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A Rapidly Progressing Native Coronary Artery to an Aneurysm Due to Distal Coronary to Ventricular Fistula- causing Anginal Chest Pain

Pooja Sethi[1], Ghulam Murtaza[2], Muhammad F. Khalid[2] and Timir K. Paul[1*]

[1]Department of Internal Medicine, Division of Cardiology, East Tennessee State University, Johnson City, TN, USA.
[2]Department of Internal Medicine, East Tennessee State University, Johnson City, TN, USA.

Authors' contributions

This work was carried out in collaboration between all authors. Author PS developed the idea presented in article and wrote the first draft of the manuscript. Authors GM and MFK managed literature review and completed the discussion section. Author TKP managed the final review and approval prior to submission. All authors read and approved the final manuscript.

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Case Study

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ABSTRACT

Coronary artery fistula (CAF) is a rare congenital or acquired coronary anomaly. Although the majority of patients with CAF are asymptomatic, many of them present with angina pectoris, atypical chest pain, palpitation, fatigue, bacterial endocarditis, congestive heart failure, pericardial effusion and sudden cardiac death. We present a case of coronary artery fistula with recurrent episodes of chest pain that were thought to be resulting from coronary artery atherosclerosis. His left heart catheterization showed right coronary artery to left ventricular fistula that was not believed to be causing his chest discomfort as it was small and was left untreated. He continued to have these symptoms until fistula was repaired. Interestingly his fistula leads to a large coronary artery aneurysm over a short period of time. Our case is important with the fact that in the symptomatic patient with untreated fistula might get complicated and would lead to persistent symptoms.

**Corresponding author: E-mail: pault@etsu.edu;*

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1. INTRODUCTION

Coronary artery fistulas (CAF) are a rare but potentially curable cause of ischemic chest pain. Based on coronary arteriography, the incidence of CAF is 0.1% [1]. While most of the patients with CAF are, asymptomatic and can be managed conservatively, the symptomatic patient should be evaluated for possible intervention. Untreated cases can develop several complications including myocardial infarction, cardiac tamponade, thrombosis, and endocarditis. Timely diagnosis and intervention are important to prevent these complications.

2. CASE REPORT

A 52-year-old gentleman with multiple cardiac risk factors including diabetes mellitus, hypertension, and family history was seen in outpatient clinic with episodes of intermittent, sharp non-radiating mid- sternal chest pain 10/10 in intensity. The patient reported relief of pain with sublingual nitroglycerin. He denied any palpitations, dyspnea on exertion, paroxysmal nocturnal dyspnea or orthopnea. Physical examination showed normal vitals. There was no jugular venous distension, normal heart sounds with no murmurs on auscultation and no pedal edema was noticed. EKG showed normal sinus rhythm, no ST-T wave changes, normal QT interval and mild left ventricular hypertrophy. Echocardiogram showed normal ejection fraction with mild left ventricular hypertrophy. He underwent a stress test which showed inducible angina with apical and septal ischemia. The patient was started on dual antiplatelet therapy and taken to cardiac catheterization laboratory for elective percutaneous coronary intervention. The cardiac catheterization showed 70% stenosis in the Left anterior descending (LAD) artery and a right coronary artery (RCA) to left ventricle (LV) fistula. The fistula was considered not causing symptoms. The LAD was stented and the patient was discharged. The patient was asymptomatic for 6 months but then started developing episodes of recurrent chest pain as he had in the past. The possibility of stent thrombosis was considered and a repeat catheterization was done which showed widely patent stent. Also noticed was the RCA to LV fistula but now the RCA was very ectatic and almost 3 times the normal size (Figs. 1 and 2). The fistula was thought to be the cause of the episodes of recurrent chest pain. The patient was scheduled for coronary artery bypass surgery. The fistula was ligated and repaired. The patient tolerated the procedure well and is currently chest pain-free 6 months after surgery.

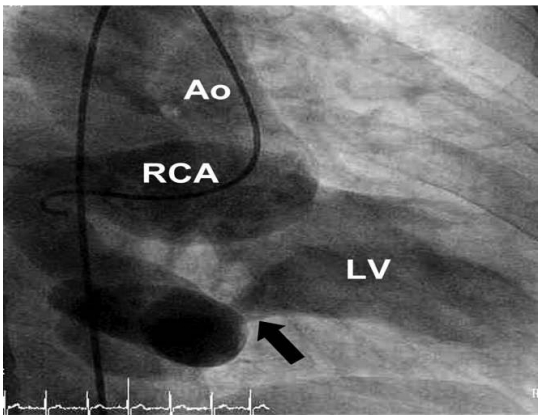

Fig. 1. Coronary angiogram showing dilated RCA to LV Fistula (arrow)

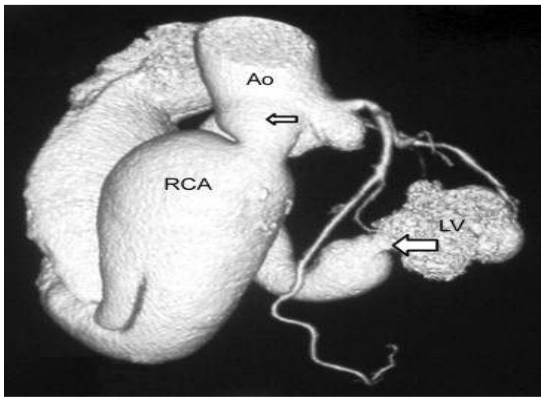

Fig. 2. CT reconstructed coronary angiogram showing RCA to LV Fistula (arrow)

3. DISCUSSION

A communication between a coronary artery and heart chamber is called coronary cameral fistula and to a segment of the systemic or pulmonary circulation is called coronary arteriovenous fistula. The majority of these fistulas are congenital that may occur as an isolated finding or may appear as in the context of other congenital cardiac anomalies or structural heart defects. Acquired cases result from cardiac surgeries such as coronary artery bypass grafting [2], myocardial biopsies or valve replacement. CAF have also been thought to

occur in severe coronary atherosclerosis due to aberrant neovascularization in which collateral vessels inadvertently terminate into the cardiac chamber [3]. Most CAF drain on the right side of the heart. Fistula draining into the left ventricle is extremely rare and account for 3% of all CAF.

Major sites of origin of the fistula are from the RCA 40-60%, left anterior descending artery 30-40%, circumflex and combination 0.5-1.9% [4]. Most fistulas terminate in venous chamber or vessel and, only rarely, into the left ventricle or the pericardium. The major sites of the termination include the right side of the heart (90%), left ventricle, left atrium and the coronary sinus. The most frequent sites of termination on the right side of the heart, in descending order, are the right ventricle, right atrium, and pulmonary vasculature.

Usually, coronary artery fistulas are small, do not cause any symptoms, and are clinically undetectable. Common symptoms that may develop include chest pain, dyspnea, and palpitation. These symptoms usually develop in patients above 40 years of age [5]. Depending upon the size and blood flow through coronary artery fistula, a murmur may be heard. This might be the only incidental finding in asymptomatic patient leading to identification and referral to the cardiologist. Continuous murmur is reported more often than a systolic and diastolic murmur [6]. When the amount of blood flow through fistula is small, a cardiac murmur may not be present. Larger fistulas are usually 3 times the size of a normal caliber of a coronary artery and can cause myocardial ischemia because of a coronary steal mechanism [7]. The pathophysiologic mechanism of coronary artery fistula is myocardial stealing or reduction in myocardial blood flow distal to the site of coronary artery fistula connection. Other complications that can happen with large fistula include congestive heart failure, infective endocarditis, aneurysm formation, and rupture or thrombosis of the fistula [8,9,10]. The factors that determine the hemodynamic significance of the fistulous connection include the size of the communication, the resistance of the recipient chamber, and the potential for development of the myocardial ischemia.

Our patient did not have any history of cardiac surgery. His CAF was most likely congenital in origin occurring in isolation without any other congenital cardiac anomaly. He did not become symptomatic until the fifth decade when presented with anginal symptoms that were thought to be resulting from coronary artery atherosclerosis. His symptoms continued to persist until fistula was repaired. Usually, patients with CAF remain asymptomatic unless fistulous communication gets enlarged to such an extent that complications develop. In a study of 185 patients hemodynamically significant fistulas resulting in clinical symptoms or sequelae was in approximately 19 percent of patients under the age of 20 compared those over the age of 20 in whom the prevalence of symptoms or complications was 63% [11]. Our patient's symptoms were related to coronary artery steal, with resultant ischemia of the segment of myocardium perfused by the coronary artery distal to the fistula causing recurrent episodes of chest pain. It is unclear that what could have caused such rapid progression of an aneurysm in coronary artery secondary to fistula in our patient. There are no previously reported cases of such rapid increase in shunt size over a short period. Significant myocardial ischemia from a large sized aneurysm is what we believe made our patient so symptomatic over short time. Our case highlights the importance of close follow-up of elderly patients with fistula being managed conservatively especially those with concomitant atherosclerotic coronary artery disease. These patients may rapidly become symptomatic with an increase in the size of fistula that would need repair as soon as possible to prevent further complications.

Precise evaluation of the anatomical origin and course, drainage site and the possible presence of an aneurysm are necessary when considering treatment indication and options.Coronary artery fistula can be detected by two-dimensional echocardiography. However, it is not as sensitive as transesophageal echocardiogram in the diagnosis and defining precise location of coronary fistula [12]. Coronary angiography is the gold standard and should be used before planning any intervention. Cardiac catheterization helps to identify the hemodynamic significance of the fistula and to provide a detailed anatomy of the fistula.

Small asymptomatic and incidentally discovered fistula can be managed conservatively [13]. Symptomatic patients can be treated either surgically or transcatheter closure device. Both of these approaches have been found equally effective and there is no significant difference in morbidity and mortality [14]. Our patient had significant aneurysmal dilatation and distally located fistula and thus underwent open surgical approach. The choice between these procedures

should be based on size, location, origin and drainage of the fistula.

4. CONCLUSION

Coronary artery fistula should be kept in mind as an important differential of ischemic chest pain besides coronary artery atherosclerosis. Once the patient becomes symptomatic, further evaluation should be done for the different curative options. In untreated cases, the fistulous connection might continue to dilate progressively resulting in persistent symptoms and development of complications. Good history and physical exam are important pillars for early diagnosis and referral to the cardiologist.

CONSENT

As per international standard or university standard, the patient's written consent has been collected and preserved by the authors.

ETHICAL APPROVAL

It is not applicable.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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