Spontaneous Hemo-pneumothorax as a Result of Venous Hemangioma
A Unique Case?

Spontaneous hemopneumothorax is a rare clinical entity that can be life-threatening, with a significant increase in mortality if not recognized and treated in time. We report the case of a young man who presented to us with nontraumatic spontaneous hemopneumothorax. Histologic examination of lung tissue showed the most likely cause of the bleeding to be venous hemangioma, which to the best of our knowledge has not been reported before. (Tex Heart Inst J 2006;33:91-2)

Case Report

A 24-year-old man of Chinese origin presented at the emergency department of our institution with sudden onset of chest pain associated with shortness of breath. A chest radiograph showed pneumothorax with an air-fluid level and a collapsed lung on the right side. An intercostal tube drained 1,200 mL of blood; this was associated with sudden cardiovascular compromise, which required fluid and blood resuscitation. A follow-up computed tomographic scan of the chest showed a large amount of fluid in the right thorax with mediastinal shift, but with no major vessel injury.

The patient was immediately taken to the operating theatre, where he underwent a posterolateral thoracotomy. Perioperatively, apart from massive blood clots, we found a bullous lesion in the right upper lobe. A small blood vessel overlying the lesion was actively bleeding (Fig. 1). There was a corresponding spurting vessel in the chest wall. These were clipped and a bullectomy was performed. Perioperatively, the patient required 4 units of blood. Postoperatively, he was moved to the intensive care unit, where he was extubated after 3 hours; the next day, he was moved to the ward in a stable condition. The chest drains were removed after a couple of days, and he was discharged from the hospital within a week. He has been doing very well since discharge. Regular follow-up at the cardiothoracic outpatient clinics have shown him to be perfectly asymptomatic and back to his normal routine.

Microscopy. Sections of lung showed bullous formation, areas of chronic inflammation, and evidence of old hemorrhage. A small collection of dilated vessels that demonstrated an uneven distribution of smooth muscle was identified adjacent to an area of chronic inflammation that contained cholesterol clefts and hemosiderin. The features were suggestive of venous hemangioma.
The spontaneous form of hemopneumothorax is reported in only 1% to 12% of spontaneous pneumothoraces. Spontaneous hemopneumothorax is defined by Ohmori and colleagues as the accumulation of more than 400 mL of blood in the pleural cavity in association with spontaneous pneumothorax.

There are said to be 3 mechanisms of bleeding in hemopneumothorax. First, bleeding can result from a torn adhesion between the parietal and visceral pleurae. According to Barry and co-authors, the site of bleeding is usually a small noncontractile vessel on the parietal pleura, where vascular adhesions exist. Second, bleeding may result from the rupture of vascularized bullae and underlying lung parenchyma. Third, it may result from torn congenitally aberrant vessels between the parietal pleura and bullae. In their series of patients with spontaneous hemopneumothorax, Tatebe and co-workers reported the pathologic examination of vascular specimens from 2 patients who had aberrant vessels. Those authors found mucoid degeneration and sclerotic changes in the arterial wall and fibrosis in the intima and media of the aberrant vessels.

To the best of our knowledge, our case of hemopneumothorax is the only one reported with the histologic diagnosis of venous hemangioma.

Spontaneous hemopneumothorax is a rare clinical entity that is life-threatening if diagnosis and intervention are delayed. This has to be borne in mind by the surgical registrar or resident when admitting the patient. Early recognition must be complemented by prompt resuscitation and monitoring, then followed as quickly as possible by thoracotomy and resection or clipping of the offending lesion. Prompt surgical intervention in this situation gives excellent results, with negligible morbidity and mortality.

References