A 38-year-old woman who had undergone an original Bentall operation in December 1995 for annuloaortic ectasia associated with ulcerative colitis underwent reoperation for ascending aortic aneurysm, coronary ostial aneurysm, and patent Cabrol trick. The initial Bentall operation included aortic root replacement using a valved conduit and reconstruction of the coronary arteries. Both coronary ostia were directly anastomosed to the composite valved graft, which was wrapped with the dilated aortic wall, and a Cabrol trick was added at the same time. She underwent reoperation for a 60-mm ascending aortic aneurysm which had been used for wrapping at the initial operation. The findings at reoperation were a patent Cabrol trick, leakage from the distal anastomosis, aneurysm of both coronary ostia, and paravalvular leakage. The repairs included graft replacement, leaving the valvular prosthesis, reconstruction of both coronary arteries by the Piehler method and Carrel patch technique, repair of the paravalvular leakage, and closure of the Cabrol trick. Her postoperative course was uneventful, and the serum concentration of C-reactive protein remained within normal limits. Strict follow-up care is required to avoid further anastomotic dehiscence. (Circ J 2005; 69: 861–864)

**Key Words:** Anastomotic dehiscence; Aortitis syndrome; Bentall operation; Reoperation

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**Case Report**

A 38-year-old woman with aortitis syndrome associated with ulcerative colitis underwent an original Bentall operation for annuloaortic ectasia with severe aortic insufficiency in December 1995. At the time of initial operation a composite Dacron graft with a 27-mm mechanical valve was used, and both coronary ostia were directly anastomosed to the composite valved graft, which was wrapped with the native aortic wall, together with interposition using a tube graft between the aorta and right atrium (Cabrol trick). Pathology revealed low-grade inflammation of the aortic wall with interrupted elastic fibers, which was compatible with a diagnosis of aortitis (Fig 1). The patient was well until 5 years later when she had shortness of breath and anemia (Fig 2). Preoperative chest computed tomography and angiography revealed a patent Cabrol trick, aneurysmal dilatation of the ascending aorta and aortic arch around the artificial graft, coronary ostial aneurysms, and paravalvular leakage (Fig 3). She had a history of steroid therapy for the ulcerative colitis.

At the time of reoperation, dense adhesions were found especially around the Cabrol trick and the ascending aorta was dilated more than 60 mm. The femoral artery was cannulated for arterial perfusion. Two-staged venous cannulation was carried out through a small right atriotomy. After implementation of cardiopulmonary bypass, the ascending aorta was cross-clamped obliquely beyond the distal anastomotic site of the previous graft. After clamping the Cabrol trick, aortotomy of the dilated ascending aorta and Dacron graft revealed that the internal aortic wall was normal color and that the native aortic wall around both coronary ostia was dilated. Cold crystalloid cardioplegic solution was antegrade administered through both coronary ostia. Anastomotic dehiscence was found at the distal anastomosis of the Dacron graft and at the non-coronary cusp site of the proximal composite graft, but not at either of the coronary ostia (Fig 4). The primary dehiscence was considered to be the distal site of the graft, resulting in aneurysmal dilatation of the wrapped aorta. Paravalvular leakage was considered to have begun 5 years after the initial operation when the inflammation deteriorated (Fig 2).

The Dacron graft was totally removed, leaving intact the sewing ring of the mechanical valve. A new 30-mm Dacron graft was then sutured in place using everting sutures of pledgeted 2-0 polyester suture placed in the sewing ring. The proximal dehiscence at the non-coronary cusp was repaired from outside the aorta using pledgeted 2-0 poly-
Fig 1. (Left) Fibrous thickening with a torn medial wall and no active inflammation; Hematoxylin-eosin. (Right) Disruption of the medial elastic fibers (Elastica van Giesson stain).

Fig 2. Changes in the white blood cell (WBC) count and C-reactive protein (CRP) values after initial operation. Inflammatory deterioration was observed 5 years after initial operation.

Fig 3. (Left) Computed tomography scan shows aneurysmal dilatation of the ascending aorta (Ao), which compressed the artificial graft, with calcification of Cabrol's trick (arrows). (Right) Angiography shows the dilated aorta, aneurysmal dilatation of the left coronary orifice (arrows), patent Cabrol's trick (arrows), and paravalvular leakage to the left ventricle (LV).
ester sutures. A 10-mm Dacron graft was anastomosed to the left coronary ostium and the new 30-mm Dacron graft (Piehler’s method). The right coronary ostium was directly anastomosed to the 30-mm Dacron graft by button technique (Carrel’s technique). The ascending aorta and the partial proximal arch were totally excised beyond the previous distal anastomotic site, and distal anastomosis was then performed with 4-0 polypropylene suture, reinforcing the aorta with a Teflon felt strip (Fig 5). After declamping, the patient was easily weaned from cardiopulmonary bypass. Histologic studies of the excised aortic wall showed chronic inflammation compatible with aortitis.

Her postoperative course was unremarkable, and she has remained in good condition with the serum concentration of C-reactive protein (CRP) remaining within normal limits during follow-up as an outpatient (Fig 2).

Discussion

Aortitis syndrome causes chronic inflammatory change in the great arteries and/or branches resulting in stenosis/occlusion or dilatation. Cardiovascular complications can be fatal. The relatively rare combination of aortitis syndrome and ulcerative colitis suggests that they may have a common pathological background and an autoimmune genetic influence in Japanese patients, but the details are still unknown. In patients with aortic regurgitation and/or dilatation of the aortic root, AVR and/or root reconstruction are indicated. Cardiovascular symptoms may occur while the gastrointestinal disease is quiescent. In the present patient salazopyrine was already being administered before the first operation, and aortitis syndrome was not diagnosed preoperatively. Because there were no active inflammatory changes in the preoperative blood examinations or the intraoperative examination of the aorta, Bentall operation was performed. However, at reoperation the histopathology of the excised specimen was compatible with aortitis and the patient has strictly been followed up as an outpatient (Fig 2).

The most serious complications after AVR or root reconstruction for aortitis syndrome are dehiscence of the anastomotic site and formation of a false aneurysm. Therefore, the surgical treatment must be carefully selected. Suzuki et al have reported 15 patients with Takayasu’s disease or Behcet’s disease who underwent various surgical procedures, depending on the pathological changes in the aortic valve, aortic root and coronary artery. Reoperation was performed in 2 patients who required translocation of the valve with coronary artery bypass grafting (CABG) because of a deteriorated annulus and re-CABG due to graft occlusion. Those authors emphasize the importance of reinforcing sutures around the aortic annulus and Carrel’s button technique for reconstruction of the coronary artery, and recommend postoperative steroid therapy for active inflammation.

Ando et al have reported that the incidence of prosthetic valve detachment after AVR in 90 patients with aortic regurgitation caused by non-specific aortitis was 40% in patients with Behcet’s disease, 33% in aortitis of unknown etiology and 4.6% in Takayasu’s arteritis! Aoyagi et al have emphasized the importance of transmural pledged sutures and perioperative steroid therapy for preventing complications after AVR, especially during the active phase of the inflammation.

The original Bentall operation with direct coronary ostial reconstruction has a potential risk of reoperation depending on the pathologic changes of the aortic root. Ito et al have reported redo root replacement in 8 patients (7 with Marfan’s syndrome and 1 with active endocarditis) for pseudoaneurysm, coronary ostial aneurysm, and endocarditis. Carrel’s button technique, the interposition technique (Piehler), and Cabrol’s technique were used for reoperation, resulting in a hospital mortality rate of 25% (2/8). The Bentall procedure using a button technique for aortic root replacement is safe and durable with a low mortality rate and high reoperation-free rate in patients with dystrophic aneurysm or Marfan syndrome. In the present patient, Bentall operation using button technique should have been performed initially; however, only a few reports have described reoperation after the Bentall procedure for aortitis syndrome. In patients with the aortitis syndrome, the potential risk of anastomotic dehiscence is much higher than in patients with dystrophic aneurysm or Marfan syndrome.
syndrome, even in the inactive phase of the inflammation. The technique of coronary reimplantation has a significant influence on the long-term results and the method of choice is the button technique using reinforcing transmural sutures, especially in the presence of a fragile aortic wall. Either Cabrol’s technique or Piehler’s technique must be used when the Carrel or original Bentall reimplantation is not feasible; for example, during redo procedures. In the present patient, redo graft replacement of the ascending aorta was chosen instead of reimplantation of a new composite valved graft because of the fragile aortic root and it was a simpler procedure. The translocated Bentall procedure may be an alternative technique for composite graft detachment. Perioperative adjunct steroid therapy is also recommended for avoiding serious anastomotic dehiscence. When aortitis is associated with ulcerative colitis, as in the present patient, coronary inflammatory findings may be masked by the inflammation of the ulcerative colitis, so the timing and duration of the adjunct steroid therapy should be considered. In the present case, reoperation was performed while the patient’s gastrointestinal disease was quiescent. Strict postoperative follow-up is required to monitor and prevent further anastomotic dehiscence.

References


