

CASE REPORT

Infective endocarditis of a bicuspid aortic valve complicated by septal aneurysm and mitral-aortic intervalvular fibrosa pseudoaneurysm

Mitral-aort bileşkesi psödoanevrizması ve septal anevrizma ile komplike olan biküspit kapak enfektif endokarditi olgusu

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Summary—Aortic valve endocarditis can lead to the destruction or perforation of the leaflets, ring abscess, fistula, or perivalvular regurgitation; less commonly, the regurgitation itself may result in secondary damage to subaortic structures. Pseudoaneurysm of the mitral-aortic intervalvular fibrosa is a rare complication of native aortic valve endocarditis. This region is a relatively avascular area and is prone to endocarditis infection and subsequent aneurysm formation. Herein, the case of a 33-year-old man who was diagnosed with bicuspid aortic valve endocarditis complicated by a pseudoaneurysm, an aneurysm, and aortic valve rupture is described.

Özet—Aort kapak endokarditi yaprakçıkların hasarına, perforasyonuna, kapak halkası apsesine, perivalvuler yetersizliğe ve daha nadir olarak kapaktaki yetersizlik nedeniyle subaortik yapılarda ikincil hasara neden olabilir. Mitral-aort kapaklar arası bileşke psödoanevrizması, doğal aort kapak endokarditinin nadir bir komplikasyonudur. Bu bölge kısmen damarsız alandır ve endokardit sırasında enfeksiyona ve anevrizma oluşumuna meyillidir. Bu yazıda, psödoanevrizma, anevrizma ve aort kapak rüptürü ile komplike olan otuz üç yaşında biküspit aort kapak endokarditli olgu sunuldu.

A pseudoaneurysm usually occurs as a result of infection, prosthetic valve surgery, chest trauma, or tuberculosis. Less frequently, it may be congenital. A pseudoaneurysm of the mitral-aortic intervalvular fibrosa (P-MAIVF) is a rare, but potentially fatal, complication that generally occurs secondary to aortic valve endocarditis.^[1]

In the present report, the case of a 33-year-old male patient who was diagnosed with an aneurysm, a large pseudoaneurysm, aortic valve rupture, and bi-

cuspid aortic valve (BAV) endocarditis is described. The aneurysm and pseudoaneurysm were visualized with cardiac computed tomography and two- and three-dimensional transesophageal echocardiography (2D-, 3D-TEE).

CASE REPORT

A 33-year-old male patient with the diagnosis of infective endocarditis (IE) was referred for treatment. The patient had been followed for a BAV for more than 10 years. He presented with weakness, shortness of breath, and a fever lasting for 2 weeks. He had received antibiotic treatment for 1 week at another medical center. On physical examination, his arterial blood pressure was 100/70 mm Hg, heart rate was 100 beats/minute, and

Abbreviations:

BAV	Bicuspid aortic valve
IE	Infective endocarditis
MAIVF	Mitral-aortic intervalvular fibrosa
P-MAIVF	Pseudoaneurysm of the mitral-aortic intervalvular fibrosa
TEE	Transesophageal echocardiography
TTE	Transthoracic echocardiography

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the arterial oxygen saturation value was 95%. He had a 3/6 diastolic murmur at the left parasternal border. No crackles were heard on pulmonary auscultation. There were no specific risk factors for IE, including drug abuse or history of a dental procedure.

Laboratory data showed an elevated sedimentation rate (58 mm/hour; reference value: 0–20 mm/hour) and C-reactive protein level (23 mg/L; reference value: 0–8 mg/L). The test results for acid-resistant microorganisms performed at the other hospital were negative. The patient had been taking parenteral levofloxacin, gentamisin, and vancomycin for 1 week upon arrival. Therefore, 3 sets of blood culture results were negative. In our hospital, the only antibiotic administered was intravenous penicillin.

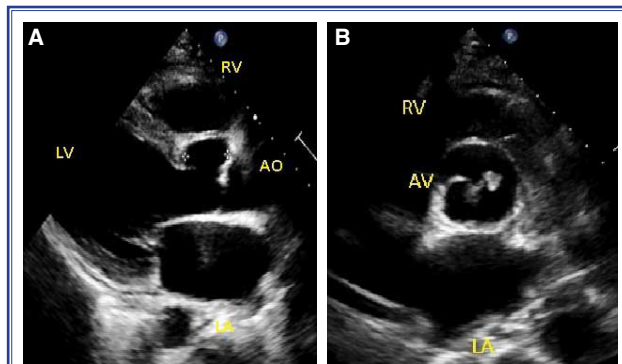


Figure 1. Transthoracic echocardiography. **(A)** Parasternal long axis view showing aortic deformation and aneurysm formation. **(B)** Parasternal short axis view displaying the bicuspid aortic valve and vegetation.

AO: Aorta; LA: Left atrium; LV: Left ventricle; RA: Right atrium; RV: Right ventricle.

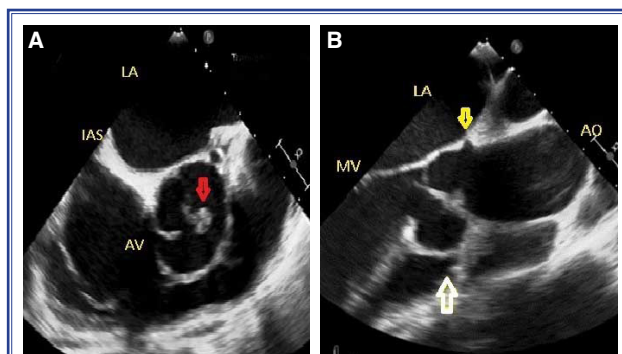


Figure 2. Two-dimensional transesophageal echocardiography. **(A)** Short axis view illustrating the ruptured aortic valve. Red arrow shows vegetation on bicuspid aortic valve. **(B)** The pseudoaneurysm extending towards the mitral-aortic intervalvular fibrosa (yellow arrow), the septal aneurysm (white arrow), and the perforation of the leaflets are seen.

AO: Aorta; AV: Aortic valve; IAS: Interatrial septum; LA: Left atrium; LV: Left ventricle; MV: Mitral valve.

Two-dimensional transthoracic echocardiography (TTE) was performed and revealed vegetation of 6x9 mm in size on a ruptured BAV bicuspid aortic valve (Fig. 1b) with eccentric severe regurgitation, a septal aneurysm (Fig. 1a), and moderate mitral regurgitation with normal left ventricular function and chamber size.

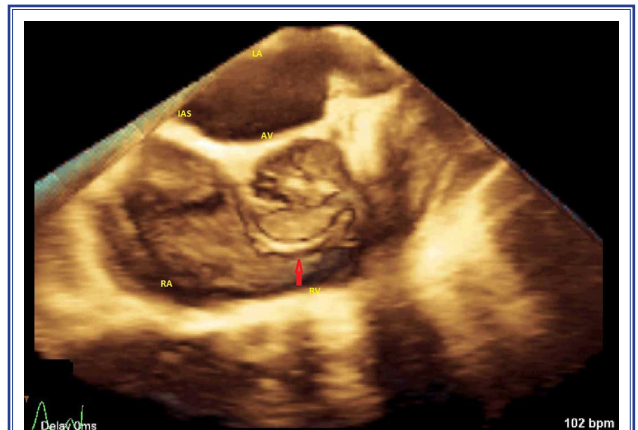


Figure 3. Three-dimensional transesophageal echocardiography. Short axis view of the ruptured aortic valve and the aneurysm extending through right ventricle (red arrow).

AV: Aortic valve; IAS: Interatrial septum; LA: Left atrium; RA: Right atrium; RV: Right ventricle.

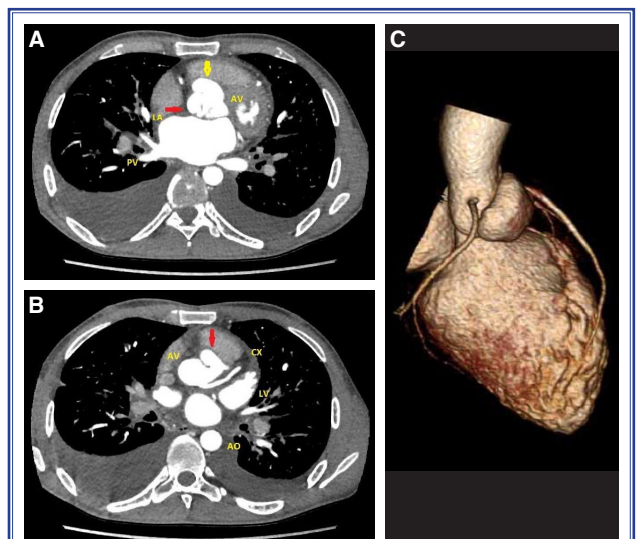


Figure 4. Cardiac computed tomography (CT). **(A)** Red arrow shows the pseudoaneurysm in the mitral-aortic intervalvular fibrosa. Yellow arrow indicates an aneurysm near the right ventricle. **(B)** Red arrow demonstrates the septal aneurysm extending to the right ventricle. **(C)** Three-dimensional cardiac CT illustrating the aneurysm extending from the left sinus of Valsalva to the left ventricular outflow tract.

Ao: Aorta; AV: Aortic valve; Cx: Circumflex artery; LV: Left ventricle (LV); PV: Pulmonary vein.

TEE showed 1 perforated leaflet of the BAV with vegetation 7x9 mm in size between the aortic leaflets (Fig. 2a), a P-MAIVF (Fig. 2b, yellow arrow), a septal aneurysm (Fig. 2b, white arrow), severe aortic regurgitation (Video 3*), and moderate mitral regurgitation. The aneurysm extending to the right ventricle was 25x10 mm in size and 8x5 mm toward the MAIVF.

Cardiac multi-detector computed tomography (Fig. 4c) confirmed a 18x38-mm aneurysm cavity with a 17-mm opening to the left ventricular outflow tract and a 12-mm opening to the left coronary sinus.

Aortic valve replacement (21-mm mechanical; St Jude Medical, Inc., St. Paul, MN, USA) and mitral valve ring annuloplasty (34-mm 3D ring; Sorin SPA, Milan, Italy/LivaNova PLC, London, England) surgery with repair of the aneurysm and pseudoaneurysm was performed 18 days later. The postoperative course was uneventful.

DISCUSSION

The literature contains few reports on the coexistence of more than one mechanical complications of infective endocarditis. In our case, multimodality imaging techniques were used to diagnose and confirm the pathology. These multimodality imaging techniques were also essential for the surgeons to be able to operate efficiently.

BAV increases the risk for cardiovascular complications like aortic valve dysfunction, aortic dissection, ascending aortic aneurysm, and IE. High rates of cardiovascular adverse events in adults with BAV were reported in 2 large-scale studies.^[2,3] The damage caused by aortic valve endocarditis can lead to the destruction or perforation of the leaflet, ring abscess, and perivalvular regurgitation; less commonly, the regurgitation itself may result in secondary damage to subaortic structures, such as the MAIVF and the anterior mitral leaflet.^[4] The MAIVF is more prone to trauma like IE because of its relatively avascular structure. Although the leading etiological causes of MAIVF aneurysms are infection and surgical trauma, aortic regurgitation jet is also a contributor to its formation.^[5] P-MAIVF should be differentiated from abscess of the aortic ring. However, in practice, it is very difficult to distinguish these 2 lesions. Han et al.^[6] reported that until imaging technology improves, the phenomenon of expansion in systole and collapse in diastole remains

the key point to distinguish P-MAIVF from aortic ring abscess. Ring abscesses are smaller, non-pulsatile, and show no inner flow on color-flow imaging.^[7]

Abscesses are more common than pseudoaneurysms. An abscess may progress to a pseudoaneurysm under the influence of systemic intravascular pressure. Once a pseudoaneurysm develops, it is prone to rupture. Such a rupture may occur in the pericardium, resulting in cardiac tamponade; in the left atrium, resulting in an eccentric jet of mitral regurgitation; or in the aorta, resulting in a fistula connecting the left ventricle to the ascending aorta.^[8]

IE can also lead to fistula formation. The Gerbode defect is characterized by a perimembranous ventricular septal defect between the left ventricle and the right atrium.

IE is associated not only with cardiac complications but also neurological, renal, musculoskeletal, and systemic complications related to infection, such as embolization, metastatic infection, and mycotic aneurysm.^[9]

An infected aneurysm is an aneurysm deriving from bacterial infection of the arterial wall. It is very rare compared with a noninfected aneurysm, but it is a very serious medical condition with a high probability of rupture. The incidence of infected aneurysm is approximately 0.9% to 1.3% among all cases of aortic aneurysm. *Staphylococcus aureus* and *Salmonella* species are the most common pathogens involved.^[10] Thrombosis, rapid enlargement, and even spontaneous regression are possible. Rupture carries an exceptionally high mortality risk.

Early diagnosis of aortic valve aneurysm is very important, as it may prevent further catastrophic complications. Surgery should be considered to prevent development of severe complications, even in asymptomatic patients.

**Supplementary video file associated with this article can be found in the online version of the journal.*

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Keywords: Bicuspid aortic valve; infective endocarditis; pseudoaneurysm.

Anahtar sözcükler: Biküspit aort; enfektif endokardit; psödoanevrizma.