

Brucella Endocarditis in Childhood and Surgical Treatment with Manouguian Procedure: Case Report

Çocukluk Çağında Brusella Endokarditi ve Manouguian Yöntemi ile Tedavisi

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ABSTRACT Brucella endocarditis is a rare complication of Brucella infection, but is responsible for the majority of deaths from this disease. When treated by antibiotics alone, this complication is usually fatal. A combination of medical and surgical therapy is necessary for the successful treatment for Brucella infective endocarditis. Herein, we present a 5-year-old boy who underwent operation with Manouguian procedure due to Brucella endocarditis following a 6 week period of medical therapy. No complication associated with Brucella infection or prosthetic valve were observed for a period of 8 years after operation.

Key Words: Brucella; endocarditis; child; heart valve prosthesis implantation

ÖZET Brusella endokarditi, brusella enfeksiyonunun nadir bir komplikasyonu olmasına karşın bu hastalığa ait ölüm sebeplerinin başında yer alır. Tedavide sadece antibiyotiklerin kullanılması genellikle ölümlü sonuçlanır. Bu yüzden medikal ve cerrahi tedavinin birlikte uygulanması gereklidir. Bu makalede 6 haftalık antibiyotik uygulamasından sonra Manouguian yöntemi ile ameliyat edilen ve postoperatif 8 yıllık izleme döneminde olan 5 yaşında erkek bir olguyu sunuyoruz. Bu süreçte brusella enfeksiyonu ve yapay kalp kapağı ile ilgili bir komplikasyon görülmemiştir.

Anahtar Kelimeler: Brusella endokarditi; çocukluk çağı; kalp kapağı replasmanı

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Brucella endocarditis is a rare complication of Brucella infection, but is responsible for the majority of deaths from this disease. When treated by antibiotics alone, this complication is usually fatal.¹⁻⁵ For successful treatment of Brucella infective endocarditis a combination of medical and surgical therapy is necessary. Surgically treated cases are rare in childhood, hence, herein we present a 5-year-old boy with diagnosis of Brucella aortic valve endocarditis who underwent surgery due to 3° aortic regurgitation and persisting vegetations following a 6 week period of medical therapy.

CASE REPORT

A 5-year-old boy was admitted to the hospital with a history of erythema, swelling and arthralgia of the left leg for 4 weeks, high fever for 10 days. He

had been treated with penicillin before admission. His blood pressure was 80/50 mmHg, pulse 132 beat per minute, oral temperature 37.4°C, respiration rate 36 per minute. His lungs were clear and a systolic murmur of grade 2-3/6 was heard over the aortic and mesocardiac areas. The liver and spleen were palpable 5 and 3 centimetres below the right and left costal margins respectively. Fundoscopic examination was normal. There was pretibial edema in both legs, and tenderness with motion in left leg. Radiology of the chest yielded normal results. An electrocardiogram (ECG) showed left ventricular hypertrophy. Abdominal ultrasonography revealed enlarged liver and spleen. Transthoracic echocardiography showed bicuspid aortic valve, aortic stenosis and incompetence and vegetation on the aortic valve (Figure 1). Laboratory results were as follows: erythrocyte sedimentation rate (ESR) 40 mm/first one hour, antistreptolysin O titre 191 TU, hemoglobin (Hb) 9.3 g/dl, hematocrit 28%, white blood count (WBC) 7800/mm³ with 40% lymphocytes and 46% polymorphs, platelets 100.000/mm³. Biochemical tests of liver and renal function were within normal limits. His prothrombin time was 13 seconds and partial thromboplastin time was 34 seconds (controls 13 and 33 seconds respectively). Urinalysis showed mild proteinuria and microscopic hematuria. Antibiotic treatment was initiated with penicillin and gentamicin with the diagnosis of infective endocarditis and/or attacks of acute rheumatic fever.



FIGURE 1: Preoperative transthoracic echocardiography shows bicuspid aortic valve, aortic stenosis and incompetence and vegetation on the aortic valve. (RV and LV=right and left ventricles, LA=left atrium, AO=aorta, kitle=vegetation).

Ten days later, *Brucella melitensis* was isolated from blood culture, and the brucella agglutination titre was 1:1280. Therefore, the antibiotics were replaced with rifampicin (300 mg/day), trimethoprim-sulfamethoxazole (TMP-SMZ, 160 mg of TPM/day and 800 mg of SMZ/day) and ceftriaxone (rocephin 1400 mg/day). An operation was indicated due to 3° aortic regurgitation and vegetation following a 6 week period of antimicrobial therapy. Cardiopulmonary bypass was instituted with the cannulation of ascending aorta, superior and inferior vena cavae. Cardiac arrest and protection was achieved with cold crystalloid cardioplegia adjunct to systemic hypothermia. At operation, multiple vegetations were seen on the right and noncoronary aortic valve cusps which were fused heavily. The aortic cusps with obvious vegetation were removed. The aortic valve was replaced with a 17mm St. Jude prosthesis after aortic root enlargement with Manouguian procedure (Figure 2). The hospital course remained uneventful and the patient was discharged with anticoagulant and antibiotic (TPM-SMZ and rifampicin) therapy. The brucella agglutination titre was 1:256 six months after surgery and antibiotic therapy was discontinued. According to the last echocardiographic findings, the gradients at the prosthetic aortic valve at rest were 28 mm/hg. No complications were seen related to either *Brucella* infection or prosthetic valve for a period 8 years after operation.

CONCLUSION

Brucella organisms commonly invade the lymphoreticular system in the liver, spleen and lymph nodes, but less commonly, the *Brucella* organisms may attack the endocardium. *Brucella* endocarditis, although a rare complication of brucellosis, is the main cause of death related to this disease.¹⁻⁵ This is usually due to heart failure resulting from acute aortic and/or mitral valvar regurgitation.² *Brucella* organisms may be implanted on a valve which has been damaged by previously rheumatic fever or a congenital malformation; but they can also infect and damage normal valves.²

It is difficult to distinguish acute brucellosis from acute rheumatic fever, especially if cardiac in-



FIGURE 2: Aortic root enlargement with the technique of Manouguian procedure.

involvement is noted. Both diseases may present with fever, arthralgia/arthritis, splenomegaly, and high erythrocyte sedimentation rate, but acute brucellosis only occasionally shows leucocytosis.⁶

Endocarditis commonly presents with pyrexia with or without cardiac murmur and sudden dramatic deterioration of cardiac function may occur. There may be acute aortic and/or mitral valve regurgitation with acute left ventricular failure which needs urgent valve replacement.² The diagnosis of *Brucella* endocarditis is based on the symptoms, the high *Brucella* antibody titers, positive blood cultures and particularly, the finding of vegetations on the heart valves at echocardiography.² Echocardiography is usually helpful in detecting valvar vegetations and we have observed vegetations of the aortic valve in our patient. The usual

presentation is with fever and varying degrees of valvular damage, usually necessitating valve replacement. Our patient was initially treated with antimicrobial drugs for 6 weeks, and then underwent surgery for 3° aortic regurgitation and vegetation following a 6 week period of antimicrobial therapy.

Although some authors⁷ claim that *Brucella* endocarditis resolves with medical therapy alone, most consider urgent or emergency surgery mandatory.^{1,3,5,8,9} The best antibiotic regimen for *brucella* endocarditis is unknown. It seems wise to add a bactericidal agent such as TMP-SMZ and/or rifampin to the classic combination of tetracycline and aminoglycoside (streptomycin or gentamicin). The optimal duration of antimicrobial treatment is still undetermined. The bacteria in *Brucella* endocarditis survive because they are in the intracellular compartment. For this reason, there is no common consensus on the duration of the antibiotic therapy after surgery. The left ventricular outflow enlargement procedure may involve several techniques. Manouguian procedure for enlargement of the narrow aortic valve ring involves extension of the aortic incision through the fibrous origin of the aortic leaflet of the mitral valve into this leaflet.¹⁰ This technique was preferred for our patient owing to its simplicity and convenience. Postoperative medication may last from 2 weeks to 13 months.^{2,3} The discontinuation criteria of antibiotic therapy in our case were the decrease in *Brucella* agglutination titers, absence of fever, and clinical improvement. We did not observe any recurrence or prosthetic valve related complications in our case.

As a result, the treatment of *Brucella* endocarditis should be multidisciplinary. Early diagnosis of *Brucella* endocarditis or its complications on the basis of clinical, serological, echocardiographic, and if required, computerized tomographic findings is essential. Early surgical intervention, combined with antibiotics provides the best chance of preventing abscess formation and reduces the risk of mortality.

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