Q fever (*Coxiella burnetii*) causing an infected thoracoabdominal aortic aneurysm

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We report a patient, which we believe is the first, with a thoracoabdominal aortic aneurysm, Crawford type IV, caused by Q fever (*Coxiella burnetii*). Treatment consisted of antibiotic therapy started preoperatively and continued postoperatively and an open repair, including resection of the infected aneurysm, replacement with a rifampin-soaked polyester graft, and an omental wrap covering the grafts. After 13 months of follow-up, the patient had no signs of infection, and results of laboratory findings were normal. (J Vasc Surg 2011;53:1402-4.)

An infected aortic aneurysm is a rare disease with an incidence of approximately 1%.[1] The most commonly involved organisms are *Staphylococcus* and *Salmonella*. However, approximately 25% of infected aneurysms remain without bacterial identification.[2] Reasons for this are various, including administrating antibiotics before cultures are taken, and infections with slow-growing or fastidious bacteria, or obligate intracellular bacteria, that are difficult to culture. One of these pathogens is *Coxiella burnetii*.

In patients with underlying vascular disease, such as an aortic aneurysm, *C burnetii* can cause chronic Q fever infection.[3,4] This is due to the affinity of *C burnetii* for cardiovascular tissues and its ability to survive within the phagolysosome after gaining entry into monocytes and macrophages that are present in the aortic thrombus.[5] To date, 26 patients have been described in the literature with *C burnetii* infection in combination with an aneurysm of the native aorta,[6-9] including two cases of a thoracoabdominal aortic aneurysm (TAAA) without confirmation of *C burnetii* DNA. In this article, we report what we believe is the first case of a patient with a TAAA caused by chronic Q fever, confirmed by molecular detection of *C burnetii* DNA in the aortic wall.

CASE REPORT

In September 2009, a 59-year-old man was admitted to a regional hospital for analysis of radiating abdominal pain and high fever (38.6° C). His relevant medical history revealed a coronary artery bypass graft in 1992 and endovascular repair of an infrarenal AAA in 2002.

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Laboratory tests showed an increased C-reactive protein of 60 mg/L (normal <5 mg/L), elevated erythrocyte sedimentation rage of 41 mm/h (normal <20 mm/h), and leukocyte level of 6.4×10^9/L (normal range, $4\text{-}10 \times 10^9$/L).

The initial computed tomography (CT) scan identified a saccular aneurysm of the TAAA, Crawford type IV, with a maximum diameter of 4.1 cm (Fig, *A*). Subsequent positron-emission tomography scanning showed an area of increased intensity of the supraceliac aneurysm, without increased intensity of the infrarenal endoprosthesis, suggestive for a mycotic aortic aneurysm. Because he lived in an area endemic for Q fever, serology for Q fever was performed and showed a chronic infection of *C burnetii* (immunofluorescent assay phase 1 immunoglobulin G (IgG) >1: 4086; phase 2 IgG >1: 4086). Antibiotic therapy was started, consisting of doxycycline (200 g/d and hydrochloroquine (200 mg orally, twice daily), and he was referred to our hospital for further treatment of the mycotic TAAA. Here, a repeat CT scan 1 month later showed a growing infected TAAA with a diameter of 6.0 cm (Fig, *B*).

In November 2009, an open repair of the TAAA type IV was performed, with replacement of a rifampin-soaked polyester graft. During the procedure, distal aortic perfusion was performed using cardiopulmonary bypass with a femoral artery canula and a femoral vein canula up to the right atrium. Motor-evoked potential recording was performed to evaluate spinal cord function during cross-clamping.[10]

Rigorous debridement was done of the infected aneurysmatic aorta, and samples were sent for pathologic as well as microbiologic examination. The left renal artery was reconstructed with an 8-mm graft, and the celiac trunk, superficial mesenteric artery, and right renal artery were anastomosed at the distal end of the main graft. An omental flap was harvested and wrapped around the thoracic graft. Pathologic examination of the aortic wall showed activity of infection, and a quantitative real-time polymerase chain reaction (rt-PCR) amplification detected DNA of *C burnetii* in the aortic wall tissue.

His postoperative course was uneventful and the intravenous antibiotic therapy with doxycycline (100 mg, twice daily) and rifampin (600 mg, twice daily) was continued for 5 weeks. After the definitive diagnosis, the antibiotics were switched to oral doxycycline (100 mg, twice daily) and oral hydroxychloro-

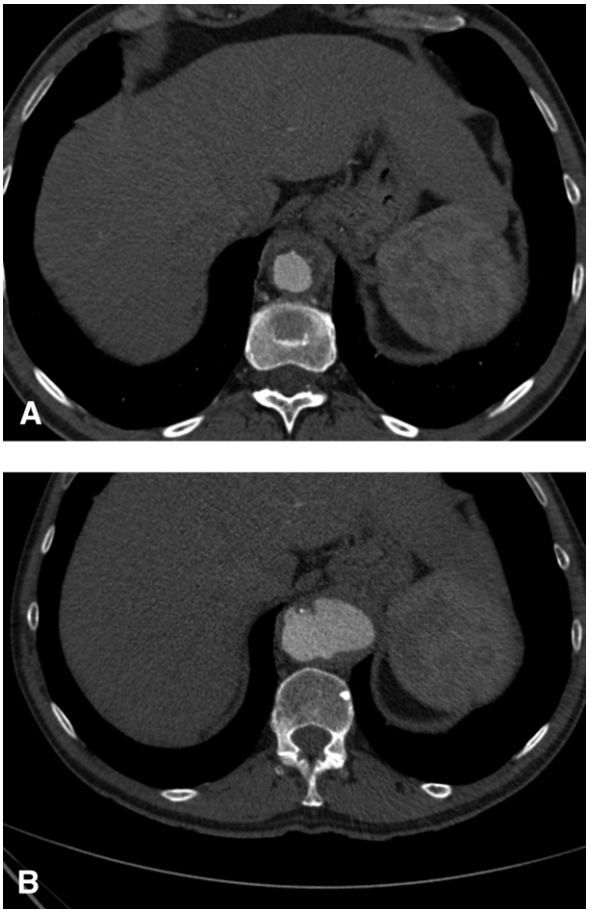

Fig. Single-image from the preoperative computed tomography (CT) scans of the mycotic thoracic thoracoabdominal aortic aneurysm (TAAA). **A,** The first CT scan showed a TAAA of 4.1 cm in diameter. **B**, The CT scan 2 months later showed a TAAA of 6.0 cm in diameter.

quine (200 mg, thrice daily) and will be continued for at least 18 months. After 13 months of follow-up, there were no complaints, and the follow-up CT scan showed a stable condition of the aorta.

DISCUSSION

This case report demonstrates the rare manifestation of an infected TAAA caused by chronic Q fever. Because of the presence of abdominal complaints, fever, increased inflammatory parameters, and radiologic findings, together with the fact that our patient lived in an area with a large Q fever outbreak,[11] an infection with *C burnetii* was suspected at an early stage.

An English literature search revealed 26 cases of chronic Q fever infection with a vascular manifestation of the native aorta, whereas in 2 of these cases the TAA was involved.[6-9] In contrast with our patient, *C burnetii* could not be isolated in the two TAAA patients[9] by PCR from the aneurysm. Their diagnosis was based on positive serology only, associated with the TAAA. In our patient, however, we could demonstrate *C burnetii* DNA in the aneurysm, confirming the diagnosis of infected TAAA by Q fever. The rt-PCR used in our patient targets the insertion element IS*1111* that is present in multiple copies in the *C burnetii* genome, ensuring sensitive detection of the bacterium.

The aerosol inhalation is the primary mode of human contamination by *C burnetii*, which may occur directly from infected animals. The organism may also spread by the wind,[12] and for this reason, Q fever may occur in patients who live in urban areas without any evident contact with animals. It is now well established that development of a chronic infection is more often due to predisposing factors in the host than the virulence of the bacterium.

Because infected aneurysms of the aorta are fatal if untreated,[13] surgical and medical treatment are both necessary. En bloc resection of both the aneurysm and the perianeurysmal infected tissue and in situ prosthetic graft replacement with reimplantation of visceral or renal arteries are the basic principles of operation for mycotic TAAA.[14,15] In situ replaced prosthetic grafts should be soaked with rifampin,[16-18] whereas it has already been demonstrated to bind significantly to gelatin-sealed Dacron grafts.[2]

Medical treatment consists of intensive antibiotic therapy, which is crucial for successful treatment and should be started perioperatively. The required duration of antibiotic therapy is not well established. Recommendations range from 6 to 8 weeks to lifelong antibiotic therapy.[16,19] These should be stopped only when all infectious signs and blood cultures are negative. The current recommendation for the treatment of Q fever vascular infection is a combination of doxycycline and hydroxychloroquine administered for a minimum of 18 to 36 months.[2,20] Regular controls with CT scans and control of infection parameters are necessary, because recurrent or persistent aortic or graft infections might occur.[16,21]

CONCLUSION

For patients with a vascular medical history who live in an endemic area for Q fever and complain of abdominal pain with fever, an infected aneurysm due to Q fever should be included in the differential diagnosis.

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