Thallium-201 Myocardial SPECT in Bland-White-Garland Syndrome: Two Adult Patients with Inferoposterior Perfusion Defect

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A myocardial Thallium-201 SPECT examination was conducted both prior to and following surgery of two adult patients with an anomalous left coronary artery originating from the pulmonary artery (Bland-White-Garland syndrome). In this syndrome, the anterior wall is usually affected first. Preoperative examination demonstrated an inferoposterior perfusion defect. In both patients, the right coronary artery was markedly dilated, and a significant left-to-right shunt formation was observed. Both patients were diagnosed as possibly having coronary steal syndrome. A prolonged inadequate blood supply to the right coronary region may cause inferoposterior cardiomyopathy. In the postoperative examination, myocardial perfusion markedly improved in one patient whose preoperative SPECT showed redistribution in a delayed scan. Thallium-201 SPECT was therefore found to be useful in assessing both the preoperative and postoperative myocardial perfusion.

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Bland-White-Garland syndrome (BWG syndrome) is a relatively rare disease condition accounting for only 0.5% of all instances of congenital heart disease (1–3). Lethal heart failure symptoms based on myocardial ischemia are not rare in infancy (4). Some patients, however, remain asymptomatic well into adulthood. In such patients, collateral vessels of the coronary artery may develop, however, the perfusion patterns of such vessels have not yet been fully elucidated. Noninvasive myocardial scintigraphy is commonly conducted to evaluate myocardial perfusion. However, only a few reports have been previously presented on this condition (5–7). We report our findings on two adult patients with BWG syndrome who were examined both before and after undergoing radical surgery.

METHOD

Thallium-201-chloride (111 MBq) was injected into the patients’ antecubital vein during peak exercise on a treadmill. Imaging began immediately after the end of exercise. The data were then collected for 15 min (30 steps per 30 sec). Three hours later, a redistribution study was recorded. Images were obtained on a rotating gamma camera with a low-energy, multipurpose collimator.

CASE REPORT

Patient 1

The patient, a 35-yr-old male, had experienced shortness of breath during exercise over the past few years, but had not undergone any treatment. Cardiac enlargement and hypertension were observed during a physical checkup, after which he was hospitalized for a complete examination. Cardiac catheterization indicated a marked dilatation of the right coronary artery. The left coronary artery was visualized in a retrograde fashion; the pulmonary artery could also be seen. The left-to-right shunt formation rate was 34%. Anastomosis was established between the left coronary artery and the aorta, and plastic surgery was performed on the pulmonary artery. The presurgical exercise myocardial SPECT image (Fig. 1A) showed accumulation defects on the anteroseptal and inferoposterior walls. Delayed images indicated slight redistribution on the anteroseptal wall, but no noticeable redistribution was apparent on the inferoposterior wall. On the postsurgical SPECT image (Fig. 1B), myocardial perfusion was improved on the anteroseptal wall, but a perfusion defect was still evident on the inferoposterior wall.

Patient 2

This patient, a 68-yr-old female, had experienced palpitations, malaise and edema since the age of about 50 and had undergone palliative treatment based on a diagnosis of mitral regurgitation. The patient, however, began to experience a loss of consciousness at the age of 67. BWG syndrome was suspected by her local physician so she was admitted to our hospital for surgery. Cardiac catheterization showed a dilated right coronary artery, left coronary artery and pulmonary artery. The shunt rate was 62%. Preoperative exercise myocardial SPECT (Fig. 2A) demonstrated an extensive defect of the inferoposterior wall and a slight decrease of accumulation on the anteroseptal wall following exercise. Delayed images indicated tracer redistribution on the inferoposterior wall, although it was incomplete. On the postoperative SPECT image (Fig. 2B), distribution was improved, particularly on the inferoposterior wall.
from childhood are considered to have increased right coronary ~ left coronary ~ pulmonary arterial shunt and eventually fall into a state of coronary steal syndrome (9). Our two patients seemed to be in such a state, in view of dilatations of the right coronary artery and distinct shunts. A chronic decrease in blood flow due to steal seemed provoked at the right coronary area, which preceded a subsequent decrease in the accumulation in the inferoposterior wall.

In addition to our patients, five cases of adult or young adult BWG syndrome who also underwent thallium imaging have been reported (5–7). These patients showed a decrease in accumulation on the anterior wall side. However, an accumulation defect on the inferoposterior wall was noted in one patient (a 44-yr-old female) (7). This finding was also considered to have been caused by coronary steal syndrome due to a large right coronary artery and significant shunt formation detected by cardiac catheterization. In the present two patients, no particular symptoms at childhood could be recalled. The decrease on the side of the anterior wall was thought to be comparatively slight because of the

**DISCUSSION**

BWG syndrome is characterized by left coronary artery bifurcation from the pulmonary artery. Therefore, ischemia is apt to occur on the side of the anterior wall in the left coronary artery, which is a perfused area. In fact, a number of previous reports, including infant and child cases, have shown decreased accumulation on the side of the anterior wall (1–3,5,6,8). We predicted such a decrease during preoperative examination for our two patients. However, contrary to our expectations, the findings in the same area were comparatively mild; on the side of the inferoposterior wall, a distinct accumulation deficit was observed. In this respect, both patients appeared to be different from previous reports.

The inferoposterior wall is essentially an area fed by the right coronary artery. In our two patients, dilatation was noted in the right coronary artery, where sufficient blood volume should have been present. Nevertheless, thallium was in a state of hypo-accumulation, and a dissociation was found between intracoronary blood flow and myocardial perfusion.

With the BWG syndrome, patients who have survived

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**FIGURE 1.** (A) Preoperative and (B) postoperative exercise thallium SPECT scans for Patient 1. On the preoperative images, perfusion defects could be seen on the anteroseptal and inferoposterior walls immediately after exercise. In the delayed images, no redistribution was apparent on the inferoposterior wall. After surgery, the tracer distribution improved on the anteroseptal wall, but the inferoposterior wall showed no definite changes.

**FIGURE 2.** (A) Preoperative and (B) postoperative exercise thallium SPECT scans for Patient 2. Preoperative images demonstrated an extensive defect and a slight decrease of accumulation on the inferoposterior and anteroseptal walls, respectively. Redistribution, though incomplete, was apparent on the inferoposterior wall. Postoperative images showed improved distribution.
good development of a left coronary shunt from immediately after birth. Among the cases of the present syndrome, the above findings are supposedly observable in patients with good development of a left-to-right shunt as a collateral pathway from early age.

Exercise-loading myocardial scintigraphy with thallium is frequently used as a noninvasive approach to detect ischemia and evaluate myocardial viability. We used this approach in Patient 1 because the preoperative examination revealed a defect in accumulation without any redistribution at the inferoposterior wall. No postoperative improvement was noted in the same area, thus suggesting myocardial necrosis. For Patient 2, redistribution was noted at the defect in the inferoposterior wall, which indicated myocardial viability. Some improvement was expected and the postoperative examination revealed a distinct increase in distribution in the same area, suggesting a noninfarcted focus.

In conclusion, there are a number of patients who present with a high degree of decreased accumulation on the side of the anterior wall. Myocardial thallium scintigraphy is useful in identifying various kinds of perfusion disturbances and in making an accurate postoperative evaluation.

REFERENCES