Valve Detachment After Aortic Valve Replacement
— A Successfully Reoperated Case With the Cabrol Technique —

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A 50-year-old Japanese male was admitted with a 1 month history of progressive shortness of breath, palpitations, and intermittent low-grade fever. On admission, a musical diastolic murmur (sea-gull murmur) was heard on the 3rd left sternal border. Blood studies showed an accelerated erythrocyte sedimentation rate, and positive C-reactive protein. Retrograde aortography revealed severe aortic regurgitation. At the operation, the aortic wall was remarkably thickened, and the aortic valve commissures between the right and left coronary cusps and between the left and non-coronary cusps had completely detached from the aortic wall. The aortic valve was replaced with a St Jude Medical valve using pledgeged sutures. Histologic studies of the aorta and aortic valve revealed no evidence of any pathologic changes. Echocardiography performed 82 days after aortic valve replacement showed severe paravalvular leakage and valve detachment. At the reoperation, the prosthetic valve was detached from the extremely edematous and fragile native aortic annulus. There was no vegetation or abscess formation. Aortic root replacement with the Cabrol technique was performed using a composite graft in which the prosthetic valve was placed 1 cm above the proximal extremity to reduce the tension on the suture line and the aortic annulus. Histologic study of the aorta after the reoperation showed non-specific inflammatory disease. The patient, who received steroid therapy, has recovered without any signs of graft detachment or pseudoaneurysm formation. (Jpn Circ J 1996; 60: 377–381)

AORTITIS syndrome, including Takayasu’s arteritis, often affects the aorta, its major branches and the aortic valve. Surgical treatment of this pathology presents many difficulties; not only those due to the extensive and severe lesions, but also those due to the manipulation of fragile and inflamed tissue. For these reasons, postoperative complications such as hemorrhage, pseudoaneurysm formation, valve detachment, and paravalvular leakage are often encountered after the initial or even after the second operation.

In this report, we describe a patient with aortitis syndrome who had valve detachment after aortic valve replacement and who was successfully managed with the Cabrol technique.

CASE REPORT

A 50-year-old Japanese male was admitted to our hospital with a 1 month history of progressive shortness of breath, palpitations, and intermittent low-grade fever. He had
had no prior history of cardiovascular disease or trauma, and regular annual physical examinations had disclosed no abnormalities. During the previous 1 year, he had occasionally been aware of a low-grade fever of around 37°C. One month prior to admission, he experienced shortness of breath and palpitations on exertion for the first time. He subsequently noticed a progressive limitation of his activity because of dyspnea.

On admission, blood pressure was 120/35 mmHg in both arms, and the pulse rate was 85 beats per minute with an irregular rhythm. A Grade 2 systolic ejection murmur and a III/VI musical diastolic murmur (sea-gull murmur) were heard on the 3rd left sternal border. While on the ward, the patient occasionally had a low-grade fever of around 37°C. Blood studies constantly showed an accelerated erythrocyte sedimentation rate, and positive C-reactive protein (CRP), while the white blood cell count was not increased. Repeated blood cultures were sterile. Echocardiography revealed eccentricity of the coaptation point of the aortic valve without thickening and or deformity of the cusps, diastolic flutter of the anterior mitral leaflet, and diastolic separation of the closure line of the aortic cusps. Severe aortic regurgitation (AR) and mild mitral regurgitation were disclosed on Doppler flow studies. Retrograde aortography showed Grade 4/4 AR. Aortic valve replacement (AVR) was urgently scheduled because of frequent anginal attacks due to AR. At the operation, the aortic wall was remarkably thickened, and the aortic valve commissures between the right and left coronary cusps and between the left and non-coronary cusps had completely detached from the aortic wall and prolapsed into the left ventricle (Fig 1). The aortic valve was otherwise normal. The valve was replaced with a 23-mm St Jude Medical (SJM) valve (St Jude Medical Inc, St Paul, MN, USA) using 17 pledged sutures. Although the diagnosis of aortitis syndrome was strongly suspected, histologic studies of the aortic wall and the aortic cusps revealed no evidence of any pathologic changes such as aortitis, syphilis, infective endocarditis, myxomatous degeneration, cystic medial necrosis, or connective tissue disorders. Echocardiography, including Doppler study, performed 1 week after the operation confirmed that the SJM valve was functioning normally and that there was no paravalvular leakage. After a 72-day uneventful recovery, except for an intermittent low-grade fever and constantly positive CRP, a routine echocardiography showed mild paravalvular leakage around the aortic valve prosthesis. Ten days later, echocardiography revealed severely progressed paravalvular leakage with valve detachment (Fig 2). Repeat blood cultures after the operation were sterile. A reoperation was planned immediately. At the reoperation, about one third of the aortic prosthetic valve
was detached from the native annulus, which was extremely edematous and fragile (Fig 3). Aortic root replacement was performed using the Cabrol technique. A 23-mm SJM valve and a 26-mm Dacron graft (Hemashield, Meadox Medicals Inc, Oakland, NJ, USA) were used to form a composite valve graft in which the prosthetic valve was placed 1 cm above the proximal extremity. Histologic study of the aorta at the second operation showed non-specific inflammatory disease. Steroid therapy (prednisolone, 20 mg/day) was started begun after the second operation. Three months after the second operation, the patient was discharged from our hospital without any signs of graft detachment or pseudoaneurysm formation on echocardiograms.

DISCUSSION

Aortitis syndrome is a panarteritis of unknown cause involving the medium- and large-sized arteries, and is seen more frequently in Japan than in other countries. This pathology is better known as Takayasu’s arteritis in the West. After the original description of Takayasu’s arteritis, epidemiologic, pathological, medical and surgical investigations and treatments have been performed, and various terms such as “pulseless disease”, “aortic arch syndrome”, “atypical coarctation of the aorta” have been used to describe this disease. However, such names do not address the entire scope of the disease, since it can involve not only branches of the aortic arch, but also the thoracic aorta, the abdominal aorta with its branches, and even the pulmonary artery. Based on these diverse clinical features, the name “aortitis syndrome” was proposed by Ueda and colleagues in 1963. In their study, aortitis syndrome included “Takayasu’s arteritis”, “pulseless disease”, “aortic arch syndrome due to aortitis”, “atypical coarctation of the aorta”, “renovascular hypertension due to aortitis”, and “aortic valve regurgitation due to aortitis”.

The cardiovascular complications of aortitis syndrome include steno-occlusive lesions or aneurysmal dilatation in the aorta and its branches, and aortic regurgitation. Among these complications, AR is seen in 5 to 19% of patients with aortitis syndrome. In these patients, the inflammation which extends to the ascending aorta and aortic valve principally involves dilatation of the aortic annulus and retraction of the aortic cusps, and consequently results in de-coaptation of the aortic cusps. However, AR due to detachment of the aortic valve commissures is rarely seen in patients with aortitis syndrome. To our knowledge, there has been only one report of a patient with detachment of the aortic valve commissure resulting from aortitis syndrome, by Yamauchi and colleagues.

AVR in aortitis syndrome deserves special attention because valve detachment, paravalvular leakage, and pseudoaneurysm formation are not rare postoperative complications. These complications have been described by several authors. Some investigators have recommended the use of pledgeted sutures or transmural buttressed sutures to prevent valve detachment after AVR. However, these reinforced sutures do not necessarily prevent the complications. In fact, we previously experienced 1 patient with aortitis syndrome who required a reoperation for paravalvular leakage of a prosthetic aortic valve. Although a new prosthetic valve was implanted with pledgeted transmural sutures in this patient, valve detachment recurred 40 months after replacement of the prosthetic valve, and the pledgeted sutures used to fix
the new prosthetic valve were observed around the sewing ring of the prosthetic valve in pseudoaneurysms which resulted from disruption of the aortic wall. To prevent valve detachment, Suzuki and colleagues emphasized that a modified Bentall procedure using a composite graft with a Teflon felt flange at the sewing ring and using a double-fixation method with reinforcement by a Teflon felt strip was useful because it reinforced the suture line and also reduced the tension on the suture line and the aortic annulus. They also recommended postoperative steroid therapy in patients who are operated upon during the active inflammatory stage and who have a histological diagnosis of aortic wall involvement. On the other hand, Yamauchi and colleagues pointed out that patients with non-Takayasu’s arteritis, especially male patients, were more susceptible to valve detachment than those with Takayasu’s arteritis. None of their patients with Takayasu’s arteritis has suffered valve detachment, and their 4 patients who developed valve detachment were successfully managed with a modified Bentall technique, which included translocation of the prosthetic valve to 1.5–2.0 cm above the proximal extremity of the vascular tube graft. Based on these results, they advocate that the key to repairing valve detachment in arteritis syndrome is to reduce the tension on the suture line and aortic annulus, and suggest that aortic root replacement with an allograft may provide favorable results for AR in arteritis syndrome.

Postoperative steroid therapy for arteritis syndrome is controversial. Although the administration of steroids in the early postoperative period has some potential hazards, such as susceptibility to infection and disturbance of the wound-healing process, we believe that steroid therapy should be started in the early postoperative period to control inflammation in patients who are operated on during the active inflammatory stage.

REFERENCES
