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Surgical Treatment of Congenital Coronary Arterial Fistula

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Introduction

Congenital coronary arterial fistula, an abnormal communication between a coronary artery and a cardiac chamber or the pulmonary artery[40], is no longer a clinically rare lesion. With the improvement in facilities for diagnosis and correction of such an anomaly, there has been an increased interest in this lesion and an increase in the number of case reports. This anomaly can now be easily and accurately diagnosed by selective coronary angiography preoperatively, and also can be operated upon successfully by various surgical procedures such as ligation, division, arteriorrhaphy and aneurysmorrhaphy. But the general lack of sufficient knowledge about the natural history and surgical prognosis can be seen in the absence of uniformity in the operative indication and procedure. Surgical treatment is definitive in most patients and carries little risk. This report reviews our experience with the surgical treatment of this anomaly.

Patients

The six patients (2 males and 4 females) reported herein were all hospitalized and operated upon in the 2nd Department of Surgery, Kyoto University Hospital, between April, 1968 and December, 1981 (Table I). The age at the operation varied between 2 and 7 years. Detailed physical examination, phonocardiograms, roentgenographic examination of the chest and electrocardiograms were all done routinely. Each patient underwent bilateral heart catheterization; two patients underwent aortography, one patient left ventriculography and three patients

Key words: Congenital coronary arterial fistula, Surgical indications, Surgical procedures, Natural history and Surgical results.
索引語：先天性冠動脈瘻，手術適応，手術術式，自然歴，手術結果.
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Table I. Presentation, Clinical Findings, Management, Complication and Follow-up in 6 Patients with Congenital Coronary Arterial Fistula

Case	1	2	3	4	5	6
Age-Sex	6 y.o. F	5 y.o. F	6 y.o. F	2.8 yo. M	7 y.o M	6 y.o. F
Preoperative symptom	frequent respiratory infection	—	—	Fatiguability	—	Fatiguability
E C G	L V H	normal	supravent. extrasystole	normal	wandering pace maker	incomplete R-BBB
Cardiac murmur	continuous 3/6 3/6	continuous 4/6 3/6	continuous 4/6 3/6	continuous 4/6 4/6	continuous 3/6 2/6	mid.Sys. Dias. 2-3/6 2/6
Thrill	palpable	palpable	palpable	palpable	palpable	none
Fistula course	LCA→RA Sinus node artery	LCA→LAD→RV (Apex)	LCA→RA Sinus node artery	LCA→Cx→RA	LCA→RA Sinus node artery	RCA→m-PA Conus artery
Cardiac catheterization	RA2,RV23/2,10 BA 120/62 Shunt L-R 10%	RA0,RV20/-1,2 FA 116/62 Shunt L-R 16%	RA10,RV38/0,13 FA 120/80 Shunt L-R 58%	RA5,RV30/-2,7 FA 100/60 Shunt L-R 18%	RA3,RV28/0,4 Ao 115/83 Shunt L-R 24.5%	RA10,RV35/0,9 Ao 90/55 Shunt L-R 64.6%
Other cardiac lesions	—	—	two ASDs(II) $7x7_{mm}$ $30x10_{mm}$	—	—	ASD(II) $28x11_{mm}$
Surgical procedures	prox. intacted dist. sutured from within the RA	prox. ligated at a point past the bifurcation of the 1st Diagonal Artery dist. sutured from within the RV	prox. ligated dist. sutured from within the RA	prox. ligated externally near the coronary sinus dist. sutured from within the RA	prox. sutured from within the left main trunk dist. divided near the RA	prox. ligated dist. sutured from within the m-PA
Surgical complication	none	none	none	none	none	none
Postop. change in ECG	T-wave: negative (V_{2-4})	no change	no change	transient elevation of ST (I, II, $_aV_F$)	flat P-wave	no change
Follow-up	13 yrs asymptomatic	10 yrs asymptomatic	9 yrs asymptomatic	6 yrs asymptomatic	1.5 yrs asymptomatic	1 yr asymptomatic

LVH left ventricular hypertrophy, R-BBB right bundle branch block, LCA left coronary artery, RCA right coronary artery, LCA left coronary artery, RA right atrium, LAD left anterior descending artery, RV right ventricle, Cx left circumflex artery, m-PA main pulmonary artery, BA brachial artery, FA femoral artery, Ao aorta, prox. proximal, dis. distal

selective coronary artery angiography. The congenital coronary arterial fistulae were all diagnosed prior to operation.

Figure 1 depicts the arteries involved and the sites of termination in these six patients.

Symptoms: One patient (Case 1) had a history of frequent upper respiratory infection, and two others (Cases 4 and 6) fatigued easily. The remaining three patients had no symptoms.

Physical examination: In five patients, a typical loud continuous murmur, located in an area atypical for a patent ductus arteriosus (PDA) was present. In Case 6, which was associated with atrial septal defect (ASD) secundum type, a midsystolic and middiastolic murmur characteristic of ASD, was heard. A thrill was present in 5 patients. The maximum palpable areas were: the 2nd right intercostal space (2 RIS), (Cases 1 and 5); apex, (Case 2); 4 RIS, (Case 3) and 5 RIS, (Case 4). These areas were in accordance with the terminal point of the fistula. The grades of murmur were between 3/6 and 4/6.

Laboratory data: Electrocardiographic findings were non-specific. In the electrocardiograms, left ventricular hypertrophy, supraventricular extrasystole and wandering pace maker were seen. The roentgenography of the chest in Case 3 showed increased pulmonary vasculatures, prominent pulmonary arteries and cardiac enlargement, resulting in a large left to right (L-R)

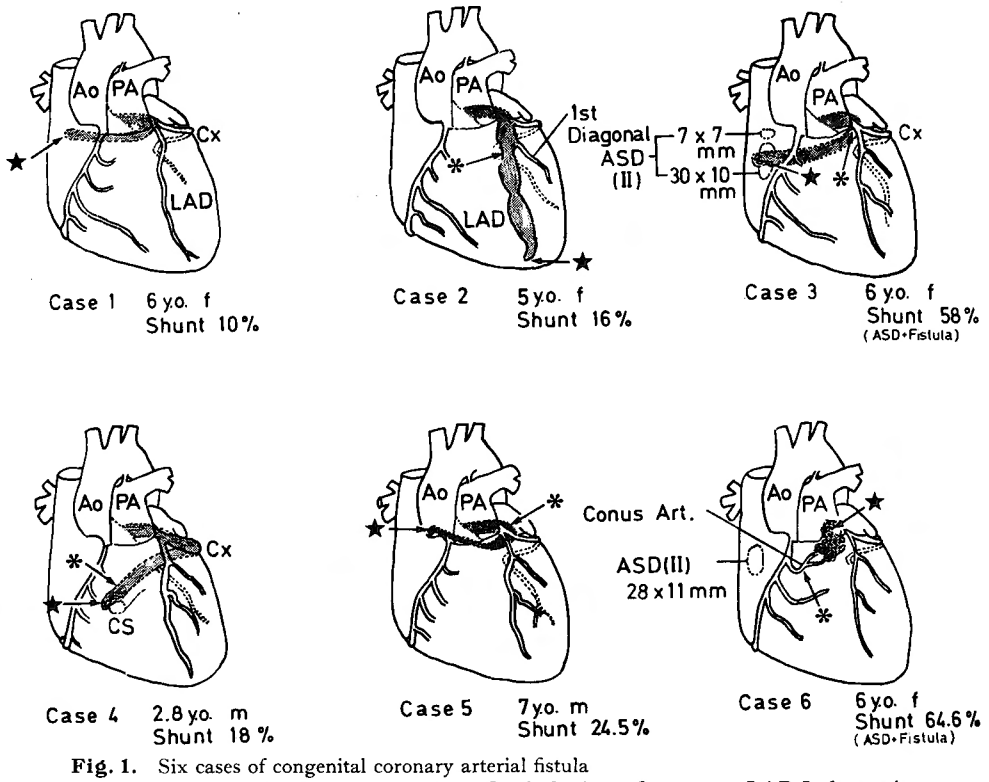

Fig. 1. Six cases of congenital coronary arterial fistula
Ao Aorta, PA Pulmonary artery, Cx Left circumflex artery, LAD Left anterior descending artery, CS Coronary sinus
★→ Distal portion closed surgically
＊→ Proximal portion closed surgically

shunt. In Case 5, ultrasonic cardiotomography revealed aneurysmatic dilatation of the main trunk of the left coronary artery, and this finding was comparable to that of selective coronary angiography of the left coronary artery.

Each patient underwent cardiac catheterization of both the right and left side of the heart; pressures in the right ventricle and pulmonary artery were not altered or only mildly elevated. In all patients, cardiac catheterization demonstrated an increase in blood oxygen saturation in the right atrium or right ventricle depending on the degree of the L-R shunt. In all cases except for Case 6, injection of contrast media into the aortic root or selectively into the coronary arteries outlined the anatomy of the fistulae.

Anatomy and surgical procedures

Case 1: The fistula branched from the main left coronary artery and drained into the right atrium close to the foramen ovale. This fistula had two small openings through which blood flowed into the right atrium; jet lesion was seen around the opening, causing stenosis. The L-R shunt through the fistula was calculated to be 10% of the pulmonary blood flow. A conti-

nuous strong thrill was palpable at the upper area of the right atrium.

Using a cardio-pulmonary bypass, the fistula was closed by suturing the opening from within the atrium. Closure of the proximal portion of the fistula was not performed because the bifurcation from the main left coronary arterial trunk could not be found.

Case 2: A tortuous and dilated left anterior descending artery drained into the apex of the right ventricle where a strong thrill was palpable.

Upon making a longitudinal incision over the apex, it was found that the opening of the abnormal left anterior descending artery into the right ventricle was between the trabeculae. The opening was closed by suture from within the right ventricle. In addition, the proximal portion of the left anterior descending artery was ligated following the test occlusion which revealed no change in electrocardiogram. The patient tolerated the procedure well and had an uneventful postoperative course. The left anterior descending artery, ligated at points proximal and distal, was not opacified in the selective coronary angiography one year postoperatively.

Case 3: The fistula branched from the main trunk of the left coronary artery and drained into the right atrium close to the two secundum ASDs. Left to right shunt of the fistula and the ASDs comprised 57.8% of the pulmonary blood flow. The sizes of the ASDs were 30×10 mm and 7×7 mm, respectively. The opening of the fistula and the ASDs were simultaneously closed by direct suture.

Case 4: The fistula branched from the circumflex artery, ran downwards, and drained into the right atrium close to the coronary sinus. A continuous thrill was palpable over the lower right atrium. The fistula had two openings in the right atrium, and was stenotic at the portion of the openings; the L-R shunt was 18%.

The fistula was closed by suture from within the right atrium and, in addition, by ligation from the outside at the distal portion close to the coronary sinus.

Case 5: This case was reported in detail in a previous paper[23]. The fistula branched from the circumflex artery, ran horizontally, and then curving sharply draining into the sinus node area of the right atrium. The fistula had a L-R shunt of 24.5%; a strong continuous thrill was palpable over the opening of the fistula.

In this case, right atriotomy was not performed, for fear of postoperative arrhythmia resulting from possible damage of the sinus node following direct closure from within the right atrium. Therefore, the closure of the fistula was performed by division in the distal portion and direct closure from within the dilated circumflex artery in the proximal portion. After the above procedures, there was no abnormal change upon electrocardiography.

A small specimen of the wall of the aneurysmatic coronary artery was removed, and upon histological study, the wall of the aneurysmatic coronary arterial fistula was found to have three layers without any inflammatory findings, but the elastic fibers were heavily ruptured.

Case 6: The fistula branched from the conus branch of the right coronary artery, and drained into the main pulmonary artery. Arterial plexus formation was seen on the main pulmonary artery and right ventricular outflow tract. The fistula had one small opening which was on a small hypertrophied pulmonary arterial wall. The ASD (secundum type), 28×11 mm, was

closed by direct suture. The conus branch of the right coronary artery was ligated and the small opening was closed from inside the main pulmonary artery by direct suture. No change was seen upon electrocardiography.

Results

All patients survived the operation and were discharged in good condition. A recent follow-up study showed that all six patients are fully active and well. Postoperatively, no cardiac murmur was heard in any of the cases.

Discussion

Though the clinical features, physiology and anatomy of coronary arterial fistula have been extensively studied, there is no agreement as to the management of patients with this anomaly [7,10,12,18,28,30,31,33,34]. Young patients with this lesion are generally asymptomatic and they rarely have related complications. For this reason, and because of the potential for surgical complications and the rare occurrence of spontaneous closure, the necessity for selective ligation of congenital coronary arterial fistula has been questioned.

The natural history of coronary arterial fistula depends on the volume of shunt and the age of the patient. In addition, complications such as congestive heart failure[6,33], coronary ischemia [19], subacute bacterial endocarditis (SBE)[6,42] and rupture of the aneurysmatic coronary arterial fistula[3,14] may occur. LIBERTHSON et al.[19] reported that the incidences of preoperative fistula-related complications were: congestive heart failure (12%), coronary ischemia (4%), SBE (3%) and rupture (1%). A large shunt may cause congestive heart failure and pulmonary hypertension [6,21,33]. Congestive heart failure is a common cause of death in untreated cases, and it is seen commonly in early infancy and also the case of after 40 years[6,21,25]. On the other hand, cases in which the small fistula closed naturally have been reported[19,24,25,35]. As seen in Cases 1, 2 and 3, the fistula draining into a cardiac chamber had multiple openings at which stenosis and subsequent jet lesion were seen. These findings may suggest natural closing. Because the cardiac muscle receives blood during diastole, a decrease in diastolic coronary arterial pressure from the coronary arterial fistula may lead to the "coronary steal phenomenon"[32], causing coronary ischemia. The incidence of SBE was 8% in one reported series of 259 cases[27]. Rupture of an aneurysm of the fistulous tract can occur[3,14], but it is rare.

The natural history of most coronary arterial fistulae involves the development of a progressive dilatation and elongation of the affected coronary artery and the vessel or chambers receiving the shunted blood, resulting from a progressive increase in blood flow[8]. However, JAFFE[16] reported that progressive dilatation and tortuousness of the affected coronary artery was not found in untreated cases. In FURUSHIMA's review[11], after surgical closure of the aneurysmatic coronary arterial fistula, there was no change in fistula diameter in 6 out of 14 cases but 5 out of 14 showed a decrease to within normal limits upon postoperative angiography[29]. As in our Case 5, when histological studies[9,25] reveal a remarkable interruptions in the elastic layers of the dilated

segment of artery, it may be unlikely that a postoperative decrease in diameter of the affected aneurysmatic coronary artery will occur. ARAYA et al.[3] reported a case in which rupture of an aneurysmatic fistula occurred 6 months after operation. In this case the aneurysmatic fistula was closed only in the distal portion.

According to KONNO et al.[17], surgery for congenital coronry arterial fistula is indicated when there is:

1) A shunt of more than 30% even though no symptoms are present.
2) Ischemic change or strain patterns in electrocardiogram
3) Progressive congestive heart failure
4) Progressive pulmonary hypertension
5) Past history of bacterial endocarditis
6) Great possibility of rupture of aneurysmatic coronary artery
7) A loud murmur which is a disadvantage in getting a job.
8) Associated cardiac lesions such as aortic valvular atresia[33], pulmonary valvular atresia [33], ASD[2,37], PDA[4,36], tetralogy of Fallot[11] and acquired heart disease[11].

Among our six cases, Cases 3 and 6 fell under one of the above conditions, but the other cases did not. However we decided to operate in order to prevent the possibility of subacute bacterial endocarditis and myocardial ischemia. All of our patients survived operation, and follow-up ranging from 1 to 13 years revealed that all were asymptomatic and clinically free of heart disease. These results, compared to the risk of potential complications of coronary arterial fistula, strongly support the advisability of surgical repair when the lesion is diagnosed.

LIOTTA[20] classified coronary arterial fistula into two types: 1) primary or associated with anomalies such as PDA or atrial/ventricular septal defects and 2) secondary or complicated type in which important congenital cardiac anomalies appear to be the main hemodynamic impediment, and the fistula does not play a major role. Cases 1, 3, 4 and 5 belong to the primary type; Cases 2 and 6 belong to the secondary type. Surgical procedure should be chosen according to the type of coronary arterial fistula. Some of the various surgical procedures for the lesion are:

1) Ligation and/or division[8,20]

The fistula of the primary type is ligated and/or divided, requiring no extracorporeal circulation. The most commonly employed technique for closure has been ligation of the involved coronary artery proximally and distally to the fistulous opening. Closure distal to the fistulous opening only may result in progressive retrograde development of thrombosis[1] and rupture of the aneurysmatic coronary artery[13]. On the other hand, proximal ligation of the contributing coronary artery is of no value in the treatment of coronary arterial fistula, as the fistula can still receive blood via collateral branches from the distal portion of the involved artery, and thus there is a real danger of producing myocardial ischemia or even infarction.

SAKAKIBARA[33] speculated that, in most cases, ligation proximally and distally to the fistulous opening does not cause myocardial ischemia, because the other coronary artery may supply blood via collateral branches. Usually, ligation of the coronary artery at points proximal and distal to the fistula may be more safely performed in young patients than in adults. Many

authors[5,31,38] have recommended that prior to ligation and/or division it is necessary to perform a temporary test occlusion of the coronary artery for 15 to 20 minutes in order to confirm by electrocardiogram that no ischemic pattern or arrhythmia will develop. But there have been cases in which myocardial damage occurred even though there were no changes in the electrocardiogram during the test period[5,31,38].

Actually, there are so many different types of coronary arterial fistulae, that a uniform classification of this lesion can not be made. Accordingly, the safest technique for the correction of these defects varies from patient to patient. LIBERTHSON et al.[19] emphasized that early elective ligation of congenital coronary arterial fistula is indicated in all patients because of the high incidence of late symptoms and complications, and the increased morbidity and mortality associated with ligation in older patients.

2) Selective ligation of the fistula[33]

This procedure may be done safely, without risking myocardial ischemia when the associated coronary artery has an orifice in the side wall; but not when multiple orifices are scattered over a wide area.

3) Tangential arteriorrhaphy[5,20,26]

COOLEY reported that, in the cases with multiple orifices scattered over a wide area, many overlapping U-mattress sutures were made on the longitudinal axis along the border of the associated coronary artery and this artery, in the functional stage, was separated from the cardiac chamber. However cases of regression following this procedure have been reported[15,26].

4) Closure from the inside of the cardiac chamber[39,41]

This procedure may be suitable for fistulae terminating in the right atrium when there is an atrial septal defect. It is difficult to close the orifices which are between the trabeculae. Also the possibility of postoperative hypofunction of the cardiac chamber, right and left, may be increased by the incision. This procedure is not recommended when the orifice of the fistula is located in the left ventricle.

5) SYMBAS' internal arteriorrhaphy[38]

In this method the associated coronary artery is incised longitudinally and the orifice is closed with sutures from the inside; the diameter of the aneurysmatic coronary artery can be decreased by aneurysmorrhaphy. This procedure not only is applicable when the orifice is located at the end or on the side of the fistula, but it also permits the blood flow to remain normal. However, LIOTTA[20] and SYMBAS[38] have pointed out that with this method a thrombus may develop at any time from an early stage to a long postoperative period. They recommended the administration of anti-coagulant drugs. The development of a thrombus in the aneurysmatic coronary artery does not depend on the existence of the fistula. The main factor in the development of a thrombus is thought to be congestion following the closure of the fistula, therefore, it is reasonable to perform aneurysmorrhaphy in the aneurysmatic fistula.

6) Ligation of the associated coronary artery in conjunction with aorto-coronary bypass[20]

This method is restricted to adult cases. It is impossible to perform on children because of patency of graft, technical problems and growth.

Causes of postoperative death reported in the literature are:[4,11,13,22,27] massive bleeding, low cardiac output syndrome in a case associated with tetralogy of Fallot, arrhythmia in a case associated with large patent ductus arteriosus and severe pulmonary hypertension, rupture of aneurysmatic coronary artery, substernal abcess and heart failure in a case associated with patent ductus arteriosus.

There have been many reports of residual shunts occurring after ligation and tangential arteriorrhaphy[11,30], but degree of residual shunt is unknown. NOGUCHI et al.[26] reported that the main causes of occurring residual shunts following operation are: remaining fistula, recanalization of closed fistula, and the characteristic structure of the coronary artery. Therefore, closures of more than triple ligation and division of the fistula are desirable together with suture-ligation from the inside of the fistula in order to prevent recurrence.

The low operative mortality and morbidity associated with closure of a coronary arterial fistula seems to justify operation, even in asymptomatic patients, to prevent the potential complications which have been already well described.

Summary

Six patients with congenital coronary arterial fistula have been operated upon during a 13 years period. All six cases are of the terminal fistula type: left coronary artery to right atrium fistula (4), left coronary artery to right ventricle fistula (1), and right coronary artery to main pulmonary artery fistula (1).

Operations in all cases were performed using cardio-pulmonary bypass. Terminal fistulae from a main coronary trunk were treated by direct external ligation or the suturing of the openings from within the chamber. The proximal portions of the fistulae were closed in 4 of 6 cases: 3 external ligation and 1 suturing of the opening from within the main left coronary artery combined with aneurysmorrhaphy. None of the cases had any residual shunt or recurrence. All patients survived the operation, and a recent follow-up survey indicated that they are doing well. Aneurysmatic coronary artery may be treated by aneurysmorrhaphy in order to prevent thrombosis, but there are many difficulties in performing this procedure. Terminal type fistula should be treated by closure at the visible proximal and distal portions using cardio-pulmonary bypass, to prevent residual shunt and recurrence.

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和文抄録

先天性冠動脈瘻の手術

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過去13年間に，先天性冠動脈瘻6例の手術が行なわれた．6例とも terminal fistula 型で，内訳は，左冠動脈一右房瘻（4例），左冠動脈一右室瘻（1例），右冠動脈一主肺動脈瘻（1例）であった．

手術は，冠動脈瘻の末梢側および中枢側の遮断を原則としている．

本症は，年長になれば症状および合併症の出現頻度が高くなることから，手術は，たとえ無症状でもより安全性の高い年少時に行なうのが有利である．