Case Report

Successful *In Situ* Reconstruction with a Prosthetic Graft in Tuberculous Pseudoaneurysm of Abdominal Aorta: Two Case Reports

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ABSTRACT

Tuberculous pseudoaneurysm (TP) of the abdominal aorta is an exceedingly rare and life-threatening disease. Here, we report two patients treated with a combination of *in situ* reconstruction with a prosthetic graft and antitubercular therapy. The first case was a 51-year-old man with an infrarenal abdominal aortic pseudoaneurysm; the second case was a 56-year-old man with an infrarenal abdominal aortic pseudoaneurysm and a paraspinal abscess at three months after endovascular stent-graft repair for abdominal aortic dissection. Both patients had a history of tuberculosis (TB) and presented with lumbar or abdominal pain. The extensive debridement of infected tissues and *in situ* reconstruction with a prosthetic graft were performed with laparotomy. Pathological examination of the periaortic and aortic wall revealed chronic inflammation with TB infection. Therefore, *in situ* reconstruction with a prosthetic graft and antitubercular therapy offer favorable results in TP of the abdominal aorta.

KEYWORDS: aorta abdominal, blood vessel prosthesis, pseudoaneurysm, tuberculosis

INTRODUCTION

Tubercular involvement of aortic wall is a rare phenomenon. With worldwide resurgence of tuberculosis due to an increasing incidence of drug-resistant tuberculosis and its association with acquired immunocompromised condition[1,2]. The incidence of tuberculous pseudoaneurysm (TP) has arisen as a significant clinical entity[3]. Symptomatic TP becomes a fatal lesion, if not diagnosed and treated promptly. This report describes two cases of successful *in situ* reconstruction with a prosthetic graft of TP of the infrarenal aorta secondary to TB. The pathogenesis, clinical features and management of TP are also reviewed.

CASE REPORT

Patient 1

A 51-year-old man presented with constant lower gastric pain radiated to the right shoulder and back lasting for one month. He did not complain of nausea, vomiting, diarrhea, or fever. He was diagnosed as a case of pulmonary TB seven months ago and received antituberculous therapy. A purulent lump was found in his left epididymis and it was surgically removed two week ago. Physical examination revealed a pulsatile mass located at the right quadrant of the abdomen reaching the level of navel with an obscure boundary, limited degree of excursion, and non-tender to palpation. Enhanced computerized tomography (CT) scan demonstrated dilation of the abdominal aortic lumen and chronic inflammatory infiltration of the vessel wall (Fig. 1a). CT angiography exhibited a saccular pseudoaneurysm measuring 8 x 6 cm in the infrarenal abdominal aorta without involving bilateral renal artery and bilateral iliac artery (Fig. 1b). An obvious laceration was observed at the lateral wall of the infrarenal abdominal aorta. The patient was diagnosed as a case of TP of the infrarenal abdominal aorta due to the history of TB and typical finding of CT scan.

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The symptom of persistent pain was believed to be the sign of impending rupture. A laparotomy was performed which confirmed the existence of a TP arising from the lateral wall of the infrarenal abdominal aorta. Once aneurysmal wall was opened, massive cheesy necrotic tissue and light coffee-colored purulent liquid poured out of the aneurysmal sac. Lymphatic vessels were found to be markedly dilated in the aortic wall, and chyle flowed out of dilated vessels intraoperatively. The observed red and white thrombi were attached to the intimal surface of the involved aortic wall. The maximal diameter of expanded aorta was approximately 3 cm, and the aortic wall around the ventage was hardened and brittle. After extensive debridment of the periaortic necrotic tissue and removal of the diseased segment of aorta, the operating field was washed repeatedly with 1.0 g of streptomycin and 0.2 g of Armazide diluted in 500 ml of saline. With macroscopically disease-free margin, *in situ* aortic reconstruction was performed by means of a prosthetic graft. The pedicle wall flap was used to cover the anastomosis site. Drainage tubes were inserted into the lower abdominal incision sites. The pathological examination (performed by the Department of Pathology, Changhai Hospital, Second Military Medical University, Shanghai, China) of the periaortic tissue and aortic wall revealed a combination of acute and chronic inflammation with multinucleated giant cell reaction. The patient received antitubercular drugs with isoniazid, rifampicin, and ethambutol on the first day following the operation. The patient's postoperative course was uneventful, and he was discharged at week two after admission. At the 6-month follow-up, the patient remained well and asymptomatic; CT angiography revealed that the periangiitis around the abdominal aorta completely

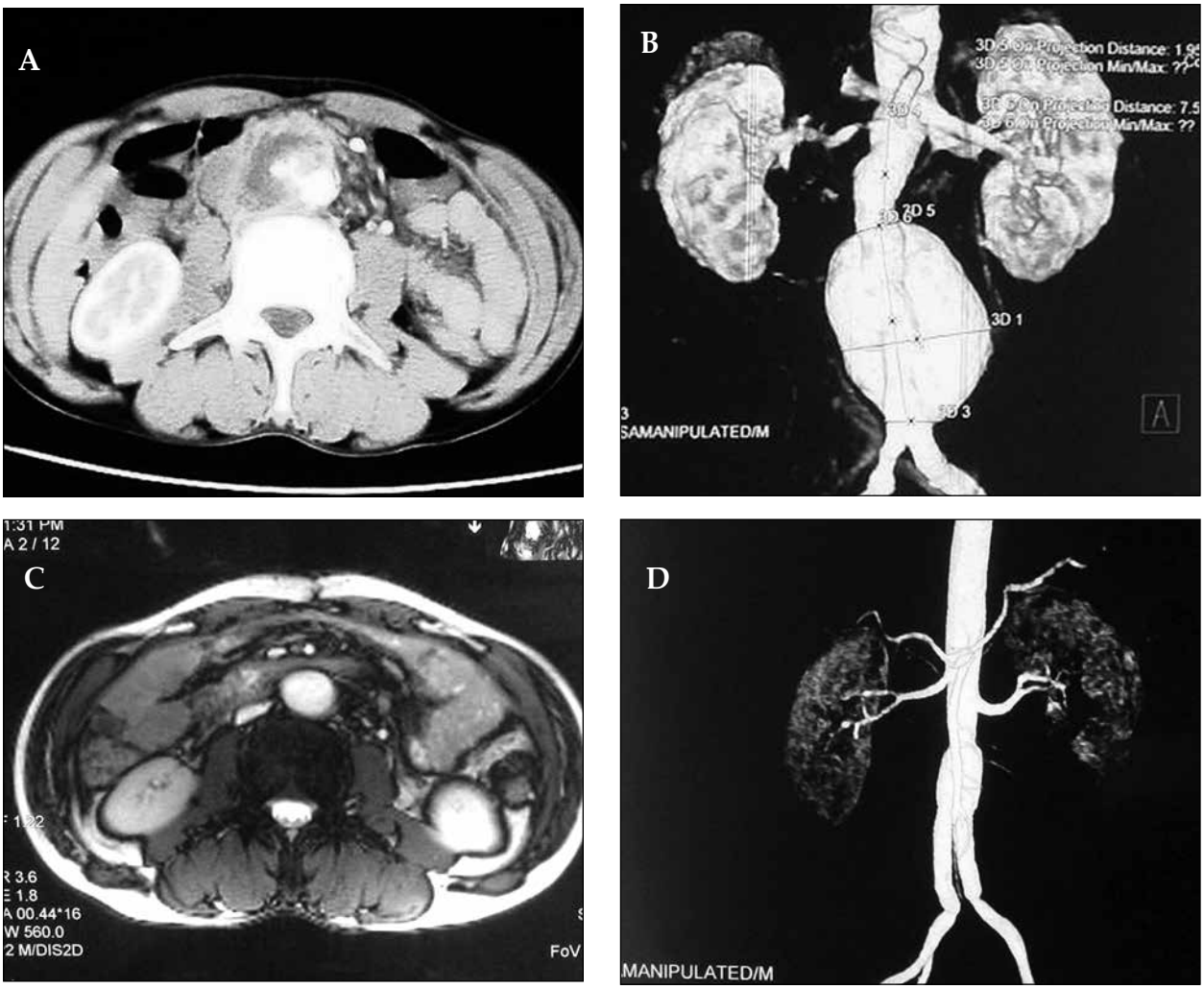

Fig. 1: Tuberculous false aneurysm of infrarenal abdominal aorta in a 51-year-old male. (a) Enhanced CT scan demonstrating the dilatation of abdominal aorta and inflammatory thickening of the vessel wall. (b) CT angiography showed a saccular false aneurysm measuring 8 x 6 cm in the infrarenal abdominal aorta without involving bilateral renal artery and bilateral iliac artery. (c) Postoperative CT scan exhibited smooth abdominal aortic wall and complete disappearance of periangiitis. (d) Postoperative CT angiography showed the normal profile of abdominal aorta.

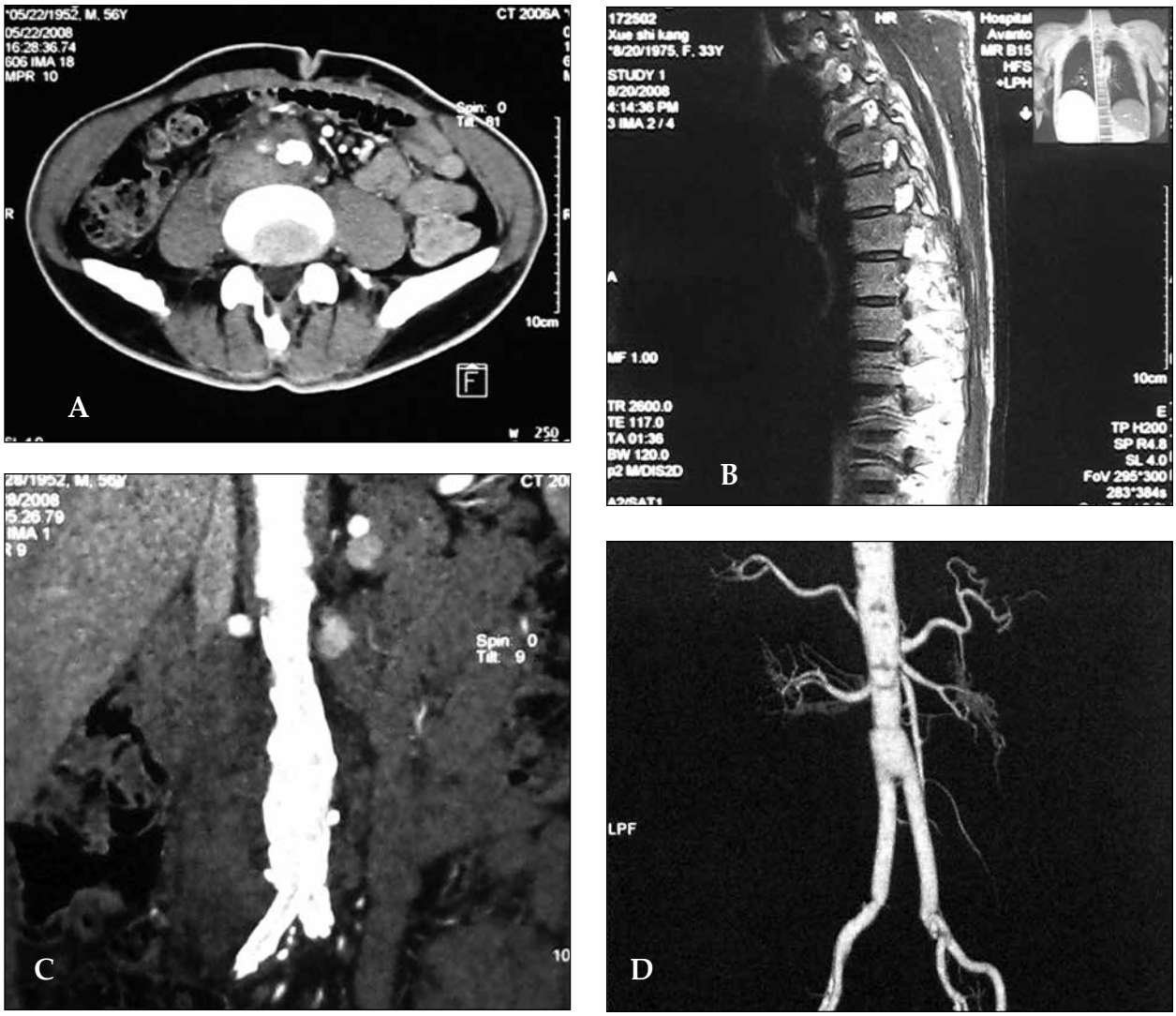

Fig. 2: Tuberculous false aneurysm of infrarenal abdominal aorta after endovascular repair for abdominal aortic dissection in a 56-year-old male. (a) The enhanced CT scan showed a huge heterogeneous enhanced mass about 25 x 20 cm in size in the right lower quadrant. (b) Magnetic resonance imaging showed a neighboring paraspinal abscess. (c) CT angiography showing a saccular false aneurysm of the infrarenal aorta without involving the bilateral iliac arteries. (d) Postoperative CT angiography demonstrated the continuity of the abdominal aorta.

disappeared and the vessel wall became smooth (Fig. 1c). A normal profile of abdominal aorta was simultaneously observed on enhanced CT scan (Fig. 1d).

Patient 2

A 56-year-old man presented with an abdominal mass persisting for 10 days at three months after endovascular stent-graft repair for pseudoaneurysm of the abdominal aortic dissection. He had been complaining of waist and back pain and intermittent fever since the stent-graft implantation. However, a pulsatile mass was detected in the right lower quadrant of the abdomen at ten day before admission, and was associated with persistent gastric pain in this region. Six days prior to admission, the mass increased markedly in size and the pain worsened. The patient developed sweating all over his body, his face turned pale, and his blood pressure dropped significantly at the same time. CT scan at the local hospital showed a large saccular mass in the right lower quadrant. A CT scan done two days later revealed that the mass had dilated significantly. The patient received an emergency transfer to our hospital due to a suspected impending rupture of the aneurysm. On admission, the patient's vital signs were stable (blood pressure: 105 / 76 mmHg, pulse rate: 106 beats/min, respiration rate: 24 breaths/min, body temperature: 36.4 °C). Physical examination revealed an increased tactile fremitus and a dull percussion sound in the right lower lung. The rest of physical examination revealed a pulsatile mass extending to the right waist and back with clear

boundary and tenderness; nevertheless, no vascular murmur was heard. Enhanced CT scan demonstrated a huge heterogeneous enhanced mass about 25 x 20 cm in size in the right lower quadrant (Fig. 2a). Magnetic resonance imaging demonstrated erosion of the anterior aspect of the fourth lumbar vertebral body on the left side with a neighboring paraspinal abscess (Fig. 2b). CT angiography demonstrated a saccular pseudoaneurysm of the infrarenal aorta without involvement of the bilateral iliac arteries (Fig. 2c).

The patient underwent an emergency operation of the TP of the abdominal aorta, ablation and vascular prosthesis reconstruction. Intraoperative observation included a few hematoceles in the abdominal cavity and retroperitoneal hemorrhage on the right side of the middle and inferior abdomen and the right iliac fossa. The right greater psoas muscle and the vertebral body were found to be eroded and destroyed, with the fractured bone being sharp. The retroperitoneal hematoma and the old organizing blood clot in the cavity was cleaned with total volume of approximately 2000 ml of normal saline. After the aortic wall was opened, the membrane of stent-graft could be seen as split, with a crevasse of about 1 cm. The periaortic necrotic tissue and bone chips were debrided. The operating field was washed repeatedly with iodophors saline and chlorhexidine. The abdominal aneurysm resection was performed with *in situ* placement of a prosthetic graft. The resected periaortic necrotic tissue was sent for pathological analysis. Analysis of the tissue sample showed florid necrotizing granulomas. The abdominal cavity was fitted with routine drainage tubes. The patient was treated with antitubercular drugs immediately after operation. The postoperative recovery was uneventful, and he was discharged two weeks later. Postoperative CT angiography was suggestive of the continuity of the abdominal aorta (Fig. 2d).

DISCUSSION

A TP of the abdominal aorta is exceedingly rare. Until recently, only two such cases could be traced in the Chinese-language literature. Han *et al*[4] reported that a patient suffered from multiple TP of abdominal aorta, and died of rupture of smaller abdominal aortic aneurysm and gastrointestinal hemorrhage after endovascular stent-graft exclusion. Zhao *et al*[5] outlined a case in which abdominal aortic aneurysm ablation and vascular prosthesis reconstruction were performed with exploratory laparotomy. In this report, we present our experience with two such patients and a brief review of pathogenesis, clinical features and management.

The two cases had the history of TB. Both complained of paroxysmal and radiating waist or abdominal pain. Other accompanying symptoms included intermittent low-grade fever and abdominal distension; nevertheless, neither presented with the manifestation of intestinal tract ischemia. Physical examination revealed a large abdominal pulsatile mass in one case, and an easily detectable non-pulsatile mass in the other. The mass pulsatility depended largely on the foundation of the pseudoaneurysm and communication between saccular mass and aorta. The final diagnosis was acquired based upon the history of TB, typical symptoms, and signs on CT or MRI. The two cases underwent combination of *in situ* reconstruction with a prosthetic graft and antitubercular therapy, and were well and asymptomatic at six-month follow-ups.

The majority of abdominal aortic pseudoaneurysms are due to trauma, infection[6], iatrogenic injury, and arteriosclerosis[7]. Aortic TP is extremely rare with a high mortality rate. Typical manifestation includes evidence of tuberculous focus or disseminated tuberculosis with one or more of the three clinical scenarios: 1) fever and persistent abdominal or back pain, 2) hypovolemic shock or other evidence of major bleeding, or 3) palpable or radiographically-visible periaortic mass, especially if expanding or pulsatile[8]. The *Mycobaterium tuberculosis* may encroach on the aortic wall in one of three ways. First, the bacilli may attach directly to the internal surface of the vessel wall. Normally, the aortic intima is very resistant to infection; however, when this protection is weakened by atherosclerotic plaque or aortic ulcers, the resistance to infection is depressed and the intimal surface may become colonized by blood-stream bacilli. Second, the bacilli may be carried to the adventitia or media by the *vasa vasorum*. Third, the bacilli are most commonly spread to the aorta by direct extension (or indirectly *via* the lymphatic system) from a contiguous focus, such as lymph nodes, paraspinal or posas abscess, vertebrae, and prostate[9]. The two cases presented herein were attributed to the extension of tuberculous infection in lymphatic system and the direct erosion from lumbar spinal TB. In fact, we postulated that all three infection routes may be implicated into the pathogenesis of TP of abdominal aorta. Caseating necrosis occurring in the entire layers of the aortic wall results in perforation, either with massive hemorrhage or with the formation of a perivascular hematoma. The latter may become encapsulated and retain communication with the lumen, in which case it is referred to as a pseudoaneurysm.

Early surgical operation in combination with perioperative antituberculous therapy has been demonstrated to offer favorable results for patient survival[10]. If the abdominal aortic aneurysm ruptures and results in hypotension, the success rate may be lower than 50%. Especially when the *arteriae aorta* is severely destroyed by TB, the rupture speed may be fairly fast[11]. Consequently, if a patient with an aortic

TP develops symptoms of persistent abdominal or back pain, the surgical operation should be performed urgently[12]. It needs to be noted here that the diameter of tuberculous false aneurysm is not a critical determinative factor for operation necessity[13]. It is also not necessary to wait for enough antitubercular drugs, which would delay the opportunity to operate, since the curative effects of the antitubercular drugs on TB in the wall of TP and mural thrombosis are fairly limited[14].

The treatment for TP is *in situ* reconstruction using a prosthetic graft[15], extra-anatomic bypass re-establishment[16], and endovascular stent-graft repair[17]. Controlling tuberculous infection and keeping distal aorta unobstructed are the most important therapeutic principles in surgical operation. The debridement of tuberculous focus and necrotic tissue and extra-anatomic bypass, such as the axillary – femoral artery bypass and avoidance of direct contact with tuberculous infection are commonly recommended[16]. However, these procedures are known to provide a lower patency rate than *in situ* reconstruction. Despite the likelihood of prosthetic graft infection by tubercle bacillus, in our cases the *in situ* reconstruction did not carry a risk of infection as evidenced at follow-up; this is likely to be due to the patient having been provided adequate antitubercular drugs. In situ reconstruction depends on the size of aortic aneurysm and the condition of the neighboring aorta. After ablation of aortic aneurysm, the cutting edges of aorta and neighboring aortic wall should be evaluated. The surgeon should rely on visual inspection of the aortic wall to decide on the extent of resection, rather than on frozen sections and histological examination. Endovascular stent-graft repair of tuberculous aortic aneurysms has been reported in three cases with limited follow-up[17]. Endovascular repair does not allow extensive debridement of the infected periaortic tissues, and thus could be associated with a high risk of infection and aneurysm recurrence, as occurred in one of the two cases in our report. Endovascular repair should be more suitable for patients with advanced age and poor health status, but long-term efficacy of this technique has not yet been established.

CONCLUSION

TP of the abdominal aorta carries a high risk of impending rupture. Once the diagnosis is made, operation should be performed urgently, even in the presence of a small pseudoaneurysm. A combination of *in situ* reconstruction with a prosthetic graft and antitubercular therapy offers favourable results for the treatment of TP of abdominal aorta.

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