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Case Report

Takayasu disease revealed by aortic insufficiency associated with an aneurysm of the ascending aorta

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Takayasu's disease is a rare inflammatory disease of unknown etiology that affects the large vessels, particularly the aorta and its major branches. A case of 29-year-old patient received for surgical management of severe aortic insufficiency was reported. Preoperative clinical and paraclinical investigations revealed a dilatation of the ascending aorta associated with a stenosis of the left subclavian artery and an inflammatory syndrome. This patient underwent a successful Banhson operation. Pathological analysis of the surgical specimen confirmed the diagnosis of Takayasu disease. In conclusion, aortic valve replacement combined with ascending aorta replacement performed in an inflammatory context was successful. However, it requires monitoring to detect complications related to tissue fragility.

Key words: Takayasu disease, aortic insufficiency, aneurysm.

INTRODUCTION

Takayasu's disease is a chronic inflammatory arteritis of unknown etiology that affects young women preferentially with segmental involvement of the aorta and its main branches. The median age at diagnosis was 28 to 40 years (Keser et al., 2018; Numano, 2002). It is characterized by thickening of the vascular wall leading progressively to stenosis, thrombosis and sometimes to the development of aneurysms (Bouzerda and Khatouri, 2016). Clinically, Takayasu's disease evolves in two phases: The acute period, called the preocclusive or systemic phase, which associates general signs, skin manifestations, pain in the arterial pathways and ophthalmological involvement: episcleritis, uveitis. The occlusive period or vascular phase is the consequence of arterial lesions (stenosis, obliteration and aneurysm) (Kerr et al., 1994; Keser et al., 2018). In the early phase, features of inflammation are present clinically and on blood tests (acute phase response). However, the disease may not present until after arterial damage has occurred (O'Connor et al., 2008). These vascular complications in untreated TA patients are the major causes of morbidity and mortality (Alali et al., 2017). Prognosis is probably improving with lower mortality rates in recent years, with survival of 97% at 10 years and 86% at 15 years, probably due to the use of more effective medical treatments as well as the use of surgical

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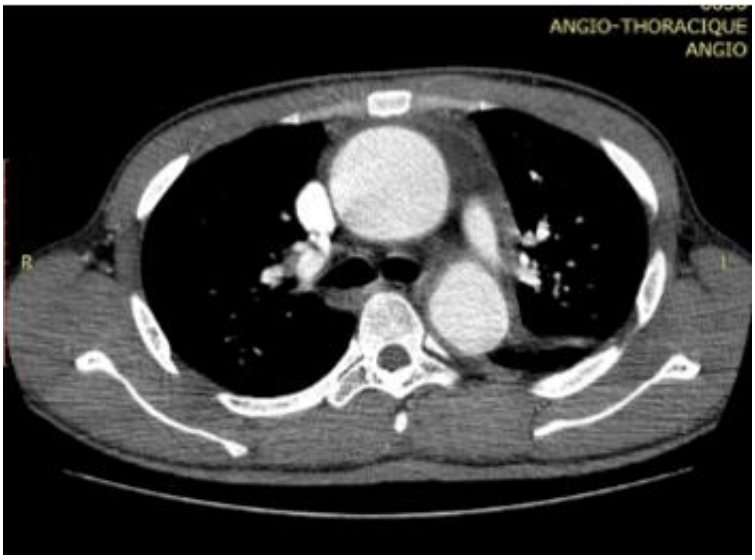

Figure 1. Angioscannographic section showing dilatation of the ascending aorta.
Source: Author

interventions when necessary and available.(Keser et al., 2018; Schmidt et al., 2013) Rarely described in black population of sub-Saharan Africa, the study reported the case of an aortic insufficiency revealing a Takayasu disease with a large arteries vasculitis.

CASE PRESENTATION

A 29-year-old male patient was presented to us with symptoms of angina and polyarthralgia; his investigations revealed aortic insufficiency for which he was referred to our hospital from Central African Republic for surgical management. His history included recurrent angina and polyarthralgia since the age of 15 years. The functional symptomatology was marked 2 years before the admission presenting as dyspnea on exertion with progressive aggravation classified as stage III, NYHA, associated with dizziness, palpitations, poly arthralgias and erectile disorders.

On admission, the patient's general condition was good (weight: 69 kg; height 1.77 m; Temperature: 36.8°C). Right and left limb blood pressure difference was noted as 154/56 and 87/58 mmHg, respectively. The radial and humeral pulses were hardly palpable at 87 beats/min. The heart sounds were regular with a diastolic murmur 4/6 maximum left lateral-sternal border. The rest of the physical examination was unremarkable. On electro-cardiogram, the rhythm was sinus with left ventricular hypertrophy of diastolic overload type.

Posteroanterior (PA) view of chest revealed cardiomegaly with a cardiothoracic index of 0.58 and an unwinding of the aorta. Cardiac echodoppler diagnosed severe aortic insufficiency with dilatation of the left ventricle with LVOT diameter of 63 mm. The LVEF was 50%, and 65 mm dilatation of the ascending aorta was noted. The angioscanner confirmed this aneurysm (Figure 1). An echodoppler of the supra-aortic trunks showed a stenosis of the left subclavian artery. The coronary angiography performed in this context did not reveal any coronary artery lesions.

The biological work-up revealed an inflammatory syndrome with a CRP of 59 mg/l, the ASLO were negative. The researchers decided to perform an aortic valve replacement along with the replacement of the ascending aorta in this patient.

The procedure was performed under cardio pulmonary bypass (CPB); the initial approach was a vertical median sternotomy. When the pericardium was opened, a serous effusion was found. The coronary arteries were flexible, the supra-coronary ascending aorta was very much dilated (Figure 2) with an inflammatory appearance confirming the probable diagnosis of Takayasu disease. The lesion assessment revealed a tricuspid aortic valve, very sclerotic and retracted. The aortic wall was very much thickened (Figure 3). Resection of the entire supra-coronary ascending aorta (Figure 4) was sent for Histopathological and bacteriological analysis. Then valve resection and implantation of a beleaflet mechanical prosthetic valve of 25 mm diameter (Saint Jude Medical®) (Figure 5) was done with a restoration of vascular continuity was done with a PTFE Prosthetic vascular tube of 28 mm of diameter (Saint Jude Medical®) (Figure 6). The postoperative course was uneventful and the patient was discharged from the hospital on the fifteenth postoperative day. Histopathological report of the surgical

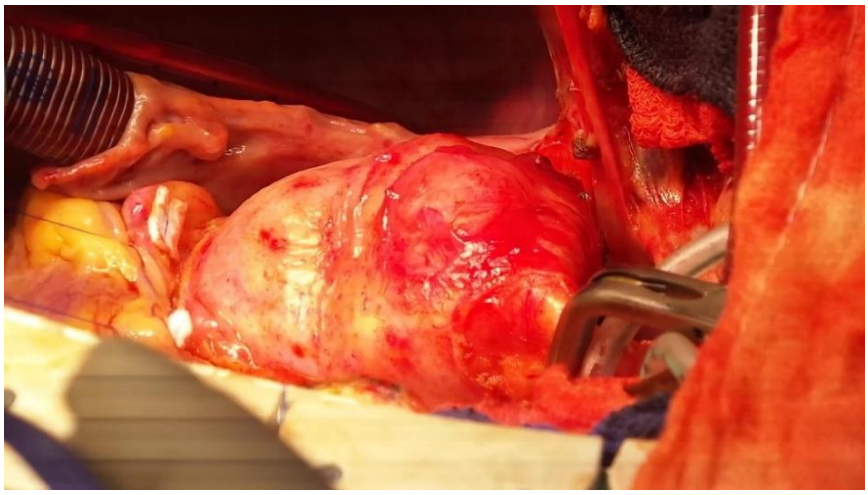

Figure 2. Intraoperative view of ascending aortic aneurysm.
Source: Author

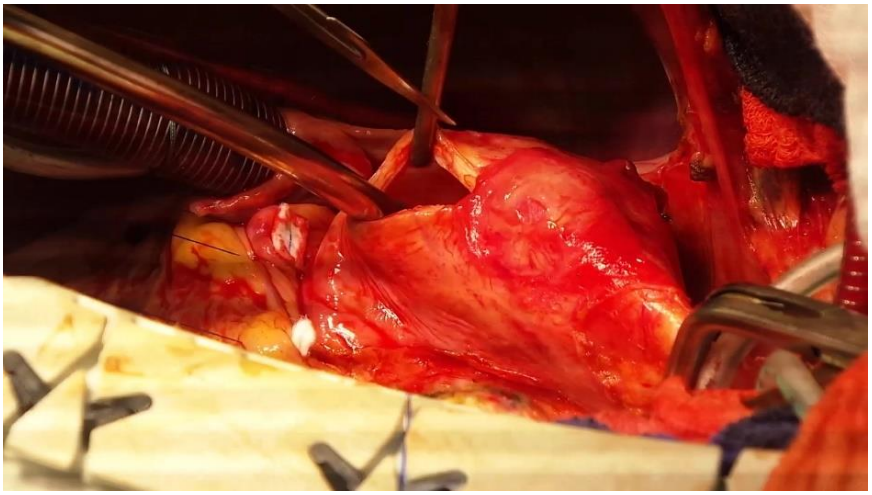

Figure 3. Intraoperative view of aortic wall thickening at the time of flattening of the ascending aortic aneurysm.
Source: Author

specimen revealed a predominantly medial adventitial granulomatous vasculitis in favor of Takayasu's disease.

DISCUSSION

Takayasu arteritis (TAK), also known as "pulseless disease," "aortic arch syndrome," or "occlusive thrombo-arthropathy," was first described by MikitoTakayasu, a professor of ophthalmology at Kanazawa University in Japan, as a case of pulseless retinal vasculitis in 1908 (Numano, 2002). Takayasu's arteritis occurs mainly in young Asian women. It has been reported worldwide with the highest prevalence in Asia (Ouali et al., 2011). Some cases of cardiac manifestations revealing Takayasu disease similar to our case have been described a case of cardiac manifestation such as coronary stenosis in North Africa (Bouzerda and Khatouri, 2016); a case revealed by aortic regurgitation associated with aneurysm in Arabia (Alali et al., 2017). However very few cases have been described in black populations in sub-Saharan Africa, hence the interest of this presentation. Clinically, Takayasu's disease evolves in two phases: The acute period, called the preocclusive or systemic phase, which associates general signs, skin manifestations, pain in the arterial pathways and ophthalmological involvement: Episcleritis, uveitis. However, these manifestations were not found in our patient. The occlusive period or vascular

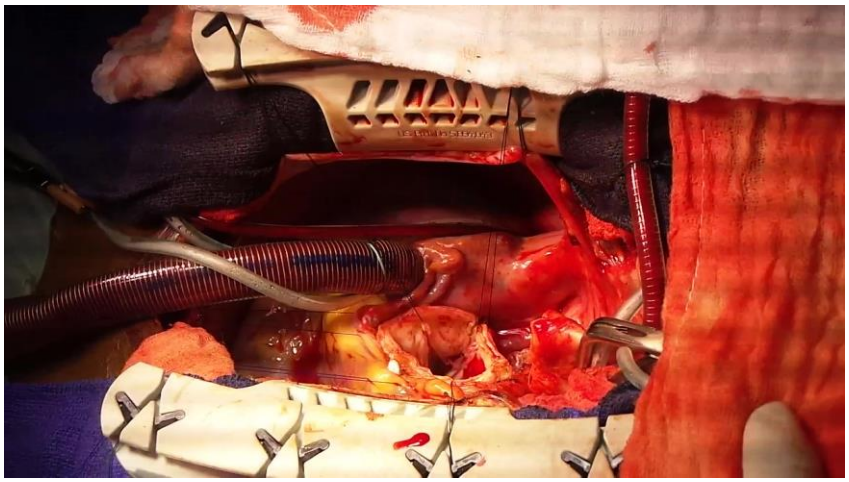

Figure 4. Intraoperative view of the resection of the entire supra-coronary ascending aorta.
Source: Author

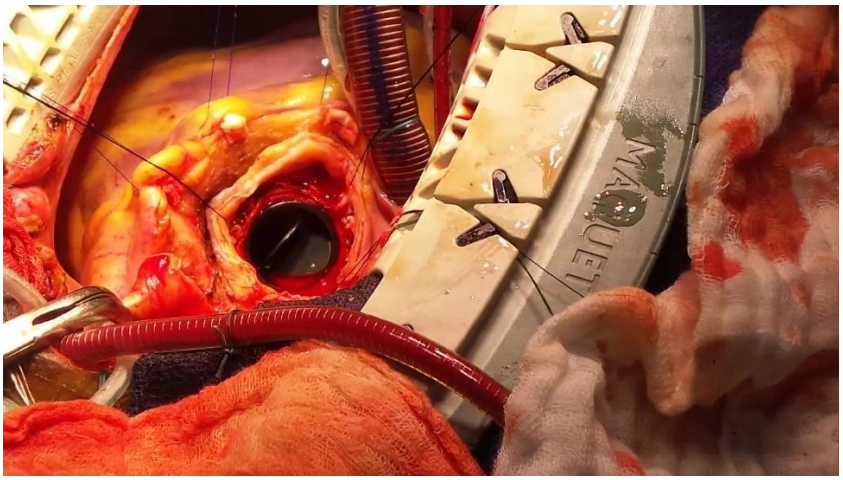

Figure 5. Intraoperative view of the implanted aortic valve prosthesis.
Source: Author

phase is the consequence of arterial lesions (stenosis, obliteration and aneurysm) (Kerr et al., 1994; Keser et al., 2018). Diagnosis is difficult in the absence of a laboratory test, and is therefore based on imaging. Echo-Doppler, angiography and nuclear magnetic resonance imaging are reliable methods of evaluating the lumen and the vessel wall.

The most frequently used diagnostic pattern in clinical and epidemiologic studies of TAK is the American College of Rheumatology (ACR) Classification Criteria, which includes five clinical and one imaging criteria (Arend et al., 1990; Podgorska et al., 2019). A diagnosis of Takayasu arteritis according to the criteria of ACR from 1990, might be made if a patient has at least three of these six criteria present:

1. Age < 40 years at disease onset
2. Claudication of extremities
3. Decreasese brachial artery pulse
4. Diference of>10 mmHg in systolic blood pressure between arms.
5. Bruit audible on auscultation over one or both subclavian arteries or abdominal aorta.
6. Pathological changes (usually focal or segmental) in aortic arteriography and/or its branches (Arend et al., 1990).

According to the criteria the diagnosis of Takayasu's arteritis was evident in the patient Cardiac involvement was found in 30 to 40% of cases and is considered one of the criteria for disease severity (Johnston et al., 2002).

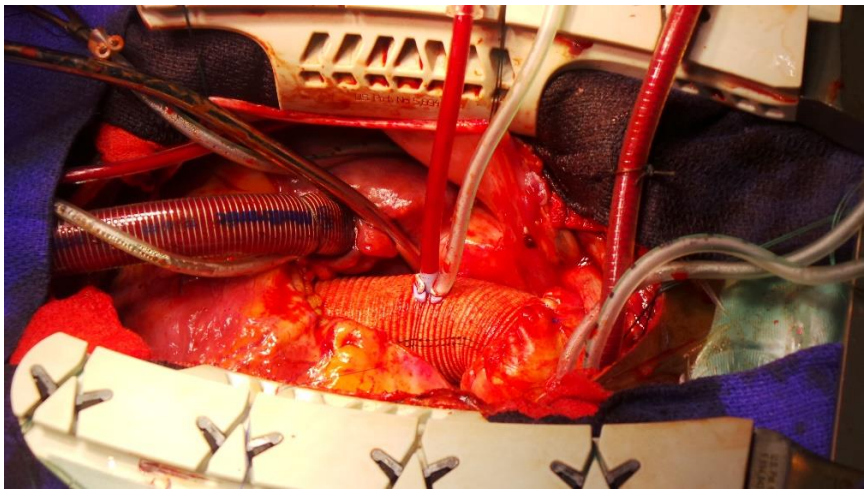

Figure 6. Restoration of vascular continuity using a prosthesis.
Source: Author

Aortic valve regurgitation has been reported as a complication in up to 25% of cases of Takayasu Arteritis. However, its association with ascending aortic aneurysm is a relatively rare complication of Takayasu Arteritis (Alali et al., 2017). In our patient, in view of the severity of the aortic regurgitation and the symptomatology, the indication for surgery was clear. However, in the presence of active inflammation decision for surgery was a difficult proposition. General recommendation is that surgery should be considered only after the initiation of medical treatment in order to reduce the inflammation to manageable levels (Ohigashi et al., 2017). Some complications may be observed postoperatively and require revision surgery (prosthetic disinsertions and pseudo aneurysms) due to tissue fragility and inflammation (Bouzerda and Khatouri, 2016). Corticosteroid therapy is the first-line treatment, in case of failure the addition of methothrexate would allow the control of the disease (Bouzerda and Khatouri, 2016; Keser et al., 2018; Sahin et al., 2019). In the patient, the medical treatment was only considered after the surgical intervention. However, pharmacological therapy is the predominant modality in 75.2% against 13.3% for surgical treatment (Field et al., 2017).

Conclusion

The incidence of Takayasu disease in black populations in sub-Saharan Africa appears to be underestimated in the literature demonstrating the importance of strengthening the health system for early diagnosis and training among physicians at the first point of care. Aortic valve replacement combined with ascending aorta replacement performed in an inflammatory aseptic setting has been successful. However, it requires monitoring to detect complications related to tissue fragility.

CONFLICT OF INTERESTS

The authors have not declared any conflict of interests.

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