

Endovascular treatment of a ruptured thoracic aortic pseudoaneurysm secondary to Pott disease during a spine surgery

A case report and a literature review

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Abstract

Rationale: The coexistence of a tuberculous aortic pseudoaneurysm and Pott disease in patients with a history of tuberculosis (TB) is relatively rare, and the treatment strategies remain still controversial.

Patient concerns: A 57-year-old female patient with a history of primary pulmonary TB presented with symptoms of breathlessness, chest pain, weight loss, and fever. Magnetic resonance imaging (MRI) and computed tomography (CT) showed a thoracic aortic pseudoaneurysm secondary to Pott disease at T11/12 level.

Diagnoses: Tuberculous pseudoaneurysm at the descending thoracic aorta associated with tuberculous vertebral osteomyelitis.

Interventions: We originally planned a combined surgery consisting of posterior spine stabilization, anterior excision of the infected field, and aortic reconstruction. When we surgically stabilized the posterior spine, unexpectedly, the pseudoaneurysm ruptured. Immediately, we terminated the surgery and appropriately placed an endovascular stent graft, which successfully rescued the patient.

Outcomes: When the patient's conditions were stable, we anteriorly debrided all infected tissues and then performed a spinal fusion by grafting autologous iliac bone. After the debridement and spinal fusion, we arranged a 1-year anti-tuberculous chemotherapy for this patient and performed a 24-month follow-up. This patient had no signs of recurrent infection during the follow-up.

Lessons: For the patients with tuberculous aortic aneurysm(s) complicated with vertebral osteomyelitis, the endovascular repair of an aneurysm(s) should be considered a conventional therapy before the spine surgery, lowering the risk of aortic aneurysm rupture. Meanwhile, minimally invasive endovascular stent graft combined with anti-tuberculosis drugs may be considered one of the therapeutic regimens for the patients whose conditions are not suitable for open surgery, such as age, weakness, or severe organ failure.

Abbreviations: ADA = adenosine deaminase, CRP = C-reactive protein, CT = computed tomography, ESR = erythrocyte sedimentation rate, HIV = human immunodeficiency virus, L = lumbar vertebra, LDH = lactate dehydrogenase, MRI = magnetic resonance imaging, T = thoracic vertebra, TAA = tuberculous aortic aneurysms, TB = tuberculosis.

Keywords: case report, endovascular repair, Pott disease, thoracic aorta, tuberculous pseudoaneurysm

1. Introduction

Tuberculous spondylitis (also known as Pott disease), one of the complications of tuberculosis (TB), is tuberculous arthritis of the vertebral body and intervertebral joints. The disease was first described by Percival Pott in 1779,^[1] and its morbidity is

approximately 1% among TB patients.^[2,3] This disease commonly occurs in immunocompromised patients, dystrophia persons, and especially, elderly patients. Recently, TB has resurged in Asia and Africa, which caused an increased incidence of Pott disease.^[4] However, it is clinically uncommon that an

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aneurysm coexists with Pott disease at the aorta near to the intervertebral joints, which greatly increases the risk of a ruptured of the tuberculous aortic aneurysm (TAA) during surgery. Indeed, 3 TAA ruptures during surgery have been reported.^[5–7]

In this report, we describe a Pott disease case of a TAA rupture at the thoracic aorta when posteriorly stabilizing the spine. This patient was successfully treated by placing an endovascular stent graft. Here, we described the therapy procedures. Along with reported Pott disease cases, we propose some treatment principles for Pott disease patients.

2. Case report

The patient consents to this case report being published. This study was approved by the ethical committee of the Affiliated Hospital of Kunming University of Science and Technology, Kunming (Ethical approval number: 7131095).

A 57-year-old woman with primary pulmonary TB presented to the department of internal medicine with symptoms of breathlessness, chest pain, weight loss, and fever. A computed tomography (CT) scan showed vertebrae lesions with a paravertebral abscess in the T11/12 vertebrae (Fig. 1A and B). The patient was transferred to the department of orthopedic surgery for further clinical examinations.

This patient exhibited slightly raised C-reactive protein levels (CRP: 10.1 mg/L) and erythrocyte sedimentation rate (ESR: 43 mm/h), positive for tubercle bacillus antibody and interferon

gamma release, but negative for anti-treponemal antibodies and human immunodeficiency virus (HIV) serology. White cell count, renal and liver function, and electrolytes were normal. Tuberculin skin test was strongly positive, and sputum cultures for acid-fast bacilli were negative. Chest x-ray showed a widened mediastinum and a left pleural effusion consistent with pathological features of pulmonary TB (Fig. 1C). Hydrothorax biochemistry analysis showed elevated total cell count ($3.15 \times 10^9/L$), predominant monocytes, raised levels of lactate dehydrogenase (LDH: 334 U/L) and adenosine deaminase (ADA: 98 U/L), and positive in rivalta test. The patient received a standard anti-tuberculosis chemotherapy treatment (po QD: Isoniazid, 300 mg; rifampicin, 450 mg; pyrazinamide, 1500 mg; and ethambutol, 750 mg). A contrast-enhanced CT scan demonstrated a left paravertebral abscess, osteomyelitis, and a $2.0 \times 2.5 \text{ cm}^2$ pseudoaneurysm of the suprarenal aorta at the T11 level (Fig. 1D). Magnetic resonance imaging (MRI) further confirmed the presence of a saccular dilatation of the posterior thoracic aorta surrounded by a paravertebral abscess (Fig. 2A and B).

Our team planned to perform a combined surgery, that is, the combination of a posterior vertebra stabilization surgery that stabilizes the spine and an anterior open-chest surgery that extensively excises infected tissues and pseudoaneurysm, aortic reconstruction, and intervertebral fusion (autologous iliac bone graft). According to this plan, we first performed a vertebra stabilization surgery by placing a double-screw and double-rod system on T9 to L2 vertebra. When the posterior vertebra

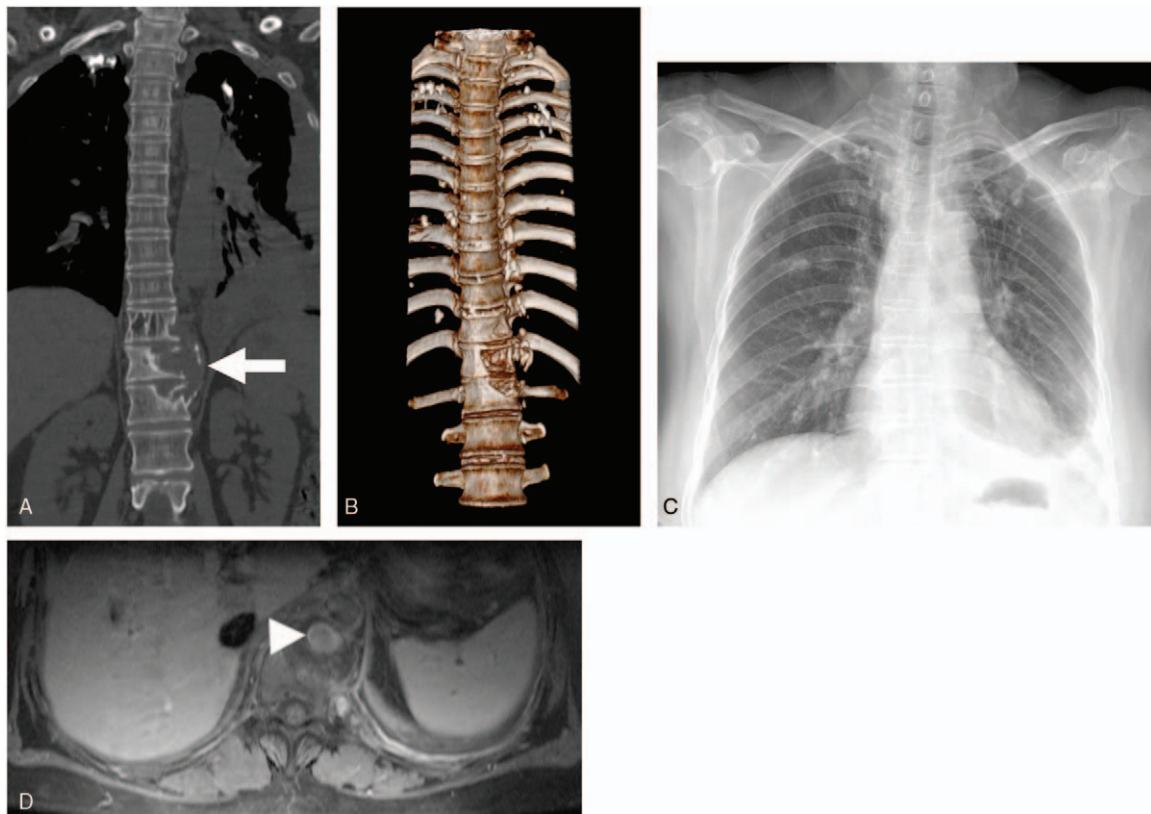


Figure 1. A, Coronal CT scan shows left-side vertebral destruction with a paravertebral abscess (arrow). B, Three-dimensional CT scan shows the vertebral destruction in the 11th to 12th thoracic vertebrae. C, Anteroposterior radiograph of the chest shows a widened mediastinum and left pleural effusion. D, Axial enhanced CT scan shows that a tuberculous pseudoaneurysm (arrow-head) of the descending thoracic aorta was surrounded by a paravertebral abscess. CT = computed tomography.

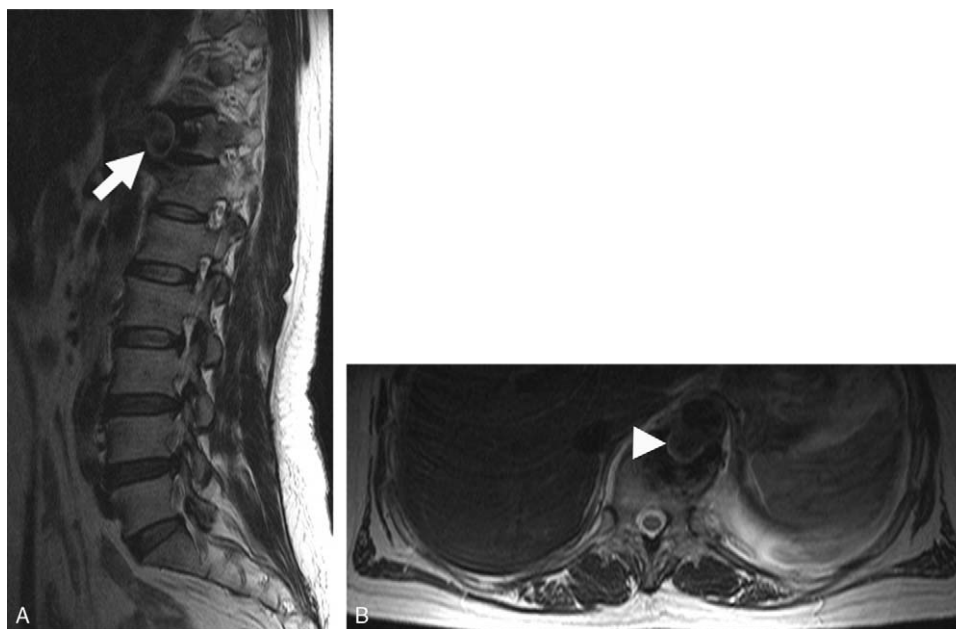


Figure 2. A, Sagittal MRI scan indicates the vertebral erosion with a paravertebral abscess in the 11th to 12th thoracic vertebrae (arrow). B, Axial MRI scan shows a descending thoracic aortic saccular pseudoaneurysm (arrow-head). MRI=magnetic resonance imaging.

stabilization was nearly finished, the patient progressively lost normal hemodynamics (heart rate increased to 150 beats/min, and blood pressure dropped to 85/50 mmHg), which was accompanied by a remarkable decline of hemoglobin from preoperative 12.1 to 4.1 g/dL, implying, tuberculous pseudoaneurysm rupture. Immediately, we administered 8 erythrocyte concentrates to this patient, and performed a thoracoabdominal angiography. We observed a pseudoaneurysm rupture on the lateral wall of the descending thoracic aorta at the T11 level (Fig. 3A). Therefore, we performed an endovascular treatment by positioning an Ankura thoracic stent graft (28 mm at diameter; 160 mm at length; Lifetech, Shenzhen, China) without covering the orifice of the bilateral renal arteries. Arteriography showed complete blockage of the orifice of pseudoaneurysm rupture with no residual blood flow to the pseudoaneurysm sac (Fig. 3B). The patient was then transferred to the intensive care unit of our hospital, and we suspended the anterior open-chest surgery.

Five days later, we sequentially performed a debridement of all infected tissues, a partial corpectomy of the T11 and T12 vertebrae, a meticulous removal of the necrotic tissues in the spinal canal, and, followed by an intervertebral fusion. Histopathological observations of the resected lesions exhibited typical caseous necrosis and chronic granulomatous inflammation with marked accumulation of epithelioid cells and multinucleated giant cells (Fig. 4A and B). The patient recovered rapidly and was discharged 18 days after the surgery. The patient was then treated with anti-tuberculous chemotherapy for 1 year, and we arranged a 24-month follow-up. During this follow-up, the patient had no signs of recurrent infection and showed a fixed vertebra and precisely placed thoracic stent on imaging (Fig. 3C and D).

3. Discussion

In patients with TB, the coexistence of TAA and tuberculous spondylitis are relatively rare. Since Long et al^[8] reviewed

advances in tuberculous mycotic aneurysm of the aorta in 1999, only 23 patients with tuberculous pseudoaneurysm were reported, of whom 22 had pseudoaneurysm secondary to tuberculous spondylitis.^[5-7,9-20] We reviewed these case reports and found only 3 patients with a TAA rupture during the surgery (Table 1).^[5-7] Together with the present case, a total of only 4 cases have been reported.

Clinically, once the complications of TAA and Pott disease are diagnosed, treatments should start immediately. The current regimen to treat TAA complicated with vertebral erosion is the combination of anti-tuberculosis chemotherapy and anterior open-chest surgery.^[15,21] This therapeutic strategy aims to stabilize the spine, extensively debride infected tissues, reconstruct aorta in situ, and perform intervertebral fusion. The anterior surgery of anterior spine stabilization and intervertebral fusion has been considered as the “gold standard” procedure, but this surgery has potentially lethal risk of a disseminated TB infection due to the use of a metallic implant (titanium mesh, screw, and rod) in the infected field. Therefore, it is necessary to optimize anterior and posterior spine stabilization and fusion, avoiding lethal risks during the therapy.^[22,23]

The case in this report showed the combination of TAA with marked tuberculous spondylitis according to our initial diagnosis. We, therefore, planned to perform the combination of posterior spine stabilization and, anterior open-chest debridement surgery. Unexpectedly, during our posterior spine stabilization surgery, TAA ruptured and caused emergency conditions of hemodynamic instability. We immediately placed a thoracic stent graft and eventually completed whole combination surgery. However, this combination surgery did not include aortic resection and reconstruction. To reduce surgical trauma, we abandoned the original surgery plan and chose to leave the thoracic stent in place on the following anterior surgery. During the 24-month follow-up, no recurrent infection occurred, no fixation loosening was found, and complete spinal interbody fusion was obtained.

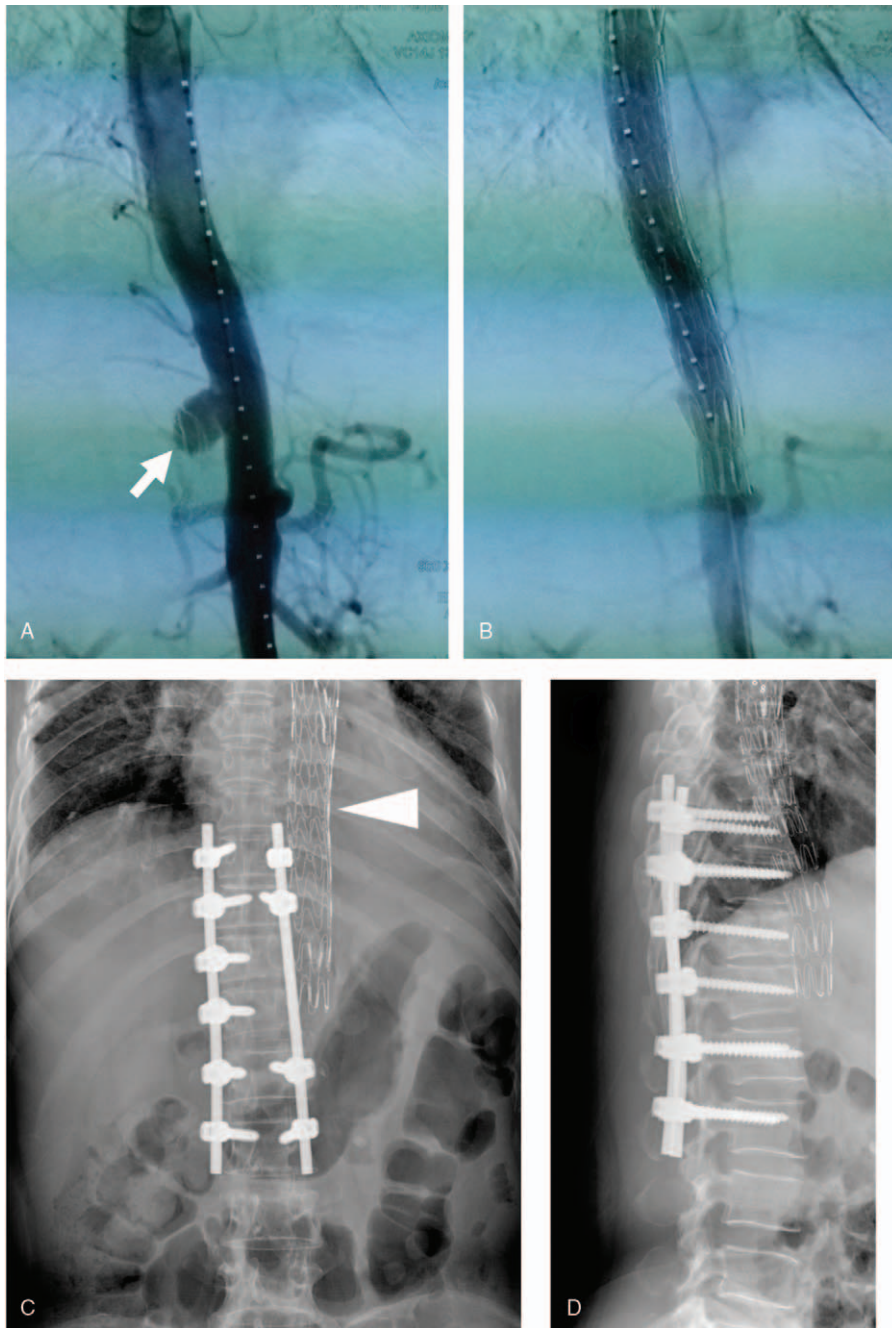


Figure 3. A, Thoracoabdominal angiography shows a ruptured tuberculous pseudoaneurysm at the descending thoracic aorta at the T11/12 level (arrow). B, Thoracoabdominal angiography shows that the pseudoaneurysm was successfully repaired by an endovascular stent graft placement. X-ray images show internal fixation of the spine and implanted endovascular stent (arrow-head) 24 months after surgery. C, Anteroposterior view. D, Lateral view.

In this case, one possible reason for tuberculous pseudoaneurysm rupture is that posterior spine stabilization surgery might somehow squeeze aortic aneurysms because TAA and tuberculous spondylitis are adjacent. In addition, the rupture of aortic aneurysms often unpredictably happens even when TAA is only 10 mm in diameter.^[24] In the situation with risk of the rupture of aortic aneurysms, it is, therefore, reasonable to repair TAA before spine surgery avoiding TAA rupture. At present, a less invasive endovascular repair device has been considered attractive in

repairing TAA.^[25,26] However, this endovascular repair does not allow extensive excision of necrotic tissues, which could restrict anti-tuberculosis medication efficiency.^[9,27] The minimal invasive endovascular therapy may be more efficient before spine surgery. Thus, the endovascular repair in combination with posterior spine stabilization, anterior open-chest debridement, and autologous iliac bone graft may represent a new promising strategy for the patients with TAA complicated with tuberculous spondylitis.

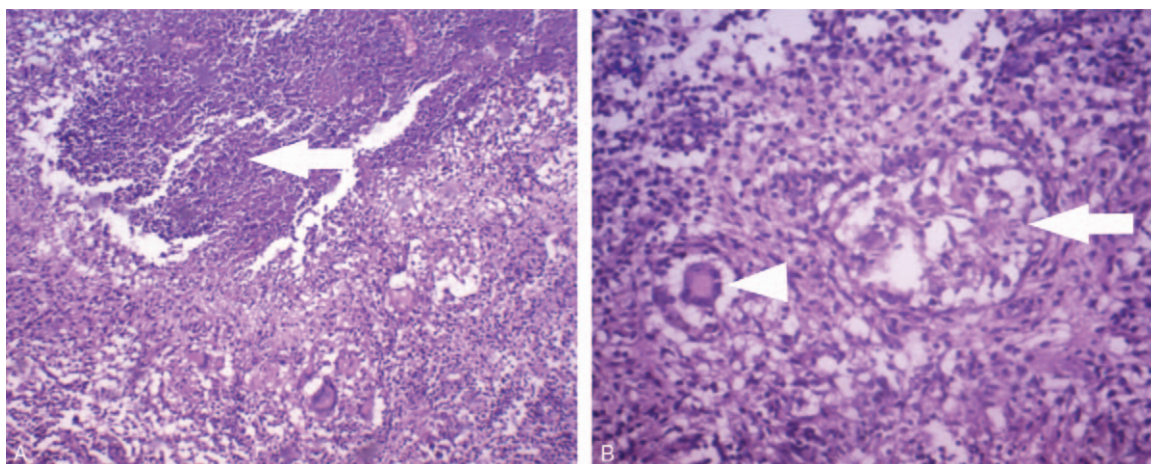


Figure 4. A, Hematoxylin & eosin (H&E) staining of the resected tissues shows typical caseous necrosis (arrow) consistent with the pathological features of spinal TB (original magnification, $\times 40$). B, Photomicrograph shows accumulation of epithelioid cells (arrow) and multinucleated giant cells (arrow-head) (H&E stain; original magnification, $\times 100$). TB=tuberculosis.

Table 1

Review of literature about a ruptured TAA secondary to Pott disease during surgery.

Patient no	First author	Year	Age/gender	Location (artery)/size, cm	Location (vertebrae)	Surgical procedure	Outcome
1	Liu WC ^[5]	2000	41/M	Abdominal aorta/3 \times 4 \times 5 cm	L1–L2	A psoas abscess drainage	Endovascular repair by stent graft
2	Xue J ^[6]	2018	33/F	Thoracic aorta/1.9 cm tear	T8–T9	Thoracic spinal surgery	Death
3	Pluemvitayaporn T ^[7]	2018	79/M	Abdominal aorta	L2–L3	Thoracolumbar vertebra surgery	Endovascular stent graft implantation

F=Female, L=lumbar vertebra, M=Male, T=thoracic vertebra, TAA=tuberculous aortic aneurysms.

4. Conclusion

The patients with Pott disease complicated with TAA may be at high risk of the TAA rupture during spine surgery. Therefore, the repairment or stabilization of TAA is necessary before spine surgery, even TAA may be repaired first. For the patients with TAA complicated with severe vertebral erosion, endovascular stent grafting should be considered as a conventional therapy before spine surgery or a vital part of the surgical procedures.

Author contributions

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