A Case with Fistulas from Both Coronary Arteries and the Left Bronchial Artery to the Pulmonary Artery

Ken Ogasawara, M.D., Tadanori Aizawa, M.D., Junichi Fujii, M.D., Hiroshi Watanabe, M.D., Eiichi Uchida, M.D., and Kazuzo Kato, M.D.

SUMMARY

A case with fistulas from both coronary arteries and the bronchial artery to the pulmonary artery is reported. We believe this is the first case report of an uncommon form of complex arteriovenous fistula diagnosed by selective angiography.

Additional Indexing Words:
Coronary arteriovenous fistula Bronchial artery-pulmonary artery fistula Coronary angiography

CONGENITAL coronary fistula is now considered to be a common anomaly. However, involvement of both right and left coronary arteries remains a relatively rare subset.

Here, we would like to present a case of bilateral coronary artery-pulmonary artery fistula combined with left bronchial artery-pulmonary artery fistula. We believe this is the first case report of an uncommon form of complex arteriovenous fistula diagnosed by selective angiography.

CASE REPORT

A 57 year old man was admitted to the Cardiovascular Institute Hospital for evaluation of a continuous murmur which had been known for 42 years. He had been asymptomatic throughout his life, but paroxysmal atrial fibrillation and hypertension had been diagnosed 2 years earlier when he was 55 years old.

On physical examination, his heart rate was 74 beats/min and irregular, and his blood pressure 130/90 mmHg. A continuous murmur (grade 2/6)
Table I. Cardiac Catheterization Data

<table>
<thead>
<tr>
<th>Site</th>
<th>Pressure (mmHg)</th>
<th>Oxygen Saturation (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>RA</td>
<td>V=3 (1)</td>
<td>63.0</td>
</tr>
<tr>
<td>RV</td>
<td>11/0</td>
<td>62.0</td>
</tr>
<tr>
<td>main PA</td>
<td>12/5 (7)</td>
<td>65.0</td>
</tr>
<tr>
<td>left PA</td>
<td></td>
<td>65.0</td>
</tr>
<tr>
<td>PCW</td>
<td>v=6 (4)</td>
<td>63.0</td>
</tr>
<tr>
<td>IVC</td>
<td></td>
<td>63.0</td>
</tr>
<tr>
<td>SVC</td>
<td></td>
<td>63.0</td>
</tr>
<tr>
<td>LV</td>
<td>98/5</td>
<td>92.0</td>
</tr>
<tr>
<td>Aorta</td>
<td>95/67 (81)</td>
<td>30.0</td>
</tr>
</tbody>
</table>

RA=right atrium; RV=right ventricle; PA=pulmonary artery; PCW=pulmonary capillary wedge; IVC=inferior vena cava; SVC=superior vena cava; LV=left ventricle; ( )=mean.

was heard at a localized area over the 3rd left inter-costal space. Other physical findings were within normal limits.

The electrocardiogram demonstrated atrial fibrillation and left high voltage. The chest roentgenogram showed neither pulmonary congestion nor cardiomegaly.

On cardiac catheterization, a step-up of blood oxygen saturation was detected at the region of the pulmonary artery and the left-to-right shunt ratio was 11% determined by Fick's method. Pressures were normal in the pulmonary artery, right ventricle, right atrium, left ventricle and aorta (Table I).

Right coronary arteriography showed abnormal, dilated coiling branches
Fig. 2. Left coronary angiogram (right anterior oblique view). Thin anomalous branches taking origins from the left main stem enter into the pulmonary trunk.

Fig. 3. Selective left bronchial arteriogram showing a large tortuous vessel anastomosing with the pulmonary trunk.

arising from the main stem of the right coronary artery and the conus branch which entered into the pulmonary trunk. Left coronary arteriography also revealed an anomalous branch arising from the left main trunk which flowed into the main pulmonary artery. Because an abnormal vessel branching off from the distal portion of the aortic arch was found on aortography, selective left bronchial arteriography was performed and dilated serpentine arteries were found to drain into the pulmonary trunk.

Surgical correction of the fistulas was not performed because of the small shunt and the absence of symptoms. Atrial fibrillation was eliminated
by quinidine sulfate administration.

The patient has been under our follow-up for 2 years without any change.

**DISCUSSION**

Because of the wide propagation of selective coronary angiography, congenital coronary fistula has been reported with increasing frequency and considered to be a common congenital anomaly. However, bilateral coronary artery fistula has remained a relatively uncommon subset of coronary fistulas. In a review of 363 cases, Levin and his co-workers reported that bilateral coronary artery fistulas accounted for 5% (19/363) of the total; the same trend was reported by Wilde and Watt. The characteristic of bilateral fistulas is that these tend to drain into pulmonary arteries. In the collective review by Wilde and Watt, 6 of 18 bilateral fistulas drained into pulmonary arteries, while 15% of the total of reported coronary fistulas terminated in pulmonary arteries.

In 1885 Brooks reported an autopsy case with right coronary artery-pulmonary artery, left subclavian artery-pulmonary artery and aortic arch-pulmonary artery fistulas, but we believe that our case is the first case report of an uncommon form of complex arteriovenous fistulas diagnosed by angiography.

Ordinary coronary fistulas are thought to be the result of abnormal persistence of fetal intra-myocardial sinusoids, but this theory cannot clearly explain the genesis of bilateral fistulas. Gobel et al suggested anomalous coronary arteries might arise from the pulmonary artery and Baim et al implied supernumerary implantation of the developing coronary arteries into the pulmonary arterial portion of the embryonic trunkus arteriosus.

On the other hand, Ernst et al referred to the possibility that these fistulas arose from the vasa vasorum of the pulmonary artery. Coronary-pulmonary artery fistulas associated with bronchial-pulmonary artery fistula in our case may support this hypothesis in combination with the fact that normal pulmonary arteries accept vasa vasorum from both coronary arteries and bronchial arteries.

**REFERENCES**

4. Brooks HJ: Two cases of an abnormal coronary artery of the heart arising from the pulmonary artery. J Anat Physiol 20: 26, 1885
5. Grant RT: Development of the cardiac coronary vessels in the rabbit. Heart 13: 361, 1926