Case Report

**Tuberculous abdominal aortic pseudoaneurysm with renal and vertebral tuberculosis: a case and literature review**

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**Abstract**

Tuberculous pseudoaneurysm of the aorta is rare and exposes patients to a very high risk of unpredictable rupture. To our best knowledge, only 32 cases have been reported related to all arterial systems from 1993 to 2013 in the literature. We report a 44-year-old male who presented with an aortic pseudoaneurysm and tuberculosis of the kidney and vertebrae. He underwent endovascular repair and antibiotic therapy for tuberculosis, combined with a bare stent implanted to seal endoleaks after endograft stenting. The postoperative course was uneventful and the patient recovered and lived well afterwards. Epidemiology, pathogenesis, presentation, management, and mortality of this entity were reviewed and discussed.

**Key words:** pseudoaneurysm; tuberculosis; infection; treatment.

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**Case Report**

A 44-year-old man who was admitted for gross hematuria and lower back pain with radiation down to both legs for six months, without accompanying fever, night sweat, or weight loss. He was not a smoker and had no history of angina, hypertension, dyslipidemia, ischemic stroke, or chronic renal failure. Vertebral body biopsy revealed acid-fast bacilli with bone destruction and granulomatous formation (Figure 1), and there was no bacteria by general bacterial culture of pus with aerobic and anaerobic conditions. Urine culture was positive for tuberculosis without other bacteria. These findings well supported the diagnosis of tuberculosis of the lumbar vertebrae and right kidney. Ultrasound showed a 39×38 mm low-echo area of the right kidney, and a 22×15 mm nodule adjacent to abdominal aorta (Figure 2A, 2B). A computed tomography angiography (CTA) scan simultaneously revealed a right renal abscess (Figure 2C) and a pseudoaneurysm sized 20×12×20 mm, which involved the posterior wall of the infrarenal abdominal aorta (Figure 2D-F). In addition, a left psoas abscess (Figure 2D) with vertebral bodies L3-L5 deteriorated (Figure 2F) was found.

After a six-month anti-tuberculosis treatment with isoniazide (0.3g per day), rifampin (0.45g per day), ethambutol (0.75g per day), and pyrazinamide (0.5g per day), the case was referred to us for digital angiographic evaluation, which revealed a saccular pseudoaneurysm of the infrarenal abdominal aorta. Therefore, endovascular abdominal aortic aneurysm repair (EVAR) was performed by implanting a straight (80 mm long and 20 mm diameter) endograft (Endurant Stent Graft, Medtronic, CA, USA), but endoleaks of the left posterior were observed after implantation. Another 80 mm long and 16 mm diameter bare stent (Sinus-XL Stent, Optimed, Ettlingen, Germany) was implanted and the endoleaks were sealed.

The patient was discharged on day 5 following interventional therapy and was instructed to take anti-tuberculosis therapy with isoniazide, rifampin, ethambutol, and pyrazinamide at the same dose for 12 months; he would afterwards require an operation for posterior stabilization of the lumbar vertebrae. On the six-month follow-up, the patient was without lumbar pain or any complication. CTA scan follow-up with contrast was performed at six months (Figure 3A-C), displaying a marked reduction of the pseudoaneurysm.
Figure 1. Pathological changes in lesion. A: Acid-fast bacilli (thick arrow) were detected in the vertebral body from the granulomatous lesion (acid-fast stain, original magnification 100×); B: Histopathological slide of vertebral body revealed bone destruction (thin arrow), and caseation necrosis (thick arrows); C: Multiple cell granulomas in the lymph node. From center out: pink section – necrosis of initially formed (thick arrow); white section – epithelioid cells (thin arrow); purple section – lymphocyte (arrow head) (hematoxyline-eosin stain, original magnification 20×).

Figure 2. Imaging before treatment. A, B: Abdomen ultrasound showed two low-echo areas of abdominal aorta (thick arrow) and the right renal (thin arrow); C: Abdominal and pelvic CTA revealed a low-density area measuring 24×30×25 mm involving the right kidney (thin arrow); D, E, F: Three-dimensional reconstruction CTA revealed a pseudoaneurysm measuring 20×12×20 mm involving the posterior wall of the infrarenal abdominal aorta (thick arrow) and a low-density area of the left psoas (thin arrow) with deteriorated vertebrae bodies (arrow head).

Figure 3. Imaging after treatment. A, B, C: Abdominal and pelvic CTA obtained six months after the initial CT examination, displaying both endo-graft and stent in the original position and marked reduction of the pseudoaneurysm.
Table 1. Review of literature about tuberculous pseudoaneurysm and treatment

<table>
<thead>
<tr>
<th>Year</th>
<th>Study</th>
<th>Age (years)/Gender</th>
<th>Location (artery)</th>
<th>Follow-up (months)</th>
<th>Anti-TB treatment (months)</th>
<th>Pseudoaneurysm therapy</th>
<th>Comments</th>
</tr>
</thead>
<tbody>
<tr>
<td>1995</td>
<td>Cross et al. [6]</td>
<td>31/Male</td>
<td>Carotid A</td>
<td>2M</td>
<td>-</td>
<td>-</td>
<td>Endovascular stenting</td>
</tr>
<tr>
<td>1996</td>
<td>Hagino et al. [7]</td>
<td>80/Male</td>
<td>Ab AA</td>
<td>6M</td>
<td>RIE/10M</td>
<td>Extra-anatomic bypass</td>
<td>Prostate cancer with radiotherapy</td>
</tr>
<tr>
<td>1996</td>
<td>Ikezawa et al. [8]</td>
<td>47/Female</td>
<td>Th AA</td>
<td>11M</td>
<td>RIES/7M; RE/4M</td>
<td>In-situ reconstruction</td>
<td>Pu TB</td>
</tr>
<tr>
<td>1998</td>
<td>Bojar et al. [9]</td>
<td>34/Male</td>
<td>Th AA</td>
<td>3M</td>
<td>RIEP/1M</td>
<td>In-situ reconstruction</td>
<td>Pu TB, HIV history</td>
</tr>
<tr>
<td>1999</td>
<td>Long et al. [10]</td>
<td>67/Male</td>
<td>Ab AA</td>
<td>-</td>
<td>RIPE/nearby 2M</td>
<td>In-situ reconstruction</td>
<td>-</td>
</tr>
<tr>
<td>1999</td>
<td>Goizarian et al. [11]</td>
<td>27/Female</td>
<td>Th AA</td>
<td>10M</td>
<td>RIP/10M</td>
<td>In-situ reconstruction</td>
<td>-</td>
</tr>
<tr>
<td>1999</td>
<td>Kao et al. [12]</td>
<td>88/Male</td>
<td>Femoral A</td>
<td>18M</td>
<td>RIE/12M</td>
<td>In-situ reconstruction</td>
<td>Ruptured femoral aneurysm</td>
</tr>
<tr>
<td>2000</td>
<td>Liu et al. [13]</td>
<td>42/Female</td>
<td>Ab AA</td>
<td>24M</td>
<td>RIP/2M; Anti-TB/7M</td>
<td>EVAR</td>
<td>-</td>
</tr>
<tr>
<td>2000</td>
<td>Deshmukhet al. [14]</td>
<td>12/Male</td>
<td>Gluteal A</td>
<td>8M</td>
<td>-</td>
<td>EVAR</td>
<td>Ruptured aneurysm</td>
</tr>
<tr>
<td>2001</td>
<td>Choudhary et al. [15]</td>
<td>5 patients</td>
<td>Th/Ab AA</td>
<td>18-36M</td>
<td>RIPE/2M and (or) RI/4M</td>
<td>In-situ reconstruction</td>
<td>Pu TB, pericardium TB</td>
</tr>
<tr>
<td>2004</td>
<td>Satokawa et al. [16]</td>
<td>77/Female</td>
<td>Celiac A</td>
<td>-</td>
<td>RI, LVFX/12M</td>
<td>Extra-anatomic bypass</td>
<td>Recurrence after reconstruction</td>
</tr>
<tr>
<td>2005</td>
<td>Kim et al. [17]</td>
<td>25/Male</td>
<td>Splenic A</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>Gastric TB, Pu TB</td>
</tr>
<tr>
<td>2005</td>
<td>Shikata et al. [18]</td>
<td>76/Male</td>
<td>Ab AA</td>
<td>6M</td>
<td>RIPE/6M</td>
<td>In-situ reconstruction</td>
<td>Pu TB</td>
</tr>
<tr>
<td>2006</td>
<td>Corby et al. [19]</td>
<td>12/Female</td>
<td>Mesenteric A</td>
<td>9M</td>
<td>RIPE/9M</td>
<td>Endo-embolization, gastrectomy</td>
<td>Gastric TB, Pu TB</td>
</tr>
<tr>
<td>2006</td>
<td>Sirvanciet al. [20]</td>
<td>65/Male</td>
<td>Th AA</td>
<td>-</td>
<td>Anti-TB/-</td>
<td>In-situ reconstruction</td>
<td>Recurrence after reconstruction</td>
</tr>
<tr>
<td>2006</td>
<td>Bavunghuet al. [21]</td>
<td>16/Male</td>
<td>Mesenteric A</td>
<td>Died</td>
<td>-</td>
<td>Endovascular embolization</td>
<td>Intestinal TB</td>
</tr>
<tr>
<td>2006</td>
<td>Leceseet al. [22]</td>
<td>69/Male</td>
<td>Femoral A</td>
<td>18M</td>
<td>RIP/12M</td>
<td>In-situ reconstruction</td>
<td>Pu TB, cutaneous TB</td>
</tr>
<tr>
<td>2007</td>
<td>Lohet al. [23]</td>
<td>63/Male</td>
<td>Th AA</td>
<td>6M</td>
<td>RIPE/6M</td>
<td>TEVAR</td>
<td>Stroke after EVAR</td>
</tr>
<tr>
<td>2007</td>
<td>Labrousset al. [24]</td>
<td>68/Male</td>
<td>Th AA</td>
<td>12M</td>
<td>RIPE/2M; RIPE/2M; RI/10M</td>
<td>TEVAR</td>
<td>Pu TB, renal cancer with nephrectomy</td>
</tr>
<tr>
<td>2008</td>
<td>Keeling et al. [25]</td>
<td>40/Male</td>
<td>Pu A</td>
<td>Died</td>
<td>RIP/0.5M</td>
<td>Endovascular embolization</td>
<td>Pu TB, Rasmussen aneurysm, cardiac arrest</td>
</tr>
<tr>
<td>2008</td>
<td>Stephen et al. [26]</td>
<td>2 patients</td>
<td>Carotid A</td>
<td>6M</td>
<td>-</td>
<td>Anti-TB/-</td>
<td>Stenting/in-situ reconstruction</td>
</tr>
<tr>
<td>2010</td>
<td>Bachmeyer et al. [27]</td>
<td>38/Male</td>
<td>Iliiac A</td>
<td>1M</td>
<td>-</td>
<td>Anti-TB/6M</td>
<td>In-situ reconstruction</td>
</tr>
<tr>
<td>2012</td>
<td>Li et al. [28]</td>
<td>62/Male</td>
<td>Th AA</td>
<td>12M</td>
<td>RIPE/1M</td>
<td>RIPE/5M</td>
<td>TEVAR</td>
</tr>
<tr>
<td>2012</td>
<td>Nakayama et al. [29]</td>
<td>84/Male</td>
<td>Th AA</td>
<td>-</td>
<td>No anti-TB</td>
<td>No anti-TB</td>
<td>TEVAR</td>
</tr>
<tr>
<td>2013</td>
<td>Villegas et al. [30]</td>
<td>72/Male</td>
<td>Ab AA</td>
<td>36M</td>
<td>-</td>
<td>Anti-TB/12M</td>
<td>EVAR</td>
</tr>
</tbody>
</table>

Th AA: thoracic aortic artery; Ab AA: abdominal aortic artery; Pu: pulmonary; R: rifampicin; I: isoniazid; P: pyrizinamide; E: ethambutol; S: streptomycin; (T)EVAR: (thoracic) endovascular aortic repair
Discussion

The etiology of aortic and peripheral aneurysms is the most clinically relevant classification system related to not only natural history, but also treatment. The main cause of aneurysm formation is medial degeneration, found in 80% of cases; dissection, which causes the aneurysm to form in response to excessive proteolytic enzyme activity and the increase of smooth muscle migration that diminishes the integrity of the arterial wall, is found in 17% of cases; finally, 3% of cases are inflammatory aneurysms such as Takayasu disease and aortitis [1]. In addition, approximately less than 1% of all aortic aneurysms [2] are associated with arterial infection, which often results in the formation of pseudoaneurysms that are differentiated from true ones specifically because they lack all three normal elements of the arterial wall, which are more often saccular than fusiform. Staphylococcus and Salmonella [1] are the major causes of primary aortic infection; tuberculosis is very rarely described. The original cause of non-tuberculous aneurysms are mainly spinal surgery and trauma, whereas pseudoaneurysms of tuberculosis are induced by the constant stimulation of surrounding tissue with tuberculosis infection. In this study, we reviewed 32 cases describing tuberculous pseudoaneurysm from 1993 to 2013 in 25 studies [Table 1].

The earliest report of a tuberculous aortic aneurysm was by Kamen in 1895 [3]. Yet the first attempt to repair a tuberculous aortic aneurysm was made in 1949, by Herndon and colleagues [4]. The patient died of a massive gastrointestinal hemorrhage on the sixth day after surgery. In 1959, Prophit [5] reported a successful resection of such pseudoaneurysm in the thoracic aorta. But tuberculous pseudoaneurysms may occur anywhere along the arterial system via different infectious pathways of the arterial wall [6,7,10,12,16,17,19,25,27]. Different positions of tuberculous pseudoaneurysm have distinct names: tuberculous pulmonary pseudoaneurysm is called Rasmussen aneurysm [25], and aortic pseudoaneurysm is called Pott’s disease [28]. It is therefore of great importance to know the different approaches through which tubercle bacilli reach the arterial wall: (i) the bacilli may implant directly on the internal surface of the vessel wall; (ii) the bacilli may be carried to the adventitia or media by the vasa vasorum; (iii) infection may reach the vessel wall by the lymphatics of the vasa vasorum; or (iv) the outside of the vessel wall may be affected by direct extension from a neighboring tuberculous lymph node, abscess, or bone. This last approach has been reported in the literature as the most common cause (75%) of infection [10]. In our case, the patient had deteriorated vertebrae bodies (L3-L5) and a left psoas abscess. Biopsy revealed caseation necrosis, acid-fast bacilli in the vertebrae bodies and their surroundings, and a granulomatous formation in the lymph node. These finding are completely in accordance with infection by direct extension from a neighboring tuberculous lymph node, abscess, or bone. Interestingly, all evidence in our case supports tuberculosis as a distinctive etiology with aortic pseudoaneurysm.

Because a tuberculous aortic pseudoaneurysm is exposed to a very high risk of unpredictable rupture with serious hemodynamic consequences and mortality (40%) [10], surgical resection with perioperative anti-tubercular therapy offers the only chance for the patient’s survival. At present, surgery of choices include open surgery (in situ reconstruction [8-11] or extra-anatomic bypass[7,16]) and interventional therapy (embolization[14,17,21,25], stenting[26], or endovascular aneurysm repair [13,23,24,28-30]) along with anti-tuberculosis therapy. The most common option is resection of the infected arterial segment, debridement of the surrounding tissues, and revascularization of the lower extremities using grafts brought through uninfected tissues remote from the infected site (extra-anatomic bypass). However, treatment of such lesions involving the juxtarenal aorta or visceral aorta is much more complicated. In our case, the best options (aortic debridement or direct revascularization) were limited for open surgery because the lesion was located near the opening of renal arteries, and the patient needed secondary orthopedic surgery for vertebra fixation. In addition, a review of the literature showed that surgical mortality ranged from 14% to 20% [10,15], and that there was some surgical morbidity, such as recurrent tuberculous pseudoaneurysm [16] and risk of graft infection [20]. Therefore, open surgery was not appropriate for this case. Percutaneous endovascular interventional therapy offers shorter hospitalization and convalescence periods, and reduces the morbidity and mortality associated with open surgery [23-26,28-30]. There are other options to remove pseudoaneurysms than surgery, such as embolization, stenting or EVAR (TEVAR), or a combination of these treatments. But it must be assumed that the primary infection can be controlled with long-term antibiotics (pre-operation and post-operation) in these circumstances. In our case, we combined EVAR and stenting to avoid endoleaks; this resulted in no prosthesis infection during the six-month follow-up.
We also considered EVAR (TEVAR) a temporizing option to treat acute complications from infected aortic aneurysms, such as uncontrolled bleeding.

Despite the use of modern chemotherapy and imaging technology, this disastrous complication still occurs, which reinforces the need for early suspicion, diagnosis, surgical resection, and anti-tubercular therapy along with close postoperative follow-up to prevent recurrence. The entire body should be examined in order to make early diagnosis and to effectively treat this life-threatening infection.

References


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