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Cover Page Footnote

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Case Report

Triple valve infective endocarditis - a late diagnosis

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Abstract

Behcet's disease is a systemic vasculitis of unknown aetiology with cardiac involvement as well as damage to other organs. Whether the sterile valvular inflammation which occurs in this autoimmune disease predisposes to bacterial adhesion and infective endocarditis is not yet established.

We present the case of a patient with Behcet disease in which transthoracic echocardiography showed mobile masses on the aortic, tricuspid, and mitral valves, leading to multivalvular infective endocarditis diagnosis, possibly in the context of valvular inflammation.

The case presented in this article confirms observation of other studies, namely that ultrasonography plays an important role in the diagnosis and evaluation of rheumatic diseases and permits optimal management in daily practice.

Keywords

: Behcet disease, infective endocarditis, echocardiography, multivalvular endocarditis

Highlights

- ✓ Behcet's disease is a systemic vasculitis of unknown aetiology, involving not only cardiac impairments but also damage of other organs
- ✓ Ultrasonography plays an important role in the diagnosis and evaluation of rheumatic diseases and permits optimal management in daily practice

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Introduction

Infective endocarditis (IE) is an infection of the endocardium usually involving heart valves. The incidence of IE in the last decade has slowly increased from 10 to 15 cases/100.000 population (1). Major bacteria involved are staphylococcus and streptococcus sp.; an ascending trend is also seen for Gram- bacteria and fungi (1). The clinical manifestations of the disease are polymorphic and include persistent fever (>38°C), the presence of heart murmurs, heart failure symptoms, and different vascular (embolic) and immunological complications. Positive blood cultures and the presence of typical IE lesions (vegetation, abscess, pseudoaneurysm, fistula, or new partial dehiscence of a prosthesis) seen on echocardiography are necessary for a diagnosis, these elements being included in the Duke criteria for diagnosis of IE (2).

Behcet's disease (BD) is a systemic vasculitis of unknown aetiology with cutaneous (recurrent bipolar aphthous ulcers), ocular, digestive, articular, neurologic, vascular, and cardiac damage (3). Whether sterile valvular inflammation in this autoimmune disease predisposes to bacterial adhesion and infective endocarditis has not been clearly established. We present a patient with multivalvular IE and Behcet syndrome, with a long medical history that led to a late diagnosis and many complications.

Case Report

A 30 years old male without previous cardiovascular disease presented with persistent lumbar and right buttock pain. The lumbar pain started 2 months prior, was initially ameliorated by corticosteroids (10 days of methylprednisolone) and intensified with movement. Other symptoms were oral ulcers, fatigue, dyspnea when doing usual physical activity, poor appetite, and 15 kg weight loss in the last months.

His medical history revealed: laminectomy for L4-L5, L5-S1 herniated disc at the age of 17 years, chronic viral B hepatitis 3 years ago in immune tolerance stage; 5 months ago, diagnosed with left frontal and maxillary sinusitis (from a dental abscess) with fistula to the left nostril for which a sinus puncture was performed and antibiotics were administered. After 3 months he had left trochlear (IV) nerve paresis and later left retinal hemorrhage. Brain MRI showed areas of demyelination in the left frontal and parietal regions interpreted as an inflammatory reaction to infection. Severe anemia and an inflammatory biological syndrome were noticed. CT scan of the thorax and abdomen identified 2 splenic infarcts and marginal bone condensation of thoracic and

lumbar vertebral bodies (T6-T10, L1-L4, L5-S1) and around sacroiliac joints.

Physical examination showed a patient with normal body mass index, fever (39°C), pale skin and mucous membranes, vasculitis lesions on limbs (Figure 1); pain and severe reduction in mobility of lumbar spine, pain on examination of sacroiliac joints; normal breathing sounds, heart rate about 90 b/min, regular rhythm, mitral regurgitation murmur (grade 3/6), tricuspid regurgitation murmur (grade 2/6), BP= 110/60 mmHg, enlarged liver, splenomegaly.



Figure 1. Vasculitis lesions located around metacarpophalangeal joints

Blood tests showed microcytic hypochromic anemia (Hb = 7.5g/dl) with normal ferritin value, a leucocyte counting with predominance of neutrophils, normal platelet count, high value of erythrocyte sedimentation rate =140mm /1h, high C reactive protein= 259 mg/dl, creatinine value was 1.32 mg/dl (eGFR= 72 ml/min/1.73m²) which increased later to 4.15 mg/dl (eGFR= 18 ml/min/1.73 m²). Urine exam revealed high numbers of erythrocytes and white blood cells and a nephrotic range proteinuria- 3.5g/24h. Immunological tests for autoimmune diseases (extended ANA –BLOT) were negative, complement fractions were normal. Multiple blood cultures (3) were positive for streptococcus sp. HLA – B51 antigen was present showing predisposition for Behcet disease.

On the patient's ECG, sinus tachycardia and voltage criteria for left ventricle hypertrophy (LVH) were seen. Transthoracic echocardiography (TTE) revealed: dilatation of the left ventricle (LV) - end-diastolic diameter 63 mm, end systolic diameter 46 mm, mild increase in thickness of interventricular septum and posterior wall (12mm). We found a LV mass of 154 g/m² and RWT= 0.38 (excentric left ventricular hypertrophy), normal global LV systolic function (LV ejection fraction 60%, LV-EDV = 160 ml, ESV= 64 ml), a mobile mass of 2.4/2.1 cm attached on the atrial side of the anterior mitral cusp (suggestive of vegetation) causing severe mitral regurgitation with an excentric jet directed towards the lateral atrial wall. The

aortic valve was bicuspid with 2 smaller mobile masses (vegetations) of 0.95 and 1 cm attached; aortic valve area (by planimetry) was 2cm², moderate aortic regurgitation was present. A large vegetation of 2.25/1.1 cm was attached on the septal cusp of tricuspid valve and caused moderate tricuspid regurgitation; pulmonary artery pressure (PAP) was slightly increased (40 mmHg), right chambers had normal diameters with good longitudinal systolic function of the right ventricle (RV) (TAPSE= 20 mm) (Figure2).

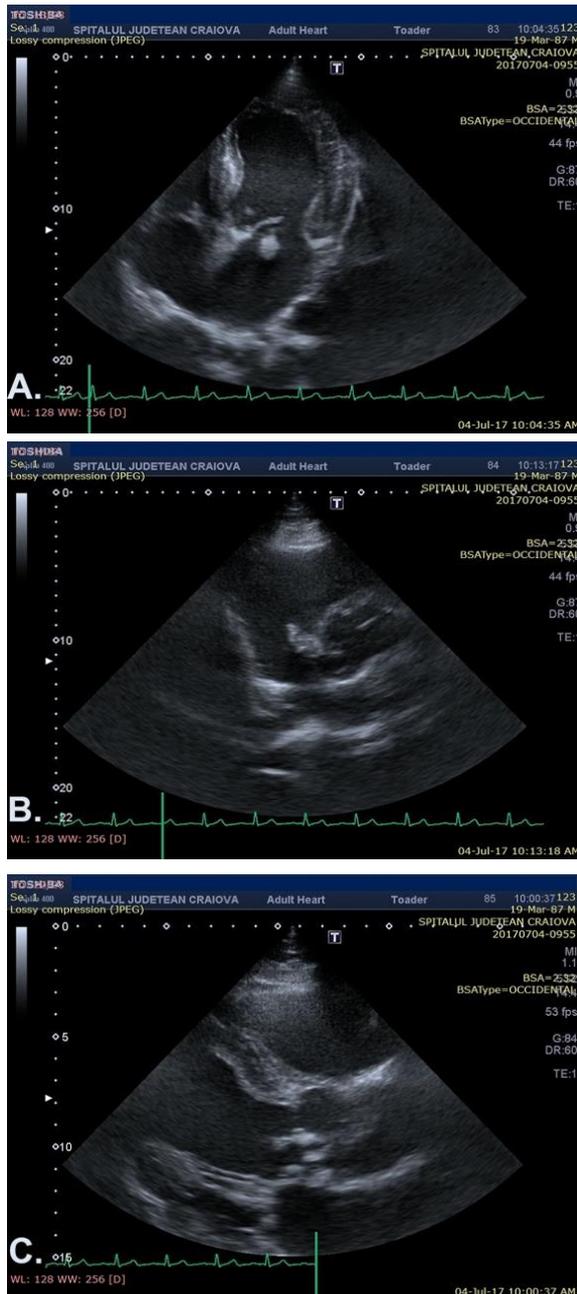


Figure 2. TTE images A. four chamber view-large vegetation attached on anterior mitral valve. B. parasternal short axis view- vegetation attached on septal tricuspid valve. C. parasternal long axis view with focus on the aortic valve-vegetations attached on aortic valve; cusps are thickened in the distal part

IE was diagnosed using the Duke criteria (2 major criteria present). Antibiotic therapy (following blood culture and sensitivity results) was initiated with vancomycin 30 mg/kg (2g/day) iv, later changed to ceftriaxone 4g/day, on which he became afebrile. He was haemodynamically stable but the large size of the mitral vegetation with its embolic risk (and previous embolic events- splenic infarcts) were indications for early surgery, so he was dispatched to another center. During transfer he had a hemorrhagic stroke (with dysarthria and right hemiparesis) that postponed surgery such that he finished the 4 weeks of antibiotic therapy prior to surgery. Mitral and aortic valves were replaced with mechanical prosthesis and tricuspid valve was repaired (partial resection of septal cusp). On the 4th day after surgery he had a massive hemorrhagic stroke that was fatal.

Discussion

The presented case illustrates the polymorphism of manifestations of a streptococcal IE that led to a late diagnosis and the rare triple valve involvement. Criteria for Behcet disease can be recurrent oral ulcerations, vascular purpura skin lesions, bilateral sacroiliitis, diplopia, demyelination in the left frontal and parietal regions, left IV cranial nerve paresis and the presence of HLA – B51 antigen. With the exception of oral ulcerations, all the other manifestations could be due to infective endocarditis.

Cardiac involvement in BD is cited as 0.62-29%, and it is represented by myocardial ischemia, myocardial infarctions, pericarditis, aseptic endocarditis, valve disease, myocarditis, endomyocardial fibrosis, and systolic/ diastolic LV dysfunction (4, 5).

In our case bacteremia began, most probably, from a dental infection, with septic local and systemic complications. Predisposing factors were bicuspid aortic valve and immunodepression due to corticosteroid therapy. Whether aseptic endocarditis due to Behcet disease was a predisposing factor for bacterial adhesion and infection remains unclear. Almost all complications of IE were found: embolic (splenic infarcts, petechiae, Roth spot, cerebral mycotic aneurysm that probably caused the hemorrhagic stroke), immunological (glomerulonephritis), and heart failure.

A review on multivalvular IE found an incidence of 17-18% of IE cases; most often it involves two valves (mitral and aortic) and in the majority of cases is due to staphylococcus sp., followed by streptococcus sp (6, 7). Valvular and congenital heart disease, intravenous drug

use, dialysis, alcoholism, and liver cirrhosis are the most frequent predisposing factors (8-10). The outcome of multivalvular IE is negative, with more frequent complications like perivalvular extension of infection and heart failure; thus, patients need valve surgery more often and mortality is higher (6, 7).

Conclusions

The case presented here confirms conclusions of other studies (11, 12), namely that ultrasonography plays an important role in the diagnosis and evaluation of rheumatic diseases and permits optimal management in daily practice.

Acknowledgment

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