Massive progression of annuloaortic ectasia in a patient with Takayasu aortitis

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A 52-year-old woman with Takayasu aortitis had undergone coronary artery bypass grafting and left subclavian artery reconstruction two years before admission to the Kinki University Medical Hospital (Osaka, Japan). On this admission, marked annuloaortic ectasia with severe aortic regurgitation was noted on echocardiography and aortography. Because of refractory heart failure, a modified Bentall operation was performed. Considering the serious cardiovascular complications of Takayasu aortitis, such as aneurysmal dilation of the aortic root, coronary artery ostial stenosis and, frequently, the need for surgical intervention, patients should be monitored closely.

Key Words: *Annuloaortic ectasia; Coronary artery bypass grafting; Modified Bentall operation; Sick sinus syndrome; Takayasu aortitis*

Takayasu aortitis is an inflammation of the aorta and its branches that leads to various vascular complications such as stenosis, occlusion and dilation. Some of these complications, such as aortic regurgitation secondary to aneurysmal dilation of the aortic root, can be fatal.

We describe a patient with Takayasu aortitis who underwent coronary artery bypass grafting (CABG) and left subclavian artery reconstruction, and subsequently required reoperation for marked annuloaortic ectasia (AAE) with severe aortic regurgitation.

CASE PRESENTATION

The patient was a 52-year-old woman who had previously been diagnosed with Takayasu aortitis based on clinical symptoms and findings such as chest pain and dizziness, as well as positive inflammatory markers, including a high C-reactive protein level. She was admitted to the Kinki University Medical Hospital (Osaka, Japan) with congestive heart failure due to severe aortic regurgitation. Two years earlier, she had undergone CABG and left subclavian artery reconstruction for coronary and subclavian artery disease. Six months after her first operation, she had received a permanent AAI mode pacemaker implantation due to sick sinus syndrome.

On admission, an examination revealed shortness of breath on exertion, palpitation and general malaise, and her blood pressure was 112/50 mmHg in both arms. She had a grade 3/6 diastolic murmur at the right sternal border, as well as abdominal bruits. Echocardiography revealed marked AAE with severe aortic regurgitation. Aortography confirmed progressive dilation of the AAE and grade IV aortic regurgitation (Figure 1). Due to the risk of rupture of the sinuses of Valsalva and the refractory heart failure, surgical repair was performed.

Évolution très importante de la maladie annulo-ectasique de l'aorte chez une patiente atteinte du syndrome de Takayasu

Une femme âgée de 52 ans, atteinte du syndrome de Takayasu a subi un pontage coronarien et la reconstruction de l'artère sous-clavière gauche deux ans avant son hospitalisation au Kinki University Medical Hospital, à Osaka, au Japon. Une maladie annulo-ectasique marquée, accompagnée d'une régurgitation aortique grave, a été observée à l'échocardiographie et à l'aortographie effectuées au moment de l'admission. Une intervention de Bentall modifiée a été pratiquée en raison d'une insuffisance cardiaque réfractaire. Compte tenu des complications cardiovasculaires graves du syndrome de Takayasu, comme la dilatation de l'origine de l'aorte, le rétrécissement orificiel des artères coronaires et le recours fréquent à l'intervention chirurgicale, la maladie appelle un suivi étroit.

During surgery with cardiopulmonary bypass, the aortic wall proximal to the previous saphenous vein and aortoaxillary bypass grafts was markedly enlarged and noted to be thin. The ascending aorta proximal to the previous aortoaxillary bypass graft was cross-clamped obliquely and the aortic root was opened (Figure 2). The aortic valve was also thin, and the aortic valve ring had expanded to 60 mm in diameter. A modified Bentall procedure using a 24 mm Dacron graft with a 21 mm St Jude valve (St Jude Medical Inc, USA) was performed. In addition, the left coronary ostium was found to be stenotic; therefore, CABG was performed, using a saphenous vein graft grafted to the left anterior descending artery. A pathological examination of the aortic wall revealed extensive intimal fibrosis and patchy infiltration of inflammatory cells, compatible with Takayasu aortitis. The patient's postoperative course was unremarkable, and she has continued to do well on adequate steroid and acetylsalicylic acid therapy. At the time of writing, she was 1.5 years postoperation without further complications of Takayasu aortitis.

DISCUSSION

Takayasu aortitis presents with various manifestations, including vascular disease affecting the aorta and its main branches, as well as aneurysm and aortic valve disease. Takayasu aortitis is induced by inflammatory changes, but the etiology remains unclear. Cardiovascular complications are serious, especially aneurysmal dilation of the aortic root with aortic regurgitation, which can be fatal. In our case, the initial operation was performed for coronary and subclavian artery stenosis and occlusive disease. The coronary arterial lesion is often located near the coronary orifice in patients with Takayasu aortitis (1,2). In the present case, the left coronary ostium was found to be stenotic in the intraoperative view, necessitating CABG.

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Figure 1) Aortography revealing progressive dilation of annuloaortic ectasia with marked aortic regurgitation (A) compared with previous aortography two years before admission (B)

In addition, our patient received permanent pacemaker implantation for cardiac conduction disturbance. A few cases of cardiac conduction disturbance due to the extension of aortitis have been reported (3,4). Watanabe et al (3) and Yokoi et al (4) reported that the possibility of conductance disturbance was caused by a myocarditis-attributable disease. Although coronary ischemia was another possible cause of cardiac conduction disturbance in the present case, a cardiac conduction complication with Takayasu aortitis should be considered in such patients.

The most serious cardiovascular complication with Takayasu aortitis is aneurysmal dilation of the aortic root with aortic regurgitation (5,6). In our case, although the inflammation associated with aortitis was controlled after the first operation, the patient required reoperation for AAE with refractory heart failure. In addition, dehiscence of the aortic prosthetic valve in patients with Takayasu aortitis has occasionally been reported (7,8). Suzuki et al (7) emphasized the importance of placing reinforcing sutures around the aortic annulus and Carrel's button technique for reconstruction of the coronary artery. Shiono et al (8) described that the technique of coronary reimplantation has a significant influence on the long-term results, and that the method of choice is the button technique using reinforcing transmurals sutures. Given the involvement of the aortic root by inflammation, radical correction of the aortic root is recommended in patients with Takayasu aortitis, even in the inactive phase of the inflammation (9).

Figure 2) A Intraoperative view of annuloaortic ectasia with previous saphenous vein graft (thin arrow) and aortoaxillary bypass graft (thick arrow). B The aortic wall and aortic valve were thin, and marked dilation of aortic valve ring was noted

In summary, surgical treatment of Takayasu aortitis presents a number of challenges because of the diversity of manifestations. Because of this, close follow-up of these patients is warranted.

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