Atrial Septal Defect Presenting With Brucella Endocarditis

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Brucellosis is a zoonosis that behaves as a systemic infection with various clinical signs and symptoms. Brucella endocarditis, although a rare complication of Brucella infection, is nevertheless responsible for the majority of deaths related to this illness. Brucella endocarditis was associated with an atrial septal defect (ASD) in a 45-year-old woman. Echocardiography showed a secundum ASD with vegetations of 0.5×0.8 cm arising from the border of the defect, and serologic analysis was positive for Brucella agglutinin, thus confirming the diagnosis. She was initially treated with a 1-month preoperative course of antibiotics. At surgery, the vegetations were excised and the defect was closed with polypropylene. Postoperative recovery was uneventful and she was discharged on the 10th postoperative day. (*Circ J* 2008; **72:** 2096 – 2097)

Key Words: Atrial septal defect; Brucella; Endocarditis

Brucellosis is a zoonosis and the organism most commonly implicated is Brucella mellitensis which is found in the Mediterranean, the Arabian Gulf, Latin America, Asia, parts of Mexico, the Indian subcontinent[1] and the Middle East. It continues to be endemic to the eastern part of Turkey.

In humans, brucellosis behaves as a systemic infection with various clinical signs and symptoms. The disease usually presents as a fever with no apparent focus, although in 20–40% of cases there are focal forms that have been described in almost all organs and systems, with the osteoarticular forms being more common and those affecting the heart and central nervous system being more severe[1].

Brucella endocarditis, although a rare complication of Brucella infection, is nevertheless responsible for the majority of deaths related to this illness. The diagnosis and treatment of endocarditis following brucellosis is difficult because of its stealthy progress and difficulties in isolating the causative organism[2]. In this report, a case of Brucella endocarditis associated with an atrial septal defect (ASD) was successfully treated with a combination of medical and surgical therapy. To our knowledge, a fossa ovalis type of ASD presenting with Brucella endocarditis in an adult patient has not previously been described.

Case Report

A 45-year-old woman was admitted with complaints of intermittent fever associated with chills and rigors, shortness of breath and weight loss for 3 weeks. She had been given several antibiotics at primary care centers. At presentation, her body temperature was 39°C, blood pressure was 85/66 mmHg, and pulse rate was 118 beats/min. Auscultation revealed a systolic ejection murmur over the left sternal edge and fixed splitting of the second heart sound.

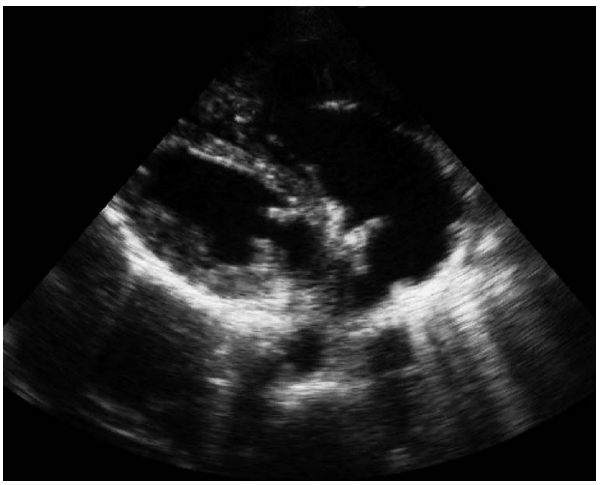


Fig 1. Echocardiogram showing the atrial septal defect associated with vegetation arising from the border of the defect.

The hemoglobin level was 12.0 g/dl, the white blood cell count was 14.200/mm^3, the platelet count was 400.000/mm^3, and the erythrocyte sedimentation rate was 65.0 mm/h. Chest radiography showed a marked widening of the pulmonary arteries, and electrocardiogram revealed atrial fibrillation and an incomplete right bundle branch block. Three blood culture specimens were obtained on separate occasions, but no microorganism was isolated in the cultures. Serologic analysis was positive for Brucella agglutinin (1/320). Echocardiography showed an ostium secundum ASD (fossa ovalis type) that was 2 cm in diameter with left-to-right shunting through the defect. There was a mobile vegetation measuring 0.5×0.8 cm arising from the border of the defect (Fig 1). The patient was offered opera-

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tion, but she declined, so we administered a 1-month course of antibiotic therapy comprising 200 mg/day doxycycline orally, 900 mg/day rifampicin orally, and 2,000 mg/day ceftriaxone intravenously. Her condition had improved sufficiently for surgery after the month, but the vegetation was still visible on echocardiography. At surgery, the areas of vegetation were excised and the defect was closed with polypropylene. No microorganisms were observed or grown from the excised material. Postoperative recovery was uneventful and she was discharged on the 10th postoperative day. Antibiotics were continued for 3 months after surgery (900 mg/day doxycycline orally and 600 mg/day rifampicin orally for 3 months, and 2,000 mg/day ceftriaxone intravenously for 1 month) and the patient was followed up weekly in the first month and then monthly for the following year. Physical examination, erythrocyte sedimentation rate and CRP level were assessed at each visit were normal. The Brucella agglutinin test became negative in the third month. Neither vegetation nor residual septal defect was observed on echocardiography performed during the follow-up period.

Discussion

A patient presenting with a heart murmur and a history of ingestion of unpasteurized milk and exposure to infected animals or animal products should be considered as having cardiac manifestations of brucellosis until proved otherwise.[3] Brucella appears to be a slowly destructive organism, with a marked tendency to cause tissue ulceration and the development of large vegetations carrying a significant risk of embolization, and it is difficult to eradicate with medical therapy alone. For these reasons, optimal treatment should achieve sterilization of infected cardiac tissue and extensive surgical debridement without delay to control acute illness and prevent complications and relapse.[3]

The diagnosis of brucellosis is based on epidemiological evidence and positive culture or serology. The manifestations of the disease, especially in the chronic stage, make the diagnosis difficult and most of the conventional diagnostic methods have their limitations. Blood culture is the only specific test, but its sensitivity ranges from 17% to 85% depending on culture conditions, antibiotic therapy and the interval between the onset of symptoms and diagnosis.[4] None of the blood cultures revealed any growth in the present case, which may have been because she was given antibiotic medications before admission. For this reason, agglutination titers, as well as clinical evaluation, should be used for the diagnosis.

However, it should be emphasized that seronegativity does not exclude the diagnosis.[5] In one study, the diagnosis of brucellosis could not be made by several blood cultures and serological assays, but the type of organism was determined by PCR.[2]

There is no consensus as to the ideal timing of surgical intervention, if indicated. Generally, antibiotic therapy is recommended for at least 6 weeks beforehand, but the presence of vegetation and congestive heart failure are indications for early surgical management.[5] Horstkotte et al suggested that early surgical intervention within 72 h of presentation is warranted in patients with thromboembolic events[6] and Hadjinikolau et al have operated on their patients within 1 week of commencing antibiotic therapy.[7] The present patient had vegetations arising from the border of the defect and we performed surgery after an initial month of antibiotic therapy, because the patient and her family refused to undergo immediate operation without medical treatment.

Bacterial endocarditis associated with ASD is rare. Accordingly, infective endocarditis prophylaxis is not recommended in cases of adult congenital ASD.[8] Rahman et al reported a sinus venosus type of ASD presenting with Brucella endocarditis.[9]

Brucella melitensis resists the bactericidal activity of normal serum and remains viable inside human polymorphonuclear leucocytes.[10] The antimicrobial agents used for Brucella endocarditis should be bactericidal and able to penetrate the infected cells and exert their activity inside the phagosome.[7] There is no consensus on the duration of postoperative antibiotic therapy;[11] the reports range from 2 weeks to 13 months.[7,11] However, the decision to stop treatment could be individualized after a thorough clinical observation and evidence of a decrease in the anti-agglutinin titer.[7] In this case, the serum agglutination titers became negative after 3 months and we ceased antibiotic therapy because of the excellent clinical condition of the patient.

Conclusion

Although ASD may not need prophylaxis for infective endocarditis in clinical practice, physicians should maintain a high degree of suspicion of Brucella endocarditis in patients with a congenital heart defect living in endemic areas. A combination of medical therapy and surgical intervention should be performed to ensure a successful definitive outcome.

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