Applying Preoperative Multidetector Computed Tomography to Bilateral Coronary Artery Fistulas

Hsiao-Ching Huang[1], Chao-Yu Liu[1], Tse-Min Lu[2], Chiao-Po Hsu[1]*

[1]Division of Cardiovascular Surgery, Department of Surgery and [2]Department of Medicine, Taipei Veterans General Hospital and National Yang-Ming University School of Medicine, Taipei, Taiwan, R.O.C.

Coronary artery fistula arising from both the right coronary artery and the circumflex coronary artery is rare. A 28-year-old woman with non-insulin-dependent diabetes mellitus and end-stage renal disease who was on regular hemodialysis complained of recent progressive exertional dyspnea. Congestive heart failure was diagnosed during work-up for simultaneous kidney and pancreas transplantation. Bilateral coronary fistulas draining into the coronary sinus were found by coronary angiography and further characterized by multidetector computed tomography followed by 3D reconstruction preoperatively. The operation was performed smoothly without heart arrest. [*J Chin Med Assoc* 2010;73(8):431–434]

Key Words: circumflex coronary artery, coronary artery fistula, coronary sinus, multidetector computed tomography, right coronary artery

Introduction

Coronary artery fistula (CAF) is a rare congenital anomaly with a reported incidence of 0.1–0.2% in the adult population referred for cardiac catheterization.[1] According to previous literature, only 4–5% of CAFs arise from both the right and left coronary arteries, and only 7% drain into the coronary sinus.[2] We present a case of multiple fistulous communication from the left circumflex artery and right coronary artery to the coronary sinus evaluated by multidetector computed tomography (MDCT) preoperatively.

Case Report

A 28-year-old woman was a victim of hepatitis B, non-insulin-dependent diabetes mellitus for 4 years with triopathy, and end-stage renal disease under regular hemodialysis for 6 months. She felt progressive exertional dyspnea and occasional chest tightness for 1 year. Because she was tired of blood sugar control, she was referred to our hospital for simultaneous kidney and pancreas transplantation. Chest roentgenography unexpectedly revealed cardiomegaly during pre-transplantation evaluation. Thereafter, radionuclide ventriculography disclosed left ventricular ejection fraction 29%, right ventricular ejection fraction 30%, and generalized hypokinesia of the left ventricle (LV) compatible with congestive heart failure. Physical examination showed a grade II systolic heart murmur over the apex. LV hypertrophy with left axis deviation was noted on electrocardiography. Further examinations were arranged to search for the cause of heart failure. Echocardiography revealed moderate to severe hypokinesis of the basal to mid anterior septum, inferior septum and inferior wall of the LV with moderate LV dysfunction, mild to moderate pulmonary hypertension and abnormal flow in the coronary sinus.

Cardiac catheterization and coronary angiography performed later revealed pulmonary-to-systemic flow ratio (Q_p/Q_s) 1.2, pulmonary artery pressure 25/9 mmHg and no stenosis or occlusion of the coronary artery. Moreover, 2 CAFs were found, 1 from the left

*Correspondence to: Dr Chiao-Po Hsu, Division of Cardiovascular Surgery, Department of Surgery, Taipei Veterans General Hospital, 201, Section 2, Shih-Pai Road, Taipei 112, Taiwan, R.O.C.
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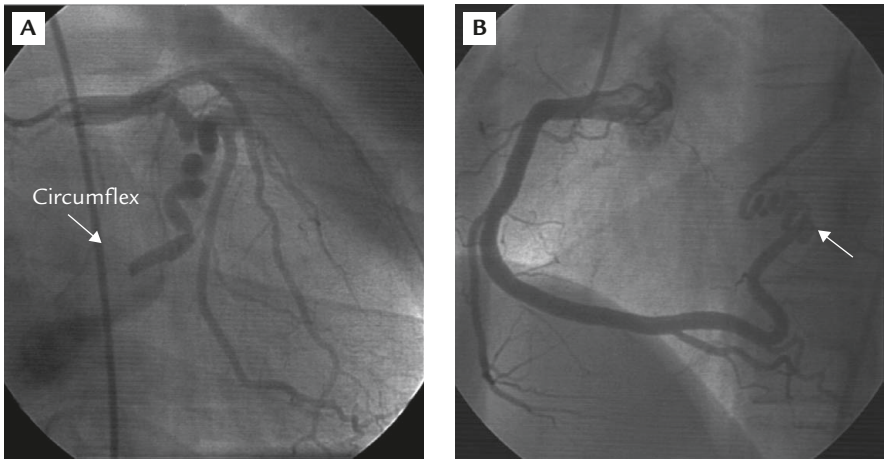

Figure 1. (A) Left coronary system in right anterior oblique view and (B) right coronary system in left anterior oblique view.

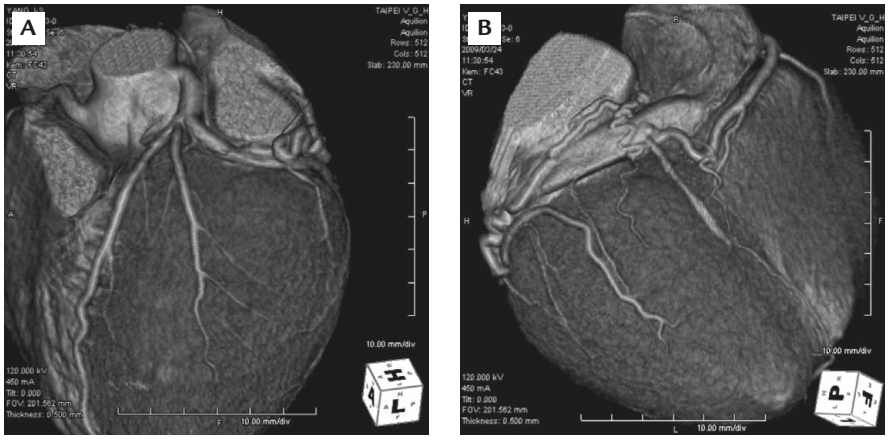

Figure 2. Three-dimensional multidetector computed tomography shows coronary artery fistulas from the left circumflex and right coronary artery to the coronary sinus: (A) left anterior oblique view; (B) posterior view.

circumflex artery drained to the coronary sinus, and the other from the right coronary artery and its atrial branch with obscure drainage site (Figure 1). Percutaneous transcatheter embolization was tried later, but failed because of the inaccessible fistulas. Surgical intervention was suggested. MDCT followed by 3D reconstruction was arranged as a preoperative guide for more precise detail about the origin, drainage site and even relative position of these 2 fistulas which could not be confirmed clearly by angiography (Figure 2). MDCT also disclosed the terminal branch of the right coronary artery on the atria which connected these 2 fistulas beyond the level of the coronary sinus.

The operation was performed under the support of cardiopulmonary bypass without heart arrest. The heart was elevated manually and the posterior surface of the heart looked exactly the same as what is shown in Figure 2B. Two fistulas were confirmed with Doppler and ligated by 5-O polypropylene mattress sutures with pledgets. Obliteration of the fistulas was confirmed intraoperatively by Doppler and transesophageal echocardiography, and no change in the electrocardiogram was observed. The postoperative course was uneventful, and the patient has not complained of any further chest discomfort.

Discussion

CAF is a connection between ≥1 coronary arteries and a cardiac chamber or great vessel. The majority of fistulas is congenital in origin but may also be traumatic, infectious, or iatrogenic. In our case, fistulas arose from both the right and left coronary arteries and drained into the coronary sinus. Previous reports showed that bilateral fistulas are rare, affecting only 4–5% of patients.[3] However, a recent study showed that Asian patients have a higher percentage (21%) of

bilateral or multiple fistulas and more left-side CAFs compared to Caucasians.[4] Congenital fistulous connection of the coronary artery into a cardiac chamber or major vessel often causes a marked dilation of the donor coronary artery leading to aneurysm formation. Aneurysmal dilatation of the coronary artery was found in 26% of CAFs.[5] Rupture of congenital aneurysmal fistula occurred more often in Asian females than in Caucasian females.[6] Fortunately, in our case, the fistulas had not progressed to aneurysm before intervention.

Seventy-nine percent of pediatric patients with congenital CAFs are asymptomatic; however, 71% of adult patients present with exertion dyspnea and chest pain.[7] Patients with CAF do not have a normal life expectancy without treatment.[8,9] The onset of symptoms and complications in untreated patients typically begins by the second or third decade of life. These complications include myocardial infarction, subacute bacterial endocarditis, aneurysm formation, and death.

Coronary angiography remains the gold standard for imaging coronary vessels. Coronary angiography can be used to reliably identify the size and anatomic features of fistulous tracts,[10] but it fails to relate the CAF to other structures or to show the drainage site of the CAF clearly.[11,12] Compared to coronary angiography, MDCT is noninvasive and can provide excellent visualization of coronary vessel morphology, including the origin, course and vessels distal to the drainage site of the CAF, the size of the receiving chamber, and additional precise anatomic details of the CAF, enabling more optimal therapeutic planning.[13,14] Magnetic resonance angiography with 3D multiplanar reconstruction may detect sites of influx and flow of abnormal blood vessels.[15] However, MDCT is more time-efficient, accessible and preferred to evaluate coronary anatomy compared to magnetic resonance angiography in patients with implantable devices.[16]

Management of CAF remains controversial. What constitutes optimal therapeutic strategy has not been clearly established. It is evident that surgical intervention is necessary for symptomatic patients and those with complications. For asymptomatic patients, indications for surgery are similar to those for other left-to-right shunts ($Q_p/Q_s > 1.5$ or right ventricular volume overload). The intervention techniques include operation and transcatheter closure of CAF. Mavroudis et al,[17] based on the literature and personal experience, outlined the requirements for satisfactory coil embolization of a CAF as follows: absence of multiple fistulas and presence of a single narrow drainage site, absence of large branch vessels that could be inadvertently embolized, and safe access to the coronary artery supplying the fistula. Under this criteria, coil embolization was possible in only 37% of the patients.

In our case, transcatheter embolization failed due to distal location. Operation was considered. MDCT disclosed vessels distal to the drainage site of the coronary sinus and the relative position of the 2 fistulas. MDCT changed our surgical planning from arrested heart to beating heart under cardiopulmonary bypass support. Without the MDCT findings, we might have left residual fistulas draining from the distal coronary artery. Advances in CT technology have yielded MDCT scanners with decreased scanning times that reduce respiratory motion artifact, allow cardiac gating capabilities, and provide superior imaging of coronary vessels. It cannot be overemphasized that MDCT can be regarded as an important imaging modality of choice for the preoperative evaluation of coronary artery diseases.

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