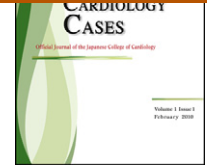


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Case Report

Coronary arteriovenous fistulas complicated by complete atrioventricular block: A case report

Kiyoo Mori (MD, FJCC)^{a,*}, Mitsuru Nagata (MD)^a, Kotaro Oe (MD)^b,
Shu Takabatake (MD)^c, Kenji Sakata (MD)^c, Katsuharu Uchiyama (MD)^c,
Masakazu Yamagishi (MD, FJCC)^c

^a Department of Internal Medicine, Houju Memorial Hospital, Nomi, Ishikawa, Japan

^b Department of Internal Medicine, Saiseikai Kanazawa Hospital, Kanazawa, Ishikawa, Japan

^c Division of Cardiovascular Medicine, Kanazawa University Graduate School of Medicine, Kanazawa, Ishikawa, Japan

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KEYWORDS

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Summary We report the case of a patient with bilateral coronary arteriovenous fistulas (CAVFs) connecting the right coronary artery and left circumflex coronary artery with the right atrium who had progression of first-degree atrioventricular (AV) block to complete AV block during a 4-year period. The His bundle electrogram revealed that the complete AV block was the result of a block at the level of the AV node. Dipyridamole stress thallium-201 myocardial imaging showed decreased perfusion in the inferoapical wall. Coronary angiography and computed tomography showed fistulas that arose from the AV nodal branch of the right coronary artery and from the distal portion of the circumflex coronary artery and drained into the right atrium. Because the fistulas were small, they were not repaired surgically, and a permanent pacemaker was implanted to treat the complete AV block. We presumed that the complication by complete AV block was due to abnormalities of the arteries feeding the AV node and chronic ischemia resulting from a coronary steal associated with the fistulas. To the best of our knowledge, this is the first report of CAVF complicated by complete AV block.

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Introduction

Coronary arteriovenous fistula (CAVF) is a rare disorder that is sometimes discovered incidentally or because of the onset

of various symptoms or signs. Over 90% of CAVFs drain into the right-sided chambers of the heart [1]. Complications include congestive heart failure due to a left-to-right cardiac shunt, myocardial ischemia due to a coronary steal phenomenon, rupture of aneurysmal fistulas, and endocarditis [1,2]. However, arrhythmias related to CAVF are extremely rare [3–5]. We report a case of CAVFs from the atrioventricular (AV) nodal branch of the right coronary artery and the distal portion of left circumflex coronary artery that drained into the right atrium and were complicated by complete AV block.

* Corresponding author at: Department of Internal Medicine, Houju Memorial Hospital, Midorigaoka 11-71, Nomi 923-1226, Japan. Tel.: +81 761 515 551; fax: +81 761 515 557.

E-mail address: k-mori@houju.or.jp (K. Mori).

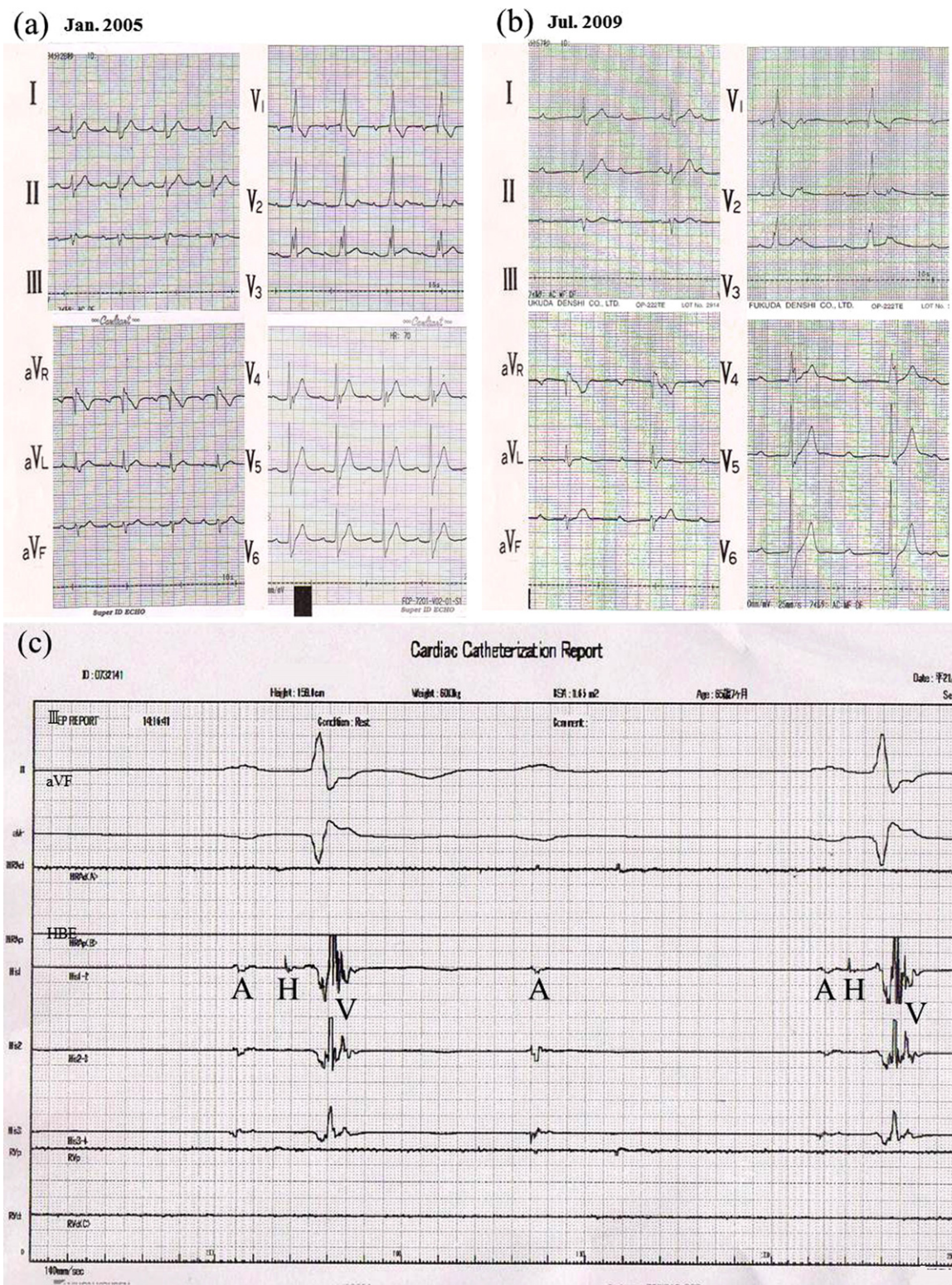


Figure 1 Electrocardiogram (January 2005) showing first-degree atrioventricular (AV) block with a PR interval of 0.28 s and complete right bundle branch block QRS pattern. (b) Electrocardiogram (July 2009) showing the development of complete AV block with a ventricular rate of 40 beats/min. (c) The His bundle electrogram shows that the A wave was not followed by a His deflection. The level of the block is proximal to the bundle of His. A, atrial deflection; H, His deflection; V, ventricular deflection.

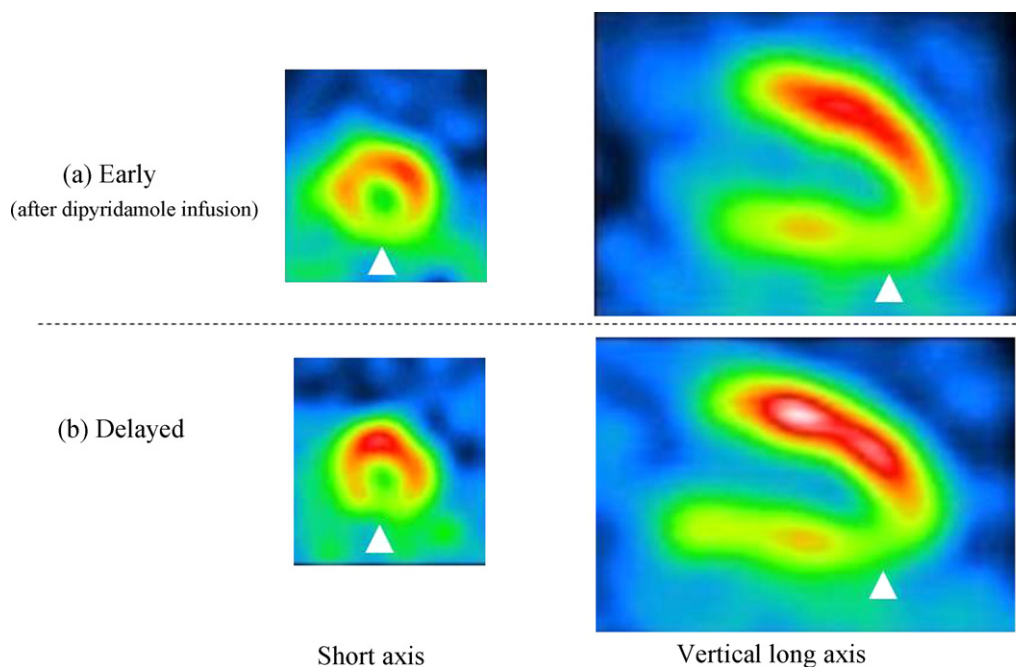


Figure 2 Dipyridamole stress thallium myocardial perfusion scintigraphy. (a) Images obtained immediately after intravenous dipyridamole infusion demonstrate markedly decreased perfusion in the inferoapical wall (arrowheads). (b) Images obtained 4 h later show no redistribution in the same area (arrowheads).

Case report

In January 2005, a 61-year-old man with a history of hypertension, type 2 diabetes mellitus, and hyperlipidemia was admitted to our hospital because of the onset of chest discomfort several months before. On admission, his blood pressure was 128/80 mmHg and pulse rate was 62 beats/min. No heart murmurs were audible. Electrocar-

diography showed first-degree AV block and complete right bundle branch block (Fig. 1a). Chest radiographs were normal. Coronary angiography revealed fistulas arising from the AV nodal branch of the right coronary artery and from the distal portion of the left circumflex coronary artery that drained into the right atrium near the ostium of the coronary sinus. Because the chest discomfort had resolved, no medication was prescribed. In July 2009, he was admitted

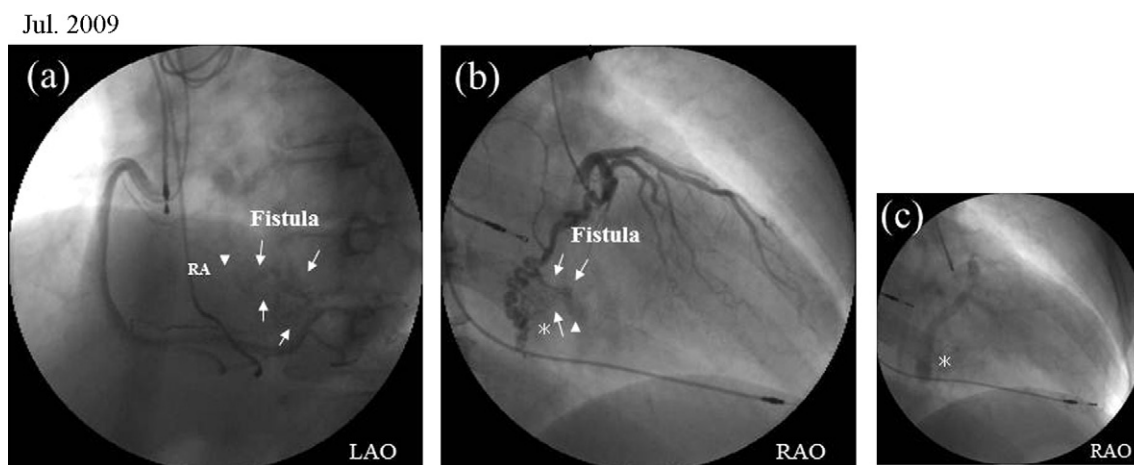


Figure 3 Selective coronary angiography. (a) The right coronary artery in the left anterior oblique (LAO) view. The atrioventricular nodal branch of the right coronary artery (arrows) is seen draining into the right atrium (RA). The arrowheads point to the site of fistulous leakage into the right atrium. (b) The left coronary artery in the right anterior oblique (RAO) view. The left circumflex coronary artery is tortuous, and its distal branches (arrows) drain into the right atrium. The arrowheads point to the site of fistulous leakage into the right atrium. (c) Opacified coronary sinus ostium (*) after injection of contrast medium into the left coronary artery in the RAO view. Bilateral coronary arteriovenous fistulas originating from the right coronary artery and from the left circumflex coronary artery terminate at exactly the same site near the coronary sinus ostium in the right atrium.

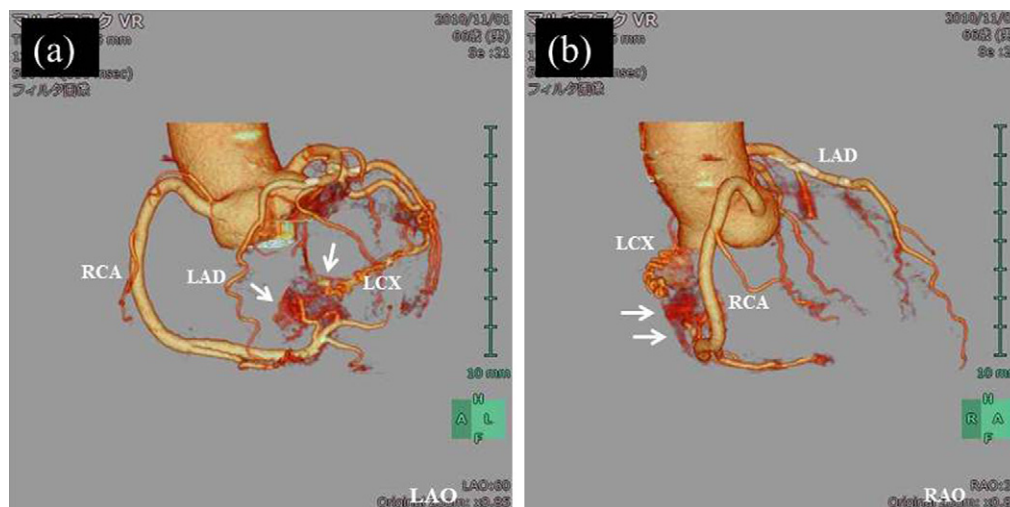


Figure 4 Computed tomography images reconstructed by the volume rendering technique in the left anterior oblique (LAO) view (a) and the right anterior oblique (RAO) view (b). Coronary arteriovenous fistulas (arrows) arising from the distal right coronary artery (atrioventricular nodal branch) and from the tortuous distal circumflex coronary artery that drain into the right atrium can be seen. RCA, right coronary artery; LCX, left circumflex artery; LAD, left anterior descending coronary artery.

to our hospital because of bradycardia. Electrocardiography showed a complete AV block with a ventricular rate of 40/min, and a complete right bundle branch block (Fig. 1b). Echocardiography revealed normal left ventricular function, an ejection fraction of 67%, and mild aortic regurgitation. The His bundle electrogram showed that the site of the complete AV block was proximal to the bundle of His (Fig. 1c). Dipyridamole stress thallium-201 myocardial perfusion imaging showed decreased uptake in the inferoapical wall and redistribution was not present (Fig. 2). Coronary angiography did not show any significant stenosis or marked changes in the fistulas in comparison with the previous study in 2005 (Fig. 3). Right heart catheterization revealed a mean right atrial pressure of 6 mm Hg and a pulmonary to systemic blood flow ratio (Q_p/Q_s) of 1.1. Computed tomography showed that both the fistula from the right coronary artery and the fistula from the left circumflex coronary artery drained into the right atrium at the same site (Fig. 4). Since the fistulous tracts were not aneurysmally dilated, and the shunts through the fistulas were small, the fistulas were not repaired surgically, and a pacemaker was implanted to treat the AV block. The patient will be followed up with close attention to the development of symptoms of complications.

Discussion

A CAVF is a connection between a coronary artery and a cardiac chamber or great vessel (pulmonary artery, coronary sinus, or superior vena cava). The majority of CAVFs are thought to be congenital, but the exact incidence of CAVFs is unknown [1]. CAVFs were found in 225 of 126,595 patients (0.18% incidence) who underwent coronary angiography at the Cleveland Clinic Foundation [6] and the incidence of CAVF in the general population is about 0.002% [7]. Most CAVFs arise from the right coronary artery or the left anterior descending coronary artery. The right coronary artery,

or its branches, is the site of the fistula in about 55% of the cases, the left coronary artery in about 35%, and both coronary arteries in about 5%, but the left circumflex coronary artery is rarely involved [1,8]. Most fistulas are single communications and multiple fistulas are rare [1]. The fistulas usually drain into low-pressure structures. Gowda et al. reported that fistulous drainage was into the right ventricle in 41% of the cases in their series, into the right atrium in 26%, a pulmonary artery in 17%, the left ventricle in 3%, and the superior vena cava in 1% [1], and Branco et al. reported that the most common sites of drainage were a pulmonary artery and the right ventricle [8]. In our case, both the right coronary artery fistula and the left circumflex coronary artery fistula drained into the right atrium, which is rare.

The most common causes of acquired AV block are idiopathic fibrosis of the conduction system, coronary arteriosclerosis, and drug toxicity [9]. In our own patient, first-degree AV block had progressed to complete AV block during a 4-year period. The AV nodal branch of the right coronary artery and the left circumflex coronary artery both communicated directly with a site near the ostium of the coronary sinus in the right atrium, and because the ostium of the coronary sinus is only a few millimeters from the AV node [10], we assumed that the fistulas drained near the AV node. Because the His bundle electrogram revealed that the AV block was the result of a block at the level of the AV node, we speculated that ischemia of the AV node was the cause of the AV block. Arrhythmias including ventricular tachycardia, sinus-node dysfunction and atrial fibrillation have been reported as complications of CAVF [3–5], but the mechanism by which the fistula might have caused the arrhythmias is unknown. Several mechanisms have been suggested, particularly ischemia caused by a steal phenomenon [1,5]. In our own patient, we speculated that there was ischemic injury to the AV node because the feeder arteries of the AV node were involved. Furthermore, dipyridamole stress thallium-201 myocardial perfusion imaging showed decreased uptake

in the inferoapical wall, and it suggested some perfusion abnormality was present in this area due to the fistula.

To the best of our knowledge, this is the first report of CAVF complicated by complete AV block. The long-term outcome of CAVF is not well known, and caution is required for complications, such as angina or congestive heart failure.

Conclusion

We have reported a case of CAVFs arising from the AV nodal branch of the right coronary artery and from the distal circumflex coronary artery drained into the right atrium in which complete AV block developed as a complication. Bilateral CAVFs that communicate with the right atrium are rare, and none complicated by complete AV block has ever been reported. Further study will be necessary to elucidate the relationship between CAVFs and AV block.

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