Nonresolving Spontaneous Pneumothorax in a 38-Year-Old Woman*

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A 38-year-old woman had a history of intermittent chest discomfort and dyspnea over many years. She never sought medical attention for these symptoms. Her only chest radiograph was taken 20 years ago when she started college. The film was unavailable.

Her present illness began during air travel when she experienced chest pain and dyspnea. Upon landing, she came to the emergency room of the hospital. An x-ray film revealed a tension pneumothorax of the left side of the chest. Also noted on the x-ray film was a faint lucent “streak-like” shadow extending into the lower zone of the left lung. The left thoracic cavity appeared to be bigger than the right cavity, with widening of the rib spaces. The tension was relieved with a catheter and a tube inserted on the left side of the chest (Fig 1). Shortly after an initial air leak, the chest tube stopped bubbling. Another x-ray film revealed a persistent pneumothorax. After chest tube occlusion was excluded, a second tube was inserted into the chest. Again, cessation of the air leak occurred very shortly after insertion just as before, but the pneumothorax persisted. Bronchoscopy ruled out any endobronchial lesion that prevented lung reexpansion.

When an 18-gauge needle was inserted into the left hemithorax, air would escape both spontaneously and with coughing. However, the chest tube would not demonstrate an air leak. Perplexed with a similar malfunction of a third chest tube, the patient underwent thoracotomy, which revealed a large hernial sac-like structure with an inner cavity into which a suction catheter could be advanced to 20 cm before encountering any resistance. Upon thoracotomy, a giant cyst was removed (see Fig 2). The patient recovered uneventfully and was discharged after 3 days.

What diagnosis can explain the patient’s clinical presentation?

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Diagnosis: Ruptured bronchogenic cyst

Bronchogenic cyst has been reported to present as a pneumothorax in only one case report. We hereby present the second documented case and, to the best of our knowledge the only one in which the cyst progressed to a size of 20 cm into adulthood. It presented as a management problem of persistent pneumothorax with repeated occlusion by the floppy, redundant cyst wall upon application of suction pressure through the chest tube (Fig 3).

Bronchogenic cysts result from an abnormal budding of the tracheal diverticulum between the 3rd and 6th week of gestation. They may be mediastinal or pulmonary in location. Most often, they are unicocular. They usually are thin-walled and tend to conform to the anatomic space in which they arise. On histologic examination, they may contain some normal bronchial elements, ie, smooth muscle, elastic tissue, fibrous tissue, and cartilage. The cyst may be filled with mucoid material and usually is lined with pseudostratified ciliated columnar epithelium (see Fig 4, 5).

In a review of 13 series comprising 2,163 patients with mediastinal lesions, the prevalence of bronchogenic cysts was 6%. Adults are mostly asymptomatic. Symptomatic cysts are seen most commonly in children. Respiratory insufficiency in children results from distortion of airways. Chest pain, dysphagia, dyspnea, cough, and hemoptysis can also occur. Infection is the most common serious complication in adults. Malignant transformation, a rare occurrence, is found in adults. Pneumothorax has been documented in a single case report.

A bronchogenic cyst is usually discovered incidentally on a routine chest x-ray film. A CT scan confirms its cystic nature (low Hounsfield units). Needle aspiration may help to support the diagnosis.

Treatment: Options available are observation, fine-needle aspiration, mediastinoscopy for aspiration and biopsy, thoracoscopy, and thoracotomy or mediastinoscopy for resection.

In adults with small asymptomatic cysts, follow-up studies may be done with periodic roentgenography of the chest. An air-fluid level suggesting tracheobronchial communication, the presence of malignancy, the development of a symptomatic recurrence, or enlargement evidenced on a follow-up roentgenographic study mandates complete surgical removal of the cyst.

Figure 2. Gross specimen showing a large thin-walled cyst.

Figure 3. Diagram of occlusion of the chest tube by the floppy, redundant cyst wall.
Because of the higher incidence of complications associated with intrapulmonary bronchogenic cysts, they usually should be removed.

**Clinical Pearls**

1. Although unusual, a ruptured bronchogenic cyst should be considered in the differential diagnosis of a persistent spontaneous pneumothorax.

2. In the event of a nonresolving pneumothorax, consider occlusion of the chest tube orifice by the floppy, redundant cyst wall.

3. Unilateral widening of the rib spaces may serve as a clue to consider space-occupying embryologic abnormalities that may lead to thoracic cavity asymmetry.

**Suggested Readings**

