

Mycotic aortic aneurysm due to brucellosis

Abdullah Alhaizaey, MD, Mohammed Alassiri, MD, FRCSC, Musaed Alghamdi, MD, FRCSC, and Mushabab Alsharani, MD, *Abha, Saudi Arabia*

Brucellosis is a multisystem zoonotic disease. Mycotic aneurysm due to *Brucella* is rare and has no clear management approach. Here, we present two cases of mycotic aortic aneurysm due to *Brucella*. The first patient was treated with surgical resection of a symptomatic infrarenal abdominal aortic aneurysm combined with lifelong doxycycline and rifampicin. The second patient improved with conservative treatment including a 6-month course of antibiotics and regular clinical and radiologic monitoring. Through these cases, we hope to draw attention to this serious adverse effect of *Brucella* and the importance of management of its local arterial complications, especially in endemic areas. (*J Vasc Surg Cases* 2016;2:50-2.)

Brucellosis is the most prevalent zoonotic disease worldwide. *Brucella melitensis* is the most common type of *Brucella* bacteria transmitted to humans through unpasteurized products.¹⁻³ Brucellosis may present with a large spectrum of clinical manifestations affecting the gastrointestinal, cardiovascular, genitourinary, hematologic, neurologic, and skeletal systems. The most common cardiovascular complication is endocarditis, which occurs in 1% to 2% of cases.⁴ *Brucella* arteritis or its local arterial complication is rare. Through this case series, we hope to draw attention to the absence of an optimal treatment for vascular brucellosis and mycotic aneurysm due to *Brucella*. The consent of the patients for publication was obtained.

CASE REPORT

First patient. A 60-year-old woman with a history of asymptomatic type III DeBakey chronic spontaneous aortic dissection for 4 years was admitted through the emergency department with severe continuous abdominal and bilateral flank pain persisting for 3 days. Her most recent thoracoabdominal computed tomography (CT) scan 6 months earlier showed stable aortic dissection without aneurysmal change (Fig 1). Physical examination on admission revealed a pulsatile abdominal mass. Urgent thoracoabdominal CT with arterial-phase administration of contrast material showed a 6- × 6-cm infrarenal abdominal aortic aneurysm that was not obvious on previous CT scans (Fig 2, A and B). She was a nonsmoker and nonalcoholic, and she had no family history of aortic aneurysm. However, she had a history of continuous low-grade fever with sweating for 8 weeks, which was accompanied by generalized joint and back pain. She was a farmer

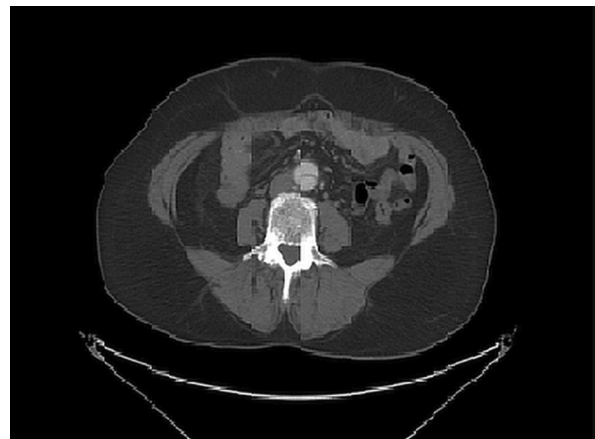


Fig 1. An axial view of computed tomography (CT) of the abdomen during arterial phase showing spontaneous infrarenal abdominal aorta dissection without change for 4 years.

and had a history of consuming unpasteurized milk daily. Her leukocyte count was 17,000/mm³ and her C-reactive protein (CRP) level was 32 mg/dL. Results of an acid-fast bacilli test were negative, whereas those of a *Brucella* immunoglobulin G (IgG) serologic test were highly positive (600).

At this point, the infectious disease team began treatment with antibiotics, including aminoglycoside, doxycycline, and rifampicin. However, the patient's abdomen was tender and her pain was continuous. Therefore, she underwent urgent mycotic aneurysm excision with a 20- × 10-mm silver-coated aortobi-iliac in situ graft. Histopathologic examination revealed a severe degenerative aneurysm with severe arteritis. Tissue cultures after a 3-week incubation period were positive for *Brucella*, which was sensitive to rifampicin and doxycycline.

Regular clinical and radiologic follow-up at 6 months showed a normal erythrocyte sedimentation rate, CRP level of <10 mg/dL, gradual decrease in *Brucella* IgG serology titer to 80, and normal aortic graft on 1-year follow-up abdominal CT (Fig 2, C). Currently, the patient undergoes regular clinical and radiologic follow-up annually.

Second patient. An 83-year-old man was referred by his primary care physician because of continuous abdominal pain persisting for 3 days. He also had a history of consuming

From the Division of Vascular Surgery, Aseer Central Hospital, King Khalid University.

Author conflict of interest: none.

Correspondence: Abdullah Alhaizaey, MD, Division of Vascular Surgery, Aseer Central Hospital, King Khalid University, PO Box 34, Abha 61321, Saudi Arabia (e-mail: abd.25@hotmail.com; aalhizaey@moh.gov.sa).

The editors and reviewers of this article have no relevant financial relationships to disclose per the Journal policy that requires reviewers to decline review of any manuscript for which they may have a conflict of interest.

2352-667X

© 2016 The Authors. Published by Elsevier Inc. on behalf of Society for Vascular Surgery. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

<http://dx.doi.org/10.1016/j.jvsc.2016.03.009>



Fig 2. Computed tomography (CT) of the abdomen during arterial phase showing an infrarenal abdominal aorta dissection complicated by a 6- × 6-cm mycotic aneurysm due to chronic *Brucella* arteritis. **A**, Axial view. **B**, Three-dimensional view. **C**, Coronal view of CT of the abdomen during arterial phase showing normal infrarenal abdominal aorta graft that was applied 1 year earlier for a patient with a 6- × 6-cm mycotic aneurysm due to chronic *Brucella* arteritis.

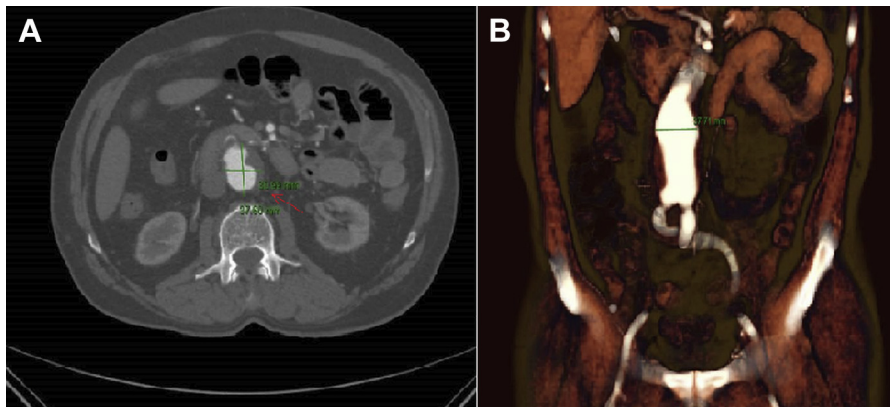


Fig 3. Computed tomography (CT) of the abdomen during arterial phase showing an infrarenal abdominal aorta aneurysm of 31 × 38 mm; the *arrow* shows wall thickening and inflammatory fat stranding changes localized at the aneurysm wall in a patient who had high *Brucella* serology titer. **A**, Axial view. **B**, Coronal three-dimensional view.

unpasteurized milk with a history of generalized joint and low back pain and low-grade fever for 3 months. On admission, his CRP level was 28 mg/dL, and results of a *Brucella* IgG serologic test were positive (320). Abdominal CT with arterial-phase administration of contrast material showed a 3.8-cm infrarenal abdominal aortic aneurysm surrounded by retroperitoneal fat stranding and inflammatory change. The other parts of the aorta appeared to have normal wall thickness surrounded by normal fatty layers (Fig 3). There was no previous abdominal CT scan because the patient did not have such symptoms previously. All of these clinical findings are highly suggestive of mycotic aneurysm.

Treatment with antibiotics including doxycycline and rifampicin was initiated. Within 3 days, his symptoms were completely resolved. Because of the patient's dramatic clinical response to antibiotic treatment and the small size of the aneurysm, we leaned toward conservative management, including a 6-month course of antibiotics and close clinical and radiologic monitoring.

Regular clinical and radiologic follow-up at 6 months showed a CRP level of <10 mg/dL, gradual decrease in *Brucella* IgG serology titer to 80, and no change in aneurysm size on abdominal CT.

DISCUSSION

Mycotic aneurysm may be caused by either local extension of an adjacent soft tissue infection or embolization of an infectious source. It is postulated that in the case of embolization, emboli reach the adventitia through the vasa vasorum, and the inflammatory response disrupts both the muscularis and adventitia, resulting in blood vessel wall weakness and pseudoaneurysm formation.⁵

Mycotic aneurysm involving the aorta or large arteries due to *Brucella* is very rare. According to a literature search, the ascending aorta, superior mesenteric artery, subclavian artery, or axillary artery may be affected. Thirty-four cases of *Brucella* endarteritis have been published.⁶ In a previous review of 25 published cases of aortic brucellosis, Kakkos et al reported that the infrarenal abdominal aorta (65%) was most affected, followed by the ascending thoracic aorta (23%).⁵

The chronic form of brucellosis usually is manifested with either local disease or constitutional symptoms in

the form of generalized bone pain, back pain, low-grade fever, or sweating. However, *Brucella* endarteritis usually has no specific clinical symptoms that may guide immediate diagnosis.⁶ Abdominal pain appears to be the most common symptom of aortic brucellosis, and it was the main presenting complaint for both of our patients. In endemic areas, we believe that brucellosis should be included in the differential diagnosis or as the leading cause of the main disease. We routinely screen for *Brucella* infection in all cases of mycotic aneurysm in addition to other suspected causes, such as tuberculosis, syphilis, and salmonellosis. *Brucella* serologic testing by enzyme-linked immunosorbent assay is the usual method of diagnosis.⁴⁻⁶ The sensitivity of *Brucella* IgG serology is 92% or more for *Brucella* endarteritis, which may be due to the long period of disease formation. Enzyme-linked immunosorbent assay and tissue histopathology also have high sensitivity for *Brucella* infection.

There is no specific management approach for *Brucella* arteritis or its local complication of mycotic aneurysm. Herrick et al reported a case of brucellosis presenting as bacteremia and aortic ulcer 18 years after *Brucella* exposure, which was treated successfully by combined surgical repair and lifelong doxycycline and rifampicin.⁶ Doxycycline (200 mg) plus rifampicin (600-900 mg) daily for a minimum of 6 weeks is the recommended treatment for brucellosis.^{7,8} A literature search on the optimal treatment for *Brucella* infection and its local complication of endocarditis revealed no reliable data regarding choice or duration of antibiotic therapy; however, there seems to be unanimous agreement that therapy should be prolonged.⁶⁻⁸ Medical treatment alone for *Brucella* endocarditis has a high mortality rate (33%) compared with combined medical and surgical treatment (7%).⁶⁻¹⁰ However, there is no comparative study on the various treatments for *Brucella* arteritis or its local arterial complications.

CONCLUSIONS

Through these cases, we hope to draw attention to this serious adverse effect of *Brucella* and the importance of management of its local arterial complications, especially in endemic areas.

REFERENCES

1. Corbel MJ. Brucellosis: an overview. *Emerg Infect Dis* 1997;3:213-21.
2. Pappas G, Akritidis N, Bosilkovski M, Tsianos E. Brucellosis. *N Engl J Med* 2005;352:2325-36.
3. Cascio A, De Caridi G, Lentini S, Benedetto F, Stilo F, Passari G, et al. Involvement of the aorta in brucellosis: the forgotten, life-threatening complication. A systematic review. *Vector Borne Zoonotic Dis* 2012;12:827-40.
4. Cakalagaoglu C, Keser N, Alhan C. Brucella-mediated prosthetic valve endocarditis with brachial artery mycotic aneurysm. *J Heart Valve Dis* 1999;8:586-90.
5. Kakkos SK, Papadoulas S, Lampropoulos G, Marangos M, Kalogeropoulou C, Tsolakis IA. Aorto-iliac aneurysm infected by *Brucella*: distinctive presentation patterns of a rare entity. *Vascular* 2013;21:307-15.
6. Herrick JA, Lederman RJ, Sullivan B, Powers JH, Palmore TN. *Brucella* arteritis: clinical manifestations, treatment, and prognosis. *Lancet Infect Dis* 2014;14:520-6.
7. Chopra I, Roberts M. Tetracycline antibiotics: mode of action, applications, molecular biology, and epidemiology of bacterial resistance. *Microbiol Mol Biol Rev* 2001;65:232-60.
8. Franco MP, Mulder M, Gilman RH, Smits HL. Human brucellosis. *Lancet Infect Dis* 2007;7:775-86.
9. Kumar N, Prabhakar G, Kandeel M, Mohsen IZ, Awad M, al-Halees Z, et al. *Brucella* mycotic aneurysm of ascending aorta complicating discrete subaortic stenosis. *Am Heart J* 1993;125:1780-2.
10. Keshtkar-Jahromi M, Razavi SM, Gholamin S, Keshtkar-Jahromi M, Hossain M, Sajadi MM. Medical versus medical and surgical treatment for brucella endocarditis. *Ann Thorac Surg* 2012;94:2141-6.

Submitted Jan 8, 2016; accepted Mar 23, 2016.