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Arterial Switch Operation With and Without Coronary Relocation for Intramural Coronary Arteries

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Background. The arterial switch operation (ASO) for the transposition of the great arteries (TGA) with intramural coronary arteries has been performed using several techniques to avoid coronary events. We mainly performed ASO without coronary relocation by creating an aortopulmonary fenestration (Imai technique). Coronary circulation was rerouted by covering the aortopulmonary window and coronary orifices with a nonfacing sinus flap. Long-term results have not been reported. We describe our early and late results.

Methods. Among 551 patients who underwent an ASO between 1985 and 2014, intramural coronary arteries were detected in 15 of them. Coronary arteries were managed using 2 techniques: the double-button method in 5 patients (with unroofing and trapdoor incision in 1 patient) and the Imai technique in 10 patients.

Results. There were 3 hospital deaths and 3 deaths after discharge, 5 of which showed coronary complications. Actual survival and freedom from coronary

complications at 15 years were 70% and 67%, respectively, with the Imai technique and 40% and 20%, respectively, with the double-button method. Late coronary intervention was performed for a long intramural coronary artery stenosis in 1 patient who underwent the Imai technique. In the others, late aortography showed good patency of the aortopulmonary window and growth of the coronary pouch after the Imai technique.

Conclusions. The Imai technique can be an option for coronary management in the presence of high-risk coronary anatomy, particularly distal intramural coronary artery stenosis and inseparable coronary arteries with an almost single orifice. Adequate neopulmonary artery augmentation must be performed to prevent right ventricular outflow stenosis.

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The arterial switch operation (ASO) is the standard technique for the transposition of the great arteries (TGA). The overall mortality rate after operation has been reported to be between 1.1% and 6.0%. However, the presence of an intramural coronary artery (IMCA) was considered to increase mortality and coronary events [1].

Several techniques have been used for avoiding coronary complications when performing ASO for TGA with an IMCA [2]. In 1978, Aubert and colleagues [3] reported ASO without coronary relocation. In this procedure, coronary arteries were left in situ and covered with bovine pericardium; perfusion was created using an aortopulmonary window. Various modifications have been described [4–7]; however, late results have not been reported.

At our institution, we mainly performed the modified Aubert procedure (Imai technique) [2, 6, 7] using a nonfacing sinus flap for covering the aortopulmonary window and coronary orifices to prevent late tunnel

obstruction and thrombus. We evaluated our early and late results.

Patients and Methods

The Tokyo Women's Medical University Institutional Review Board approved this retrospective study; the need for informed consent was waived. We retrospectively reviewed the medical records, operative notes, echocardiographic findings, cardiac catheterization reports, and multislice computed tomographic angiography findings of 551 patients who underwent ASO for TGA or double-outlet right ventricle at our institution between November 1985 and December 2014.

Fifteen patients (2.8%) with an IMCA were identified. There were 12 boys and 3 girls, including 7 neonates. At the time of operation, their median age was 81 days (11–804 days) and their weight was 3.8 kg (2.5–10.5 kg). Ten patients had TGA with an intact ventricular septum, 3 patients had TGA with a ventricular septal defect (VSD), and 2 patients had a Taussig-Bing anomaly. Associated anomalies included aortic coarctation in 2 patients, muscular VSD in 1 patient, straddling mitral valve in 1 patient, bicuspid pulmonary valve in 1 patient, and

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Abbreviations and Acronyms

- ASO = arterial switch operation
- CABG = coronary artery bypass grafting
- IMCA = intramural coronary artery
- LAD = left anterior descending
- LCA = left coronary artery
- RCA = right coronary artery
- TGA = transposition of the great arteries
- VSD = ventricular septal defect

bicuspid aortic valve in 1 patient. Five patients had undergone previous palliative procedures, 4 patients had undergone pulmonary artery banding with a Blalock-Taussig shunt for left ventricle training before ASO, 1 patient had received a subclavian flap using pulmonary artery banding for coarctation repair, and no patients received antiplatelet treatment after operative treatment.

Coronary Anatomy

The coronary anatomy was determined based on the description in the operative records. IMCA stenosis was found in 5 patients. The following 3 anatomic types were identified (Fig 1):

1. Type A (n = 6): In 4 patients, the left anterior descending (LAD) artery arose from the right-facing sinus and passed intramurally behind the facing commissure. In the other 2 patients, the LAD artery

originated above the facing commissure. The right coronary artery (RCA) and the left circumflex artery originated centrally in the right-facing sinus. One patient had a single orifice (type A1) and another patient had 3 orifices (type A3). The other 4 patients had 2 orifices (type A2).

2. Type B (n = 8): The left coronary artery (LCA) originated from the right-facing sinus and coursed intramurally behind the facing commissure. The RCA originated centrally from the right-facing sinus. Six patients had 2 orifices (type B2). The other 2 patients had a single orifice (type B1).
3. Type C (n = 1): The right major ventricular branch was intramural and coursed anteriorly between the aorta and the pulmonary artery. The right major ventricular artery, RCA, and LCA originated separately from the right-facing sinus.

Surgical Technique

Through a median sternotomy, cardiopulmonary bypass was established by aortic and bicaval cannulation. Cardioplegia was induced. The ascending aorta was divided above the sinotubular junction. The coronary arteries were carefully inspected before deciding on coronary management. The pulmonary artery was divided just before the bifurcation. Coronary arteries were managed using 2 techniques (Table 1).

1. Double-button method: The facing commissure of the aorta was detached. All coronary orifices were excised

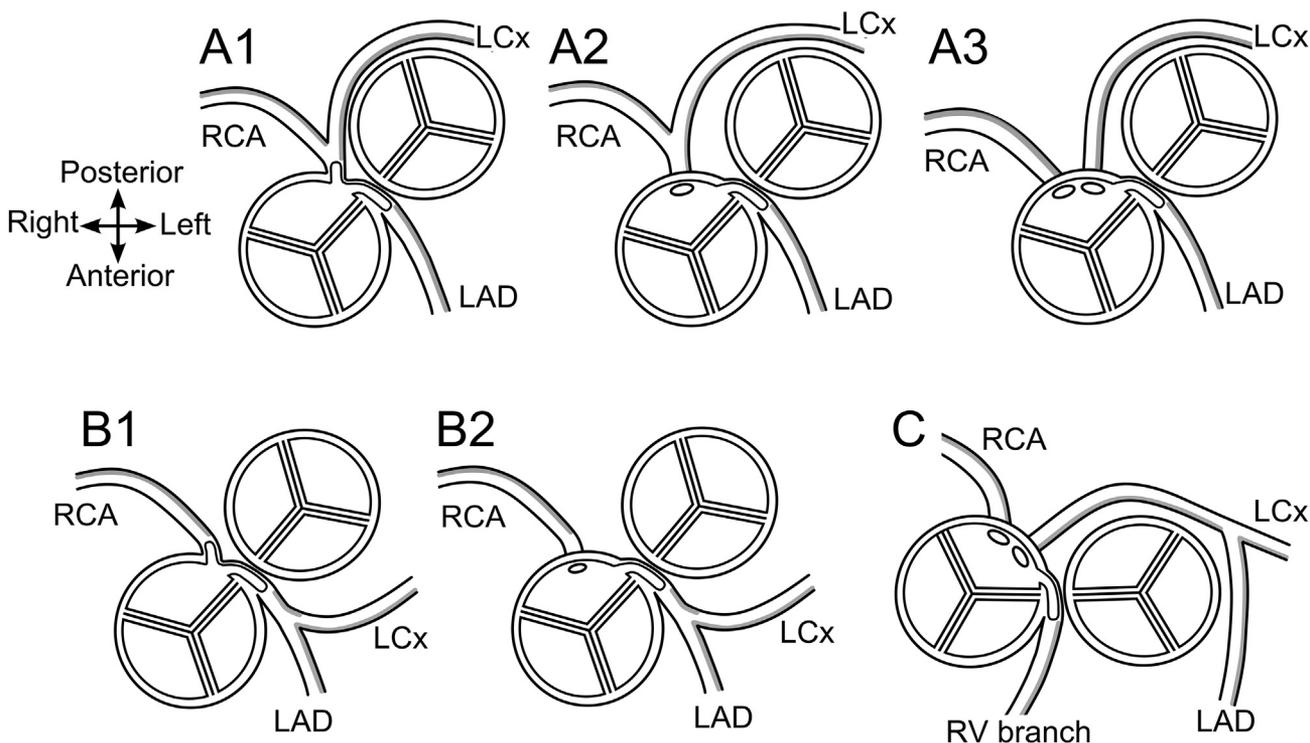


Fig 1. Coronary artery anatomy types. (LAD = left anterior descending; LCA = left coronary artery; LCx = left circumflex; RCA = right coronary artery.)

Table 1. Patient Profiles and Outcomes

Patient No.	Diagnosis	Coronary Pattern	Surgical Technique	Outcome (Time to Death)	Cause of Death	Reoperation/Remarks
1	TGA IVS	A2	Double button	Alive	...	RVOTR LAD artery occlusion
2	TGA VSD COA	B2	Double button	Hospital death (213 d)	Myocardial infarction	...
3	TGA IVS	B2 (intramural stenosis)	Double button	Hospital death (1 day)	Myocardial infarction	...
4	TGA IVS	A3 (intramural stenosis)	Double button (Asou)	Early death (63 d)	Myocardial infarction	...
5	Taussig-Bing anomaly	A2	Double button	Alive
6	TGA IVS	B2	Imai	Alive
7	TGA IVS	A1	Imai	Alive
8	TGA IVS	B1	Imai	Alive	...	RVOTR
9	TGA IVS	B1	Imai	Alive	...	RVOTR
10	TGA VSD	A2 (intramural stenosis)	Imai (Cutback)	Early death (48 d)	Myocardial infarction	...
11	TGA IVS	A3	Imai	Alive
12	TGA VSD COA	B2 (long intramural stenosis)	Imai	Alive	...	CABG, RVOTR, MVR
13	TGA IVS	B2	Imai	Alive
14	TGA IVS	C	Imai	Hospital death (16 d)	Mediastinitis	...
15	Taussig-Bing anomaly	B2 (intramural stenosis)	Imai	Late death (7 y)	Heart failure	CABG, MVR

CABG = coronary artery bypass grafting; COA = coarctation of the aorta; IVS = intact ventricular septum; MVR = mitral valve replacement; RVOTR = right ventricular outflow reconstruction; TGA = transposition of the great arteries; VSD = ventricular septal defect.

as a large piece of a button before separating into 2 coronary buttons. In 4 patients, each coronary artery was translocated by vertical incisions or punch holes in front of the proximal pulmonary artery. One patient with orifice stenosis of the IMCA underwent the Asou technique (medially hinged trapdoor incisions and unroofing the intramural segment) [8].

2. Modified Aubert procedure (Imai technique): The right-facing sinuses of the great arteries were excised in a semicircular shape, 4 to 5 mm in width, leaving a margin of a few millimeters above the coronary orifices. Both defects were sewn to make an aortopulmonary window. In the first patient, a 2.5-mm-wide aortopulmonary window was made by punching and sewing holes as described by Aubert and colleagues [3]. However, this technique was difficult in neonates; in addition, the provision of enough coronary flow and long-term patency was uncertain. Therefore we modified the technique as described earlier. The nonfacing sinus was excised 1 mm above the aortic valve ring with the aortic wall just above the facing commissure as a hinged pedicled flap. This flap was flipped on the bottom of the right-facing sinus and was sutured along the base of the right-facing sinus to cover coronary orifices, creating a coronary pouch. The suture line over the IMCA was skipped or was sewn extremely thinly to avoid injury. In some instances, a probe was inserted into the IMCA to facilitate sewing. The aorta was brought down posteriorly to the pulmonary artery and was anastomosed in an end-to-end manner with the neo-aorta and cranial margin of the coronary pouch. The coronary orifices opened into the neo-aorta through the aortopulmonary window; the coronary arteries were left in situ to maintain the exact coronary geometry. The distal pulmonary artery was sewn on the coronary pouch, a little under the suture line between the pedicled flap and distal aorta, to avoid double sewing that may make the sutures fragile. The defect of the nonfacing sinus was covered and enlarged with a distal pulmonary artery, an autopericardial patch, or a bovine pericardial patch. The right proximal pulmonary artery, which is occupied by the coronary pouch, was also enlarged in most cases to provide enough space for the supraventricular neopulmonary artery. In the latter part of the series (n = 3), the procedure was modified. If the IMCA orifice was open within 2 mm of the facing commissure, the facing commissure was partially detached and the pedicled flap was sewn 3 mm away from the coronary orifice to avoid injury to the IMCA. Next, the commissure was reattached to the pedicled flap (Fig 2).

Statistical Analysis

Data were expressed as mean ± standard deviation and range. Time-related survival date and freedom from coronary events were analyzed using the Kaplan-Meier method. All data analyses were performed using JMP 11 software (SAS Institute, Cary, NC).

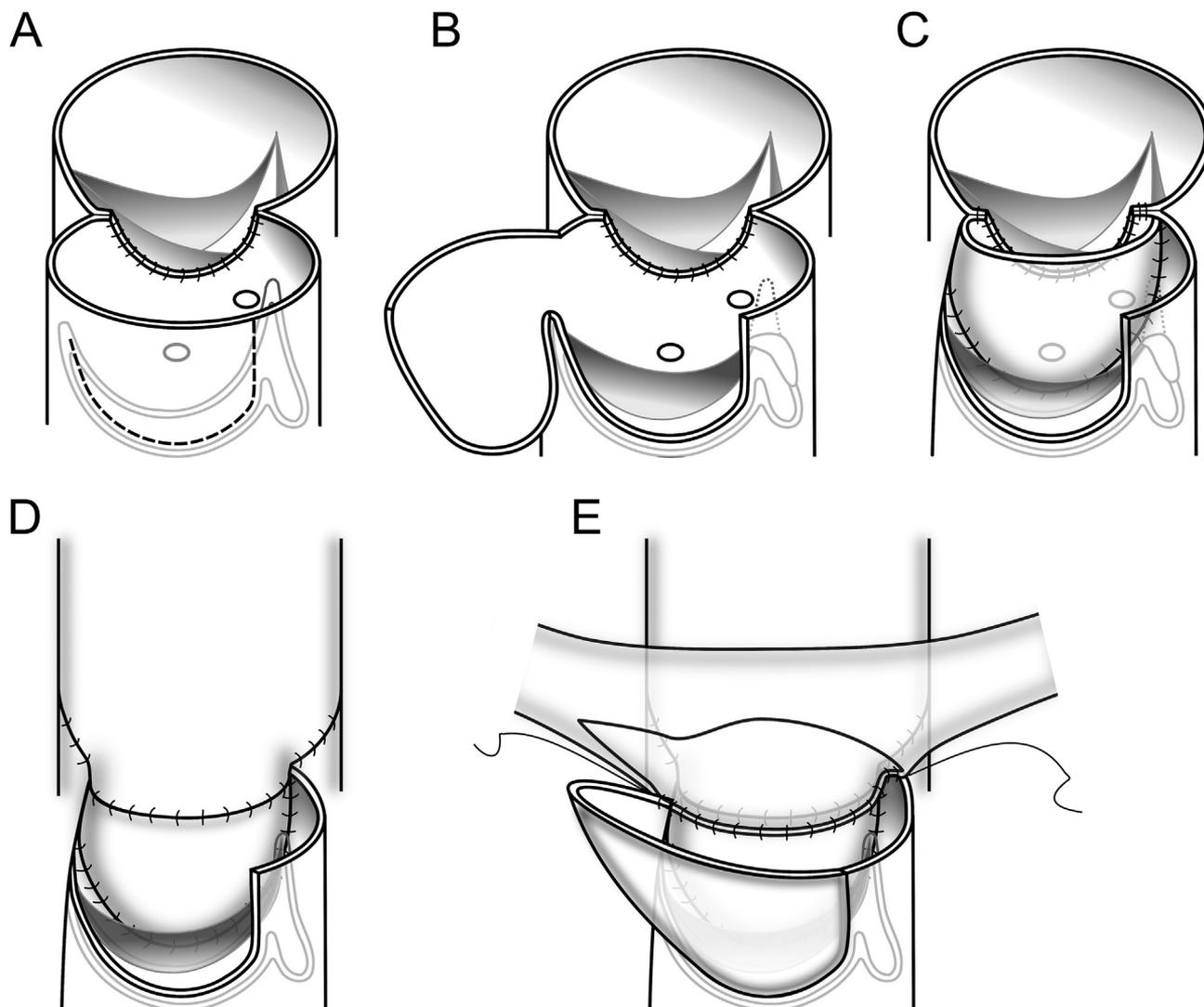


Fig 2. Imai technique. (A) Right-facing sinuses are excised and sewn. (B) Nonfacing sinus is excised as hinged pedicled flap. Partial detachment of facing commissure is made as necessary. (C) Pedicled flap is sutured along base of right-facing sinus. (D) Aorta is anastomosed with coronary pouch. (E) Right pulmonary artery and defect are covered and enlarged with patch.

Results

Intraoperative Results

An unstable hemodynamic status was observed after aortic declamping in 3 of the 15 patients. In 3 other patients (patients 4, 8, and 12), coronary perfusion was insufficient and the procedure was immediately revised. The first of these patients underwent the Asou technique. Direct pulmonary reconstruction compressed the LCA; therefore it was removed and the coronary button defect was covered with a large pericardial patch. The second patient underwent the Imai technique; however, coronary pouch compression resulting from supra-avalvular pulmonary stenosis resulted in severe pulmonary hypertension and coronary ischemia. Thereafter the neopulmonary artery was removed, and a large rhomboid patch was inserted on the left side of the anastomosis. The third patient with bicuspid pulmonary valve underwent

the Imai technique. Gradual deterioration of left ventricular function was noted before neopulmonary artery reconstruction. Each coronary orifice was pericommissural and contralateral because of the bicuspid pulmonary valve. The coronary pouch size was inadequate to cover the 2 contralateral coronary orifices; therefore it was enlarged using an autopericardial patch. All 3 patients achieved stable hemodynamic status after the revised procedures. The sternum was left open in 5 patients. Patient profiles and outcomes are summarized in [Table 1](#).

Early Results

There were 3 hospital deaths. Two of the 3 patients who died underwent the double-button method and had coronary complications on day 1 (acute myocardial infarction) and day 213 (heart failure because of myocardial

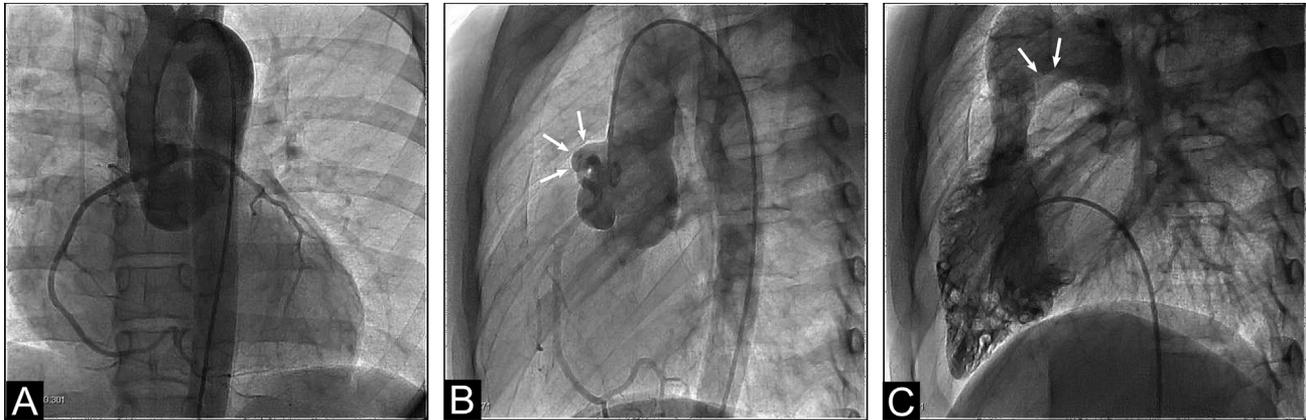


Fig 3. Postoperative angiography 13 years after Imai technique (patient 13). (A and B) Aortography shows good patency of aortopulmonary fenestration and growth of coronary pouch. (C) Right ventriculography shows adequate width of suprapulmonary portion. Coronary pouch with nonfacing sinus flap (white arrows) is shown in B and C.

infarction) after the procedure. Another patient died from a noncoronary cause (mediastinitis).

There were 2 early deaths after discharge resulting from myocardial ischemia on postoperative days 63 (patient 4) and 48 (patient 10). In both patients, cutback or wedge resection of the stenotic IMCA orifices was performed intraoperatively. In patient 10, pathologic findings showed an IMCA orifice that was narrowed by intimal hypertrophy on the cutback site. There was no thrombus formation.

In 1 patient who had Taussig-Bing anomaly with straddling and cleft mitral valve (patient 15), early reoperation was performed twice. The patient underwent coronary artery bypass grafting (CABG) for myocardial ischemia on day 1 after the procedure and mitral valve replacement for mitral valve regurgitation on day 36 after surgical intervention.

Echocardiography before discharge showed good ventricular function in all but 2 (patients 1 and 15). Only patient 15 had moderate neo-aortic regurgitation. Four patients who underwent the Imai technique had mild supralvalvular pulmonary stenosis.

Late Results

There was 1 late death because of chronic heart failure 7 years after the operation. The actual survival rate with the Imai technique 1, 5, and 15 years after surgical intervention was 80%, 80%, and 70%, respectively. The actual survival rate with the double-button method 1, 5, and 15 years after operation was 40% for all.

The 9 survivors were followed for a mean duration of 20.6 ± 3.6 years (14.9–24.8 years). One patient (patient 1) was lost to follow-up 19 years after the operation. All survivors were in New York Heart Association class I at the latest follow-up. There were 7 reoperations and 1 catheter intervention in 4 survivors. These procedures included pacemaker implantation for atrioventricular block because of catheter intervention in 1 patient; CABG for IMCA stenosis, mitral valve replacement for mitral regurgitation resulting from recoarctation, and stenting for recoarctation

in 1 patient each; and right ventricular outflow reconstruction for pulmonary stenosis in 4 patients.

All survivors underwent coronary evaluation by cardiac catheter angiography ($n = 7$) and multislice computed tomographic angiography ($n = 2$). Two patients had coronary lesions. In patient 1, cardiac catheterization before hospital discharge showed total occlusion of the LAD artery and hypokinesia of the anterior left ventricle caused by a previous myocardial infarction. In patient 12, coronary angiography showed 90% stenosis of the intramural LCA, and myocardial scintigraphy showed myocardial ischemia 4 years after the operation. Therefore the patient underwent CABG (free right internal thoracic artery to LCA grafting), which led to a good result during the follow-up period. Intraoperative findings showed that a 1-mm probe could not be passed through the intramural segment because the stenosis had progressed. The other patients had no coronary lesions after a mean follow-up duration of 12.3 ± 9.7 years (26 days–22.1 years). Late aortography showed good patency of the aortopulmonary fenestration and growth of the coronary pouch after the Imai technique (Figs 3A, 3B).

All survivors underwent postoperative cardiac echocardiography after a mean follow-up of 19.1 ± 3.3 years (14.7–24.8 years). Left ventricular function was normal in all but 1 survivor who had a previous anterior myocardial infarction. Three patients had mild aortic regurgitation, 1 patient had mild mitral regurgitation, 4 patients had mild pulmonary regurgitation, and 5 patients had mild pulmonary stenosis.

Coronary Events

Coronary events occurred in 7 patients, including coronary-related deaths ($n = 5$), nonfatal myocardial infarction ($n = 1$), and asymptomatic coronary stenosis requiring CABG ($n = 1$). The actual freedom from coronary events with the Imai technique 1, 5, and 15 years after operation was 78%, 67%, and 67%, respectively. The actual freedom from coronary events with the double-button method 1, 5, and 15 years after operation was 20% for all.

Supravalvular Pulmonary Stenosis After the Imai Technique

Echocardiography before discharge showed mild supravalvular pulmonary stenosis in 4 patients and no stenosis in 5 patients. Three patients underwent right ventricular outflow reconstruction for supravalvular pulmonary stenosis 4, 5, and 12 years after the operation, respectively. Mild supravalvular pulmonary stenosis was noticed in the other 4 patients; except for 2 late deaths, these patients remained stable and did not require repeated intervention. Late right ventriculography showed an adequate width of the supravalvular pulmonary portion (Fig 3C).

Comment

The presence of an IMCA during ASO for TGA has been reported to increase mortality and coronary events [1]. Several techniques have been reported to restore adequate coronary perfusion, and these were classified into 3 surgical techniques, including coronary relocation using double coronary buttons with unroofed intramural course [8], coronary relocation using a single coronary button [9, 10], and ASO without coronary relocation by creating an aortopulmonary fenestration [3-4, 7].

An anomalous aortic origin of a coronary artery from the opposite sinus with an intramural course has potential risks for myocardial ischemia after coronary transfer because of coronary artery kinking caused by an acute-angled takeoff, stenosis of the intramural course, and compression between the great arteries [1, 11]. In most studies, the Asou technique [8] results in good early and late results [1, 8, 12] using a medially hinged trapdoor technique to prevent coronary transfer from kinking and to release potential coronary compression between the great arteries; in addition, it may solve the late potential for IMCA stenosis by the resection of a proximal intramural segment. Therefore the technique was recently considered a standard for TGA with an IMCA. However, the technique could not be performed in patients with the most challenging coronary patterns, including inseparable coronary arteries because of a single orifice or if the distance of the coronary orifices was inadequate, despite unroofing of the intramural portion [13]. In this setting, coronary relocation using a single coronary button or ASO without coronary relocation was applied.

Coronary relocation using a single coronary button was first described by Yacoub and Radley-Smith [10]. Coronary arteries were excised as a single button. Next, the superior rim of the button was anastomosed to the proximal ascending aorta, and the other rim was sewn to the distal ascending aorta. The technique required turning the button upside down, which led to a risk for torsion of the coronary branches. In Yacoub type C arteries, the risk for coronary complications was considerably high because of coronary artery kinking [9]. Some authors have reported on the modification [1, 14]; with this technique, the rim, other than the superior rim of the button, was covered with pericardium to minimize rotation and torsion of the coronary arteries and maintain the in situ geometry of the coronary branches. However,

Metton and associates [1] reported that although the intramural portion was unroofed, this technique caused a high incidence of coronary complications. The reason may be associated with residual stenosis of the distal intramural part, in which kinking or twisting may occur even with slight rotation. An exact coronary geometry may not be maintained. Therefore, they did not recommend it as a routine technique. Furthermore, this technique also had a risk of coronary button compression by the pulmonary artery. The appropriate technique for these cases has been reported to be ASO without coronary relocation [13].

Aubert and associates [3] first described ASO without coronary relocation. The technique had no kinking or distortion because the coronary arteries were left exactly in situ. Compared with other techniques, there was no risk for injury because there was no need for coronary artery excision [14]. The aortopulmonary fenestration was created by suturing each hole of the ascending aorta and pulmonary trunk. The fenestration and coronary ostia were then covered with a polyester patch. The coronary circulation was rerouted from a neo-aorta through the fenestration to the coronary ostia. They preferred to use a polyester patch because a pericardial patch was prone to shrink. However, this technique has a potential risk for late tunnel obstruction by the nongrowing patch and thrombus formation. Moat and colleagues [5] described a modified technique using bovine pericardium and reported late sudden death from late tunnel obstruction secondary to thrombus formation in the aortopulmonary tunnel [15]. Takeuchi and Katogi [4] described another modified technique using the nonfacing sinus as a flap for rerouting coronary flow. The aortic flap had a growing material, and the intima was advantageous in preventing thrombus formation. With this modification, the surgeon must decide on the technique before transecting the aorta, although the recognition of an IMCA by external examination remains difficult [1].

Since 1989, we have mainly performed the Imai technique for TGA with an IMCA. We selected the nonfacing sinus as the hinged pedicled flap because it was useful for late tunnel patency and thrombus prevention. The technique enabled decision making after the usual aortic transection and careful internal examination of the coronary arteries. Therefore we believed that our technique was the most efficient and simplest method among the several modifications of ASO without coronary relocation.

Many studies have pointed out concerns of late tunnel obstruction, late ostial stenosis, myocardial ischemia by compression of the great arteries, and late right ventricular outflow stenosis because of an oversized coronary pouch after ASO without coronary relocation [2, 16]. In the present study, there was no late tunnel obstruction, thrombus formation, or myocardial ischemia in the 6 survivors. Late coronary evaluation showed good patency of the aortopulmonary fenestration and growth of the coronary pouch in these patients. Late coronary complications occurred in 2 patients with an IMCA stenosis after the Imai technique. In 1 patient, an intramural portion was cut back because it had an ostial stenosis. The initial

operative and postoperative courses were uneventful, but the patient died suddenly from myocardial infarction caused by orifice stenosis secondary to intimal hypertrophy of the cutback site. In the other patient, a long IMCA stenosis was detected using preoperative coronary angiography. The initial postoperative course was uneventful, and myocardial scintigraphy showed no myocardial ischemia until 4 years after the operation. However, the IMCA stenosis progressed and required CABG. Asou and coworkers [8] have reported that the distal site of a localized ostial stenosis is significantly larger than the orifice. Therefore an unroofed IMCA provides adequate coronary perfusion. In the first patient, coronary ischemia may have been avoided by additional unroofing of the IMCA and administration of antiplatelet treatment, including aspirin. However, the unroofed procedure could not always release the distal intramural segment stenosis where the coronary leaves the aorta [1], as demonstrated in 1 of our patients. Metton and colleagues [1] performed additional coronary patch ostioplasty intraoperatively after the Asou technique because of inadequate coronary perfusion from residual distal stenosis. However, coronary patch ostioplasty or CABG was technically challenging in neonates. In such cases, the Imai technique should be performed initially to maintain an exact coronary geometry, followed by the careful observation for the signs of myocardial ischemia.

A late echocardiogram showed stable and mild supra-valvular pulmonary stenosis in 4 survivors, except that 3 patients required reoperation with the Imai technique, implying that an appropriate size of the nonfacing sinus pedicled flap and sufficient augmentation of the supra-valvular pulmonary portion prevents serious right ventricular outflow stenosis. Although ASO with single coronary button relocation has compressive effects on the coronary pouch, only pulmonary artery pressure contributes to this effect. Therefore adequate pulmonary patch augmentation can lessen the risk of coronary artery pouch compression.

ASO for an IMCA was mostly accomplished using the Asou technique. However, IMCAs have variations in their intramural course and stenosis range. Most surgeons considered that it is difficult to accomplish the Asou technique in the following situations and that the Imai technique could be more appropriate.

1. The first was an IMCA passing in between the great arteries and with an almost single orifice, even if the intramural portion was resected. The coronary arteries were inseparable because there was inadequate tissue between the orifices on which buttons could be made to prevent the risk of torsion and kinking during coronary transfer. In such situations, if the coronary arteries were divided into 2 buttons, the medial suture line exactly corresponded to the thin and fragile coronary artery wall and was near the orifice, which cannot move apart from another coronary orifice. This situation may occur in coronary ostial stenosis.
2. The second consisted of a long IMCA stenosis or distal IMCA stenosis that required patch angioplasty or CABG, both of which are technically challenging procedures in neonates.

In conclusion, the Imai technique can be an option during coronary management for high-risk coronary anatomy, particularly distal IMCA stenosis and inseparable coronary arteries with an almost single orifice. Good aortopulmonary fenestration patency, coronary pouch growth, and no thrombus were shown in late aortography. Adequate neopulmonary artery augmentation must be performed to prevent coronary pouch compression and right ventricular outflow stenosis. Careful follow-up should be mandatory.

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