

MULTIPLE FISTULA EMPTYING INTO THE LEFT VENTRICLE THROUGH THE ENTIRE LEFT VENTRICULAR WALL

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Coronary artery fistulae are usually identified during invasive coronary angiographies. However, in this case, we made the early detection of coronary artery fistulae during non-invasive transthoracic echocardiography, by demonstrating diastolic multiple abnormal color Doppler flows on the entire left ventricular walls including left ventricular free wall, interventricular septum and apex, which were mimicking firecracker on the whole left ventricle. Fistulous communication from the coronary artery to the left ventricle is rare. Moreover, a case of multiple coronary fistulae emptying into the left ventricle through the entire left ventricular walls including left ventricular free wall, interventricular septum and apex is uncommon. We report a case of a 31-year-old woman who was diagnosed with multiple fistula communicating with entire left ventricular wall.

KEY WORDS: Multiple fistula · Left ventricle · Firecracker.

INTRODUCTION

Coronary artery fistulae are direct connections from one or more coronary arteries to cardiac chambers or a large vessel. Congenital anomalies are more common than acquired,^{1,2)} being found in up to 0.2% of routine angiocardiology.³⁾ Most reported cases are about coronary artery fistulae to right cardiac structure,⁴⁾ but the fistula between coronary artery and left ventricle is considerably rare.⁴⁻⁶⁾ Moreover, a case of multiple fistula emptied into the left ventricle through the entire left ventricular walls including left ventricular free wall, interventricular septum and apex is uncommon. The following we presented is such a case, with impressive echocardiographic images which demonstrating diastolic massive abnormal color Doppler flows on the entire left ventricular walls which were mimicking firecracker on the whole left ventricle.

CASE

A 31-year-old woman was referred from local clinic for further investigation of cardiac murmur. Past health status was good. Physical examination revealed a blood pressure of 107/43

mmHg, and regular pulse of 86 beats/min. There was grade III/VI diastolic murmur from base to apex of heart. Electrocardiography demonstrated biphasic T wave in lead V1-3 and borderline prolongation of QT interval (Fig. 1). Transthoracic echocardiography demonstrated a normal functioning left ventricle with hypertrophy and trabeculation of left ventricular free wall and interventricular septum (Fig. 2). Imaging with color flow Doppler demonstrated multiple sparkling colors Doppler flow through left ventricular free wall and interventricular septum during diastole (using a 4-6 MHz-frequency transducer and high Nyquist limit: 74), and these multiple sparkling color flows demonstrated multiple fistula formation from the epicardial surface into the entire left ventricular cavity through the hypertrophied segment of myocardium during diastole (Fig. 2). These communications have plexiform pattern forming arteriosinusoidal vessels along interventricular septum. It also showed suspicious non-compacting myocardium at apical infero-posterior wall of left ventricle. We observed two separated layers of myocardium with a thin, compacted, epicardial layer and a thick, non-compacted endocardial zone.

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The ratio of non-compacted myocardium to compact myocardium at the end of systole was 2.9 : 1 with mean non-compacted myocardium of 1.3 cm and compacted myocardium thickness mean of 0.4 cm. For detailed anatomical information, multidetector computed tomography coronary angiography is carried out. The computerized tomographic (CT) angiography also identified multiple fistula from the left coronary artery towards the entire left ventricular walls including left ventricular free wall, interventricular septum and apex. Coronary artery had markedly tortuous and dilated morphology from left main coronary artery to left anterior descending artery. There was no coronary anomaly in other cardiac structures (Fig. 3).

DISCUSSION

Coronary artery fistulae are rare and are most often diagnosed by angiography incidentally. In our case, because of the prominent firecracker on the whole left ventricle, we could make the early detection of coronary fistulae by noninvasive transthoracic echocardiography without invasive coronary angiography. Imaging with color flow Doppler, using a 4-7 MHz-frequency transducer and high Nyquist limit revealed unusual multiple high velocity color-flows from the epicardial surface into the left

ventricular cavity during diastole. It seemed that those multiple diastolic mosaic flows reflect the presence of fistula between epicardial coronary system and left ventricular cavity. Left ventricular walls including left ventricular free wall and interventricular septum had sinusoidal appearance and mesh like structure which were plexus of multiple small vessels emptying exclusively into the left ventricle through the whole left ventricular wall, and also she had marked trabeculations and deep sinusoidal recesses involving the apex, apico-lateral and apico-inferior walls. This exam revealed two separated layers of myocardium with the ratio of non-compacted to compacted myocardium was greater than 2, measured at end systole in the parasternal short-axis view. The definition and diagnostic criteria of non-compaction is not yet fully estab-

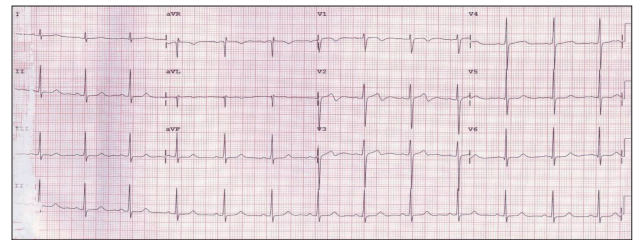


Fig. 1. Electrocardiography showing biphasic T wave in lead V1-3.

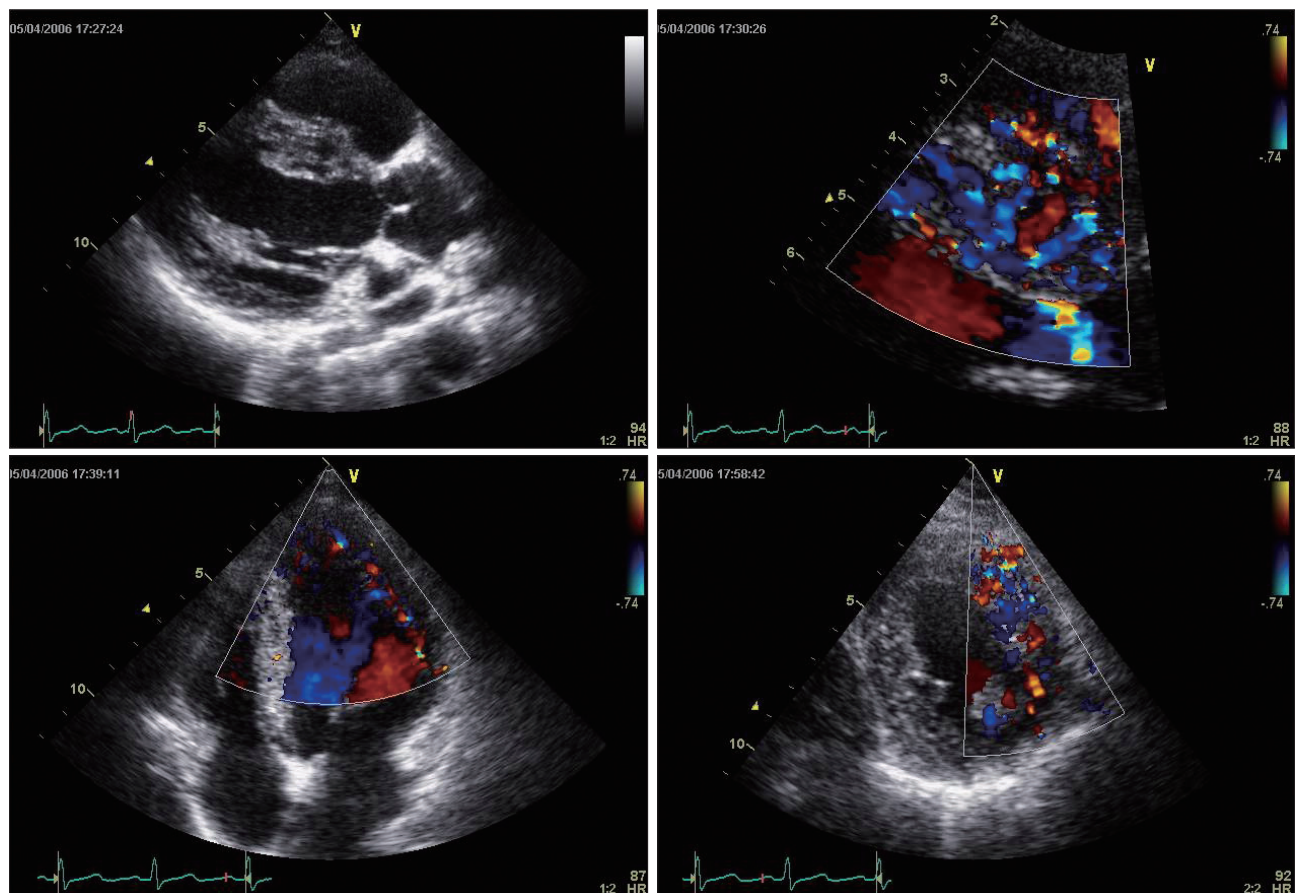


Fig. 2. Transthoracic parasternal long axis, short axis and apical four chamber view: Note the sinusoidal mesh like structure in the basal anteroseptal wall, and multiple sparkling color Doppler flows through the entire left ventricular walls during diastole.

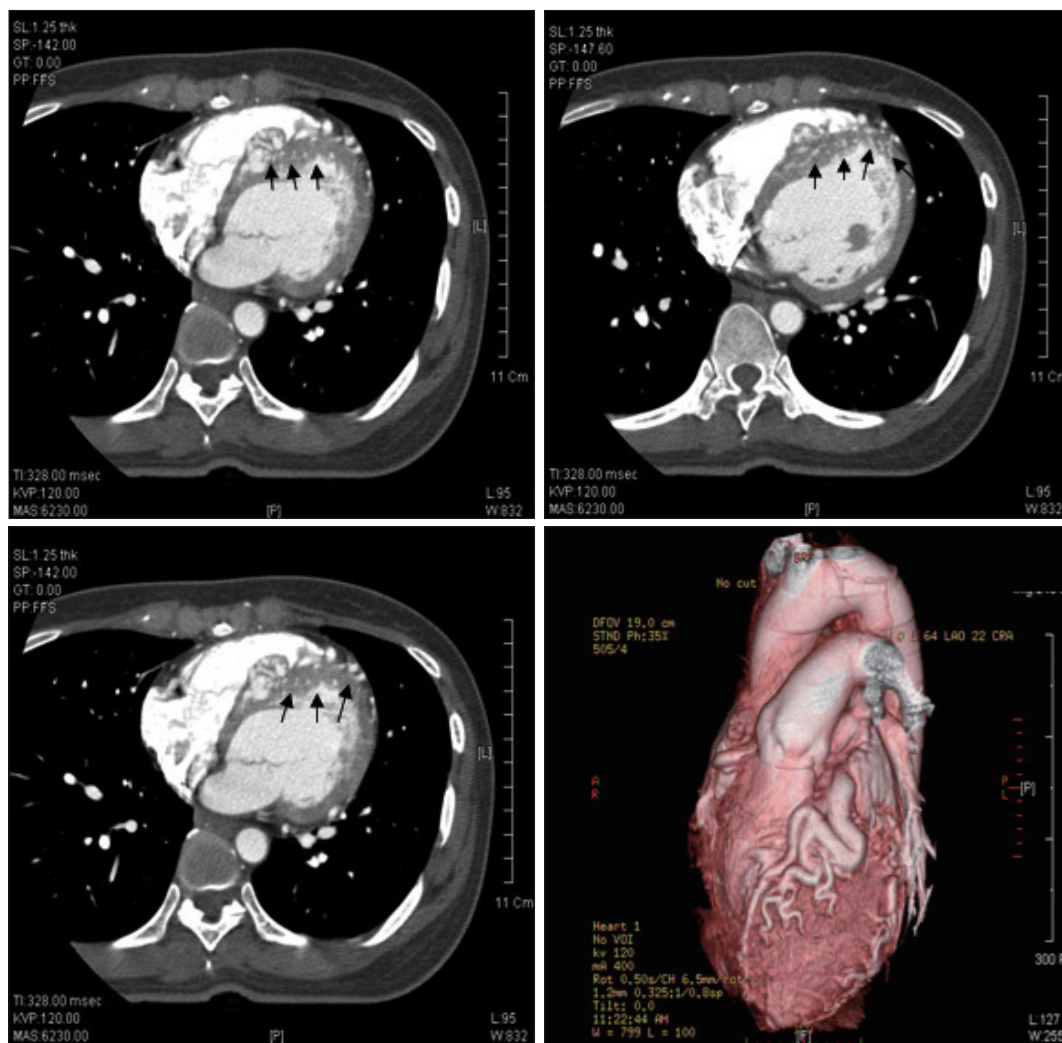


Fig. 3. The computerized tomographic angiography showing multiple fistula draining into left ventricular cavity through interventricular septum and apex (arrows): Coronary artery had markedly tortuous and dilated morphology from left main coronary artery to left anterior descending artery.

lished, but non-compaction is generally characterized by continuity between the ventricular cavity and the intertrabecular recesses, which are filled with blood from the ventricle and which have no communication with the epicardial coronary system. However, in the presented case, which had multiple direct communications with the epicardial coronary system through the entire left ventricular wall. In that point, we concluded this case is more compatible with multiple fistula than non-compaction. Although the etiology of non-compaction has not been fully investigated, our case is thought to result from an arrest in the regression of embryonic sinusoids during the embryonic period. The Arrest of myocardial compaction that occurs in the early stages of fetal development result in the persistence of intertrabecular spaces and incomplete formation of capillaries. In this case, we believed that this process have a role in the formation of multiple fistula with the ventricular cavity and epicardial coronary circulation. But there was no conclusive proof of the etiology of this case. The main

limitation of our case was that we could not perform angiography to confirm coronary artery fistula. But CT angiographic finding was also compatible with multiple fistula draining into the left ventricle. In the presented case, the left ventricular functions were preserved, although the fistulae were very extensive. In a patient with coronary artery fistulae, myocardial ischemia and diastolic volume overload of the left ventricle can be caused by left-to-left shunt: however, the clinical and hemodynamic consequences are incompletely understood. In the presented case, the patient had no symptom and refused further evaluations including coronary angiography and catheterization. With regard to this special case, due to the fact of patient's refusal and absence of angina, it was decided clinical follow-up of the patient. She remained asymptomatic and no complicate events were recorded in the next 5-year follow-up. In conclusion, this case report represents multiple fistula emptied into the left ventricle through the entire left ventricular walls including left ventricular free wall, interventricular sep-

tum and apex, confirmed by transthoracic echocardiography and CT angiography.

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