

SURGICAL TREATMENT OF CORONARY ARTERY FISTULAS AND SEBSEQUENT NONSPECIFIC ECG CHANGES

E. ÖZAL, MD,
M.H. US, MD,*
A.T. YILMAZ, MD,
F. CİNGÖZ, MD,
B.S. ÖZ, MD,
U. DEMİRKILIÇ, MD,
H. TATAR, MD,

From:

Gülhane Military Medical
Academy,
Department of
Cardiovascular Surgery,
Ankara and *İstanbul,
Türkiye

**Adress for
reprints:**

Dr. Ertuğrul Özal
Gülhane Askeri Tıp
Akademisi
Kalp ve Damar Cerrahisi
Ana Bilim Dalı
06018 Etlik, Ankara
Tel : +90 312 3045222
e-mail: ozals@tr.net

Coronary artery fistulas are rare anomalies and their diagnosis is usually difficult mostly due to the absence or atypical presentation of symptoms. Current practice still have problems in both medical and surgical therapeutic approaches due to the limited number of cases. We report our experience on surgical treatment of 19 patients with the diagnosis of coronary artery fistula and point out the postoperative ECG changes that may be seen following surgery.

Between 1990 and 2000, nineteen patients underwent surgical closure of coronary artery fistulas. Mean age was 30.8 ± 14 (ranging from 3 to 51 years). The fistula was originating from the right coronary artery in 9 (47%) and from left coronary artery in 10 (53%) cases. Fifteen (79%) of the patients were male while remaining 4 (21%) were female. Site of fistulous connection was at the right ventricle in 5 (26%), right atrium in 6 (31%), pulmonary artery in 6 (31%) and coronary sinus in 2 (10%) cases. Four of the cases had additional cardiac lesions and underwent surgical procedures related to these lesions. Cardiopulmonary bypass was used in 9 (47%) of the cases during surgical repair.

There was no operative mortality. Two patients (10%) had nonspecific ECG abnormalities during postoperative period. Postoperative control angiographic studies revealed normal coronary arteries in these patients.

Surgical intervention should be performed for preventing coronary artery fistula related complications. Mortality and morbidity of coronary artery fistula surgery significantly decrease when repair is made before fistula related complications develop. Postoperative ECG changes may be seen following surgical repair. Although they become normal after a period, postoperative control angiography should be performed to evaluate coronary anatomy.

Key words: Coronary artery fistula, surgical closure of fistula, ECG changes

Coronary artery fistula (CAF) is quite rare with an incidence of 0.16% to 0.20% in patients undergoing coronary angiography (1,2). Although the majority of CAFs are congenital, it has been reported that they may be related to

chest trauma (2), cardiac surgery (3,4) or coronary angioplasty (5). They are usually asymptomatic especially in the first two decades, but complications such as congestive heart failure, dyspnea, fatigue, angina or chest pain due to myocardial ischemia, infective endocarditis, formation and rupture of aneurysm have been reported (6-9). Also conduction tissue disorders and rhythm disturbances, especially atrial fibrillation may be associated with CAF (8). Although coronary arteriography is the essential method in diagnosis to accurately delineate the anatomy and plan the surgical repair, the fistula may be demonstrated by color-flow echocardiography as well (10). Successful transcatheter coil embolism of CAF has been reported (11,12), but still the surgical closure is the preferred therapy (13). Although it has been reported that myocardial ischemia may occur after surgical closure because of coronary artery obstruction (14), current knowledge of the literature can not explain the postoperative nonspecific ECG changes that may be encountered following surgical closure of CAFs in cases in which postoperative coronary arteriographies reveal normal coronary anatomy. In this paper, we report our experience on surgical closure of CAFs in 19 patients and point out the postoperative nonspecific ECG changes we identified in 2 cases.

MATERIALS AND METHOD

Nineteen patients underwent surgical closure of CAF between 1990 and 2000 in Ankara and İstanbul Hospitals of Gülhane Military Medical Academy. Mean age was 30.8 ± 4 , ranging between 3 to 51 years. Fifteen (79%) of the patients were male while remaining 4 (21%) were female. The fistula was originating from the right coronary artery in 9 (47%) and from left coronary artery in 10 (53%) patients. Site of fistulous connection was at the right ventricle in 5 (26%), right atrium in 6 (31%), pulmonary artery in 6 (31%) and coronary sinus in 2 (10%) cases. Four of the cases had additional cardiac lesions and underwent surgical procedures related to these lesions. Cardiopulmonary bypass was used in 9 (47%) of the cases during surgical repair. A great majority of the

patients were asymptomatic and the diagnosis has been made due to the continuous murmur detected in physical examination. Asymptomatic patients were younger in age (21 ± 9) when compared to symptomatic patients (39.7 ± 8). Of the 9 patients who were asymptomatic, fistula was originating from right coronary artery in 6, and from left coronary artery in 3 patients. The most common symptom of the patients was effort dyspnea which was the main complaint of 5 patients (case 3,8,9,11,16). Palpitation or/and atrial arrhythmias were common in patients in whom fistula was originating from the right coronary artery (case 6,13,15). Two children who were 3 and 4 years old had this diagnosis eventually because of the continuous murmur which was detected in routine physical examinations (Case 1,2). Two patients presented with the complaints of angina pectoris (case 7,17). There was associated lesion of the occlusion of the left anterior descending artery in one of these patients and coronary artery bypass surgery was performed in addition to the closure of the fistula while coronary angiography of the other patient revealed only CAF. Only 1 patient with associated mitral stenosis had the history of bacterial endocarditis and she underwent mitral valve replacement in addition to the closure of the fistula (case 13). The data of all patients is shown on Table 1.

RESULTS

All of the patients underwent closure of the fistulas and 4 had additional surgical procedures related to their associated lesions. Surgical approach was through median sternotomy in all patients. Cardiopulmonary bypass (CPB) was used in 9 of the patients. Ligation of the fistula was the most common surgical technique which was applied in 11 of the patients. CPB was not used in ten of these 11 patients, while remaining 1 patient required CPB because of additional procedure of CABG. During ligation technique "the feeding" coronary artery very close to the fistulous connection was closed by one or two suture ligatures placed around it. The closure was kept temporary for several minutes, after being confirmed that there was no ECG change the suture was tied down. In 4 of the

Table 1. Patient Data

Case	Age /Sex	Diagnosis	Associated lesion	-Symptom -Sign	Surgical technique	CPB	ECG changes
1	3/M	Cx → RV	Cx aneurysm	-Asymptomatic -Murmur, cardiomegaly	Aneurysmectomy + transarterial closure	+	-
2	4/ F	Cx→ CS	ASD	-Asymptomatic -Murmur	Transchamber closure + ASD primer closure	+	-
3	20/M	LAD → PA	-	-Effort dyspnea -Murmur, cardiomegaly	Ligation + transchamber closure	-	-
4	21/M	RCA → RV	-	-Asymptomatic -Murmur	Ligation	-	-
5	21/M	D1 → PA	-	-Asymptomatic -Murmur	Ligation + transchamber closure	-	-
6	37/M	RCA → RA	-	-Palpitation, -Murmur, atrial fibrillation	Ligation	-	-
7	43/M	D1 → PA	LAD stenosis	-Angina pectoris -Heart failure	Ligation + CABG (LIMA-AD)	+	-
8	38/M	D1 → PA	-	-Effort dyspnea, fatigue -Heart failure	Ligation	-	+
9	42/M	LAD → PA	-	-Effort dyspnea, fatigue -Heart failure	Ligation + transchamber closure	-	+
10	35/F	RCA → RA	-	-Asymptomatic -Murmur, atrial premature beats	Transchamber closure	+	-
11	51/F	Cx → RV	-	-Effort dyspnea -Angiographic diagnosis	Transarterial closure	+	-
12	39/M	RCA → RA	-	-Asymptomatic -Murmur, cardiomegaly	Ligation	-	-
13	41/F	RCA → RA	Mitral Stenosis	-Palpitation, infective endocard. -MS related	Transchamber closure +MVR	+	-
14	21/M	RCA → RV	-	-Asymptomatic -Murmur	Transchamber closure	+	-
15	34 /M	RCA → RA	-	-Palpitation -Atrial fibril	Ligation	-	-
16	41/M	LAD → PA	-	-Effort dyspnea -Angiographic diagnosis	Ligation + Transchamber closure	+	-
17	50/M	Cx → CS	-	-Angina -Angiographic diagnosis	Transchamber closure	+	-
18	19/M	RCA → RV	-	-Asymptomatic -Murmur	Ligation	-	-
19	26/M	RCA → RA	-	-Asymptomatic -Atrial extrasystole	Ligation	-	-

cases in which ligation technique was performed, additional running suture to enclose all the involved vessels and the underlying walls was placed under ECG control.

In fistulas entering the pulmonary artery, the preferred method was transchamber closure through pulmonary arteriotomy and additional ligation of the feeding artery without CPB. In one case (case11) the fistula was between the circumflex artery and right ventricle and the artery was very large and continuing beyond the fistula. We preferred transarterial closure technique for this patient, opened the artery

above the fistula, closed the fistulous connection with a running suture and closed the arteriotomy. Six patients required transchamber closure on CPB, because it was difficult to identify the feeding arteries. Two of them had additional surgical procedures (1 ASD closure, 1 MVR). Right atriotomy was performed in all of these patients and cold cardioplegic solution was infused to identify connection site and it was closed by over and over sutures and supplemented with pledget mattress sutures. To test the security of the closure cardioplegic solution was infused and additional sutures were placed if the shunt was

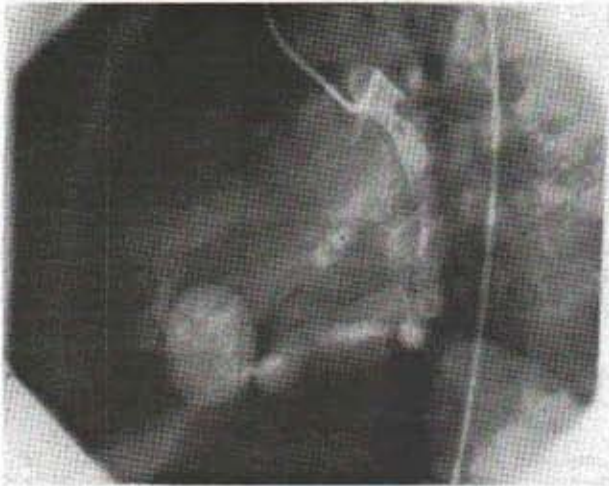


Figure 1. Cineangiogram of the patient with CAF from the circumflex artery to the right ventricle associated with coronary artery aneurysm.

still existing.

In case 1, there was a large aneurysm of the circumflex artery and the fistulous connection was to the right ventricle after the aneurysm (Figure 1). The artery was not continuing beyond the fistula, the aneurysm was excised, afferent and efferent ends of the fistula were closed by pledget sutures in this patient. In case 2, which had circumflex to coronary sinus fistulous connection and associated ostium secundum type ASD (Figure 2), first the fistulous connection was closed from coronary sinus (transchamber closure) and ASD was closed by primer suture later. In case 7, fistulous connection was between the first diagonal artery and pulmonary artery and the patient had associated occlusion of the left anterior descending artery (LAD). In this



Figure 2. Cineangiogram of the patient with CAF from the circumflex artery to the coronary sinus.

patient, at the first stage of the operation the fistulous connection was closed by ligation of the feeding artery, and then LIMA-LAD coronary bypass was performed on cardioplegic arrest.

Two patients with CAF connecting to the pulmonary artery showed nonspecific ECG changes one week after the surgery. One of them had a fistula originating from the first diagonal artery (case 8, Figure 3), and the other one from the LAD. Early postoperative ECG recordings of these 2 patients were normal, but the ECGs at the postoperative 7th day revealed inferior and lateral ischemia. Control angiography was performed for these patients and coronary arteries were found normal (Figure 4). There was no hemodynamic problem in these two patients

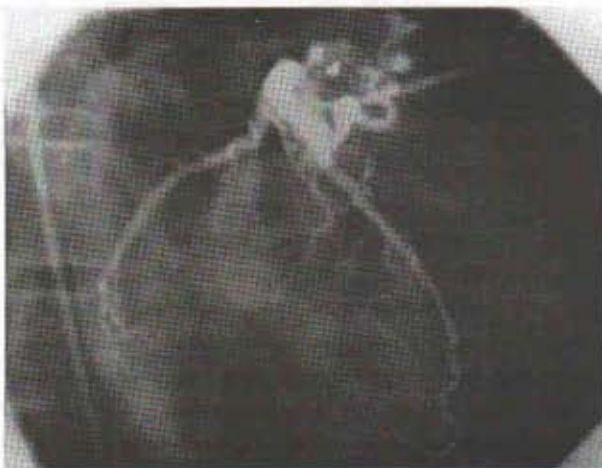


Figure 3. Cineangiogram of the patient with CAF from the first diagonal artery to the pulmonary artery.



Figure 4. Postoperative control cineangiogram of the patient with nonspecific ECG changes revealing normal coronary anatomy

during preoperative period and the ECG changes turned to normal 2 months after the surgery.

There was no operative mortality or morbidity. Four patients required inotropic support. None of the patients had significant arrhythmias. Postoperative course was uneventful in the other cases. Mean follow-up duration was 3.1 ± 1.2 years. All of the patients had functional capacity of Class I (NYHA) during follow-up period. All of the patients were asymptomatic during follow-up period.

DISCUSSION

Although the majority of CAFs are congenital anomalies, they may be acquired related to chest trauma, cardiac surgery or coronary angioplasty (2-5). Clinical features are similar in both forms. Most patients are asymptomatic in infancy and childhood and present for treatment later in life because of the onset of fistula related complications such as congestive heart failure, dyspnea, fatigue, angina or chest pain due to myocardial ischemia, infective endocarditis, the formation and rupture of an aneurysm (6-9). In their study, Lowe and colleagues (15) indicated that 45% of CAFs are asymptomatic and diagnosis can frequently be suspected due to a continuous murmur or ECG findings if it shows right or left ventricular overload or ischemia. Likewise, 9 patients in our series were asymptomatic and the diagnosis was made because of the murmur which was detected in physical examination.

The right coronary artery or its branches are the site of CAF in 50 to 55% of the cases while the left coronary artery or its branches are the involved vessel in 40 to 45%. The fistula was originating from the right coronary artery or its branches in 9 (47%) and from the left coronary artery or its branches in 19 (53%) patients in our series.

Site of the fistulous connection is as important as its origin for its hemodynamic effect. About 40% of CAFs drain to the right ventricle, 25% to the right atrium, 15 to 20% to pulmonary artery, 7% to coronary sinus and 8% to left heart chambers (16,17). In our series, the

connection site was to the right ventricle in 5 (26%), right atrium in 6 (31%), pulmonary artery in 6 (31%), and coronary sinus in 2 (10%) cases.

The three parameters that effect hemodynamics are origin, size and connection site of CAF. Main pathology in CAFs connecting to the right sided chambers is left to right shunt and they cause right ventricle and pulmonary volume overload, and left ventricle volume overload later. The CAFs connecting to the left chambers cause left ventricle volume overload like it is observed in aortic insufficiency (18). The onset of congestive heart failure in patients with CAF may be related to the shunt itself or to the steal phenomenon which occurs because of the direction of coronary blood flow through the low pressured cardiac chamber. At the beginning, the steal phenomenon causes ischemia in the involved coronary artery's area. But global myocardial ischemia may develop later because of the collateral circulation through the other coronary arteries (15,19). In fistulas in which the site of connection is into the left ventricle, blood flow is mostly during diastole and this causes an increase in left ventricle end-diastolic pressure, decrease in perfusion pressure and myocardial ischemia. Signs of congestive heart failure are common in fistulas draining into the coronary sinus and arterialization of the coronary sinus is blamed to be the cause. The increase in coronary blood flow rate through fistulous artery increases shearing stress which causes premature atherosclerosis to develop (18,20).

Indication for operating on small fistulas is still not clear. Some believe that operation is not indicated if the patient is asymptomatic. Although spontaneous closure of the fistula or remaining asymptomatic for years has been reported in some cases (21), in the view of the probability that at least some of the fistulas increase in size and produce fistula related complications, many authors recommend surgical treatment (14,16,20). Keeping the reports about CAF causing sudden death and fistula related complications in mind, we accept the diagnosis of CAF as an indication for operation (22,23). Fistula related

complications include thrombosis, distal emboli and aneurysm rupture (23). In Case 1, although the patient was 3 years old, there was a huge aneurysm of the circumflex artery and it would be expected to rupture in the future if not surgically excised. Today, surgical mortality and morbidity of CAF are considerably better compared to those of the natural course especially when the repair is performed before the onset of complications (15). It has also been reported that mortality and morbidity of the surgical intervention is higher when it is performed after the onset of complications (13). We believe that closing the fistula in asymptomatic patients will prevent complications and the life expectancy of the patient will be higher.

There is little known and reported about postoperative nonspecific ECG changes that may be encountered following the closure of CAF. Although they were nonspecific and did not effect the hemodynamic status of the patients, we have found negative T waves and ST depression in two cases. These ECG changes made us think about any damage to the native arteries during surgical repair and we performed coronary arteriography in these patients. Coronary arteriography of the both patients revealed normal coronary anatomy without any stenosis or occlusion and the successful closure of the fistulas.

It has been reported that a decrease in sympathetic activation occurs both in the proximal and distal arterial beds in arteriovenous fistulas and in these lesions, the increase in blood flow following sympathectomy is less than that of the normal vessels (24). Sudden closure of the fistula tract causes a reflex sympathetic hyperactivation and an increase in vascular resistance. It also causes a sudden decrease in increased blood flow of the proximal artery. In addition, increased vascular resistance of the distal artery may lead to subepicardial ischemia. If the ischemic T waves on the ECG following the closure of the fistula were caused by the above-mentioned mechanism of the increase of vascular resistance, the patient would be expected to be symptomatic. But, there was no ischemic sign in any of our 2 patients. Postoperative control angiographic studies

have revealed normal coronary arteries but an increase in resistance against blood flow. Keeping these normal coronary angiograms and asymptomatic status of the patients in mind, these ischemic T waves can be accepted as the "nonspecific T wave abnormalities" which are related to numerous clinical disorders such as anxiety, hyperventilation, tachycardia, cold exposure, electrolyte disturbances, cerebrovascular disease and anemia. The T wave abnormalities in our two patients turned into normal in control ECG at the postoperative 2nd month.

The goal of the surgical intervention is to close fistulous connection while keeping coronary artery anatomy. It may be safely closed without CPB when the fistula is a discrete one, and location and size are appropriate. However, it must be kept in mind that the fistulous artery will give secondary branches by time and one of these secondary branches may be ligated during surgery. In any suspect of coronary ischemia related to surgical procedure, coronary angiography should be performed to eradicate the possibility of damage to coronary anatomy.

As a result, coronary artery fistulas may result in coronary ischemia, congestive heart failure and endocarditis. Surgical intervention should be performed for preventing CAF related complications. The mortality and morbidity of the surgery significantly decrease when repair is made before fistula related complications develop. Postoperative ECG changes may be seen following surgical repair. Although they do not effect the hemodynamic status of the patient and turn into normal after a period of time, postoperative control angiography should be performed to eradicate the possibility of any damage to coronary anatomy during surgery.

REFERENCES

1. Bailtaxe HA, Wixon D. The incidence of congenital anomalies of the coronary arteries in the adult population. *Radiology* 1977;122:47.

2. Gillebert C, Van Hoof R, Van de Werf F, Piessens J, De Geest H. Coronary artery fistulas in the adult population. *Eur Heart J* 1986;7:437.
3. McNamara JJ, Gross RE. Congenital artery fistula. *Surgery* 1969;65:69.
4. Goebel N, Gander MP, Steinbrunn W: Small coronary artery fistulae. *Ann Radiol* 1979;22:277.
5. Meng RL, Harlan JL. Left anterior descending coronary artery-right ventricle fistula complicating percutaneous transluminal angioplasty. *J Thorac Cardiovasc Surg* 1985;90:387.
6. Wilde P, Watt I. Congenital coronary artery fistulae; six new cases with a collective review. *Clin Radiol* 1980;31:301.
7. St John Sutton MG, Miler GA, Kerr IH, Trail TA. Coronary steal via large coronary artery to bronchial artery anastomosis successfully treated by operation. *Br Heart J* 1980;44:460.
8. Mc Namara JJ, Gross RE. Congenital coronary artery fistula. *Surgery* 1969;65:59.
9. Haberman JH, Howard JL, Johnson ES. Rupture of the coronary sinus with hemopericardium. *Circulation* 1963;228:1143.
10. Dağalp Z, Erol Ç, Berkalp B, Telli H, Ömürlü K, Akgün G, Oral D. Noninvasive diagnosis of coronary artery fistulas; color-flow echocardiographic study. *T Clinical Cardiol* 1993;3:34.
11. Reidy JF, Jones ODH, Tynan MJ, Baker EJ, Joseph MC. Embolism procedures in congenital heart disease. *Br Heart J* 1985;54:184.
12. Reidy JF, Anjos RT, Qureshi SA, Baker EJ, Tynan MJ. Transcatheter embolization in the treatment of coronary artery fistulas. *J Am Coll Cardiol* 1991;18:187.
13. Karagöz HY, Zorlutuna YI, Babacan KM, Taşdemir O, Yakut C, Kütük E, Bayazıt K. Congenital coronary artery fistulas. Diagnostic and surgical considerations. *J Heart* 1989;30:685.
14. Hiraishi S, Misawa H, Horiguchi Y, Fujino N, Takeda N, Nakea S, Kasahara S. Effect of suture closure of coronary artery fistula on aneurysmal coronary artery and myocardial ischemia. *Am J Cardiol* 1998;81:1263-7.
15. Lowe JE, Oldham HN, Sabiston DC Jr. Surgical management of congenital coronary artery fistulas. *Ann Surg* 1981;194:373.
16. Housman LB, Morse J, Litchford B, Mazur J, Starr A. Left ventricular fistula as a cause of intractable angina. Successful surgical repair. *JAMA* 1978;240:372.
17. Horiuchi T, Abe T, Tanaka S, Koyamada K. Congenital coronary arteriovenous fistulas. *Ann Thorac Surg* 1971;11:102.
18. Rittenhouse ED, Dotty DB, Ehrenhaft JL. Congenital coronary artery-cardiac chamber fistula. Review of operative management. *Ann Thorac Surg* 1975;20:468.
19. Hillis DL, Cohn PF. Non-atherosclerotic coronary artery disease. Second edition. Edited by Cohn PF, Martinus Nijhoff. Boston, 1985, pp 497.
20. Jaffe RB, Glancy L, Epstein SE, Brown GB, Morrow AG. Coronary-arterial-right heart fistula. Long term observation in seven patients. *Circulation* 1973;47:133.
21. Roughneen PT, Bhattachajee M, Morris PT, Nasse M, Reul GJ. Spontaneous thrombosis in a coronary artery fistula with aneurysmal dilatation of the sinus of Valsalva. *Ann Thorac Surg* 1994;57:232-4.
22. Rose AG. Multiple coronary artery fistulae. *Circulation* 1978;58:178-82.
23. Hirose H, Amano A, Yoshida S, Nagao T, Sunami H, Takahashi A, Hagano N. Coronary artery aneurysm associated with fistula in adults: collective review and a case report. *Ann Thorac Cardiovasc Surg* 1999;5:258-64.
24. Cronpvent JL, Lindenaver SM. Hemodynamic effects of sympathectomy in ischemic canine limbs. *Surgery* 1980;87:417-21.