In the final analysis, we based our diagnosis of rheumatoid disease on histologic evaluation of the pulmonary and subcutaneous lesions together. The pulmonary lesion had extensive fibrosis, as well as necrosis, and appeared to be a late relatively quiescent lesion. Angiitis was found within but not outside this lesion. Granulomatous infection was unlikely because cultures of the sputum and lung biopsy failed to grow mycobacterial or fungal organisms and because the pulmonary nodules slowly multiplied and enlarged over ten years without other evidence of localized or disseminated infection. Thus, although the findings in the lung alone were consistent with either Wegener's granulomatosis or a rheumatoid nodule, such late scarred lesions are best described as nondiagnostic; however, in combination with the subcutaneous lesion, we believe that the diagnosis of rheumatoid nodule is most appropriate for both lesions.

We considered the possibility that our patient first developed limited Wegener's granulomatosis and later developed unrelated subcutaneous nodules and rheumatoid factor. The combination of rheumatoid disease and Wegener's granulomatosis has been reported previously in three cases: however, all had arthritis, and none had pulmonary parenchymal lesions. Although we cannot exclude the above combination, the absence of typical vasculitis outside the scarred lesion in our patient's lung militates against it. Thus, we believe that our case represents the unusual development of pulmonary rheumatoid nodules over ten years in the absence of clinical features of rheumatoid arthritis.

ACKNOWLEDGMENT: We wish to thank Dr. Edward A. Gaensler for his review of this manuscript and Ms. Victoria M. Bailey, Mr. Herbert W. Jones, and Ms. Martha J. LaFlamme for their technical assistance.

REFERENCES


Normal Single Coronary Artery and Myocardial Infarction *

LT S. E. Warren, MC, USNR; LCDR J. S. Alpert, MC, USNR; LCDR W. V. R. Vieweg, MC, USN; and CAPT A. D. Hagan, MC, USN

A single left coronary artery was found in an asymptomatic 21-year-old man who initially had electrocardiographic and vectorcardiographic evidence of anterolateral myocardial infarction. The single left coronary artery, which supplied the distribution of both the left and right coronary arteries, was free of disease at catheterization. There has been no previous association of a normal single left coronary artery and anterior myocardial infarction. Patients with the finding of a single coronary artery should be watched closely, as this may represent a potentially fatal condition.

More than 85 cases of a single coronary artery have been reported. 1 A review by Allen and Snider 2 in 1966 revealed an incidence of 58 percent in men and 42 percent in women. These authors found the frequency of a single right coronary artery (49 percent) to be nearly the same as that of a single left coronary artery (51 percent). The age of patients ranged from 83 years. A recent review by Tomaru and Reid 4 documented only ten cases in which a single coronary artery

From the Cardiology Division, Department of Internal Medicine, Naval Regional Medical Center, San Diego, Calif. The opinions or assertions contained herein are the private ones of the authors and are not to be construed as official or as necessarily reflecting the views of the Medical Department of the US Navy or the Naval Service at large. Reprint requests: Dr. Warren, Naval Regional Medical Center, San Diego 92134

540 WARREN ET AL

CHEST, 72: 4, OCTOBER, 1977
A 21-year-old white man on active duty in the US Air Force was admitted to his base's hospital on Feb 9, 1976, after suffering a transient episode of dizziness while running. A routine electrocardiogram revealed poor R-wave progression (Fig 1), compatible with an anterolateral myocardial infarction. For three days the patient's ECG did not change. Three determinations of the levels of cardiac enzymes gave normal results, and there were no complicating symptoms or arrhythmias. On Feb 19, 1976, the patient was transferred to the Naval Hospital at San Diego, Calif, for further evaluation.

The patient gave no history of chest pain, hypertension, heart murmur, syncope, or palpitations. He smoked one-half pack of cigarettes daily, took no medications, and had indulged in vigorous physical activity throughout adolescence without difficulty. His father died at the age of 61 years from a myocardial infarction, and his mother had a history of hypertension. Six brothers and sisters were alive and well.

The results of physical examination were normal, except for the presence of a fourth heart sound at the apex. A chest x-ray film showed left ventricular prominence. An ECG confirmed the absence of R-wave progression in the lateral precordial leads, extreme left axis deviation, and a delay in intraventricular conduction (Fig 1). An echocardiogram revealed slightly enlarged left ventricular dimensions and paradoxical septal motion (Fig 2). Continuous electrocardiographic monitoring (Holter monitor) showed sinus bradycardia with multiple premature ventricular contractions, isolated episodes of sinus pause followed by atrioventricular dissociation with junctional rhythm, and a brief episode of ventricular tachycardia. A vectorcardiogram was consistent with the ECG and revealed an anterolateral myocardial infarction. The patient exercised through three minutes of stage 4 (Bruce protocol) on a treadmill, achieving 76 percent of his maximum predicted heart rate. Transient ST-T elevation in
the precordial leads occurred in the period immediately after exercise and was not accompanied by clinical symptoms.

On Feb 24, 1976, right and left cardiac catheterization was performed. Right-sided pressures were normal. The mean pulmonary arterial wedge pressure was 16 mm Hg; the left ventricular end-diastolic pressure rose from 16 to 24 mm Hg following left ventriculographic studies. The ventriculogram showed apical akinesia. The ejection fraction was 30 percent. Injection of the right sinus of Valsalva demonstrated no evidence of a right coronary ostium. A normal left dominant left coronary arterial system was visualized, with the left anterior descending artery giving off a branch to the right ventricle and the circumflex artery supplying the diaphragmatic surface of the heart (Fig 3). The vessels were free of occlusive disease. The patient was started on therapy with digoxin (0.25 mg daily) and quinidine sulfate (200 mg every six hours). He is currently asymptomatic.

**DISCUSSION**

In 1950 Smith4 classified patients with a single coronary artery into three types: (1) type 1, a single coronary artery covering the normal distribution of one coronary artery only; (2) type 2, a single coronary artery covering the distribution of two vessels; and (3) type 3, in which the distribution is atypical. Table 1 illustrates the previously reported cases of a single coronary artery and myocardial infarction.5,6,8-10 Only one case conclusively demonstrated an absence of coronary thrombosis.8 That was a 54-year-old white man who initially had clinical acute myocardial infarction and who died in ventricular fibrillation. Autopsy showed posteroseptal infarction and a single right coronary artery that, although narrowed by atheroma, could not be shown to be occluded at any point. The investigators3 suggested that patients with a single right coronary artery might be prone to sudden death if the vessel were atheromatous or anatomically subject to compression between the great vessels (or both). In our case the single coronary artery did not pass between the great vessels.

The prognosis for patients with a single coronary artery is not known. Where it is associated with other congenital anomalies (such as tetralogy of Fallot, transposition of the great vessels, endocardial fibroelastosis, and others), the presence of a single coronary artery is reported to have little effect on the outcome; the prognosis is apparently more a function of the severity of the associated defect. Nevertheless, at least 15 percent of the patients without other abnormalities will develop cardiac disease before the age of 50 years.11 Allen and Snider2 reported that the incidence of myocardial infarction in patients with a single coronary artery was less than 5 percent before the age of 40 years and 22 percent afterward.

Nicod12 has pointed out that certain single coronary arteries have a superior course, thus possibly rendering them susceptible to systolic compression by the nearby aorta and pulmonary artery. The overwhelming prevalence of a single right coronary artery associated with infarction argues for the latter explanation, but the present case casts doubt on this hypothesis.

**Table 1—Previously Reported Cases of a Single Coronary Artery and Myocardial Infarction**

<table>
<thead>
<tr>
<th>Patient's Reference</th>
<th>Age, Years</th>
<th>Sex</th>
<th>Smith's Type</th>
<th>Thrombus</th>
<th>Infarct</th>
</tr>
</thead>
<tbody>
<tr>
<td>Alexander and Griffith4</td>
<td>74, M</td>
<td>?</td>
<td>?</td>
<td></td>
<td>Posterior (old)</td>
</tr>
<tr>
<td>Dent and Fisher6</td>
<td>64, M</td>
<td>3 (R)</td>
<td>Right coronary artery</td>
<td></td>
<td>Left ventricle</td>
</tr>
<tr>
<td>Smith and Graber1</td>
<td>46, M 2 (R)</td>
<td>Right main; anterior descending plaque</td>
<td>Anterolateral (old)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Roberts and Loube8</td>
<td>62, M 2 (R)</td>
<td>Right ventricle; right atrium</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Allen and Snider2</td>
<td>54, M 1 (R)</td>
<td>-</td>
<td>Posterior septal</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Tomaru and Reid4</td>
<td>77, F 1 (R)</td>
<td>+</td>
<td>Left ventricle (various ages)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Davis and Compton9</td>
<td>48, M 1 (R)</td>
<td>+</td>
<td>Anterolateral</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Chapman and Peterson10</td>
<td>24, M 2 (L)</td>
<td>?</td>
<td>Inferior</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Present case</td>
<td>21, M 2 (L)</td>
<td>-</td>
<td>Anterolateral</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

*Type 1, single coronary artery covering normal distribution of one coronary artery only; type 2, single coronary artery covering distribution of two vessels; and type 3, single coronary artery with atypical distribution. R, Right; and L, left.
Two other postulates regarding the significance of a single coronary artery can be entertained. The first is that some reported cases of infarction may represent ineffective (albeit unobstructed) blood flow to the myocardium and that slow "controlled" ischemia and the death of cells can occur over months or years. The second postulate is that these patients may, in fact, represent idiopathic cardiomyopathies. Halperin et al.13 described a 27-year-old woman who initially had congestive heart failure in her sixth month of pregnancy. Some weeks after delivery, the patient was studied and was found to have a normal single left coronary artery. An ECG demonstrated a lateral wall myocardial infarction. Six months later, the patient developed increasing heart failure and fatal condition. Autopsy revealed diffuse interstitial fibrosis of both ventricles with marked atrophy of the myocardial fibers. It is difficult to decide whether the pathologic changes in this case represent ischemia or cardiomyopathy. The resemblance of this latter case to our patient is striking. Whether one invokes explanations of spasm due to compression of a great vessel, ischemia, or associated cardiomyopathy, it is clear that a patent single coronary artery, either left or right, can initially be manifested by myocardial infarction, congestive heart failure, syncope, or ventricular arrhythmia and may represent a potentially fatal condition.

REFERENCES

8 Roberts JT, Loubé SD: Congenital single coronary artery in man: Report of nine new cases, one having thrombosis with right ventricular and atrial (auricular) infarction: Am Heart J 34:188-197, 1947
10 Chapman DW, Peterson PK: Unusual forms of coronary disease as demonstrated by percutaneous coronary arteriography. Med Rec (Houston ) 87:320, 1964

Splenic Echinococcal Cyst Burrowing into Left Pleural Space*

A. Barsalai, M.D.; S. Pollack, M.D.; J. K. Kafitori, M.D.; M. Soudry, M.D.; and D. Barsalai, M.D.**

We describe a rare case of hydatid cyst in the spleen which communicated to the left pleural cavity. The patient presented with respiratory distress, characterized by nonproductive cough and dyspnea. The difficulties in diagnosis, using standard laboratory and radiologic techniques, were overcome by the use of ultrasound examination diagnosing both the cysts and the supradiaphragmatic extension.

Splenic hydatid disease is rare; its occurrence even in endemic areas is less than 5.0 percent of the total incidence of echinococcosis.1-4 Since the first cases of splenic hydatid cysts described by Berthelot in 1790 and Luderson in 1808,1 about 100 cases have been reported.1-4 A review of 36 cases of echinococcal cysts at Rambam hospital in the last 15 years revealed only one other case of splenic hydatid cyst. More unusual is the complication of rupture of splenic hydatid cyst into the thorax and involvement of lung and pleura. As far as we know, no such case has been diagnosed preoperatively by ultrasound examination.

CASE REPORT

A 78-year-old woman was referred to our hospital for further investigation of cough, dyspnea, left-sided pleurisy and a palpable mass in the left upper quadrant of the abdomen. She had been treated in the past with digitalis because of chronic atrial fibrillation.

On examination, we found a thin, but healthy looking woman. Her temperature was 37° C (98.6°F), the pulse rate was 88 and irregular, and respiratory rate was 20/min. Blood pressure was 110/70 mm Hg. Heart sounds were irregular but normal. There was dullness on percussion of the left lower lung and decreased fremitus. Diminution of breath sounds was audible over the left lung. On abdominal examination, a large spleen, 8 cm below the left costal margin, was palpable.

Laboratory examination revealed a hematocrit of 37 percent, 9,000 leukocytes with 77 percent polymorphonuclear cells and 8 percent eosinophils. Casoni test was negative, but complement fixation test for Echinococcus antibodies (Weinberg test) was positive. Liver function tests were normal.

Chest roentgenograms showed a large dense homogenic shadow in the lower half of left lung (Fig 1). There was no calcification noted on plain abdominal films. Upper gastrointestinal tract series with barium meal revealed displacement of the stomach by a huge spleen.

Microscopic examination of pleural fluid revealed hooklets.

*From the Surgical Department A, Internal Department C, and the Department of Diagnostic Radiology, Rambam Hospital and the Aba-Khoushy Medical School, Haifa, Israel.
**Established Investigator, Chief Scientist’s Bureau, Ministry of Health of Israel.

CHEST, 72: 4, OCTOBER, 1977

SPLENIC ECHINOCOCCAL CYST 543