

Surgical Closure of Coronary Cameral Fistula Draining into the Left Ventricle via a “Fistula Lake”

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Abstract

A left-sided lesion of a coronary cameral fistula (CCF) is extremely rare. Surgical closure of the fistula is indicated when symptoms emerge or as a preventive strategy, while surgical approaches depend on the individual anatomical structures. In particular, a CCF forming a “fistula lake” with multiple inflow vessels is so unique that few studies have focused on the technique to close it. We report the successful management of a CCF originating from multiple coronary arteries and draining into the left ventricle via a “fistula lake” by ligation and clipping of associated communication tracts. On the postoperative coronary computed tomography angiography, the fistula lake and the small vessels entering it had all disappeared.

Keywords

Coronary Cameral Fistula, Fistula Lake, Surgical Repair

1. Introduction

A coronary cameral fistula (CCF) must be closed when any symptoms emerge or as a preventive strategy. Most CCFs originate from right coronary arteries, and >90% of CCFs drain into the right heart structures [1]. A CCF connecting multiple coronary arteries and the left ventricle is thus quite rare. Though several types of surgical techniques reported to date include the ligation or division of the fistula, closing the fistula directly, and closing it with an autologous pericardial patch from the inside of the cardiac chamber or pulmonary artery, few have referred to how to manage a CCF forming a “fistula lake” with multiple inflow vessels explicitly [2]. According to Lowe and colleagues, the diffusely distributed

fistulas are best obliterated by opening the recipient cardiac chamber while on cardiopulmonary bypass to completely close all fistulous tracts [3]. Mangukia and colleagues have insisted if a “fistula lake” is found, all of the vessels entering the lake must be identified and ligated or clipped [2]. Here we present an extremely rare case of a CCF originating from multiple coronary arteries and draining into the left ventricle via a “fistula lake”. The CCF was successfully closed by ligation and clipping of the communication tracts without opening ventricles.

2. Case Presentation

The patient was a 52-year-old Japanese man with a medical history of dyslipidemia and chronic heavy cigarette smoking, and was prescribed with Rosuvastatin. He presented to our cardiovascular outpatient department in the morning complaining of worsening exertional chest pain over approximately the prior 1 month. Vital signs and the physical examination were completely normal including auscultation of the heart and lungs. Electrocardiography showed non-specific ST-segment depression in leads II, III, and aVF. Acute coronary syndrome was suspected, and emergent coronary angiography confirmed significant stenosis in the left anterior descending artery (LAD) and a hypoplastic right coronary artery (RCA) (**Figure 1(A)**). A stream of contrast agent was seen originating predominantly from the left circumflex artery (LCx) and slightly from the branches of the LAD and RCA and draining into the left ventricle via a fistula lake (**Figure 1(A)**). Transthoracic echocardiography showed partial hypokinesis of the basal anterior and septal walls, with preserved left ventricular function.

We diagnosed unstable angina pectoris with a culprit lesion of the LAD and a CCF. The heart team decided to perform coronary artery bypass grafting (CABG) and surgical closure of the CCF. Preoperative coronary computed tomography angiography (CCTA) revealed multiple fistulous tracts predominantly

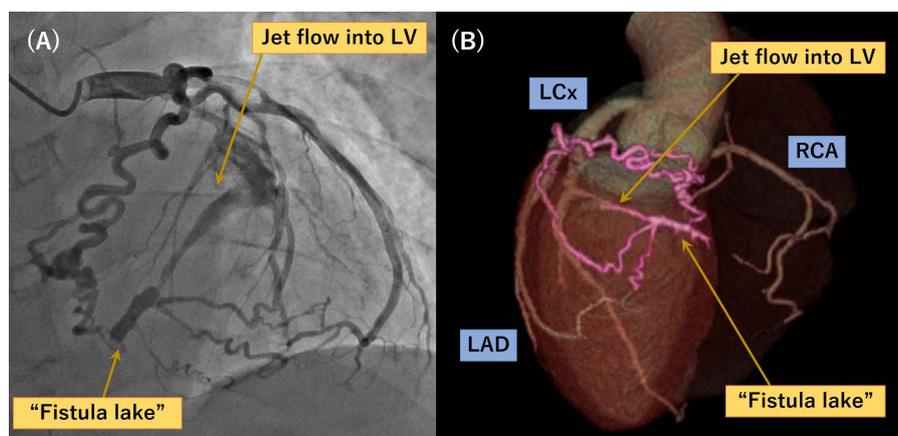


Figure 1. (A) Coronary angiogram showing multiple coronary arteries draining into the LV via a “fistula lake”; (B) Preoperative coronary computed tomography angiography showing the fistulous tracts that had converged to form a “fistula lake” and then entered the base of the LV posterior wall myocardium. LV, left ventricle.

from the LCx, with smaller vessels emanating from the LAD and RCA. The fistulous tracts converged to form a fistula lake and then entered the base of the left ventricular posterior wall myocardium (**Figure 1(B)**). Preoperative transesophageal echocardiography with color Doppler demonstrated a flow jet appearing from the base of the left ventricular posterior wall.

Through a median sternotomy, cardiopulmonary bypass was established by ascending aorta and bicaval cannulation. After the on-pump beating-heart CABG, comprised of grafting the *in situ* left internal thoracic artery to the LAD, both the aorta and left internal thoracic artery were clamped, and antegrade cardioplegia solution was administered to achieve cardiac arrest. Following the careful dissection of epicardial fat with the use of an ultrasonic scalpel, visual inspection of the left lateral to posterior aspect of the heart revealed multiple small branches of coronary arteries converging to form a fistula lake. Through further dissection around the lake, we observed the fistulous tract entering the myocardium just adjacent to the coronary sinus. Both ligation and clipping of the outflow tract were performed. Inflow vessels that could be identified were clipped as well.

Weaning from the cardiopulmonary bypass was uneventful, and the bypass graft showed good flow. Intraoperative transesophageal echocardiography demonstrated good contractility of all segments, and no fistula jet flow could be detected. On postoperative day 9, CCTA revealed that all fistulous tracts as well as the fistula lake had disappeared (**Figure 2**), and a widely patent graft to the LAD was observed (**Figure 2**). The patient was discharged uneventfully 13 days after the surgery.

3. Discussion

A coronary cameral fistula (CCF) is an uncommon anomalous communication

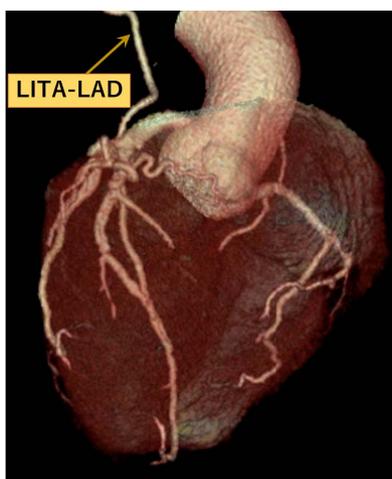


Figure 2. Postoperative coronary computed tomography angiography confirming that all of the fistulous tracts including the fistula lake had disappeared, along with a widely patent graft to the LAD. LAD, left anterior descending artery; LCx, left circumflex artery; LV, left ventricle; RCA, right coronary artery.

between an epicardial coronary artery and a cardiac chamber. Most cases of CCF are asymptomatic, detected incidentally, and conservatively managed with continual follow-up [4]. However, CCFs must be surgically closed when any hemodynamically significant situation, such as myocardial ischemia or congestive heart failure, emerges. Surgical closure may also be performed as a preventive strategy, to preclude aneurysm formation, infective endocarditis, and other cardiac disorders [2]. Our patient presented with worsening exertional chest pain, and coronary angiography showed significant stenosis of the LAD and a CCF originating predominantly from the LCx and draining into the left ventricle, which suggested the coronary steal phenomenon.

Since its first description by Biörck and Crafoord in 1947 [5], surgical repair of a coronary fistula has been the most effective treatment [6]. There are reports of CCFs being treated by transcatheter procedures, including coils, plugs, or detachable balloons [7], but these devices carry the risk of migrating within the coronary artery branches or to the extracoronary vascular structures [8]. Although various surgical techniques have been reported for fistula repair, few studies have focused on the procedure for a fistula lake with multiple inflow vessels. In our case, the lake had multiple inflow vessels but only one outflow vessel. As the inflow tracts were small and assumed to be difficult to completely detect, we assigned the highest priority to closure of the outflow tract. The outflow vessel was identified and closed by both ligation and clipping, followed by clipping of the inflow vessels to the extent possible. As a result, postoperative CCTA revealed the disappearance of all fistulous tracts as well as the fistula lake.

Since the site surrounding the fistula lake was the posterior aspect of the heart just adjacent to the coronary sinus, we chose to repair the fistula with the patient under cardiac arrest using cardiopulmonary bypass. This made sufficient heart displacement possible so that we could safely reach the posterior wall just adjacent to the coronary sinus and fully define the entire fistulous network.

Although surgical management remains the most effective treatment for CCFs, Basit *et al.* reported a case of surgical failure 6 years after closure of a CCF, indicating that CCFs can persist or recur after surgical ligation [9]. Longer and more rigorous follow-up of patients with CCFs is thus necessary.

This case report has potential limitations. Our strategy aimed at closing all of the fistulous tracts detectable which resulted in the disappearance of the left-to-right shunt. Since intervention may become more difficult or impossible when the fistulas are diffuse, despite the ischemia in the patient, sometimes intervention should not be considered. Symptoms can be relieved by therapy with metoprolol or calcium channel blockers [10].

4. Conclusion

We surgically repaired a CCF originating from multiple coronary arteries, including the LAD, LCx, and RCA, and draining into the left ventricle via a fistula lake. The complete obliteration of all fistulous tracts was accomplished by the li-

gation and clipping of the outflow vessel from the lake into the left ventricle and the multiple inflow vessels originating from the coronary arteries.

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Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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