Aneurysm of a Pulmonary Artery Branch: An Uncommon Cause of a Coin Lesion*

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We report the case of a 26-year-old woman with ventricular septal defect associated with a typical coin lesion as shown on the chest roentgenogram. Selective angiography revealed the coin lesion to be caused by an aneurysm of a tertiary pulmonary arterial branch. In addition, two smaller aneurysms were noted. It is postulated on clinical and histologic grounds that the aneurysms were caused by congenital weakness of the pulmonary arterial wall in conjunction with increased pulmonary flow and slightly elevated pulmonary arterial pressure.

Aneurysms of the pulmonary arteries are rare, usually involving either the main pulmonary artery or its primary branches.1,2 Reviewing the literature up to 1961, Charlton and Du Plessis3 collected reports of 30 cases of aneurysms of secondary or tertiary branches. Only a few case reports were added later.4,5,6 In the majority of these the aneurysms were of mycotic origin, resulting from an episode of bacterial endocarditis in the presence of a congenital cardiac defect.1,3-5,6 In about 25 percent, the aneurysms were considered congenital. Recently we encountered a case of a patient with ventricular septal defect associated with multiple aneurysms of tertiary pulmonary artery branches, the largest of which appeared as a typical coin lesion on the plain

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Table 1—Catheterisation Data

<table>
<thead>
<tr>
<th>Location</th>
<th>Pressures, mm Hg</th>
<th>Mean Oxygen Saturations, %</th>
</tr>
</thead>
<tbody>
<tr>
<td>Superior caval vein</td>
<td>12/0 (3)</td>
<td>66.6</td>
</tr>
<tr>
<td>Right atrium</td>
<td></td>
<td>72.9</td>
</tr>
<tr>
<td>Inferior caval vein</td>
<td></td>
<td>74.0</td>
</tr>
<tr>
<td>Right ventricle</td>
<td>40/1</td>
<td>99.4</td>
</tr>
<tr>
<td>Main pulmonary artery</td>
<td>40/5 (12)</td>
<td>98.4</td>
</tr>
<tr>
<td>Pulmonary capillary</td>
<td></td>
<td>98.4</td>
</tr>
</tbody>
</table>

Oxygen consumption 220 ml/min.
Hb 14.6 gm/100 ml.

Figure 1. Plain chest roentgenogram, anteroposterior view. Note chest lesion in left lower zone.
subsequently, the patient was referred to our hospital. On admission she was in no apparent distress. Physical examination revealed the typical findings of ventricular septal defect. The liver was slightly enlarged, but no other sign compatible with cardiac failure was noted. Auscultation of the lungs revealed no abnormalities. The electrocardiogram and vectorcardiogram showed moderate right ventricular and right atrial hypertrophy. On the chest roentgenogram the findings were identical with those noted previously (Fig 1). Laboratory findings were basically normal except for a moderate elevation of the serum \( \gamma \) globulin. Cardiac catheterization and angiography were performed. The data are summarized in Table 1. A ventricular septal defect with a large left-to-right shunt was demonstrated. An injection of contrast material into the branch of the pulmonary artery that supplied the region of the round opacity made the shadow fill immediately and densely (Fig 2). More distally, two small fusiform dilatations of the pulmonary arterial branch were noted. The lesions were interpreted as aneurysms of the pulmonary artery. Operation was performed on Nov 27, 1973. A large ventricular septal defect was closed with a Dacron patch. No other cardiac abnormalities, in particular no signs of previous endocarditis, were detected. Resection of the left lower lobe was performed. The course was uneventful after operation.

Pathologic Examination

The specimen consisted of a left lower lobe, unremarkable on external examination. On cut section three aneurysms of the pulmonary arteries were seen ranging in size from 8 to 25 mm (Fig 3). The pulmonary artery to the posterior segment showed cylindrical dilatation of the lumen proximal to the aneurysm. Histologically, the wall of the smallest aneurysm in the apical segment showed fragmentation and disarrangement of elastic fibers and locally a decreased thickness of the

Figure 2. Injection of contrast medium into pulmonary artery branch supplying left posterobasal segment. Round opacity fills immediately and densely with contrast medium. Distal to this lesion, note two small fusiform aneurysms.

Figure 3. Cut sections of left lower lobe. At left, aneurysms are in apical and lateral segments. At right, aneurysm is in posterior segment.
media (Fig 4). Between the elastic fibers there was no increased collagen deposition nor were there signs of cystic medial necrosis. Intimal fibrosis and thrombosis were absent. In the two largest aneurysms the media were interrupted in the proximal part. Distally, it was replaced by a broad layer of fibrous tissue. Only remnants of elastic laminae were found. At one site there was a transition between the fibrotic layer and an organizing thrombus in the lumen. There was no inflammation in the wall of any of the aneurysms.

The pulmonary parenchyma was normal. No inflammatory reactions of fibrosis were noted in the areas adjacent to the aneurysms. The elastic configuration of the walls of elastic pulmonary arteries directly proximal to the aneurysms was normal. Distal to the aneurysms some elastic and muscular pulmonary arteries arose, which exhibited pronounced intimal fibrosis as a continuation of the fibrous layers within the aneurysms. The muscular pulmonary arteries elsewhere in the lobe showed, apart from mild medial hypertrophy consistent with the moderate elevation of the pulmonary arterial pressure, no abnormalities. The pulmonary veins were unaltered.

DISCUSSION

Aneurysms of tertiary or smaller pulmonary arterial branches present serious diagnostic problems. In the cases reported more often than not the correct diagnosis was established only at operation or necropsy. The radiographic picture comprises a variety of single or multiple aspecific opacities ranging in size from so small as to escape detection to large shadows. However, in the literature we found only one case in which the chest roentgenogram showed a coin lesion as in our case. One can only speculate, however, as to the number of cases in which an incorrect diagnosis is made. Pulmonary angiography is required to establish the diagnosis, and we suggest this be performed whenever the nature of a pulmonary opacity is not entirely clear. Selective injections of contrast medium into the pulmonary artery branch supplying the lung segment involved may be helpful in detailing the malformations, as was demonstrated in our case.

Although septic processes represent the prevailing cause, we do not believe that the aneurysms were of mycotic origin in this case. First, the history revealed no episodes of endocarditis, and at operation no sequelae of endocarditis were noted. Moreover, on pathologic examination no evidence of inflammation could be detected either in the vascular walls or in the adjacent pulmonary parenchyma. A thrombotic origin is also unlikely, as histologic examination showed one aneurysm with one localized area of abrupt and extreme thinning of the wall in the absence of fibrosis (Fig 4) and failed to reveal thrombotic processes elsewhere in the lung segment. Marfan’s syndrome as a possible cause of changes of the pulmonary arterial wall leading to aneurysm formation should be rejected on clinical grounds. In view of the abnormal elastic configuration in the walls of the elastic pulmonary arteries, both at the site of the aneurysms and proximally, we presume that a congenital weakness of the pulmonary artery was present. This, associated with increased pulmonary flow and somewhat increased systolic pulmonary arterial pressure, may have resulted in aneurysm formation. This would also explain the cylindrical dilatation of the arterial segment proximal to one of the aneurysms.

The possibility remains that the marked, partly organized, thrombotic mass in the largest aneurysm developed after puncture, but as similar alterations were found in one of the other aneurysms this explanation is unlikely.

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REFERENCES

Melioidosis: Recrudescence Associated with Bronchogenic Carcinoma Twenty-six Years following Initial Geographic Exposure*

Edward E. Mays, M.D., F.C.C.P.,** and Edward A. Ricketts, M.D.†

Melioidosis was diagnosed in association with lung cancer in a World War 2 veteran 26 years after geographic exposure. The case history illustrates the prolonged latency, difficult diagnosis, and resistance to therapy of chronic melioidosis. Implications are that chronic forms of the disease will continue to surface in veterans of Southeast Asian conflicts for decades.

Melioidosis is a tropical bacterial disease with primary endemic foci in Southeast Asia, northern Australia, and Central America.1-4 Our recent military presence in Vietnam has resulted in a number of cases of overt and subclinical infections in native Americans.5,6 Serologic surveys performed on normal returnees indicate an apparent infection rate ranging from 1 to 9 percent.5,7 A potential for prolonged latency and recrudescence of clinical infection of varying severity has been clearly demonstrated.2-5

This report documents a 26-year interval between probable exposure and confirmed diagnosis of melioidosis, the longest latent interval recorded. Also implicated, for the first time, is lung cancer as the predisposing condition to relapse.

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The opinions or assertions contained herein are the private views of the authors and are not to be construed as official or as reflecting the views of the Department of the Army or the Department of Defense.

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CASE REPORT

A 54-year-old veteran was transferred to Letterman Army Medical Center from another hospital in September, 1971 with right chest wall pain of 13 months' duration, and a productive cough and 25 pound weight loss over the preceding six months. Evaluation elsewhere had included the observation of a small right 6th rib lump, and multiple small granulomas in the mid and upper lung field on chest roentgenograms. Open lung biopsy revealed multiple surface nodules up to 2 cm diameter, which histologically were healed granulomas. No diagnosis or therapy were given. Pain persisted, and re-evaluation revealed a new, thick walled, 6 cm diameter, right upper lobe cavity with an air-fluid level and shaggy internal margins (Fig 1). The bronchoscopist described chronic right endobronchitis and extrinsic narrowing of the bronchus intermedius, biopsy of which was non-diagnostic. Multiple other smears, cultures, and cytologies gave negative results.

The patient had served in the Philippines in 1945 without illness. He recalled severe pneumonia of unknown cause in Tokyo in 1948. Medical records disclosed that triple therapy had been instituted for a roentgenographic diagnosis of active tuberculosis in 1956, despite negative sputum and gastric cultures. In 1959, on dual therapy, he developed pyogenic pneumonia with right upper lobe cavities, treated empirically with short courses of tetracycline and chloramphenicol. Repeated bacteriologic isolation from sputa of B subtilis, and an unidentified gram-negative, motile rod were believed to be contaminants. Diagnostic thoracotomy revealed chronic granulomatous inflammation without demonstrable organisms. He improved, was returned to duty, served an uncomplicated tour in Korea in 1962, and remained well until the

FIGURE 1. Chest roentgenogram showing large multicystic cavity with small fluid level in anterior segment, right upper lobe. Note several 1-2 cm diameter and larger densities in background on the right, and scattered fibrinomlar disease in left middle and lower lung fields. Right fifth rib had been resected previously at diagnostic thoracotomy.