

Unusual Communication between the Pulmonary Artery and Vieussens' Arterial Ring Causing Infective Endocarditis

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Coronary artery fistula is an abnormal communication between the coronary artery and the cardiac chambers. In particular, an abnormal connection between the conus branch of the right coronary artery and the proximal left anterior descending coronary artery is defined as Vieussens' arterial ring. Coronary artery fistulas are usually asymptomatic, but some can cause complications such as infective endocarditis. Here, we report a case of Vieussens' arterial ring causing infective endocarditis with severe mitral regurgitation.

Key words: 1. Coronary vessel anomalies
2. Fistula

Case report

A 55-year-old man presented to the emergency room of Kosin University Gospel Hospital with fever and headache. Physical examination revealed that the patient's blood pressure was 90/60 mm Hg, his pulse was 63 beats/min, and his body temperature was 38.4°C. Laboratory tests showed a decreased platelet count (29,000), a decreased serum albumin level (2.6 g/dL), an increased white blood cell count (29,750), an elevated high-sensitivity C-reactive protein level (25.415 mg/L), and an elevated troponin I level (252 ng/mL). Brain magnetic resonance diffusion and fluid-attenuated inversion recovery imaging revealed the presence of a multifocal acute infarction that included the frontal, occipital, and right temporal lobes and the left cerebellum and was accompanied by hemorrhagic transformation. Transthoracic echocardiog-

raphy (TTE) was performed to identify the presence of any intracardiac vegetation, and a 1.05×1.45-cm area of vegetation in the P1 segment of the artery was observed (Fig. 1A) with severe mitral regurgitation. Follow-up transesophageal echocardiography was performed and revealed a fistula in the pulmonary artery trunk (Fig. 1B). Multidetector computed tomography (CT) was also performed and showed that the fistula originated from the right conus artery and the proximal left anterior descending artery (LAD), which communicate with each other, and drained into the main pulmonary artery (MPA) with a 6.9-mm aneurysm (Fig. 2). The right conus branch of the proximal right coronary artery (RCA) displayed an abnormal connection with the proximal LAD, which was compatible with the diagnosis of Vieussens' arterial ring (VAR). Due to the presence of a coronary artery fistula (CAF) including

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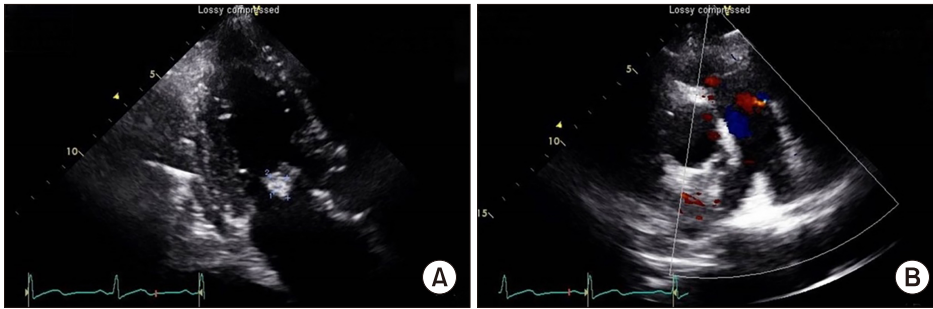


Fig. 1. Preoperative transthoracic echocardiography shows mitral valve vegetation (A), and transesophageal echocardiography shows abnormal blood flow caused by a suspected fistula in the pulmonary artery trunk (B).



Fig. 2. Three-dimensional reconstruction of computed tomography coronary angiography. The right coronary branch and the proximal left anterior descending coronary artery formed Vieussens' arterial ring (arrow), which showed aneurysmal dilatation prior to entrance into the main pulmonary artery.

vegetation, we decided to perform mitral valve replacement with external fistula ligation.

The operative findings showed vegetation on the P1 leaflet, which was then removed (Fig. 3A), and mitral valve replacement was performed. The fistula between the coronary artery and the MPA was obliterated through external ligation (Fig. 3B). Follow-up TTE conducted 1 week after the operation showed a well-functioning mechanical mitral valve without a residual fistula. We planned to perform a post-operative coronary angio-CT a few months later, but the patient was lost to follow-up.

The patient provided written informed consent for the publication of clinical details and images.

Discussion

CAFs are abnormal connections between the coronary artery, heart chambers, and great vessels. CAFs were previously diagnosed using invasive coronary angiography. However, recent advances in coronary CT angiography have shown that the prevalence (0.9%) of this condition is somewhat higher than previously reported [1].

Most patients have no symptoms; however, among symptomatic patients, chest pain (39%) and dyspnea (25%) are common. Fistula most commonly arises in the left main artery/LAD (84%), followed by the RCA (38%), and it most commonly terminates in the MPA (89%) [2]. In our case, the fistula originated from the communicating artery between the proximal RCA and the proximal LAD, which constitutes VAR. VAR was first described by Raymond de Vieussens in 1706 and is thought to be an embryological remnant of an abnormal connection between the conus branch of the RCA and the LAD [3]. Aneurysms of VAR have been previously reported; however, reports of a fistula between VAR and the MPA are rare. Although Lee and Cho [4] reported a single case similar to our case, the patient in that paper did not suffer from infective endocarditis.

The patient in our study had never been treated with dental therapy, and his hygiene was good. It was difficult to identify the factors causing endocarditis in this patient, and given his circumstances, VAR was presumed to have caused this condition. CAF with a right-sided origin can cause infective endocarditis of left-sided valve through migration of infectious emboli via the pulmonary artery [5].

Imaging studies of CAFs include echocardiography, coronary angiography, and coronary angio-CT. Coronary angio-CT is far less invasive and more cost-effective than coronary angiography and is now widely used

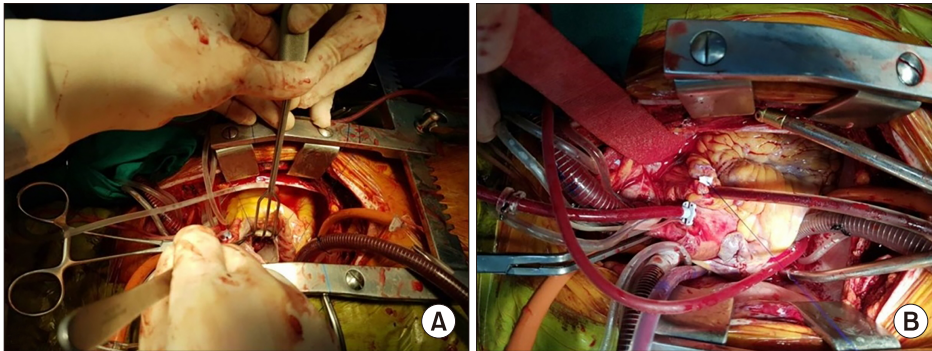


Fig. 3. Vegetation was visible on the P1 area of the mitral valve (A). Multiple pledgeted sutures were performed on the coronary artery fistula for external ligation (B).

as a diagnostic tool. In our case, because P1 vegetation due to infective endocarditis was observed, we chose CT over coronary angiography.

It is well established that patients with symptomatic CAF should undergo closure of medium to large fistulas. Latson [6] reported that it is better to close even small fistulas if: (1) the patient is considered to be at high risk for later endocarditis, (2) longitudinal follow-up is not feasible, or (3) the patient is undergoing an invasive procedure for another cardiac problem.

When considering the management of VAR, the treatment options are not markedly different from those for CAF. Both surgical correction or transcatheter embolization are available. In our patient, surgical fistula closure was considered to be an appropriate next step because of the vegetation of the mitral valve due to infective endocarditis.

Conflict of interest

The authors declare that they have no competing interests.

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