Coronary-pulmonary Artery Fistula with Anomalous Vessels Arising from the Right Coronary Sinus Detected by 64-MDCT

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Abstract

We describe a case of aneurysmal coronary-pulmonary artery fistula and a communicating anomalous vessel arising from the right coronary sinus that was clearly demonstrated by 64-multidetector row computed tomography (MDCT). MDCT angiography was more useful than cardiac catheterization for planning the surgical strategy.

Key words: coronary artery fistula, aneurysm, multidetector row computed tomography

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Introduction

Coronary-pulmonary artery fistula is detected in 0.1 to 0.2% of cardiac catheterization procedures (1, 2). These fistulas are usually found incidentally on cardiac catheterization or at autopsy, since most patients are initially asymptomatic. Some, however, may present with myocardial ischemia, myocardial infarction, congestive heart failure, or sudden death (3).

Multidetector row computed tomography (MDCT) is an important resource for the evaluation of known or suspected coronary artery disease, and additionally it can be utilized to assess anomalies of the aorta, pulmonary artery, other vascular structures, and cardiac chambers (4). In the present case, as cardiac catheterization alone may not have been adequate for evaluating the anatomy, MDCT was useful for clarifying the precise anatomy of the coronary artery fistula and anomalous vessel.

Case Report

A 59-year-old woman was referred to our hospital complaining of chest pain on exertion for the previous 5 years, which was relieved with nitrate. The incidence of these symptoms was increasing. She had a history of hyperten-

sion, but no history of dyslipidemia, diabetes mellitus, or cigarette smoking. A continuous murmur at the upper left sternal border had been pointed out previously. Echocardiogram showed normal left ventricular systolic function, and revealed an aneurysm of the left anterior descending (LAD) coronary artery and abnormal flow from the LAD to the main pulmonary artery. Further investigation was performed using MDCT (Aquilion64, TOSHIBA, Tokyo, Japan) and a workstation (ZIO STATION, ZIO SOFT, Tokyo, Japan). The volume-rendered image showed coronary aneurysms in the LAD and an anomalous vessel arising from the right coronary sinus (RCS) which was connected to the aneurysm (Figs. 1A, 1B). Axial enhanced CT images revealed the aneurysmal fistula between the LAD and pulmonary artery (Fig. 1C). Exercise stress myocardial scintigraphy using thallium-201 showed hypoperfusion in the anterior region (not shown). While subsequent cardiac catheterization also confirmed the MDCT findings, it was difficult to identify the precise location of the drainage site (Fig. 2A). Further hemodynamic study revealed an oxygen saturation step-up at the left pulmonary artery (shunt ratio 30.2 %, Qp/Qs 1.42). In this case, since she had experienced chest pain, and aneurysmal coronary-pulmonary artery fistula and myocardial ischemia were demonstrated, surgical intervention was considered necessary to prevent myocardial ischemia or rupture of the coronary aneurysm. Successful ligation of the coro-

Figure 1. 3D volume-rendered images and axial image by 64-MDCT. (A) Left anterior oblique cranial view shows aneurysmal coronary artery fistula (open arrow). (B) Left anterior oblique view clearly shows the anomalous vessel (①) arising from the right coronary sinus, with a separate origin from the right coronary artery. (C) Axial image shows the jet (②) of fistula between aneurysm (open arrow) and MPA. Ao: aorta, MPA: main pulmonary artery, LAA: left atrial appendage, LM: left main trunk, LCX: left circumflex artery

Figure 2. Preoperative cardiac catheterization. (A) Right anterior oblique cranial view of the left coronary artery shows coronary artery fistula with aneurysm (open arrow) draining into the MPA. (B) Catheter contrast injection at the right coronary cusp shows an anomalous vessel arising from the RCS into the aneurysm (open arrow). LAD: left anterior descending artery, MPA: main pulmonary artery, RCS: right coronary sinus

nary aneurysm and closure of the anomalous vessel arising from the RCS were performed, and these were confirmed by postoperative MDCT and cardiac catheterization (Fig. 3B).

Discussion

Coronary artery fistula is a rare anomaly that is character-

Figure 3. Postoperative cardiac catheterization. (A) Postoperative cardiac catheterization confirmed that the aneurysm was not found after surgical ligation of the aneurysm. (B) Postoperative 3D volume-rendered image. Postoperative MDCT showed the successful closure of coronary-pulmonary artery fistula and the communicating anomalous vessel arising from the RCS. LAD: left anterior descending artery, MPA: main pulmonary artery, RCS: right coronary sinus

ized by a direct communication between a coronary artery and another vascular structure or cardiac chamber. It may be either congenital or acquired. Acquired coronary-pulmonary artery fistulas are most often due to complications after coronary artery bypass surgery or after cardiac transplantation. They can also result from percutaneous coronary interventions. Our patient had not received any of these treatments and thus may have had a congenital anomaly.

Coronary artery fistula arises from the right coronary artery (RCA), left circumflex (LCX) coronary artery, and both RCA and LCX in approximately 50%, 42%, and 5% of patients, respectively. The common drainage sites are the right ventricle, right atrium, pulmonary artery, coronary sinus, left atrium, left ventricle, and superior vena cava in approximately 41%, 26%, 17%, 7%, 5%, and 3%, respectively (5).

The clinical symptoms of coronary artery fistulas may vary depending on their anatomy, the relative size of the fistula to other vascular structures, and their flow reserve. Even if a patient is initially asymptomatic, symptoms such as fatigue, dyspnea and chest pain are seen in the course of time. In 80% of patients over 50 years old, who had a high flow reserve fistula, congestive heart failure and angina pectoris are seen due to myocardial ischemia (6). Furthermore, cardiomyopathy, atrial fibrillation, endocarditis, and rupture can occur as late complications. In a follow-up study of 101 patients reported by Hobbs et al (7), no patients with medical treatment had serious complications or mortality. In the present case, however, chest pain due to myocardial ischemia as proven by stress myocardial scintigraphy was present, and was not controlled by vasodilator such as nitrate or nicorandil. Therefore, surgical ligation was considered to be indicated to prevent myocardial ischemia or rupture of the aneurysmal fistula.

The presence of coronary artery fistula can be suspected on the basis of transthoracic and transesophageal echocar-

diography findings (8), and is conventionally diagnosed on cardiac catheterization. However, it is usually impossible to adequately fill an aneurysmal coronary artery fistula with contrast media, and it is difficult to clarify the distal site of coronary artery fistula and the relationship between coronary artery fistula and other cardiac structures, as shown in Fig. 2A. MDCT is an emerging imaging technique for the evaluation of the coronary artery because of its high temporal and spatial resolutions. In this case, 3D reconstruction with viewing at any angle enabled detection of the fistula in the best projection without additional radiation exposure and contrast media, and enabled assessment of the precise anatomical relationship of the coronary-pulmonary artery fistula (Fig. 1).

In conclusion, although cardiac catheterization is the gold standard diagnostic method for determining the origin and the course of coronary artery fistulas, MDCT angiography may be an alternative method for determining the accurate relationship of anatomic structures because of its excellent spatial resolution. In the present case, MDCT angiography was the main modality used to evaluate coronary artery anomalies, and was more useful than cardiac catheterization for planning the ligation of an aneurysmal coronary artery fistula.

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The authors of this manuscript have certified that they comply with the Principles of Ethical Publishing in the International Journal of Cardiology (9)

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