Case Reports

A Case of a Traumatic Systemic-Pulmonary Arteriovenous Fistula

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SUMMARY

A case of systemic-pulmonary arteriovenous fistula is described. Chest trauma with multiple bone fractures 9 years prior to this admission was the suspected cause. Three years after the trauma, chest X-rays revealed abnormal vessels in the right lung. Eight years after the trauma, the patient developed congestive heart failure. This has been well controlled with digitalis and occasional diuretics.

Additional Indexing Words:
Rib fractures Pulmonary contusion Pulmonary infection Congestive heart failure Continuous murmur

A systemic-pulmonary arteriovenous fistula is a rare condition. Only 13 cases are reported in the literature. We report a case of systemic-pulmonary arteriovenous fistula which is considered to have been caused by chest trauma and aggravated by pulmonary infection.

CASE REPORT

A 57-year-old man was admitted to Tokyo University Hospital in February, 1973. He had suffered from "tuberculosis of the hilar node" in 1941 and in 1945. In 1955, he fell from a motorcycle and struck his right lateral chest heavily. In 1960, he underwent subtotal gastrectomy because of a gastric ulcer, without any subsequent complication. In December, 1964, while drunk, he fell to the ground, fracturing his right clavicle and right 4th and 5th ribs.

In January, 1967, he was admitted to Hitachi Hospital for the first time complaining of slight fever (about 37°C) and expectoration of fresh blood of 1 month

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Fig. 1. Phonocardiogram showing a continuous murmur at the second intercostal space on the right parasternal line.

duration. Chest X-ray films revealed a homogeneous shadow in the right upper lung field which was connected by abnormally dilated vessels to the hilar area. He was diagnosed as having pulmonary infection and was treated with cephaloridine. The pulmonary infection subsided within 6 months, leaving fibrocystic changes and marked pleural thickening in the right upper lung field. The abnormally dilated pulmonary vessels remained unchanged. In February, 1972, he was admitted to Hitachi Hospital for the second time because of facial and pretibial pitting edema. Physical examination revealed numerous engorged and pulsating vessels over the right upper chest wall. Chest X-ray films showed generalized cardiomegaly and slight pulmonary congestion. The diagnosis of congestive heart failure due to A-V fistula was made and treatment was started with digoxin and furosemide. The edema subsided in a week. Since then, he has taken digoxin.

On admission to Tokyo University Hospital in February, 1973, his blood pressure was 110/60 mmHg in the right arm and 130/40 mmHg in the left arm. The heart rate was 70 beats per minute and regular. There were many engorged, tortuous and pulsating subcutaneous blood vessels on the right lateral chest wall, where thrills were felt. A grade II/VI systolic ejection murmur was heard at the apex. A grade IV/VI continuous murmur was heard with the point of maximum intensity in the right second intercostal space near the sternum (Fig. 1). No cutaneous telangiectasis, clubbing of the fingers or cyanosis was noticed. The laboratory findings on admission were: hemoglobin, 12.9 Gm/100 ml; hematocrit, 41.0%; red blood cell, 374×10⁴; white blood cell, 5,800 with normal differential; erythrocyte sedimentation rate, 3 mm per hour; C-reactive protein, negative; blood chemistries, normal; urinalysis normal. The electrocardiogram revealed tall R waves, depressed ST segment and peaked T waves in the precordial leads indicating left ventricular diastolic overload (Fig. 2). Chest X-ray showed healed fractures
of right clavicle, and of the right 4th and 5th ribs. Slight but generalized cardiomegaly and marked pleural thickening in the right upper lung field were noted. Coarse reticular changes were noted in the upper and middle lung fields of both

Fig. 2. Electrocardiogram suggesting left ventricular diastolic overload.

Fig. 3. Laminogram showing the abnormally dilated pulmonary blood vessels connecting the hilus and the right lateral thickened pleura.
sides (more marked in the right). The abnormally dilated pulmonary vascular shadows connecting the hilar region and the pleura were more prominent than before. These vascular shadows were shown more clearly in laminograms (Fig. 3). A dye dilution curve with the injection of dye into the main pulmonary artery showed the left to right shunt pattern. Right heart catheterization revealed raised pulmonary arterial wedge pressure (45/25 mmHg) with an arterial pressure pulse curve in the right upper lobe. The pressure was higher than that of the main pulmonary artery (Table 1). The oxygen content of the blood taken from the right pulmonary artery was 14.9 vol% and was slightly higher than that of systemic arterial blood. Pulmonary angiograms with the contrast medium injected into the main pulmonary

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<td><strong>Pressure</strong> (mmHg)</td>
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Fig. 4. Pulmonary angiogram. The contrast medium was injected into the main pulmonary artery, and it densely opacified only the left pulmonary artery.
Fig. 5. Aortogram with the catheter tip positioned at the ascending aorta. The right axillary, lateral thoracic and internal mammary arteries are dilated. The right pulmonary arterial branches were opacified at the same time.

Fig. 6. Aortogram with the catheter tip positioned at descending aorta, showing dilated and tortuous right intercostal arteries from the first through the 9th.
artery densely opacified the left pulmonary artery and its peripheral branches, while only a trace of medium exclosed the right pulmonary artery (Fig. 4). This suggested that the flow direction is reversed in the right pulmonary artery because of the shunted blood from the A-V fistula. Retrograde aortograms showed dilatation of the right first through 9th intercostal arteries. The right axillary artery and right lateral thoracic artery were markedly dilated (Figs. 5 and 6). These arteries and the right internal mammary artery fed the dense network of small vessels in the right lateral chest wall and thickened pleura. The right pulmonary artery was filled via these shunting channels. Scanning of the lung with $^{131}$I MAA showed impaired perfusion in the anterior superior portion of the right lung (Fig. 7).

The patient refused surgery, and has been maintained in good health with digitalis.

**DISCUSSION**

The present case has a past history of right lateral chest trauma with fractures of the right clavicle and the right 4th and 5th ribs together with suspected pulmonary contusion in 1964. In retrospect, it is likely that he already had the systemic-pulmonary arteriovenous fistula in 1967, when the abnormally dilated vessels connecting hilus and pleura were recognized. Probably the bone fractures and pulmonary contusion caused the extensive intrathoracic vascular lesion which led to formation of the network of systemic arteriovenous shunts and eventually to development of the systemic-pulmonary arteriovenous shunt.

There have been 13 reported cases with systemic-pulmonary arteriovenous fistula in the literature. Two of them had signs of right to left shunt, i.e. cyanosis, clubbed fingers, and polycythemia.$^{1,2}$ In these cases, arteriovenous
fistulas were considered to be primary lesions and systemic-pulmonary shunts were established as the result of pleural adhesions due to infections. In 2 other cases the primary lesions were thought to be congenital. One of these had a variation of pulmonary sequestration, and the other had a congenital internal mammary arteriovenous fistula. Cox and coworkers reported a case in which a bruit was noted in the anterior chest wall 1 year after treatment with closed tube thoracotomy for spontaneous pneumothorax. Upon surgical operation, it was found that the lung had partially adhered to the chest wall and the intercostal arteries communicated with the pulmonary artery in the area. There is also 1 case of a thoracotomy with resection of the right upper and middle lobes because of bronchiectasis. Eleven years later the patient complained of exertional dyspnea. A bruit was noted in the chest wall. At the second operation, a vascular malformation was recognized at the sites of healed rib fractures. It was assumed that rib fractures at the first operation caused intercostal arteriovenous fistulas. The systemic arteriovenous fistula was considered to be connected eventually to the pulmonary artery via calcific fibrothorax with extensive pleural adhesion. In the remaining 7 cases, arterial blood supply to the fistulas was from the bronchial arteries, internal mammary artery, intercostal artery or aorta. However, their primary lesions were not described.

Systemic-pulmonary arteriovenous fistulas without clinical symptoms are considered not uncommon. Betenga described anastomoses between the intercostal and the pulmonary arteries in 67% of the cases of pulmonary tuberculosis in the apical region, and occasionally such anastomoses were found in other diseases with pleural adhesion. However, cases extensive enough to cause a loud bruit or congestive heart failure are rare.

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REFERENCES