

[CASE REPORT]

A Coronary Artery-left Ventricular Fistula through the Sinusoid

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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

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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

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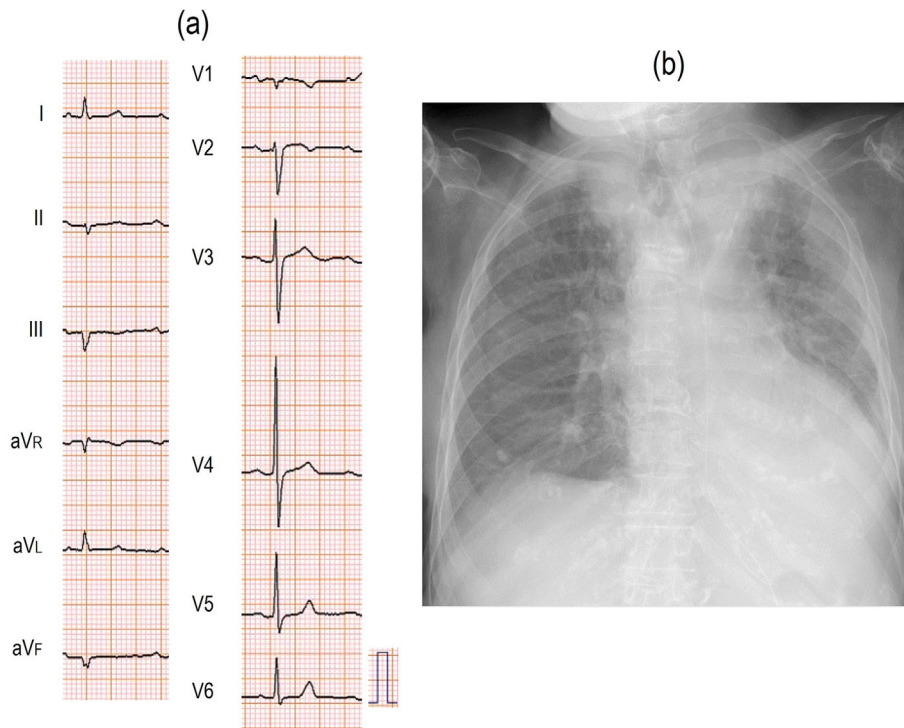


Figure 1. (a) A 12-lead electrocardiogram showed a sinus rhythm with a QS pattern in leads II, III, and aVF. (b) A chest radiograph showed pleural effusion.

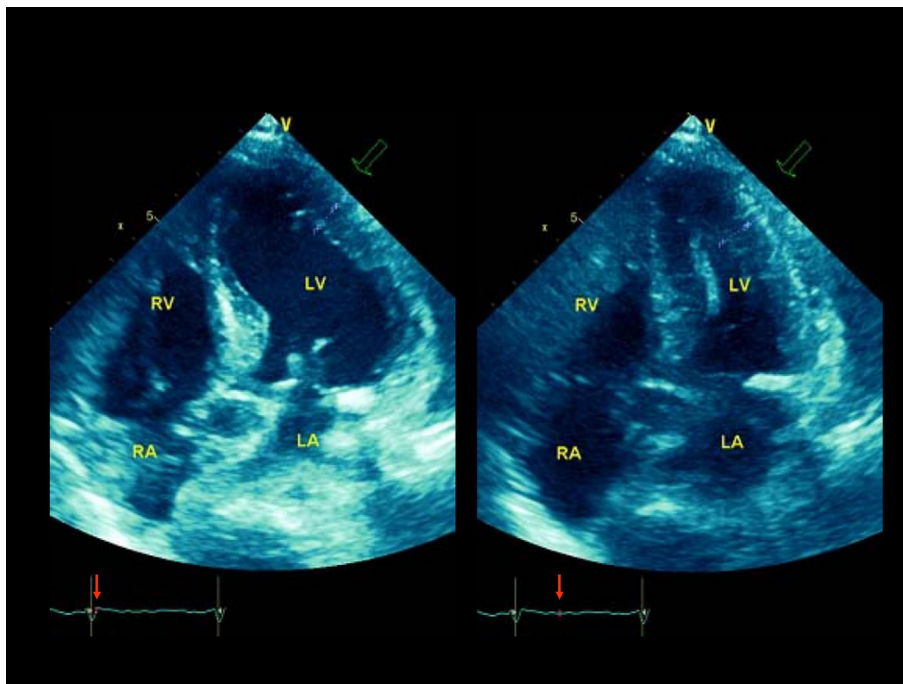


Figure 2. An apical four-chamber view showed a normal LV wall motion with hypertrophy at the apical lateral wall (arrows).

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.

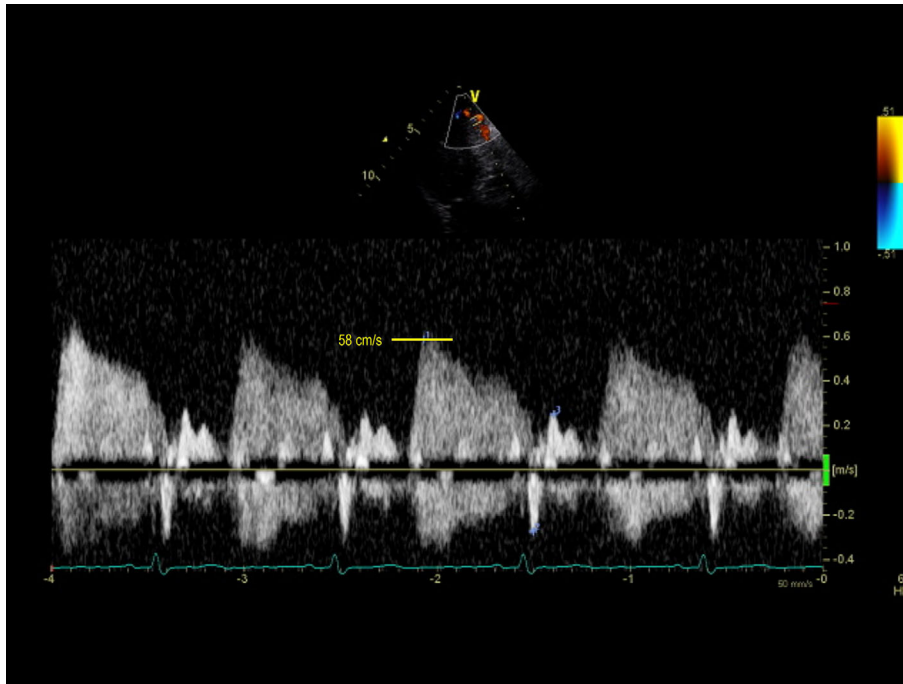


Figure 3. Pulsed-wave Doppler echocardiography at the coronary artery showed a peak diastolic flow velocity of 58 cm/s, early systolic reversal flow of 28 cm/s, and peak systolic velocity of 25 cm/s.

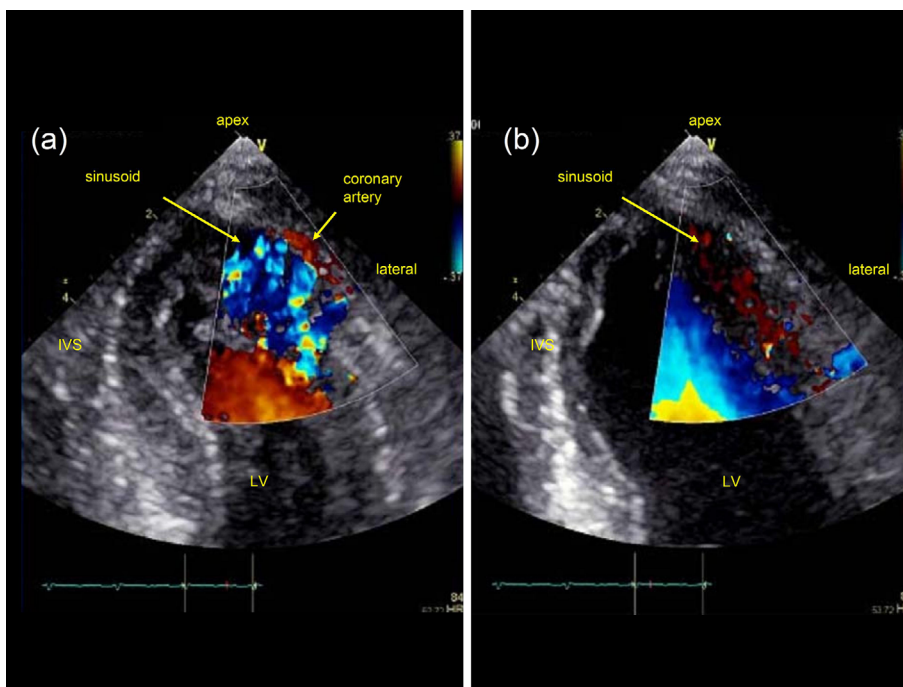


Figure 4. Color flow imaging at the hypertrophic region showed several tubular flow signals from the epicardium to the LV chamber during diastole (a). During early systole, the opposite flow from the LV lumen to the hypertrophied region was observed (b).

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

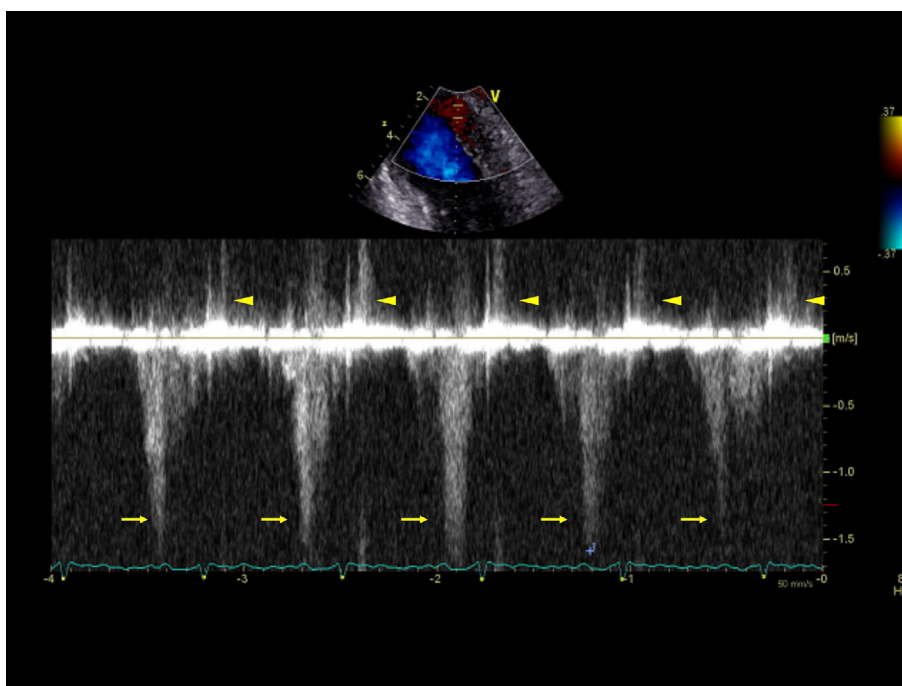


Figure 5. Pulsed-wave Doppler echocardiography at the sinusoid showed a peak diastolic flow velocity of 1.6 m/s (arrows), which was higher than the coronary artery flow. In early systole, the reversed flow from the LV cavity to the compacted layer was observed (arrowheads).

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 atherosclerosis 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

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