Coronary Arteriovenous Fistula as a Cause for Reversible Thallium-201 Perfusion Defect

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Exercise ²⁰¹TI SPECT imaging has become routinely accepted and utilized as a major screening test for atherosclerotic coronary artery disease. In appropriate clinical situations, an abnormal ²⁰¹TI study usually will require a subsequent coronary angiogram to confirm the presence of an abnormality and to define its pathologic anatomy. Although most reversible thallium defects will prove to be secondary to significant coronary artery atherosclerosis, congenital coronary or cardiac anomalies can occasionally be responsible, and it is useful to be aware of these, particularly in the evaluation of relatively young symptomatic patients. We report, for the first time, a coronary arteriovenous fistula as the cause for an exercise-induced reversible ²⁰¹TI perfusion abnormality.

Key Words: coronary artery disease; thallium-201; arteriovenous fistula

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Congenital fistulae of the coronary arteries communicating with one of the cardiac chambers or great vessels are uncommon. Although their reported incidence is approximately 2% in the adult population, the true incidence will vary depending upon whether the reported series is based upon clinical, angiographic or autopsy studies. The most common type is an anomalous origin of the left coronary artery from the pulmonary artery, the Bland-White-Garland syndrome, and there have been several publications discussing the utility of ²⁰¹Tl scintigraphy in differentiating this from congestive cardiomyopathy in infants and young children (1-3), or in the diagnostic evaluation of adults with this condition (4-7).

There is a wide variety of less common congenital coronary artery anomalies. The majority of these patients remain asymptomatic (δ), however when these patients present with symptoms which mimick other more common forms of cardiac disease, the diagnosis can be elusive. Our patient presented with minimally abnormal symptoms and her abnormal exercise thallium perfusion study was definitely instrumental in prompting us to perform the diagnostic coronary arteriogram. To our knowledge, this represents the first case of a coronary arteriovenous fistula to be reported in association with an abnormal ²⁰¹Tl perfusion scintigram.

CASE REPORT

A 70-yr-old obese female was admitted for recurrent paroxymal atrial fibrillation with a rapid ventricular response. The frequency of these episodes had been increasing over a 3-yr period. At an outlying hospital, she had previously been converted to a normal sinus rhythm with a combination of Lanoxin and Verapamil. Most of her episodes resolved spontaneously within several minutes. She denied any prior history of congestive heart failure, syncopal episodes or high blood pressure.

On initial physical examination, her BP was 130/80, and her pulse was 60 and regular. Cardiac auscultation disclosed a grade I systolic-ejection murmur at the left sternal border. No gallups or rubs were identified, and peripheral pulses were unremarkable. She recently had normal thyroid function studies. Her CBC and electrolytes were all within normal limits. A two-dimensional echocardiogram disclosed mild bi-atrial enlargement, mild left ventricular hypertrophy and uniform wall motion with an estimated ejection fraction of 60%. In order to rule out the possibility of underlying ischemic heart disease, it was felt that exercise testing would be worthwhile. Her exercise electrocardiogram was nondiagnostic secondary to resting ST segment abnormalities. An exercise SPECT 201 TI scan was performed, with the patient reaching 90% of MPHR on the treadmill. Thallium-201 (3.0 mCi) was injected intravenously 1 min prior to the termination of exercise and SPECT imaging was then performed within 15 min. Images were obtained on a rotating gamma camera with a low-energy, high-resolution collimator. After initial imaging, 1.0 mCi of ²⁰¹Tl was re-injected and repeat SPECT imaging of the heart was performed 3 hr later. The study showed a prominant reversible perfusion defect involving the antero-apical portion of the LV (Figs. 1 and 2), which was felt to be most consistent with a significant atherosclerotic coronary lesion. Consequently, 4 days later she underwent coronary angiography. The circumflex coronary artery was noted to be large and dominant, the right coronary was small and nondominant, and both were otherwise normal. However the proximal left anterior descending coronary artery supplied an anomalous branch filling an arteriovenous fistula (Fig. 3), which could be seen draining directly into the pulmonary artery (Fig. 4). It was felt that this was creating a "steal" phenomenon, accounting for the exercise-induced thallium defect.

DISCUSSION

Anomalies of the coronary arteries are most broadly divided into three categories: anomalies of origin, anomalies of course and anomalies of termination (9). The inci-

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FIGURE 1. A series of short-axis exercise thallium SPECT images, oriented from the apex on the left, towards the mid-portion of left ventricule on the right. Exercise images (top row, A) and delayed images (bottom row, B) show a significant reversible perfusion defect involving the anterior wall of LV.

dence of these anomalies in adults is approximately 2%, and the most frequently observed type is one of origin, in which the left coronary artery arises from the pulmonary artery (Bland-White-Garland syndrome). It most commonly results in death in early infancy, but survival into adulthood can occur if collateral coronary flow is sufficient. These individuals may then present later in life with a variety of signs and symptoms, which occasionally may mimick ischemic coronary artery disease. Several patients with the Bland-White-Garland syndrome have been reported in which thallium scintigraphy has been utilized in the diagnosis and also in the postsurgical follow-up of their condition (1-7). In addition, two slightly-different congenital anomalies of origin have been reported in association with thallium scintigraphy (10, 11).

Coronary arteriovenous fistulae (abnormalities of termination) are rare congenital lesions in which the fistula arises more commonly from the RCA, and which empty into the right heart in 92% of cases, with the right ventricle being the most frequent recipient chamber followed by the right atrium and finally the pulmonary artery (8), as seen in our



FIGURE 3. RAO projection during contrast injection into left coronary artery. The proximal LAD supplies an anomalous, tortuous and aneurysmal vascular network. (LCX = left circumflex, LAD = left anterior descending coronary artery).

patient. It is believed that these anomalies develop from the persistence of a large intratrabecular vascular network present in the embryonic myocardium. Although normally shrunken to capillary caliber by the developing myocardium, it is felt that in these anomalies a localized developmental arrest of the growing myocardium occurs, allowing the arteriovenous fistula to evolve (12).

Most patients are asymptomatic, and often these anomalies will be discovered incidentally during coronary artery catheterization. However, depending on the size of the A-V fistula, the involved artery and the chamber of termination, the patient may present with atrial arrhythmias (our patient), chest pain, shortness of breath, angina pectoris or acute myocardial infarction. Although our patient did not present with symptoms of myocardial ischemia precipitated by the usual factors, the results of her exercise thallium study



FIGURE 2. Horizontal long-axis exercise thallium SPECT images (near anterior wall) illustrate a reversible perfusion defect also involving apical portion of left ventricle. Exercise images (top row, A) delayed images (bottom row, B).



FIGURE 4. Later angiographic film (RAO projection) shows a thin jet of contrast (arrows) from arteriovenous fistula filling main the pulmonary trunk (arrowheads). The thin contrast jet and abnormal pulmonary artery opacification are easily visible on the dynamic cine study, but reproduce poorly on the static photograph.

clearly indicated that she did have myocardial ischemia unmasked by treadmill exercise. The pathogenesis for this is felt to be coronary "steal" with diversion of blood flow into a low-resistance channel (13).

As illustrated by our report, and by other reports of patients with Bland-White-Garland syndrome, exercise thallium studies can occasionally play an important role in the diagnostic evaluation of patients with these rare anomalies. Therapeutic management of patients is somewhat controversial, but certainly needs to be individualized, based on the severity and nature of the patients' symptoms. Our patient continues to do well with conservative medical management 18 mo after initial diagnosis.

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REFERENCES

- Kim SM, Park CH, Intenzo CM, Baht AGM. Thallium-201 imaging in anomalous left coronary artery originating from pulmonary trunk. *Clin Nucl Med* 1989;14:492-493.
- 2. Petrozzo PJ, Woodard ML, Vitullo DA, Bekerman C, Blend MJ, Pinsky

SM. The use and diagnostic significance of thallium-201 SPECT imaging in a case of anomalous left coronary artery. *Clin Nucl Med* 1988;13:629-631.

- Gutgesell HP, Pinsky WW, DePuey EG. Thallium-201 myocardial perfusion imaging in infants and children. *Circulation* 1980;61:596-599.
- Moodie DS, Cook SA, Gill CC, Napoli CA. Thallium-201 myocardial imaging in young adults with anomalous left coronary artery arising from the pulmonary artery. J Nucl Med 1980;21:1076-1079.
- Anguenot TJ, Bernard YF, Cardot JC, Boumal D, Bassand Maurat JP. Isotopic findings in anomalous origin of the left coronary artery from the pulmonary artery: report of an adult case. J Nucl Med 1991;32:1788-1790.
- Moodie DS, Fyfe D, Gill CC. Anomalous origin of the left coronary artery from the pulmonary artery (Bland-White-Garland syndrome) in adult patients: long-term follow-up after surgery. Am Heart J 1983;106:381-388.
- Katsuragi M, Yamamoto K, Tashiro T, Nishihara H, Toudou K. Thallium-201 myocardial SPECT in Bland-White-Garland syndrome: two adult patients with inferoposterior perfusion defect. J Nucl Med 1993;34:2182-2184.
- McNamara JJ, Gross RE. Congenital coronary artery fistula. Surgery 1969; 65:59–69.
- Greenberg MA, Fish BG, Spindola-Franco H. Congenital anomalies of the coronary arteries: classification and significance. In: Miller SW, ed. Radiol Clin North Am. Philadelphia: WB Saunders, 1989:1127-1146.
- Mintz GS, Iskandrian AS, Bemis CE, Mundth ED, Owens JS. Myocardial ischemia in anomalous origin of the right coronary artery from the pulmonary trunk. Am J Cardiol 1983;51:610-612.
- 11. Mustafa I, Gula G, Radley-Smith R, Durrer S, Yacoub M. Anomalous origin of the left coronary from the anterior aortic sinus: a potential cause of sudden death. *J Thorac Cardiovasc Surg* 1981;82:297-300.
- Hobbs RE, Millit HD, Raghavan PV, Moodie DS, Sheldon WC. Coronary artery fistulae: a 10-year review. Cleve Clin Quarterly 1982;49:191–197.
- Gonda RL, Gutierrez OH, Moss AJ, Lee HJ. Multiple coronary artery-left ventricular fistulas: a pattern of anomalous coronary micro-vascularization. *Cardiovasc Intervent Radiol* 1988;11:313-318.