CORONARY STENOSIS AND STEAL PHENOMENON
IN CORONARY-PULMONARY FISTULA
—Assessment with Stress Thallium Tomography After
Coronary Angioplasty and Fistulectomy—

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We present a 46-year-old male with unstable angina and bilateral coronary-to-
pulmonary artery fistulae in whom reversible myocardial ischemia was detected
by exercise-stress thallium-201 single-photon emission computed tomography
(SPECT). Coronary angiography revealed a 99% stenosis at the proximal site
of the left descending coronary artery and bilateral coronary-to-pulmonary
artery fistulae with a saccular aneurysm. Percutaneous transluminal coronary
angioplasty abolished chest pain and electrocardiographic changes. However,
definitely improved, but still present, stress-induced perfusion abnormalities
were demonstrated by an exercise-stress thallium-201 SPECT study. Myocardial
ischemia was the only serious complication related to the coronary fistulae,
and after they were surgically resected, the reversible perfusion abnormality
was no longer observed. These findings suggest that coronary-to-pulmonary
artery fistulae potentiated the myocardial ischemia in patient with coronary ste-
nosis leading to unstable angina and prolonged the presence of coronary perfu-
sion abnormality on stress thallium scans probably through a coronary steal
phenomenon.

(Jpn Circ J 1993; 57: 1021–1026)

Coronary artery fistula, which was thought to be a relatively rare cardiac
anomaly!–2 seems to be more easily detected by current coronary angiography. It has
been suggested that coronary fistulae are related to angina pectoris or atypical chest
pain, probably through complicated coronary artery disease, a coronary steal phe-
nomenon, or both. However, coronary steal phenomenon has only been demonstrated in
multiple coronary-left ventricular fistulae using stress thallium scans3–6 and not in any
other type of coronary fistula. We encoun-
tered a patient with coronary stenosis and bi-
lateral coronary-to-pulmonary fistulae in
whom stress thallium-201 single photon emission computed tomography (SPECT)
demonstrated myocardial perfusion abnormality which was probably induced both by
coronary stenosis and by the coronary fistu-
lae. In the report, the clinical implications of

Key words:
Thallium-201 single-photon emission computed tomography
Bilateral coronary-to-pulmonary artery fistulae
Percutaneous transluminal coronary angioplasty
Coronary steal phenomenon
Unstable angina

(Received October 31, 1992; accepted March 24, 1993)
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Japanese Circulation Journal Vol.57, October 1993 1021
coronary stenosis and fistulae in a patient with unstable angina are discussed.

A 46-year-old male patient was admitted with progressively manifested exertional chest pain. Although he had a Grade 2 continuous murmur in the pulmonary area, no definite abnormality was detected in his chest radiograph or baseline electrocardiogram. With the patient in stable condition after intense medication with nitrate, a calcium channel blocker, and $\beta$-blockade, exercise-stress myocardial SPECT with thallium-201 (148 MBq) was performed. This study revealed reversible perfusion abnormalities in the anterior wall and interventricular septum (Fig. 1-A). During the ergometer stress test, the patient complained of chest pain and his electrocardiogram showed a negative U wave in leads V3–6 and an ST-segment depression in leads V5–6 at a load of 50 watts. In addition to a 99% coronary stenosis at the proximal site of the left anterior descending coronary artery, coronary angiography disclosed 2 fistulae between a main pulmonary artery and 2 coronary arteries; one was connected to the diagonal branch of the left anterior descending coronary artery and the other was connected to the conus branch of the right coronary artery (Fig. 2-A). The fistulae had a saccular aneurysm close to the pulmonary artery which measured $15 \times 21 \times 8$ mm on a contrast computed tomographic scan. The left-to-right shunting as calculated by oximetry was small (12.4%). From these findings, the diagnosis of bilateral coronary-to-pulmonary artery fistulae with a saccular aneurysm and unstable angina probably due to coronary stenosis was established.

Percutaneous transluminal coronary angioplasty (PTCA) was successfully performed and left no residual stenosis (Fig. 2-B). Under an increased stress load (75 watts), thallium-201 SPECT performed 2 weeks later showed improvement of the perfusion abnormality and the absence of chest pain.

*Japanese Circulation Journal  Vol.37, October 1993*
and electrocardiographic changes suggestive of myocardial ischemia. However, less, but still significant, myocardial ischemia was present in the coronary territory (Fig. 1-B), although no significant coronary lesion was detected when a coronary angiogram had been performed after PTCA. Because these findings suggested that myocardial ischemia was associated with the coronary fistulae, probably through a coronary steal phenomenon, the fistulae and aneurysm were surgically removed. When exercise-stress thallium-201 SPECT was performed after the surgical treatment, no definite exercise-induced perfusion abnormality was detected (Fig. 1-C) and no coronary event has subsequently been documented.

**DISCUSSION**

Coronary artery fistula, first reported by Krause², is a relatively rare vascular anomaly¹. The incidence of coronary fistula detected by coronary angiography ranges from 0.16% to 1.2%⁸⁻⁹. It has been reported that approximately 17% of coronary fistula connect to the pulmonary artery¹⁰ and that bilateral coronary-to-pulmonary artery
fistulae are very uncommon. Furthermore, bilateral coronary-to-pulmonary artery fistulae with a saccular aneurysm, as described in the present report, appears to be even more unusual. The etiology of the chest pain which is often observed in a patient with a coronary fistula has not been fully explained, and angina pectoris in patients with electrocardiographic changes seems to be independent of the shunt volume. Several previous reports have suggested a coronary steal phenomenon and complicated coronary artery disease or hypoplasia of the coronary artery distal to the fistula as mechanisms of chest pain or myocardial ischemia. The coronary steal phenomenon has been suggested as a mechanism of chest pain based on the relief of angina after ligation of the coronary artery fistula. Coronary spasm seems unlikely to be the mechanism in the present case because drug therapy with nitrate, calcium channel blockade, and their combined use failed to improve the residual thallium perfusion abnormality detected after PTCA. Furthermore no residual coronary stenosis or coronary hypoplasia was demonstrated after coronary angioplasty and surgical treatment, although thallium redistribution was still present, but apparently improved, after PTCA. It has been reported that, in a fraction of patients, the recovery of an abnormal thallium perfusion scan after coronary angioplasty is delayed and that some period of time after successful angioplasty is needed to normalize impaired coronary flow reserve. The thallium perfusion abnormality observed in our case might have resulted from a late improvement rather than from coronary steal phenomenon. However, the presence and mechanism of the delayed recovery of abnormal coronary perfusion reserve is still controversial and stress thallium SPECT was performed 14 days after coronary angioplasty in the present case. Therefore, it is likely that coronary steal through the coronary-to-pulmonary artery fistulae potentiated the effect of associated coronary artery disease leading to unstable angina, and to some extent contributed to prolonged abnormalities of coronary perfusion or thallium perfusion scan after coronary angioplasty.

There have been a few reports that, in cases of coronary fistula complicated by coronary artery disease, either both coronary artery bypass grafting and surgical resection of the fistula (e) were performed or PTCA alone was carried out. However, evidence of myocardial ischemia related to coronary fistula has not been specifically reported. In the present case, by comparing exercise-stress thallium-201 SPECT images before and after PTCA or fistulectomy, myocardial perfusion abnormality induced both by coronary stenosis and by coronary fistulae was clearly demonstrated. There have been several reports of scintigraphically detected myocardial ischemia in which the coronary steal phenomenon was proposed as a possible mechanism of myocardial ischemia. It is of interest that all of the abnormal thallium perfusion scans in these studies were obtained in patients with multiple coronary-left ventricular fistulae and surgical treatment of the fistula was not performed because of technical and anatomical difficulties, which indicates the difficulty in determining an etiological relationship between a coronary steal phenomenon related to a coronary fistula and exercise-induced perfusion abnormality. On the other hand, there have been no previous reports which demonstrate coronary steal phenomenon due to coronary-to-pulmonary fistula detected by scintigraphic study. In the present case, the presence and improvement of myocardial perfusion abnormality was demonstrated by serial stress thallium SPECT imagings before and after PTCA and surgical repair.

Indications for PTCA in a patient with both coronary stenosis and coronary fistula (e) are (1) serious coronary stenosis apparently related to chest pain and myocardial ischemia, and (2) a coronary lesion suitable for PTCA. A coronary fistula per se is not necessarily an indication for surgical treatment if there is chest pain but no cardiomegaly, heart failure, large-volume shunt, pulmonary hypertension, bacterial endocarditis, or large aneurysm. Although rupture of an aneurysm is a serious complication of coronary fistula (e) spontaneous rupture of a small aneurysm (such as that seen in the present case) has not been reported, and PTCA could be readily recommended in our patient who had unstable angina induced by a single coronary artery stenosis and who

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met the above-mentioned criteria.

Thus, exercise-stress thallium-201 SPECT demonstrated that both coronary artery stenosis and coronary-pulmonary fistulae were associated with myocardial ischemia and suggested that coronary steal via the fistulae potentiated the patient's angina pectoris, leading to unstable angina. The exercise stress thallium SPECT technique contributed to a better understanding of the pathophysiological roles of complicated coronary artery stenosis and coronary fistula in unstable angina and to a determination of the therapeutic strategy in the present case.

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