中文題目：病例報告：一位重度二尖瓣狹窄的病患，其冠狀動脈血管攝影診斷左心耳之血栓合併左迴旋枝-左心房廔管

英文題目：A case of Mitral Stenosis with Left Atrial Thrombi and Coronary-cameral Fistula Between Branches of Left Circumflex Artery and Left Atrium

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Background: Coronary-cameral fistula (CCF) is a rare anomalous communication between a coronary artery and cardiac chamber, congenitally or secondary to trauma, seen in 0.1% of patients undergoing coronary angiography. For significant mitral stenosis, the CCF is considered a specific sign for left atrial thrombi from some case reports and studies. Here we present a case of severe mitral stenosis (MS), with thrombus at left atrial appendage (LAA) from transesophageal echocardiography (TEE), and the patient's coronary artery angiography (CAG) showed fistula from branches of left circumflex artery artery (LCx) to left atrium (LA).

Method: Here we report a case of rheumatic mitral stenosis, whose coronary angiography revealed coronary-cameral fistula from atrial and ramus branches of left circumflex artery. A 61-year-old woman with a history of rheumatic mitral stenosis and paroxysmal atrial fibrillation, experienced embolic event with ischemic stroke. Transesophageal echocardiography revealed thrombi at left atrial appendage. She underwent a coronary angiography before mitral valve replacement, and the left coronary angiography showed coronary-cameral fistula from atrial and ramus branches of left circumflex artery draining into left atrium, and the thrombi at left atrial appendage was enhanced by contrast staining.

Result: A 61-year-old female patient, with a history of mitral stenosis and paroxysmal atrial fibrillation on continuous oral anticoagulant therapy with Warfarin, who presented with exertional dyspnea and had regular follow at out-patient department for years. On physical examination, her blood pressure was 117/79 mmHg and the heart rate was 69 beats per minute with regular heart beats. A diastolic rumbling murmur was also detected. However, she developed embolic event with left cerebellar infarction in 2016. Electrocardiogram revealed normal sinus rhythm and chest X-ray film revealed left atrial dilation and mild increased pulmonary vascularity during hospitalization. Transthoracic echocardiography revealed normal systolic function, enlargement of left atrium with spontaneous echo contrast, and severe MS with a mitral valve area of 1.13 cm^2 by pressure half time; transesophageal echocardiography showed thrombus-like mass about 1 x 3 cm in size at left atrial appendage (Figure 2). She received cardiac catheterization for pre-operative evaluation, and the coronary angiography revealed coronary-cameral fistula,

involving the atrial branch and ramus branch of left circumflex artery emptying into left atrium (Figure 1A). Fluoroscopy also revealed LA appendage contrast stain in left atrium (Figure 1B). She undergone open mitral valve replacement later, and thrombi in left atrial appendage was found during surgery (Figure 3).

A

B

Figure 1 (A) Left coronary artery angiography revealed revealed neovascularization and fistula (white arrow) from atrial branch of left circumflex artery in right anterior oblique projection. (B) Stasis of contrast delineating the thrombus at left atrial appendage

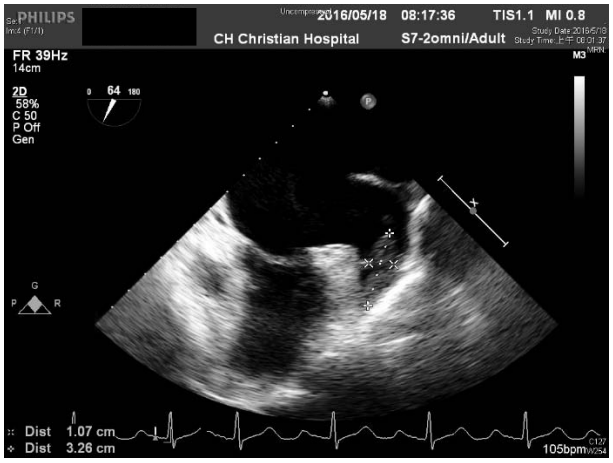

Figure 2
Transesophageal echocardiography revealed thrombus at left atrial appendage

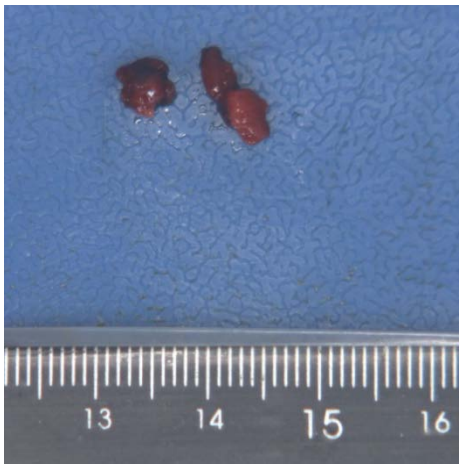

Figure 3
Thrombi taken from left atrial appendage at surgery

Conclusion: Many studies for coronary neovascularity and fistula in mitral stenosis patients were conducted before the extensively use of TEE, and reported with high specificity and low sensitivity for diagnosis of left atrial thrombus by TTE or coronary

angiography. We believed that by combination of transthoracic, transesophageal echocardiography and the coronary neovascularity, including coronary-cameral fistula, would further improve the sensitivity and specificity of detection of left atrial thrombi. However, the mechanism for thrombi and formation of coronary artery fistula or neovascularization is not very clear for now.

Table 1. Case reports with mitral valve stenosis and coronary artery fistulas

Author	Age	Sex	Origin of fistula	Drainage site	Evidence of LA thrombus before surgery	Left atrial thrombus during surgery
1975, King(1)	71	M	LAD	LA	-	+
1977, Soulen(2)	72	F	Atrial branches of both RCA and LCA	LA	4 (angiography)/6	4/6
	57	F	Atrial branches of both RCA and LCA	LA		
	59	F	Atrial branches of both RCA and LCA	LA		
	63	F	Atrial branches of LCA	LA		
	65	F	Atrial branches of LCA	LA		
1996, Sakamoto(3)	56	F	LCA	LA	+ (CT)	-
1996, Iida(4)	53	M	RCA and LCA	PA	+	+
	52	F	LCA	PA	-	-
	65	M	RCA	PA	-	-
1996, McClung(5)	56	F	Proximal Circumflex	LA	+ (TEE)	+
1997, Lin(6)	50	F	LAD	PA	-	-
2001, S. Harikrishnan(7)	62	F	Distal LCx (single coronary artery with RCA arising from LAD)	LA	-	-
2003, Gunduz(8)	56	F	LCx	PA	-	-
2006, Tanriverdi(9)	54	M	LAD and conus branch of RCA	PA	-	-
2006, Neema(10)	29	F	RCA	PA	-	-
2006, Said(11)	78	F	LCx	LA	+ (TTE and TEE)	+
2011, Chakraborty(12)	52	F	LCx and RCA	LA	+ (TTE and TEE)	+
2012, Hammami, R.(13)	43	M	LCx	LA	+ (TTE and TEE)	

2013, Uğur GÜRCÜ(14)	51	M	LCx	LA	+ (TTE and TEE)	+
2014, Fennich(15)	57	F	LCx	PA	-	-
2014, Sharma(16)	45	-	OM	LA (CTA)	-	-
2014, Gholoobi(17)	74	M	LCx	LA	+ (TTE)	+
2015, Nagre(18)	45	F	LCx	LA	+ (TEE)	+