A Coronary Artery-left Ventricular Fistula through the Sinusoid

Koji Sato¹, Ikuo Misumi¹, Miwa Nagano¹, Ryohei Arima¹, Shota Ehara¹, Takashi Sakamoto², Hiroki Usuku³, Koichi Kaikita³ and Kenichi Tsujita³

Abstract:
A 78-year-old woman was referred to our hospital because of repetitive suppurative arthritis at the artificial left knee joint. Her plasma brain natriuretic peptide level was 122 pg/mL. A 12-lead electrocardiogram showed a QS pattern in the inferior leads. A two-dimensional echocardiogram revealed hypokinesis at the inferior wall and hypertrophy at the apical lateral wall. Color flow imaging revealed this hypertrophic region to be a myocardial sinusoid, demonstrating diastolic coronary to left ventricular flow and early systolic flow vice versa. This was a very rare case of coronary to left ventricular fistula through a sinusoid without cyanotic congenital heart disease or severe coronary artery disease.

Key words: coronary artery-left ventricular fistula, sinusoid, systolic regurgitant flow

(Intern Med 60: 3755-3758, 2021)  
(DOI: 10.2169/internalmedicine.7454-21)

Introduction

A coronary cameral fistula (CCF) is an abnormal communication between one of the coronary arteries and a cardiac chamber. CCF is a rare entity reported in 0.08% to 0.3% of unselected patients undergoing diagnostic coronary angiography (1). Most cases involve the right coronary artery with a fistula draining into the right-sided chambers of the heart; a connection between the left coronary arteries and the left ventricle occurs in only 10% of cases (2).

We herein report a rare case of CCF that drained into the left ventricle through a sinusoid remnant.

Case Report

A 78-year-old woman was referred to our hospital because of swelling of the left knee joint. She had suffered from repetitive suppurative arthritis at the artificial left knee joint for 5 years. A physical examination showed her body temperature was 37.1 °C, and her blood oxygen level measured using a pulse oximeter was 95%. Her blood pressure was 122/80 mmHg, and her pulse rate was 84 beats per minute. Auscultation of heart sounds showed that there was a fourth sound and II/VI systolic murmur at the apex. Pretibial edema was also noted.

Results of blood analyses showed a high white blood cell count of 10,900/μL, mild anemia, mild liver injury, a high C-reactive protein level of 7.2 mg/dL, and a high plasma brain natriuretic peptide level of 122 pg/mL. A 12-lead electrocardiogram (ECG) showed sinus rhythm with a QS pattern in leads II, III, aVF (Fig. 1a). A chest radiograph showed pleural effusion (Fig. 1b). A two-dimensional echocardiogram in a parasternal short-axis view showed hypokinesis at the inferior wall, suggesting old inferior myocardial infarction responsible for a QS pattern in the 12-lead ECG. An apical four-chamber view showed a normal left ventricular (LV) wall motion with hypertrophy at the apical lateral wall (Fig. 2, arrows); the right ventricular wall motion was normal.

Color flow imaging showed moderate aortic regurgitation. Color flow and pulsed-wave Doppler echocardiography at the coronary artery showed an enlarged coronary artery and peak diastolic flow velocity of 58 cm/s, suggesting no significant coronary artery stenosis and a high coronary flow (Fig. 3). Color flow imaging at the hypertrophic region

¹Department of Cardiology, Kumamoto City Hospital, Japan, ²Department of Orthopedics, Kumamoto City Hospital, Japan and ³Department of Cardiovascular Medicine, Kumamoto University School of Medicine, Japan

Received: March 3, 2021; Accepted: April 12, 2021; Advance Publication by J-STAGE: May 29, 2021

Correspondence to Dr. Ikuo Misumi, misumi.ikuo@cityhosp-kumamoto.jp
showed several tubular flow signals from the epicardium to the LV chamber during diastole and an opposite flow from the LV lumen to the epicardium during systole. As these findings are not observed in LV noncompaction and are specific for sinusoid, the apical lateral region was deemed to have been formed by a porous sinusoid (Fig. 4) (3). Pulsed-wave Doppler echocardiography at the sinusoid showed peak diastolic flow toward the LV cavity velocity of 1.6 m/s (Fig. 5, arrows) and a reverse flow from the LV cavity (Fig. 5, arrowheads). Coronary angiography or coronary computed angiography was not performed because of severe infection.

Although the present patient underwent debridement of the infected knee joint, she ultimately died of sepsis. Consent for an autopsy was not obtained.
Figure 3. Pulsed-wave Doppler echocardiography at the coronary artery showed a peak diastolic flow velocity of 58 cm/s, early systolic reversal flow of 28 cm/s, and peak systolic velocity of 25 cm/s.

Figure 4. Color flow imaging at the hypertrophic region showed several tubular flow signals from the epicardium to the LV chamber during diastole (a). During early systole, the opposite flow from the LV lumen to the hypertrophied region was observed (b).

Discussion

Wearn et al. classified the CCF into two categories: arterio-luminal and arterio-sinusoidal vessels (4). In the present case, coronary to left ventricle shunt during diastole and left ventricle to epicardium shunt through the sinusoid were observed (5). Before birth, an LV sinusoidal pattern is initially present in normal fetuses, and myocardium is perfused directly from the cavity to a sinusoidal network. As vasculogenesis of the coronary artery is completed, the myocardium is compacted, and the primitive sinusoids disappear. Persistent myocardial sinusoids may be present in newborns with other congenital heart diseases, such as pulmonary atresia or hypoplastic left ventricle (6). In adults, sinusoid may be present in rare cases of severe coronary artery
Figure 5. Pulsed-wave Doppler echocardiography at the sinusoid showed a peak diastolic flow velocity of 1.6 m/s (arrows), which was higher than the coronary artery flow. In early systole, the reversed flow from the LV cavity to the compacted layer was observed (arrowheads).

stenosis (7). Although its significance is unclear, some studies of transmural laser revascularization technique for the alleviation of ischemia have been performed (3).

The present case was a very rare case of coronary to LV fistula through a sinusoid without cyanotic congenital heart disease or severe coronary artery disease at the sinusoid. In our patient, collateral flow from the lateral wall to the inferior wall might have been present. Thus, coronary steal phenomenon through collateral flow might have caused myocardial ischemia and sinusoid formation at the lateral wall. Chronic inflammation might have also contributed to athrogenesis and myocardial ischemia (8). Although this anomaly may be asymptomatic throughout a patient’s life, chest pain, exertional dyspnea, and fatigue may develop in some cases due to myocardial ischemia from coronary steal or heart failure from diastolic overload (9). The present patient had suffered from repetitive suppurative arthritis for several years, and this chronic infection as well as CCF into the left ventricle and old inferior myocardial infarction might have caused heart failure that was demonstrated by a high plasma brain natriuretic peptide (BNP) level, general edema, and pleural effusion.

Conclusion

This was a rare case of coronary artery to left ventricle fistula through a sinusoid without any other congenital heart disease or myocardial ischemia in the region.

The authors state that they have no Conflict of Interest (COI).

Acknowledgement

I would like to thank Mr. Yoshiharu Saito and Mr. Yosuke Emura for capturing the echocardiographic images.

References

1. Vavuranakis M, Bush CA, Boudoulas H. Coronary artery fistulas in adults: incidence, angiographic characteristics, natural history. Cathet Cardiovasc Diagn 35: 116-120, 1995.
2. Fragakis N, Giazitzoglou E, Katritsis DG. A case of coronary-cameral fistulae involving all three major coronary arteries. Circulation 131: e380-e381, 2015.
3. Tsang JC, Chiu RC. The phantom of “myocardial sinusoids”: a historical reappraisal. Ann Thorac Surg 60: 1831-1835, 1995.
4. Wearn JT, Mettier SR, Klumpp TG, Zschiehsche LJ. The nature of the vascular communications between the coronary arteries and the chambers of the heart. Am Heart J 9: 143-146, 1933.
5. Hammond GL, Moggio RA. Function of microvascular pathways in coronary circulation. Am J Physiol 220: 1463-1467, 1971.
6. Peraira JR, Burgueros M, Esteban I, Garcia-Gurtera L, Rubio MD, Alvarez-Ferreirae FJ. [Pulmonary atresia with intact ventricular septum associated with severe aortic stenosis]. Rev Esp Cardiol 56: 1235-1248, 2003 (in Spanish, Abstract in English).
7. Nogales-Asensio JM, Merchant-Herrera A, Lopez-Minguez JR, Gonzalez-Fernandez R. [Collateral circulation due to persistent myocardial sinusoids]. Letter to the editor. Rev Esp Cardiol 60: 886-887, 2007.
8. Mehta JL, Li DY. Inflammation in ischemic heart disease: response to tissue injury or a pathogenetic villain? Cardiovasc Res 43: 291-299, 1999.
9. Sohn J, Song JM, Jang JY, et al. Coronary artery fistula draining into the left ventricle. J Cardiovasc Ultrasound 22: 28-31, 2014.