of mediastinal shift (Fig 2). Dynamic bronchoscopy revealed severe reduction in the airway lumen due to diffuse collapse of the anterior cartilaginous structures consistent with crescent type of tracheobronchomalacia (Fig 3) extending throughout the lower trachea, right main bronchus, and bronchus intermedius. Rigid bronchoscopy was performed in order to insert an 18 × 40-mm studded silicone stent (Bryan Corporation; Woburn, MA) into the right main bronchus and bronchus intermedius as well as an 18 × 50-mm ringed silicone stent (Hood; Pembroke, MA) into the trachea (Fig 3). After several flexible bronchoscopies performed for therapeutic removal

![Figure 3. Dynamic bronchoscopy shows flattening of the cartilaginous rings at the carinal level leading to near complete occlusion of the right mainstem bronchus during expiration (top left, A). The distal trachea and the entrance to the right mainstem bronchus are visualized during bronchoscopy after silicone stent insertion. There is significant improvement in the airway cross sectional area (top right, B). Dynamic CT after stent insertion shows maintenance of patent tracheal (bottom left, C) and bronchial (bottom right, D) lumen during expiration.]()
of secretions during the first few weeks following stent insertion, inflammatory changes resolved, secretions cleared, and the patient has shown significant clinical improvement without hospitalization for the past 8 months.

**Clinical Pearls**

1. **Tracheobronchomalacia** is characterized by weakening of the tracheobronchial cartilaginous wall, resulting in narrowing of the airway lumen during expiration with or without excessive dynamic airway collapse.

2. Diagnosis of tracheobronchomalacia should be suspected in the presence of predisposing conditions or risk factors in patients with symptoms mimicking asthma or chronic bronchitis. Dynamic radiologic studies such as paired inspiratory/expiratory CT or cine MRI and dynamic bronchoscopy confirm the diagnosis and help define the severity and extent of disease.

3. The adult forms of tracheobronchomalacia may be idiopathic or secondary. Long-standing compression of the tracheobronchial tree due to postpneumonectomy syndrome may account for rare cases of secondary tracheobronchomalacia.

4. Postpneumonectomy syndrome is a rare, delayed complication after pneumonectomy, which is caused by marked mediastinal shift and compression of the airways by the thoracic spine, descending thoracic aorta, ligamentum arteriosum, or pulmonary artery.

5. Treatment for postpneumonectomy syndrome-related tracheobronchomalacia should be individualized based on severity, extent, and morphology of disease. Airway stent insertion is a reasonable consideration that may improve symptoms, ventilatory function, and quality of life.

**Suggested Readings**


Murgu SD, Colt HG. Tracheobronchomalacia and excessive dynamic airway collapse: novel diagnostic tools clarify the issues. Pulm Perspect 2005; 22:7–10