A Rare but Potentially Lethal Case of Tuberculous Aortic Aneurysm Presenting with Repeated Attacks of Abdominal Pain

Yao-Min Hung, Yun-Te Chang, Jyh-Seng Wang, Paul Yung-Pou Wang and Shue-Ren Wann

Abstract

Tuberculous aortic aneurysm is an extremely rare disease with a high mortality rate. The clinical features of this condition are highly variable, ranging from asymptomatic with or without constitutional symptoms, abdominal pain to frank rupture, bleeding and shock. We herein report the case of a 56-year-old man with a large tuberculous mycotic aneurysm in the abdominal aorta with an initial presentation of repeated attacks of abdominal pain lasting for several months. Due to the vague nature of the initial symptoms, tuberculous aortic aneurysms may take several months to diagnose. This case highlights the importance of having a high index of suspicion and providing timely surgery for this rare but potentially lethal disease.

Key words: abdominal aortic aneurysm, abdominal pain, aneurysm, aorta, mycotic aneurysm, tuberculosis

Introduction

Tuberculosis (TB) remains a major global health problem. This disease causes ill-health in millions of people each year and ranks as the second leading cause of death from infectious disease. The latest estimates from the World Health Organization include 8.6 million new TB cases and 1.3 million TB deaths worldwide in 2012 (1). TB may have a wide range of clinical presentations due to its disseminative properties, with both contiguous and hematogenous routes. Aortic involvement secondary to TB is exceedingly rare and may occur in the form of arteritis or aneurysm formation (2, 3). Whether by direct extension or via the blood stream, a focus of aortitis becomes established, and the aortic wall experiences destruction (2). Symptomatic tuberculous mycotic aneurysm is an extremely rare but uniformly fatal lesion if not diagnosed promptly (3, 4). Abdominal pain is a common chief complaint among patients who visit the emergency department (ED). We herein report the case of a 56-year-old man who presented to the ED with repeated attacks of abdominal pain. Following a preoperative evaluation, including clinical and radiological assessments, he underwent successful repair of a mycotic aneurysm in the abdominal aorta with excision and replacement of the diseased aorta. A postoperative biopsy showed a tuberculous aortic aneurysm, and he was started on antituberculous chemotherapy based on the histological findings.

Case Report

A 56-year-old man was brought to our ED with abdominal pain lasting for 10 days. He had a history of hypertension under medication control. According to his statement, he had no previous tuberculous exposure. He also denied any possibility of relevant household contact, eating habits or ownership of unusual pets, such as reptiles. He had visited several clinics and local hospitals for help within the past two to three months, receiving various diagnoses. He had also been treated by general practitioners, but never had
a complete workup. His vital signs were as follows: blood pressure= 168/98 mmHg, pulse rate= 75 beats per minute, respirations= 16 per minute and body temperature= 37.5°C. A physical examination of the chest was normal, and the abdomen was soft and mildly tender diffusely with no evidence of rebound or guarding. A rectal examination revealed hard stools, but no masses or hemorrhoids. The laboratory data obtained on admission are shown in Table; only increased an C-reactive protein (CRP) level was found. A routine urinalysis was negative, and an electrocardiogram showed a normal sinus rhythm at 87 beats/min. Chest and abdomen roentgenograms showed no abnormalities; however, abdominal computed tomography (CT) revealed a 6-cm mycotic aneurysm involving the abdominal aorta, infrarenal type (Fig. 1, 2). As typical organisms, such as the Salmonella species or Staphylococcus aureus, were suspected at the time, the patient received empirical intravenous antibiotics and was admitted to the intensive care unit. Therefore, the patient underwent surgery, and a biopsy showed features of chronic granulomatous inflammation and focal caseous necrosis, consistent with TB infection, which confirmed the diagnosis (Fig. 3, 4). He was thus started on a four-drug antituberculous regimen (isoniazid, rifampin, ethambutol, pyrazinamide), and the aneurysm cultures eventually grew mycobacterium TB resistant to isoniazid. He has since been followed up regularly at our outpatient department. Further CT scans showed negative findings.

**Discussion**

Aneurysms are a very rare complication of TB. With respect to extrapulmonary TB, tuberculous aortic aneurysm formation is considered a rare form of TB. The largest series review from 1945 to 1999 found only a total of 39 cases in the published literature (3), and Kamen first reported this devastating complication of TB in 1858 (5). Volini et al. later described the pathophysiology of aortic TB, in which a
as the differential diagnosis also included several kinds of nonspecific antibiotics and was admitted to the intensive care unit, surgery. Our patient initially also received empirical intravenous diagnoses should be considered and ruled out before surgery. In particular, the differential diagnosis includes various infected mycotic aneurysms that should be ruled out before surgery. In addition, the diagnosis of tuberculous aortitis is very difficult to establish, as the disorder is exceedingly rare and can mimic Takayasu arteritis (11). For these reasons, the diagnosis may be delayed and the patients may be treated with immunosuppressive therapy before the diagnosis of TB is established. Therefore, the possibility of tuberculous aortitis should be considered in patients with aortitis or atypical aortic aneurysms who have a history of pulmonary or extrapulmonary TB and/or chronic immunosuppression (8). In the current case, a definitive diagnosis was not obtained before surgery, and infection with typical organisms, such as Salmonella species or Staphylococcus aureus, was favored based on our previous experience. In addition, in a past case report, the rapid progression of bacterial aortitis to an ascending aortic mycotic aneurysm was documented on transesophageal echocardiography within six days (12).

Managing tuberculous aortic aneurysms is challenging, and the standard therapeutic strategy has not yet been established (13). Open aortic reconstructive surgery is generally considered to be the standard of treatment for aortic aneurysms associated with aortitis, although endovascular techniques have recently been employed, with early reported successes (13-15). While endovascular treatment has the theoretical advantage of avoiding extensive manipulation of inflamed aortic tissue, thus preventing the morbidity and mortality associated with open surgery, there are downsides in patients with TB-infected aneurysms. In such cases, despite placement of the stent graft at the site of primary infection, prospective complications include persistent bacteremia, reinfection, delayed rupture, paraplegia, distal emboli and surgical conversion. Some authors maintain that endovascular stenting is feasible if antibiotic suppression results in negative blood cultures before the intervention (16). Hence, the use of endovascular repair remains controversial (13), and there have been no head-to-head trials of the optimal strategy for managing aortic aneurysms in patients with aortitis.

Infectious aortitis is a rare clinical entity most often associated with abdominal aortic aneurysms. However, very few cases of tuberculous aortic aneurysm have been reported; to our knowledge, there are only 75 cases in the English lan-

Figure 4. Histopathology of the aortic aneurysm showing positive staining for acid-fast bacilli (Hematoxylin and Eosin staining, ×1,000).
guage literature on PubMed from 1945 to 2011 (13). Another point deserving attention is there are two extremes in the disease presentation: initial nonaneurysmal aortitis and mycotic aneurysms (17). It is unclear how long patients develop tuberculous aortic aneurysms after the initial onset of possible nonaneurysmal aortitis. Nevertheless, the small number of cases of nonaneurysmal infectious aortitis reported in the literature and rarity of the disease in patients with TB can be attributed to underdiagnosis and delayed diagnosis.

It is very important to establish an early diagnosis of infectious aortitis using adequate image studies in patients presenting with unexplained fever and repeated attacks of abdominal pain. The pathophysiology of this disease suggests that infectious aortitis and mycotic aneurysms represent extremes along the spectrum of the same disease. Although establishing a diagnosis of aortic infection before aneurysm formation or rupture is very difficult, it is essential for preventing devastating complications.

The authors state that they have no Conflict of Interest (COI).

References

1. WHO. Tuberculosis (TB). Geneva; World Health Organization, 2013. GLOBAL TUBERCULOSIS REPORT 2013. [Internet]. [cited 2014 Mar 18]. Available from: https://www.who.int/tb/publications/global_report/gbr12_main.pdf
2. Volini FJ, Olfield RC Jr, Thompson JR, Kent G. Tuberculosis of the aorta. JAMA 181: 78-83, 1962.
3. Long R, Guzman R, Greenberg H, Safneck J, Hershfield E. Tuberculous mycotic aneurysm of the aorta. Chest 115: 522-531, 1999.
4. Kolhari VB, Bhairappa S, Prasad NM, Manjunath CN. Tuberculosis: still an enigma. Presenting as mycotic aneurysm of aorta. BMJ Case Rep (in press).
5. Kamen L. Aortenruptur auf tuberculouer Grundlage. Beitr Pathol Anat 17: 416-419, 1895.
6. Volini FJ, Olfield RC Jr, Thompson JR, Kent G. Tuberculosis of the aorta. JAMA 181: 78-83, 1962.
7. Vaglio A, Buzio C. Chronic periaortitis: a spectrum of diseases. Curr Opin Rheumatol 17: 34-40, 2005.
8. Gornik HL, Creager MA. Aortitis. Circulation 117: 3039-3051, 2008.
9. Vaglio A, Salvarani C, Buzio C. Retroperitoneal fibrosis. Lancet 367: 241-251, 2006.
10. Scheel PJ Jr, Feeley N. Retroperitoneal fibrosis: the clinical, laboratory, and radiographic presentation. Medicine (Baltimore) 88: 202-207, 2009.
11. Gajaraj A, Victor S. Tuberculous aortoarteritis. Clin Radiol 32: 461-466, 1981.
12. Wein M, Bartel T, Kabatnik M, Sadony V, Dirsch O, Erbel R. Rapid progression of bacterial aortitis to an ascending aortic mycotic aneurysm documented by transesophageal echocardiography. J Am Soc Echocardiogr 14: 646-649, 2001.
13. Wang Y, Zhang J, Yin MD, Wang SY, Duan QZ, Xin SJ. Endovascular repair of a tuberculous aneurysm of descending thoracic aorta. Chin Med J (Engl) 124: 2228-2230, 2011.
14. Labrousse L, Montaudon M, Le Guayader A, Chourkroun E, Laurent F, Deville C. Endovascular treatment of a tuberculous infected aneurysm of the descending thoracic aorta: a word of caution. J Vasc Surg 46: 786-788, 2007.
15. Liu WC, Kwak BK, Kim KN, et al. Tuberculous aneurysm of the abdominal aorta: endovascular repair using stent grafts in two cases. Korean J Radiol 1: 215-218, 2000.
16. Celia R, Colin B, Ravul J, Nicholas C, Mohamad H. Endovascular stenting of peripheral infected aneurysms: a temporary measure or a definitive solution in high-risk patients. Cardiovasc Intervent Radiol 31: 1228-1235, 2008.
17. Lopes RJ, Almeida J, Dias PJ, Pinho P, Maciel MJ. Infectious thoracic aortitis: a literature review. Clin Cardiol 32: 488-490, 2009.

© 2015 The Japanese Society of Internal Medicine
http://www.naika.or.jp/imonline/index.html