Intrapleural Rupture of Pulmonary Arteriovenous Aneurysm*
Report of a Case

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Pulmonary arteriovenous aneurysm remains a rare, but well-defined entity. The clinical picture of cyanosis, clubbed fingers and localized bruit associated with roentgenologic evidence of a pulmonary parenchymal mass is well known. In the usual case, the diagnosis is confirmed by angiocardiography and resection performed with excellent results. However, the intervention of complications may so confuse the clinical picture as to make diagnosis and treatment quite difficult. One such complication, intrapleural rupture, is the subject of this report.

In contrast to intrabronchial rupture which produces hemoptysis and occurs in approximately 25 per cent of reported cases, intrapleural rupture is decidedly rare and has been reported on only eight previous occasions.* The rarity of this complication, as well as the emergent nature of the operative management of such patients, prompted the following case report.

Case Report:
E. M., a 64-year-old white woman, was admitted to St. Mary’s Hospital on November 17, 1965 with a history of rapid onset of dyspnea and left chest pain. She had been known to have hereditary hemorrhagic telangiectasia for ten years and for that period of time, had been known to have a mass in her left lower lobe on x-ray film which was presumed to be a pulmonary arteriovenous aneurysm (Fig. 1). She had previously declined any further diagnostic or surgical procedures. Upon admission, chest x-ray films were obtained which revealed massive left pleural effusion with mediastinal shift and impingement upon the right lung (Fig. 2). Aspiration of the left hemithorax revealed gross blood and a chest catheter was placed which drained approximately 250 ml of blood, but despite this, the patient’s condition failed to improve and the diagnosis of clotted hemothorax was presumed.

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Figure 1: Erect PA and left lateral chest x-ray films showing the pulmonary arteriovenous aneurysm in left lower lobe (arrows).
On November 18, 1965, following mild deterioration of her condition, emergency left thoracotomy was performed and approximately 1,000 ml of clotted blood was noted to be present in the left hemithorax.

Because of the patient's extremely precarious position, standard left lower lobectomy was performed without mobilization of the lobe, following identification of the ruptured aneurysm in the lower lobe. The pulmonary vessels were ligated in the fissure prior to mobilization. The patient tolerated this procedure well, did well postoperatively and was discharged from the hospital on November 25, 1965. The pathologic report was pulmonary arteriovenous aneurysm (Fig. 3). She has continued to do well and is now asymptomatic eight months after surgery. At present, she is doing well and her postoperative chest x-ray films show only the usual changes and no evidence of additional arteriovenous aneurysms is noted (Fig. 4).

**COMMENT**

This patient presented an unusual, frequently fatal, complication of pulmonary arteriovenous aneurysm. Her precarious condition precluded definitive diagnostic angiocardiographic studies. The diagnosis was founded on the basis of the physical findings of hereditary hemorrhagic telangiectasia coupled with the rapid onset of massive hemothorax. Mediastinal shift produced by the clotted hemothorax failed to yield to intrapleural catheter drainage and ultimately forced surgical intervention.

Lobectomy was performed because it was thought to be safe and expedient. Unless there are multiple aneurysms, it is recommended as the treatment of choice by Burford and Ferguson. Proximal vessel control was established prior to dissection in the region of the aneurysm because of its being adherent to the diaphragm. In this manner, further hemorrhage from the ruptured aneurysm which was tamponaded was avoided. Local excision has been recommended by Bosher and associates, and
### Chart 1—Summary of Reported Cases of Intrapleural Rupture of Pulmonary Arteriovenous Aneurysms

<table>
<thead>
<tr>
<th>Reference No.</th>
<th>Authors</th>
<th>Year</th>
<th>Age and Sex</th>
<th>Hereditary Hemorrhagic Telangiectasia</th>
<th>Location of Aneurysm</th>
<th>Operation</th>
<th>Results</th>
<th>Necropsy</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Wilkening</td>
<td>1917</td>
<td>23 yrs. - Female</td>
<td>Yes</td>
<td>Multiple (3), bilateral</td>
<td>No</td>
<td>Sudden death 5 yrs. after diagnosis</td>
<td>1500-200 ml blood in left pleural cavity</td>
</tr>
<tr>
<td>2</td>
<td>Verse</td>
<td>1943</td>
<td>3 days - Male</td>
<td>No</td>
<td>Multiple, bilateral</td>
<td>No</td>
<td>Sudden death</td>
<td>Massive left hemothorax</td>
</tr>
<tr>
<td>3</td>
<td>Erfg, et al</td>
<td>1949</td>
<td>22 yrs. - Male</td>
<td>No</td>
<td>Solitary, left upper lobe</td>
<td>No</td>
<td>Sudden death after exertion; 2 yrs. after diagnosis</td>
<td>1000 ml blood in left pleural cavity Aorto-pulmonic septal defect</td>
</tr>
<tr>
<td>4</td>
<td>Armentrout and Underwood</td>
<td>1950</td>
<td>25 yrs. - Female</td>
<td>Yes</td>
<td>Solitary, left lower lobe</td>
<td>No</td>
<td>1500 ml blood aspirated from left chest. Doing well at age 46</td>
<td></td>
</tr>
<tr>
<td>5</td>
<td>Heyde</td>
<td>1954</td>
<td>53 yrs. - Female</td>
<td>Yes</td>
<td>Solitary (?) left lower lobe</td>
<td>No</td>
<td>Sudden death 7 months after diagnosis. Chest x-ray showed opaque left hemothorax. (Presumptive diagnosis)</td>
<td>No</td>
</tr>
<tr>
<td>6</td>
<td>Purdue and Muras</td>
<td>1957</td>
<td>28 yrs. - Female</td>
<td>Yes</td>
<td>Solitary right upper lobe</td>
<td>Apical and Anterior Segmental Resection</td>
<td>Sudden onset, 1500 ml right hemothorax. Recovered</td>
<td></td>
</tr>
<tr>
<td>7</td>
<td>Hodgson, et al</td>
<td>1959</td>
<td>16 yrs. - Male</td>
<td>Yes</td>
<td>Not stated</td>
<td>No</td>
<td>Sudden death</td>
<td>Massive hemothorax</td>
</tr>
<tr>
<td>8</td>
<td>Brummelkamp</td>
<td>1961</td>
<td>37 yrs. - Female</td>
<td>Yes</td>
<td>Solitary left upper lobe</td>
<td>Local Excision</td>
<td>Sudden onset 1000 ml left hemothorax. Recovered</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Dalton</td>
<td>1965</td>
<td>64 yrs. - Female</td>
<td>Yes</td>
<td>Solitary left lower lobe</td>
<td>Lobectomy</td>
<td>Sudden onset 1000 ml left hemothorax. Recovered</td>
<td></td>
</tr>
</tbody>
</table>
doubtless is the procedure of choice in cases subjected to elective resection.

In a recent review of the literature, only eight previously reported cases could be found. It will be noted that two of these (cases 4 and 5) are presumptive diagnoses only. The case of Armentrout and Underwood (in which the diagnosis was based on aspiration of blood from the pleural space 21 years prior to evaluation by the authors) is excluded. The only survivors are those treated by surgery. Thus, the only acceptable method of management in this condition is emergency surgical resection.

Not included in this group are two case reports of hemothorax due to rupture of an actual pulmonary (subpleural) telangiectasis. This was first reported by Bowers11 in 1936, and an additional case was reported by Livingston and Carr12 in 1956. These are mentioned in this report in the interest of completeness.

In conclusion, it should be re-emphasized that once the diagnosis of intrapleural rupture of a pulmonary arteriovenous aneurysm is suspected, the patient should be prepared for thoracotomy as quickly as possible, as this will prove to be diagnostic and, in all reported cases, life saving. The occurrence of this lethal complication, even though rare, serves once again to underscore the importance of early elective resection of pulmonary arteriovenous aneurysms as soon as the diagnosis is made.

REFERENCES
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CONGENITAL DIAPHRAGMATIC HERNIA

Approximately 75 per cent of infants born with diaphragmatic defects have defects of the pleuropertitoneal membrane. Approximately 75 per cent of these infants die before the first month of life if untreated. The majority within the first 24 hours. Cyanosis, dyspnea and mediastinal shift, corroborated by roentgenograms of the chest and abdomen demonstrating herniated bowel in the pleural cavity and the absence of small bowel patterns in the abdominal cavity makes the diagnosis certain. Transthoracic approach and repair of the defects have given satisfactory results in the authors' experience. The treatment of the compression atelectasis must be conservative and attempts of forceful expansion of the atelectatic lung should not be made.