
症 例

Aortitis Syndrome Treated by Apico-Aortic Bypass Procedure: Case Report

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Introduction

Aortitis syndrome (Takayasu's disease) is a non-specific inflammatory process of unknown etiology which affects segments of the aorta and its main branches. The end result of marked fibrosis and thickening of the aortic wall is usually an occlusion or stenosis, and occasionally a saccular aneurysm.

The clinical features of aortitis syndrome are variable, depending on the site, mode and grade of aortic involvement. Therefore, in the surgical treatment of this disorder various procedures have been employed. Standard treatment was prescribed for the systemic hypertension with aortic occlusion. In fact, there are some limitations in selecting the sites for anastomosis of bypass graft because of active pathological lesions and calcification of the aortic wall.

We encountered a case in which extensive calcification was found in the ascending and thoracic descending aorta. Therefore, we selected the apico-aortic bypass method; to our

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knowledge, this is the first report describing this method in the surgical treatment of aortitis syndrome.

Case Report

The patient was a 46-year-old female; since 1952 she had a history of hypertension and cardiomegaly. Nasal bleeding suddenly developed in 1969 and continued for 3 days. The patient was admitted to a hospital, where she received blood transfusion which was effective in stopping the nasal bleeding. The duration of her admission was two months, at which time differences between right and left brachial arterial pressures were first noted; visual disturbance of the left eye, dyspnea on effort, palpitation, headache and dizziness also developed. She was subsequently referred to a university hospital where angiography disclosed stenosis of the left renal artery; she was treated with depressor agents. Visual disturbance of the right eye also developed suddenly in 1980; this was diagnosed as hypertensive retinopathy (Grade K-W III) with right retinal bleeding.

In 1982, in order to control the hypertension, she was admitted to Kyoto University Hospital, where right and left heart catheterization and angiography were performed. She was diagnosed as aortitis syndrome, and underwent an operation to control the hypertension.

Clinical features:

Body weight was 48.2 kg, height 150.7 cm, body temperature 36.5°C, and pulse rate 72/min. Blood pressures were as follows: right brachial artery 264/120-84 mmHg, left brachial artery 130/84 mmHg, right tibial artery 164/80 mmHg and left tibial artery 160/84 mmHg. Pulsation was remarkably felt on both of the common carotid artery and right brachial artery. Upon palpation aneurysms were noted in both of the common carotid arteries, their sizes being 1.5 × 2.0 cm (right side) and 2.0 × 3.5 cm (left side). Systolic bruits were heard widely in the anterior and posterior thoracic regions and the upper abdomen. A grade 1-2/6 early diastolic murmur was heard in the 2nd and 3rd left sternal borders, IIa was accentuated, femoral sound was heard, and pulsation in both of the dorsal pedic arteries were well palpable. Liver and spleen were not palpable. Laboratory findings are shown in Table 1. Values of LDH (141) and γ -globulin (22.2%) were high. Values of serum iron (26 μ g/dl), total cholesterol and β -lipoprotein were low. Erythrocyte sedimentation rate was slightly elevated (mean: 21 mm). The C-reactive protein value was negative and the anti-streptolysin-O titer was within normal range. The serological tests for syphilis (VDRL and TPHA) were non-reactive as was Mantoux reaction; urinalysis was normal.

Electrocardiogram showed a severe left ventricular hypertrophy ($Sv_1 + Rv_5 = 9.6$ mV) and normal axis (60°). QS pattern was noted in Leads V1-3 and ST depression (2.4 mV) was noted in Leads V5-6. Chest roentgenography revealed an enlarged cardiac shadow (CTR=0.57) and severe calcium deposits on the ascending aorta, aortic arch and descending aorta. In renogram, RPF was low (405 ml/min), with GFR and FF being 91 ml/min and 0.22, respectively. Ultrasonic cardiography showed a marked hypertrophy of septum and left ventricular wall (2.4-3.0 cm), and a small left ventricular cavity. Hypertensive retinopathy (Grade K-W III)

Table 1. Laboratory Findings

1) Complete Blood Count		4) Liver Function Test	
RBC	357 × 10 ⁴ /mm ³	GOT	12 IU/L
WBC	3800/mm ³	GPT	7 IU/L
Hb	10.7 g/dl	LDH	141 IU/L
Ht	31.6%	ALP	36 IU/L
PLT	3.1 × 10 ⁴ /mm ³	CRE	0.7 mg/dl
2) Serological Test		BUN	12 mg/dl
CRP	(-)	CPK	26 IU/L
ASLO	<20×	TP	7.2 g/dl
RAT	(-)	Alb	54.2%
3) PSP-test		α_1	3.0
15 min	40%	α_2	6.8
30	Σ57	β	13.6
60	Σ78	γ	22.2
120	Σ92	A/G	1.18
		5) Cu	144 μg/dl
		Fe	26 μg/dl

was noted in both of the fundi. Preretinal bleeding (right eye) and subretinal bleeding (left eye) were noted. Arteriosclerotic degeneration was seen but there was no aneurysmal formation. Both visual disturbances were remarkable; RV=0.01 (-1.0 D 0.02), LV=0.1 (-0.5 D 0.2).

Cardiac catheterization data are shown in Table 2. High systemic pressures were found in the left ventricle, ascending aorta and aortic arch. The pressure gradient between the aortic arch and abdominal aorta was 132 mmHg (Fig. 1). Angiography revealed a stenotic descending aorta, and aneurysmal formation in both carotid arteries. In addition, there was severe occlusive lesions of the pulmonary arterial branches, particularly on the left side. Occlusion of the left subclavian and vertebral arteries were noted. A grade 1/4 aortic regurgitation was also seen.

Renin activity (at rest) was within normal limit in the inferior vena cava and both renal veins.

Table 2. Data of preoperative cardiac catheterization*

Sites	Syst.	Diast.	Mean	Oxygen (Vol%)
r-PC			10	
r-PA	40	19	27	11.85
m-PA	40	17	22	
RVout	36	0(12)		11.30
RVinf	44	0(12)		
RAm			5	
LVout	279	0(24)		
Ao asc	287	73	154	
FA	171	72	115	

Cardiac output 3.6 L/min. Stroke volume 33 ml/beat.

Cardiac index 2.5 L/min/M²

PAR 267 dynes sec cm⁻⁵

* see Fig. 1 for intraaortic pressures.

() : End-diastolic pressure

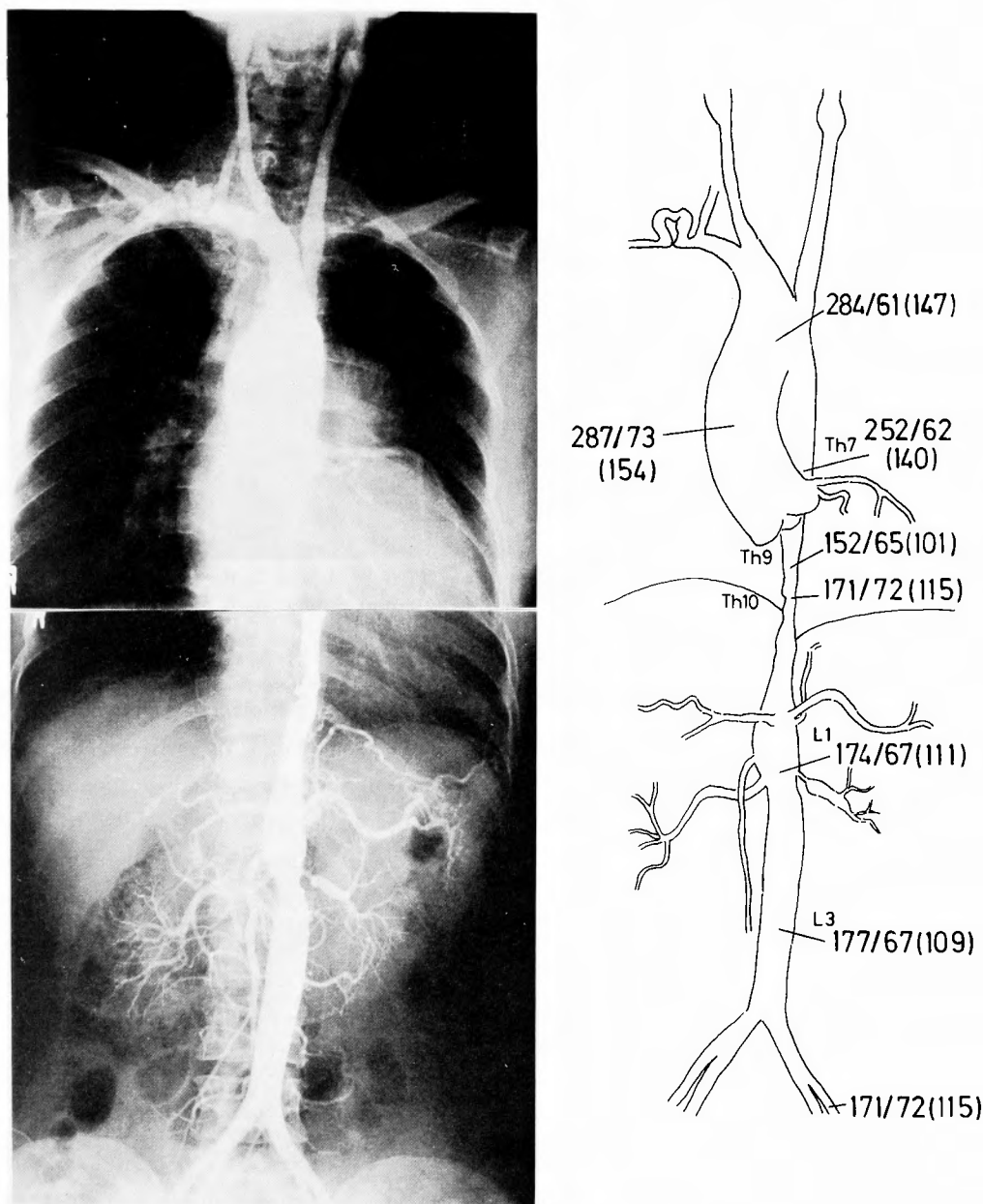


Fig. 1. Aortogram and intraaortic pressures

Serum aldosterone was elevated in the left renal vein.

CT-scan of the chest revealed a severe calcium deposit in the walls of the ascending and descending aorta, but no mural thrombosis was found (Fig. 2). The ascending aorta and aortic arch were slightly dilated; the descending aorta was stenotic. In the dilated brachiocephalic artery, calcium deposit but no thrombosis was noted. Pulmonary arterial trunk was remarkably dilated.

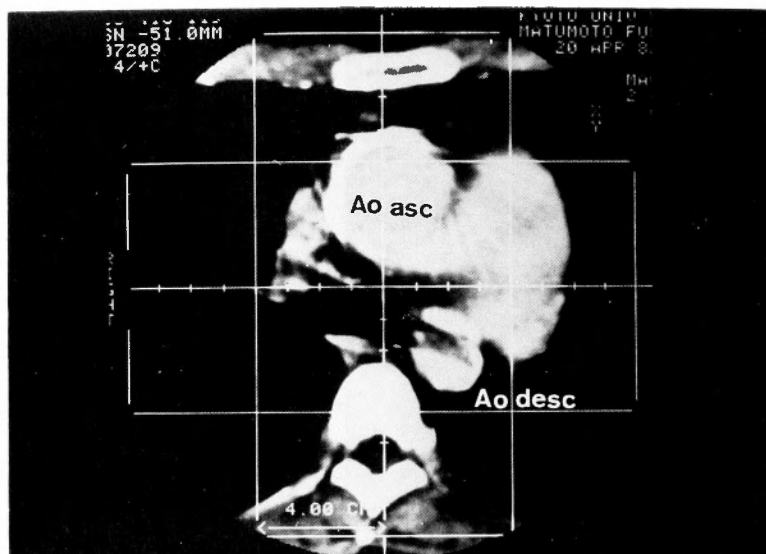


Fig. 2. CT-scan of the chest reveals a severe calcium deposit in the walls of the ascending and descending aorta, but no mural thrombosis is found.

Surgical technique (Fig. 3):

A midline sternotomy, extending into the abdomen, was performed. Diameters of the ascending aorta and main pulmonary artery were 40 mm and 32 mm, respectively. Extensive calcification was found throughout the ascending aorta, except for a small portion near the aortic arch into which aortic cannulation was performed. The decision was made to employ a conduit graft, apico-aortic bypass. The narrow segment, starting from the descending aorta, ended at the level of the celiac artery. Carpentier-Edwards bioprosthetic valved conduit was anastomosed end-to-side to the abdominal aorta at the level of the celiac artery, the wall of which had hypertrophied intima and media. Cardiopulmonary bypass was done using hemodilution and a membrane oxygenator, separating arterial cannulations into the ascending aorta and left external iliac artery. Aortic cross-clamping was unavoidably performed with two Cooley's aortic clamping forceps, despite the fear that these forceps might crush the calcified aortic wall; fortunately rupture of the aortic wall did not occur. Young's solution and MIK solution¹²⁾ were used as a cardiac arresting agent and a cardioplegic solution, respectively. An incision was made in the apex of the left ventricle, into which was inserted a rigid inlet tube of Pyrolite carbon^{*} connected to a Teflon sewing ring with a Dacron fabric graft (14 mm in diameter), this graft was sutured to the ventricular incision. Another incision was made in the diaphragm adjacent to the left ventricular apex and an identical graft was anastomosed end-to-end to the abdominal graft. The total length of the graft was 12 cm, and the total duration of the aortic clamping was 43 minutes.

The myocardial muscle was easily torn and needed many U-stay sutures to stop the bleeding. Soon after declamping the apico-aortic bypass, the pressure gradient was 30 mmHg between the right radial artery and the right pedic artery. But the pressure gradient disappeared about

Fig. 3. Apico-aortic by-pass performed in this case

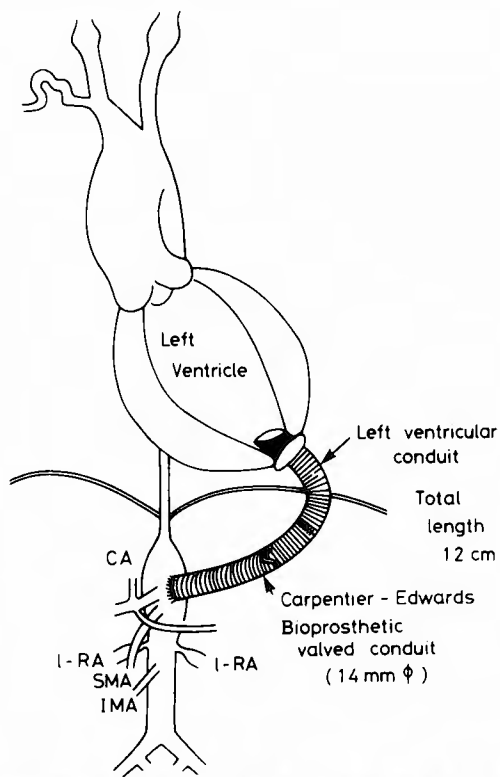


Fig. 4. Marked left ventricular hypertrophy (30 mm) in the center portion of the anterior wall resulting from concentric hypertrophy. Blood coagula (approximately 230 gm) was found, especially around the apex of the anastomosis.

6 hours later.

Though the patient complained of general malaise and dyspnea, the postoperative course was uneventful for the first 10 days. On the 11th postoperative day, chest roentgenogram revealed an increased mediastinal shadow with $CTR=0.68$ and moderate pericardial effusion. Blood pressure was 120/80 mmHg, and central venous pressure was 8 cm of water. In addition to dyspnea and general malaise, the patient complained of headache and palpitation; daily urine volume decreased to less than 1000 ml with leg edema. As low cardiac output syndrome was suspected, dopamine ($5 \mu\text{g}/\text{min}/\text{kg}$) and dobutamine ($5 \mu\text{g}/\text{min}/\text{kg}$) were administered, which resulted in a daily urine volume of 1500 ml.

On June 1, the patient died suddenly. Extrathoracic cardiac massage was not effective in resuscitating the heart.



Fig. 5. Thickening of the wall and severe calcium deposits in the ascending aorta, aortic arch and thoracic descending aorta
Minimum diameter of the thoracic descending aorta was 7 mm. Relatively new thrombi were found between the ascending aorta and the abdominal aorta at the level of renal artery. Left subclavian artery was completely occluded. The wall of the abdominal aorta appeared normal.

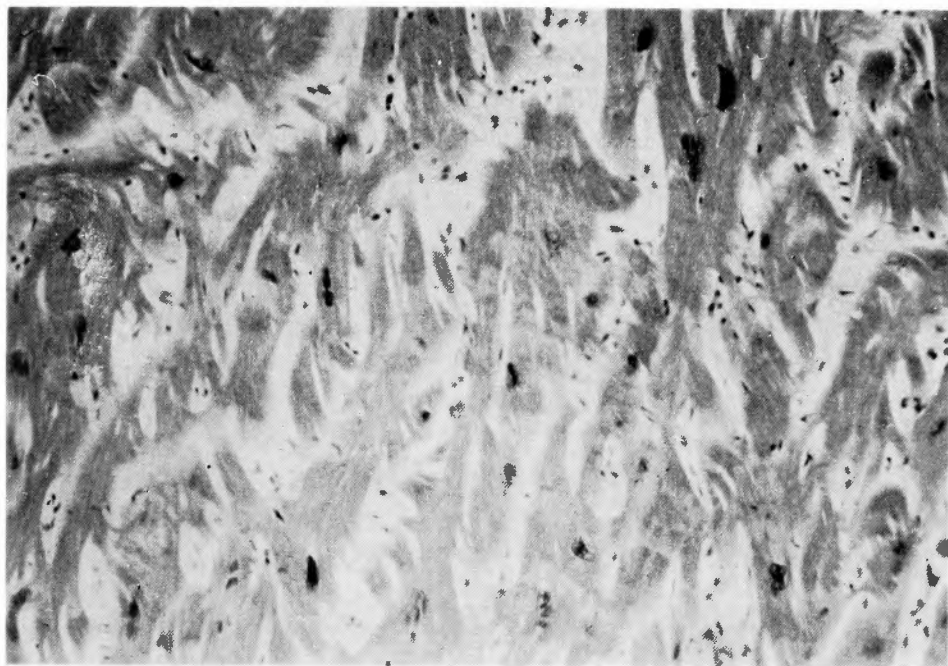


Fig. 6. Marked hypertrophy and disarray of the myocardial fibers. (H-E staining)

Autopsy findings:

Autopsy revealed cardiac tamponade, which was due to massive pericardial coagula (230 gm), possibly at the anastomosis site between the left ventricular apex and the graft (Fig. 4). Only

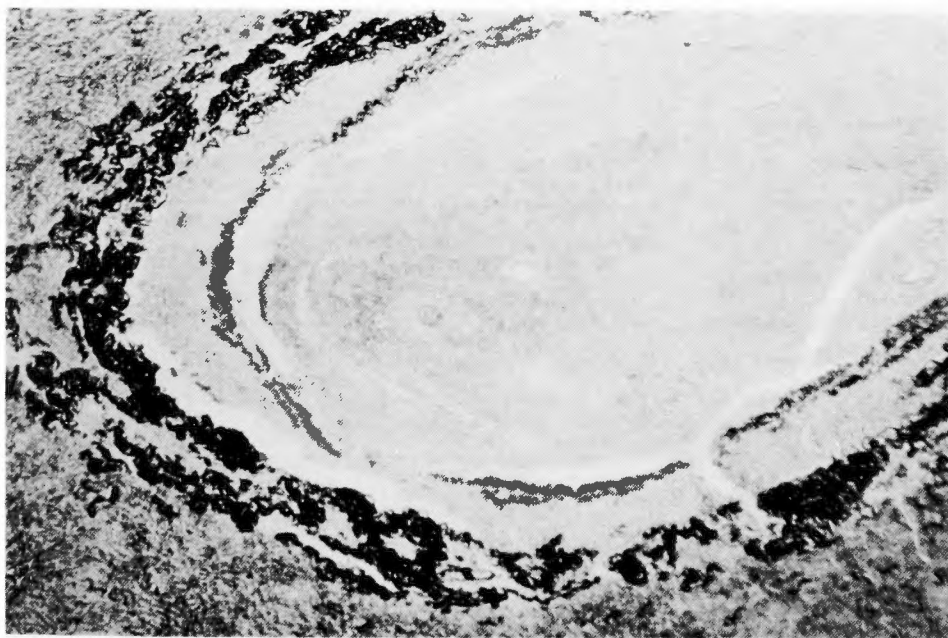


Fig. 7. Histological feature of left subclavian artery. Thickening in all layers of the wall and rupture of elastic fibers were found. Thickening of the intima completely occluded the lumen. (Van-Gieson Staining)

the intima of the ascending aorta, double-cross-clamped by Cooley's forceps, was slightly torn and the site of aortic cannulation was completely closed (Fig. 5). Subdural and intraponeal bleeding without clear onset were noted but other parenchymal organs were normal. In addition, scattered necrotic infarctions were found on the anterior wall of the left ventricle. Histological features of the cardiac muscle and the left subclavian artery are shown in Figures 6 and 7.

Discussion

The apico-aortic bypass method as a left ventricular "vent" is not a new procedure. In 1955, SARNOFF et al.¹⁴⁾ used a Lucite tube containing a Lucite Hufnagel valve to direct the entire cardiac output from the left ventricle to the descending thoracic aorta in dogs. COOLEY et al.⁴⁾ reported on the unpublished work of TEMPLETON, in 1962, who was the first to clinically apply this method in five patients with severe aortic stenosis. In 1975, BERNHARD et al.²⁾ reported a surgical technique in which a stainless steel conduit containing a Hancock valve and graft was anastomosed to the descending thoracic aorta.

In 1976, COOLEY et al.⁴⁾ applied this bypass method to nine patients; in seven of them, previous attempts were made to correct the stenosis. One patient, a 72-year-old man with coronary artery occlusive disease, calcific aortitis and valvular stenosis, had extensive calcification of the aorta. In this patient, two saphenous vein grafts were attached to the conduit distal to the valve, passing through incisions in the diaphragm, and were anastomosed to the left anterior descending coronary artery and the obtuse marginal branch of the circumflex coronary artery.

COOLEY⁴⁾ pointed out the following advantages of this method: 1) No postoperative anticoagulant therapy is needed. 2) No ill effects were noted from the "plugging" of the ventricular apex, probably because this is the thinnest, least vascular, and least irritable the ventricle. Furthermore, there appeared to be no deleterious effects from fractionalization of the left ventricular output. 3) This method is more advisable when there have been previous surgical procedures on the aortic annulus and ascending aorta or the densely cicatrized superior mediastinal structure. 4) When malfunction of the heterograft valve in the apico-aortic bypass occurs, its replacement presumably would be relatively simple.

The clinical features of aortitis syndrome are variable depending on the site, mode and grade of aortic involvement. Therefore, in the surgical treatment of this syndrome, various procedures have been employed such as various bypasses for relieving hypertension, reconstruction of common carotid or renal arteries^{7,18)}, aorto-coronary bypass for coronary arterial occlusion²³⁾, aortic valve replacement for aortic regurgitation^{5,8,19)}, and implantation of an artificial graft for aneurysmal aorta¹⁸⁾.

Most hypertension causing lesions are atypical coarctation of the aorta, in which the following bypasses are performed in order to relieve hypertension: A) Aorto-aortic bypass 1) the thoracic descending aorta to descending aorta or abdominal aorta (ASANO¹⁾, WADA²¹⁾), 2) the ascending aorta to abdominal aorta (ASANO¹⁾, LIOTTA¹⁰⁾), and B) Long bypass 1) the ascending aorta to common iliac artery (WADA²¹⁾, KAZUI⁹⁾), 2) the brachiocephalic artery to abdominal aorta (INOKUCHI⁷⁾), 3) the axillo-iliac bypass (SUEHIRO¹⁷⁾), 4) the subclavian-iliac bypass (SHIRA-

KATA¹⁶⁾ (SHIRAKATA¹⁶⁾), and 5) the axillo-femoral bypass (BLAISDELL³⁾, MANNICK¹¹⁾).

The surgical damage resulting from aorto-aortic bypass by thoracotomy and/or laparotomy is more extensive than that from the long bypass. Furthermore, in aortitis syndrome, the poor development of collateral arteries greatly increases the damage caused by aortic clamping. Postoperatively, the rapid rise of the abdominal blood pressure may lead to paradoxical hypertension and internal bleeding of the abdominal organs. On the other hand, SUEHIRO¹⁷⁾ pointed out the following advantages of the various long bypass methods: 1) Both-sided thoracotomy and positional change during surgery can be avoided, 2) Anastomosis can be performed on not-diseased vessels, and 3) It is easy for these bypasses to branch off to visceral arteries if necessary. Therefore, he recommended that the long bypass method be used for elderly patients with a long segment of atypical coarctation, and in whom lesions are widely spread over the thoracic aorta. However, the disadvantages of these methods involve: 1) the duration of patency, 2) the depressant effect, and 3) the kinking of the anastomosed vessels and bending of the long bypass graft.

When lesions of the renal arteries also cause hypertension, additional surgical procedures for the these arteries must be performed. In fact, in aortitis syndrome with renal arteries lesions and atypical coarctation, the postoperative depressant effect is unexpectedly poor²⁰⁾. The problem of whether the surgical procedures with respect to the renal arteries should be primarily or secondarily performed, has not been resolved^{20, 21)}.

It is reasonable to avoid operation during the active inflammatory stage, which can be determined by the erythrocyte sedimentation rate, values of γ -globulin and C-reactive protein, and leucocyte count, except when heart failure occurs rapidly. The anastomosis should be performed at the sites where inflammatory findings are not noted. In aortitis syndrome, the sites of anastomosis are limited by the condition of the aortic wall. Therefore, in cases where extensive calcification involves a wide area of the thoracic area, the aorto-aortic bypass is unsuitable. In our case, it was reasonable to perform the apico-aortic bypass. However, in this case, the two major concerns were whether the ascending aorta had tolerance for cross-clamping during cardiopulmonary bypass, and the site of aortic cannulation. The absence of extensive calcification in the distal and left portion of the ascending aorta of our patient permitted cross-clamping, however, when cross-clamping is not possible, the apico-aortic bypass must be performed without interrupting the coronary circulation. In our case, CT-scan was very useful in diagnosing the site and severity of calcification of the aortic wall.

Furthermore, another problem was whether the myocardial muscle around the left ventricular apex would be tolerable for anastomosis. This problem depends on the patient's age, the duration of symptoms, the extent of this disease and the thickness of the cardiac muscle.

SAITO¹⁵⁾ reported that the majority of calcification of aortic wall was found in patients with the duration of symptoms more than 5 years, and dilatation of the ascending aorta and aortic regurgitation was found in all patients with the duration of symptoms more than 10 years. In OHNO's¹³⁾ case with very extensive calcification of the thoracic aorta, the duration of symptoms was more than 20 years, such as in our case. The cardiac muscle of our patient was very thick and easily torn. Thus, many U-stay sutures were needed to stop the bleeding around the apex.

It is likely that these U-stay sutures resulted in myocardial necrosis around the rigid inlet of the graft causing the delayed cardiac tamponade.

The greatest complication of this apico-aortic bypass is bleeding, thus bleeding during operation and cardiac tamponade in the postoperative course should be carefully monitored. In our patient, during the first 10 postoperative days, an excellent "venting" effect for hypertension was seen, with no pressure gradient between the upper and the lower portions of the body. Therefore, this apico-aortic bypass method should be added to the other surgical techniques for aortitis syndrome, especially for patients with an extensively calcified ascending aorta.

Summary

A 46-year-old female with aortitis syndrome, underwent an apico-aortic bypass; because of extensive calcification covering a wide area of the aorta, an aorto-aortic bypass could not be performed. The first 10 days of the postoperative course were uneventful, but 22 days postoperatively she died suddenly of delayed cardiac tamponade, which was revealed on autopsy.

Successful suturing of the left ventricular apex to the graft, and careful screening of patients would make this apico-aortic bypass a useful technique as a "vent" of hypertension in the left ventricle not only in various types of aortic stenosis but also in aortitis syndrome with extensive calcification of the thoracic aorta.

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和文抄録

広汎な大動脈壁石灰化を伴った大動脈炎症候群に対する Apico-aortic Bypass 術の1例

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46才，女性，異型大動脈縮窄から高度高血圧症を来した広汎な大動脈壁石灰化を伴った大動脈炎症候群に対し Apico-aortic Bypass 術を施行した。術後10日間は順調な経過をたどった。降圧効果は良好で，胸・腹部大動脈圧差は消失した。しかし，術後22日目心タ

ンボナーデで急死した。左室心尖部グラフト吻合部からの出血に依るものであった。本法は，大動脈狭窄症のみならず，広汎な大動脈壁石灰化を伴った大動脈炎症候群に対しても左心室減圧に有用な術式である。