Tuberculous aortitis

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Tuberculous aortitis (TA) is a rare entity that is invariably indicative of disseminated tuberculosis. TA is associated with aneurysm formation in about half of cases. Another possible complication is perforation of adjacent structures.[1-7] Both abdominal and thoracic aorta are involved with equal frequency.[7-9] Fatal outcomes are frequently reported even after antituberculosis chemotherapy and surgical intervention. We present a case of tuberculous aortic aneurysm (TBAA) that underwent surgical resection and graft replacement in the bed of the infected aorta. Following an apparently successful chemotherapy, the patient died suddenly. We postulate that reactivation of the un-eradicated bacilli precipitated graft failure. Similar cases in the literature are reviewed. We propose lifelong suppressive therapy with antituberculosis agents to prevent such a catastrophic event.

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Case

A 63-year-old Saudi man, known to be hypertensive, was referred from the local hospital for further management of a markedly dilated aortic arch. He had occasional chest pain, shortness of breath, and a dry cough with no hemoptysis. He reported a history of fever, night sweats, anorexia, and a weight loss of 8 kg over 4 months. There was no history of tuberculosis before, or contact with, patients with tuberculosis. A few months earlier, he was told that he had an aortic aneurysm, but had declined surgery. His temperature was 37.4˚C, blood pressure 120/80 mm Hg, respiratory rate 20/min, and his pulse rate was 100/min. His body weight was 65 kg. He had no lymphadenopathy or signs of heart failure. Laboratory results showed a WBC of 5.0 x 10^9/L, hemoglobin 140 gm/L, and platelets of 123 x 10^9/L. Urinalysis and renal, hepatic, lipid, and coagulation profiles were normal. Chest x-ray revealed a huge aneurysm of the aortic arch bulging into the left lung. The descending aorta had mild-to-moderate ectasia and tortuosity. There was bilateral pleural effusion and the cardiac size was normal. Echocardiography showed a dilated aortic arch at the junction of the descending thoracic aorta and pericardial effusion. Computed tomography of the chest confirmed the huge focal dilatation of the aortic arch measuring 9 centimeters with mural thrombosis, pericardial effusion and a few calcified and enlarged mediastinal lymph nodes. Serology for human immunodeficiency virus and syphilis was non-reactive. The patient underwent surgical resection of the aneurysm and interposition of a tube graft through a posterolateral thoracotomy approach. Intra-operatively, gross examination of the aorta revealed a huge aneurysm involving the arch as well as the proximal part of the descending aorta, another aneurysm in the middle of the descending thoracic aorta, a thickened pericardium and pleural nodules. The thoracic aneurysm was replaced from the arch down to the descending aorta. Histopathology from tissues of the aortic aneurysm, pericardium, and lymph nodes showed a granulomatous disease with central necrosis suggestive of tuberculosis. All smears were negative for acid-fast bacilli. Antituberculosis chemotherapy was started on

Table 1. Tuberculous aortitis involving the aortic arch.

Author	Year	Number of patients	Age (years)	Clinical presentation and diagnosis	Type of aortitis	Type of surgery	Outcome
Scully[12]	1975	1	63	Hemoptysis, miliary tuberculosis	Isolated aortic arch, true mycotic aneurysm	Resection	Died after 2 days
Efermidis[7]	1976	1	60	Fever, weight loss, mediastinal lymphadenopathy	Isolated aortic arch, true mycotic aneurysm	Resection*	Alive and good after 2 months follow up. No recurrence
Felson[6]	1977	3	73 59 63	2 miliary TB, 1 hemoptysis, pulmonary cavity	Isolated aortic arch, true mycotic aneurysm	2 cases graft replacement* 1 surgical resection*	Two died. First, 2 days after surgery, of cardiac arrest. Second, 3 weeks later, of fatal hemoptysis. Third, had miliary TB and graft replacement. Lived with no recurrence
Current case		1	63	Chest pain, pleuro-pericardium, disseminated TB	Aortic arch, true mycotic aneurysm and descending thoracic aorta	Resection* and graft replacement	Died, recurred ruptured aneurysm 13 months after stopping anti-TB

*Patients received anti-TB therapy prior to or after surgery

the first postoperative day in the form of isoniazid, rifampin, pyrazinamide, ethambutol and vitamin B6. Specimens from the tissue of the aortic aneurysm, the pericardium, the lymph nodes, the tracheal aspirates, and pleural tissue grew *Mycobacterium tuberculosis* sensitive to isoniazid, rifampin, pyrazinamide, and ethambutol. The subsequent course was notable for excellent tolerance and adherence to antituberculosis agents. Pyrazinamide and ethambutol were discontinued after the first two months. The patient reported general improvement in his well-being, he gained weight, and had resolution of radiological findings on subsequent imaging. Repeat computed tomography of the chest and echocardiography revealed no aortic dilatation. Isoniazid and rifampin were continued for a total of 18 months. Three months after discontinuing isoniazid and rifampin, the patient was feeling well and gained 3 further kilograms. Ten months later, the patient had sudden onset chest pain and collapsed. He had very low blood pressure in a local hospital. Immediate computed tomography of the chest showed hemothorax. The patient died soon thereafter. No post-mortem was performed.

Discussion

Before the antibiotic era, the majority of TA were diagnosed incidentally at autopsy.[10] With increasing clinical awareness, improved imaging and successful therapy early diagnosis has increased.[8] Common clinical presentations include the constitutional symptoms and signs of tuberculosis with or without an aneurysmal mass effect, depending on the location. Tuberculous aortic aneurysm usually manifests as a pulsatile or palpable mass, chest pain, dysphagia, hoarsenees, abdominal pain, back pain, and if complicated, by a fistula, perforations, bleeding, and rupture. Our patient presented with a 4-month history of tuberculous symptoms of fever, night sweats, and weight loss. The chest pain and dry cough were indications for chest radiographic imaging, which led to recognition of the aortic aneurysm at the local hospital. Aneurysm with fever and clinical suspicion of tuberculosis should raise the suspicion of TA.[3,7,10] We think the most likely mechanism of TA was direct extension from a nearby focus of infected lymph nodes; less likely is direct implantation on the inner vessel wall and hematogenous spread to the adventitia by the vasa vasarum.[7-9,11] Tuberculosis was not suspected in our case, preoperatively. The working diagnosis was an aortic aneurysm planned for surgical intervention. We have histopathological and microbiological confirmation of the involvement of *M. tuberculosis* with the aneurysm. The disease was disseminated, involving all mediastinal tissues and tracheal secretions.

TA usually involves the distal junction of the aortic arch along with the descending aorta, which is ad-

jacent to the mediastinal structures. Involvement of the ascending aorta is rarely reported. In a few cases, involvement of the aortic arch was reported.[6, 7, 12, 13] Table 1 summarizes aortic arch tuberculous aortitis cases previously reported. There are about 14 cases of successful surgical resection of the tuberculous aortic aneurysm of the descending thoracic aorta.[2,14] In the English literature, less than 50 cases of tuberculous aortic aneurysm were reported between 1945 and 2003.[8,10,11,14-17]

The duration of chemotherapy for disseminated tuberculosis is usually one year. For this patient, treatment was extended for 18 months because of concerns of graft infection. The extent of *M. tuberculosis* involvement in the aneurysm was so large that the newly inserted graft was basically placed where *M. tuberculosis* bacilli were abundant. However, because of a lack of previously published experience on duration of therapy, the susceptibility data of the isolate, and the excellent response to therapy, the 18-month course was considered sufficient. We postulate that the sudden death of the patient indicates probable rupture of the aortic aneurysm. The finding of hemothorax on computed tomography of the chest is supportive. This could have happened after reactivation of *M. tuberculosis* bacilli and dehiscence of the graft. Graft infection and its attendant complications may appear postoperatively or may be the cause of the recurrence at the operative site even with prolonged chemotherapy.[3,9] Without autopsy, the final events could never be confirmed. We would, therefore, consider lifelong suppression therapy with isoniazid after completing the treatment course. The goal is to prevent further bacillary growth at the non-viable graft material with the hope of prolonging its survival and functionality.

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