Mediastinal Teratoma With Pulmonary Involvement Presenting as Massive Hemoptysis in 2 Patients

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Massive hemoptysis is described in many disease processes. However, a mediastinal teratoma is rarely considered in a patient presenting with massive hemoptysis. Since a mediastinal teratoma has no specific symptoms, its definitive diagnosis is difficult before surgical intervention. Flexible bronchoscopy can be diagnostic in cases of a mediastinal teratoma with involvement of the bronchial tree. We report 2 cases of hemoptysis caused by mediastinal teratoma with bronchial communication. Key words: mediastinal teratoma with airway communication; hemoptysis; trichoptysis. [Respir Care 2010;55(8):1094–1096. © 2010 Daedalus Enterprises]

Introduction

Mediastinal teratomas with pulmonary involvement are extremely rare. They usually do not produce symptoms and are often discovered on screening chest radiograph. We encountered 2 cases of this rare condition, both presenting with massive hemoptysis. Both patients underwent surgical resection with histological diagnosis. After surgery their symptoms resolved.

Case Report 1

A 58-year-old woman presented to the emergency department with massive hemoptysis. Her history included repeated admissions, over 20 years, for massive hemoptysis. Twenty years before she was noted to have a left-upper-lobe lung lesion. She underwent an exploratory thoracotomy that found inflammation without evidence of malignancy. Over the decades she continued to have hemoptysis and was treated medically. On admission she

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The authors have disclosed no conflicts of interest.

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underwent an extensive diagnostic evaluation, including sputum cultures and cytology, which revealed no evidence of tuberculosis or malignancy.

Chest imaging (Fig. 1) revealed an elevated left hemidiaphragm, destruction of the left upper lobe, and a soft tissue density with cavity formation. A few days after cessation of the hemoptysis, flexible bronchoscopy (see Fig. 1) revealed hair-like structures within a mass lesion. Teratoma was highly suspected. She underwent a left posterolateral thoracotomy. The tumor was difficult to resect, and severe adhesions were noted. An intra-pericardial approach was chosen, with extreme care taken to preserve vital structures such as the phrenic nerve, recurrent laryngeal nerve, aorta, and subclavian vessels. The ill-defined mass was connected to the thymus, which required resection along with the left upper lobe.

Pathology revealed a mature teratoma mainly composed of adipose tissue. There were some spaces lined with skin tissue, with epidermis, hair follicles, sebaceous glands, and sweat glands. No immature component was found within the tumor. Some atrophic thymic tissue was noted around the teratoma. The teratoma had compressed the nearby lung tissue and protruded into the bronchial lumens, where keratin substance and hair shafts were seen. There was bronchiectasis with marked acute and chronic inflammatory cell infiltration in the bronchial wall, mucous gland hyperplasia disruption of muscle layer, and squamous metaplasia of respiratory epithelium. The lung tissue around the bronchi and teratoma also displayed congestion and hemorrhage. The peribronchial lymph nodes showed reactive hyperplasia. Almost 8 years after surgical excision, she has had no repeat of the hemoptysis.

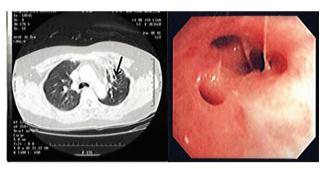


Fig. 1. Patient 1. Computed tomogram (left) shows communication of a ruptured mediastinal cystic teratoma with the left upper bronchial tree (arrow). Bronchoscopy (right) shows hair-like material at the orifice of the anterior segment of the left upper lobe.

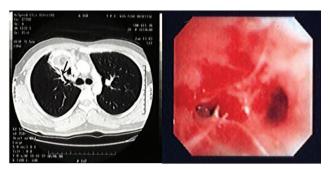


Fig. 2. Patient 2. Computed tomogram (left) shows a ruptured mediastinal cystic teratoma with intrapulmonary bronchial invasion (arrow). Bronchoscopy (right) shows a hair-like material at the orifice of the anterior segment of the right upper lobe.

Case Report 2

A 28-year-old man was admitted through the emergency department with massive hemoptysis. He had had hemoptysis at the age of 14. At that time a lung abscess was diagnosed and he was treated medically. Eight years later he had another massive hemoptysis but did not seek medical attention. Over the next 6 years he had intermittent episodes of hemoptysis, which progressively worsened.

On admission his vital signs, physical examination, and laboratory results showed no abnormalities. Chest radiography demonstrated a mass in the right upper lobe, without mediastinal widening. Computed tomography (Fig. 2) revealed a 4.7×6.8 cm heterogeneously attenuating mass with a fat component in the right anterior mediastinum with extension into the right upper lobe. Flexible bronchoscopy (see Fig. 2) revealed irregular mucosa and white hair-like material from the orifice of the anterior segment of the right-upper-lobe bronchus. Mediastinal teratoma with bronchial communication was highly suspected.

A right upper lobectomy and total excision of the mediastinal tumor and thymus were performed. Gross examination showed a brown, elastic, $7 \times 7 \times 4.5$ cm tumor

involving the right upper lobe and the mediastinum. Pathology results indicated a mature cystic teratoma, mainly lined by stratified squamous epithelium, but occasionally lined with respiratory epithelium. There was adipose tissue, hair follicles, sweat glands, sebaceous glands, and smooth-muscle bundles on the cyst wall. There were also focal intestinal mucosa. No immature element was found in the teratoma, but residual thymic tissue was observed near the teratoma. Five years after surgery he has not had a repeat of the hemoptysis.

Discussion

Massive hemoptysis has been described in many pulmonary diseases and some cardiovascular diseases. Mediastinal teratoma is asymptomatic in up to 53% of cases, and is frequently discovered incidentally on chest radiograph.^{1,2} The usual symptoms of mediastinal teratoma are pain, cough, and dyspnea. The most common symptom is chest pain. Cough is presumably the result of compression or irritation of the airways. An unusual but pathognomonic symptom is the expectoration of hair (trichoptysis). Hemoptysis is rare, but may result from bronchial bleeding from irritation by the teratoma cyst.3 Mature teratomas in the mediastinum are typically located in the anterior aspect of the mediastinum, and approximately 13% of these teratomas extend to other mediastinal components.4 Mediastinal teratoma with pulmonary involvement is rare. Mediastinal teratoma is most often recognized in adolescence or early adulthood, although occasional cases in older individuals have been reported.

Most mediastinal teratomas with pulmonary involvement have, for unknown reasons, a predilection for the left upper lobe.⁵ Mature mediastinal teratomas are benign, do not infiltrate adjacent organs, and can be resected completely with good results. These tumors are characterized by cysts lined with mature stratified squamous epithelium, and by bronchial and intestinal walls lined with epithelium, and possibly containing various mature tissues. Immature mediastinal teratomas are characterized by the presence of various tissues that histologically resemble embryonic structures. They are rare malignant neoplasms that can metastasize and recur.⁶

Our patients had no trichoptysis, and their main symptom was hemoptysis. Chest radiograph showed a mass lesion in the anterior mediastinum. Computed tomography disclosed a mass and bronchial tree communication. Prior to tissue diagnosis in surgical intervention, the finding of hair-like material during bronchoscopy can confirm the diagnosis of mediastinal teratoma with pulmonary involvement.

Mediastinal teratomas are difficult to diagnose preoperatively because these patients usually present with non-specific symptoms.⁷ A computed tomogram showing a tumor within the bronchial tree, in addition to broncho-

scopic findings of hair within the mass, strongly suggests a mediastinal teratoma with pulmonary involvement. Surgical excision is the optimal treatment for intractable hemoptysis.⁸

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