Preoperative Demonstration of Aortocardiac Fistula Caused by Aortic Valve Endocarditis Using Real-time Three-dimensional Echocardiography — A Case Report

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Despite advances in diagnostic modality and treatment, infective endocarditis is still a life-threatening disease with significant morbidity and mortality. Endocarditis of native aortic valve may invade the perivalvular tissue, causing a severe valvular regurgitation, conduction disturbances, and less commonly, the intracardiac fistula. We here report a 52-year-old woman with infective endocarditis caused by *Staphylococcus capitis*. Echocardiographic examination revealed severely destructed aortic valve with perivalvular invasion, resulting in an aortocardiac fistula into the right atrium. The initial hemodynamic status of the patient was so stable that a full-course of antibiotic treatment for 4 weeks could be completed. However, just one day before the scheduled operation, sudden onset of free aortic regurgitation resulted in an acute decompensated heart failure and prompted the patient to an emergent operation. Accurate preoperative diagnosis by 3D-echocardiography made the emergent surgical intervention smooth and successful.

Key Words: Infective endocarditis • Aortocardiac fistula • 3D echocardiography

INTRODUCTION

Coagulase-negative *Staphylococcus*, known as a major etiology of endocarditis of the prosthetic valve, may still cause 3 to 8 percent of the endocarditis of native valve.[1] *Staphylococcus capitis* (*S. capitis*), a coagulase-negative staphylococcus, is a much less frequent cause of native valve endocarditis. Most of the cases reported in the literature could be successfully managed with medical treatment during the acute stage,[2] suggesting the low virulence of this organism. However, it may severely damage the valves, with ominous results. Endocarditis of native aortic valve is often associated with perivalvular invasion that could cause severe valvular insufficiency, conduction disturbances, and intracardiac fistula formation.[3,4] Acute deterioration in hemodynamics always decompensates the cardiac function and prompts the patient to a surgical emergency. Echocardiography plays an important role in the diagnosis of infective endocarditis. Recent technological advances of real-time three-dimensional (3D) echocardiography can shorten the time for image acquisition. When patients are uncooperative with respiratory distress or arrhythmia, this is important, and makes the images more accurate and reliable than those obtained from reconstruction 3D methods. The off-line analysis can provide precise anatomical information and allow the examiner to delineate the pathologic structure with appropriate planes of view, which may be impossible or difficult to obtain from the images acquisition in a conventional 2-dimensional echocardiography. During the treatment of infective

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endocarditis, any new clinical deterioration deserves immediate echocardiographic evaluation for determination of surgical strategy.

CASE REPORT

A 52-year-old woman presented with fever, chills and palpitation for one week. Her significant medical history included chronic hepatitis B, chronic glomerulonephritis, hypertension, and hypercholesterolemia. She had received regular hemodialysis via arteriovenous fistula over the left forearm since 5 months before. On admission, initial assessment revealed a body temperature of 36.8 °C, blood pressure of 120/90 mmHg, regular pulse with rate of 120 beats/min, and respiratory rate of 20 breaths/min. There was a grade III/VI systolic murmur with faint diastolic blowing component audible along the left sternal border, apex and aortic area. The remainder of the physical examination was unremarkable. The electrocardiogram showed sinus tachycardia with prolonged PR interval up to 232 ms. Laboratory data revealed a white blood cell count of 12,520/μL, hemoglobin level of 9.6 g/dL and platelet count of 315,000/μL. Transthoracic echocardiography (TTE) showed normal left ventricular and left atrial size with good systolic function. Large vegetations were noted over the aortic valve with heterogeneous echotexture over the aortic annulus. The aortic valve was destructed, with severe aortic regurgitation. Subsequent transesophageal echocardiography (TEE) also revealed those vegetations, attaching to and disrupting aortic valve with flailed leaflets (Figure 1), with severe aortic regurgitation. The perivalvular tissue was involved and complicated with a fistula from the infected noncoronary cusp (NCC) into the right atrium (RA), where the vegetations also grew (Figure 2). Color flow mapping demonstrated a left-to-right shunt from aorta to right atrium (Figure 3). The patient was treated with empirical intravenous vancomycin (500 mg every 3 days) because she was allergic to penicillin. Five sets of blood culture within 2 days showed a growth of *S. capitis*. These isolates were susceptible to oxacillin, clindamycin, vancomycin and gentamicin by the disk diffusion method. After van-

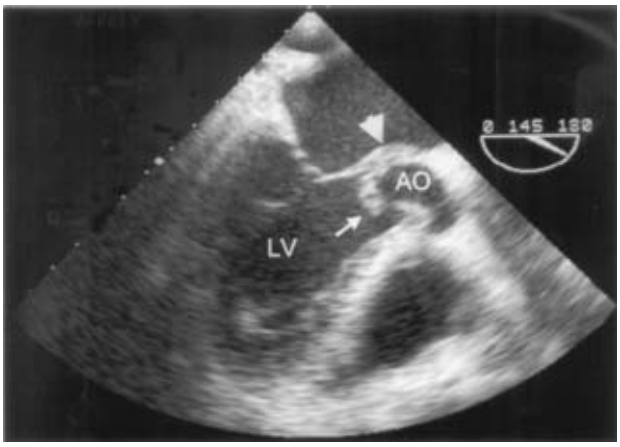

Figure 1. *Transesophageal image in long-axis view of the aortic valve showing flailed aortic valves (arrow) with vegetation and infectious extension to the perivalvular tissue (arrow head). AO, aortic ring; LV, left ventricle.*

Figure 2. *Transesophageal image in short-axis view of the aortic valve suspecting a fistula formation (arrow head) near noncoronary cusp with vegetation (arrow) in the right atrium (RA).*

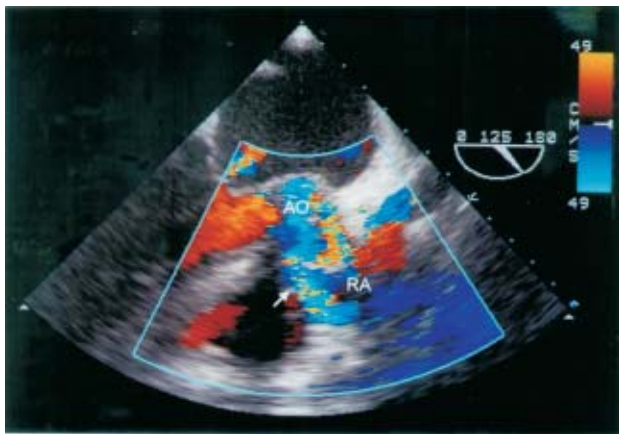

Figure 3. *Color flow map demonstrating a left-to-right shunt (arrow) from aorta (AO) to right atrium (RA).*

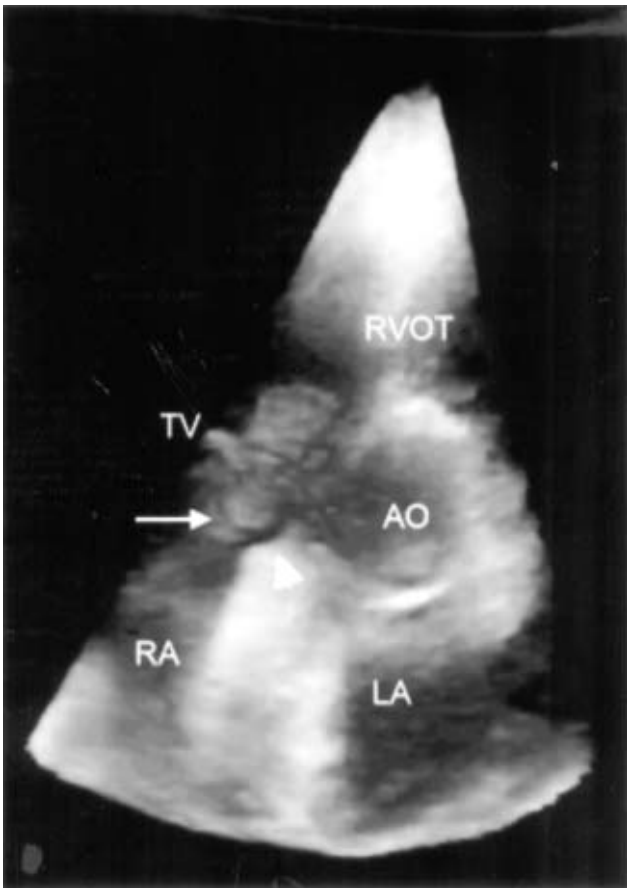

Figure 4. *Real-time three-dimensional echocardiography with short-axis projection showing an aortocardiac fistula (arrow head) beneath the noncoronary cuspid with vegetation (arrow) in the right atrium (RA). The tricuspid valve (TV) is spared from infection. AO, aortic ring; LA, left atrium; RVOT, right ventricular outflow tract.*

comycin was administered, all infectious parameters were improved. There was no evidence of immunologic or embolic complication during antibiotic therapy. The patient was scheduled for aortic valve replacement and fistula repair after 4 weeks of antibiotic therapy. The further real-time 3D echocardiography precisely demonstrated the anatomy of damaged structure one week before scheduled operation. The aortocardiac fistula occurred through the aortic annulus near NCC, sparing the tricuspid valve (Figure 4). Unfortunately, dyspnea with acute lung edema suddenly ensued one day before the scheduled operation. A repeat TTE showed flailed NCC, free aortic regurgitation and an obviously deteriorated left ventricle systolic function. The invasive arterial blood pressure monitoring showed a drop of diastolic blood pressure to around 40 mmHg that was similar to the pulmonary wedge pressure measured in the operation room before the emergent surgery. Although stationary in the initial 4-week course of medical treatment, sudden onset of fully valvular disruption can cause free aortic regurgitation, promptly decompensating the heart with acute lung edema. Emergent surgical debridement, repair of fistula and replacement of the aortic valve with bileaflet mechanical valve was successfully performed. The findings of echocardiography were confirmed at surgery.

DISCUSSION

Despite advances in diagnostic modality and treatment, infective endocarditis is still a life-threatening disease with significant morbidity and mortality.[5] The clinical manifestations of infective endocarditis are highly variable. In the clinical setting, endocarditis is diagnosed based on modified Duke criteria that combines echocardiographic, laboratory, and clinical findings.[6,7] Echocardiography is an essential component in evaluating patients with infective endocarditis. The echocardiographic assessment of vegetation, valvular structure, cardiac function and possible complications provides accurate diagnosis and optimal strategy for treatment.[8]

S. capitis, a coagulase-negative staphylococcus, is a normal flora of the skin, scalp, and forehead. The first description of native valve endocarditis caused by *S. capitis* dates back to 1992 by Juan et al.[9] There were at least 10 cases of endocarditis due to this organism reported since then.[2] Except two cases with mortality due to poor underlying disease, others had favorable clinical outcome with medical treatment, and two of them received elective valve replacement. Despite the low virulence of *S. capitis*, our patient had severe aortic valve endocarditis, highlighting that this microorganism still may have the ability to destroy cardiac valve.

In infective endocarditis, the spread of infection from its usual site on a cardiac valve to the perivalvular structure often occurs with aortic valve involvement.[4] Aortocardiac fistula is an uncommon complication. Infective endocarditis associated with perivalvular invasion and intracardiac fistula formation sometimes gives

rise to unpredictable clinical events. Even if infection is successfully controlled with antibiotics, sudden onset of decompensated heart failure secondary to increased severity of aortic regurgitation may occur. Serial echocardiographic examinations allowed us to early detect the cause for hemodynamic deterioration.

The recently developing real-time 3D echocardiography, possibly not an ideal imaging tool for initial assessment, is very helpful to construct the anatomy of lesion, guiding the treatment strategy according to the clinical assumption raised by conventional echocardiography. Real-time 3D echocardiography has many advantages. Firstly, the acquisition time for complete imaging is short, especially convenient when patients have respiratory distress or are uncooperative. Secondly, the real-time acquisition, although with a smaller angle of scanning, makes the images more accurate and reliable than those obtained from reconstruction 3D methods when the patients are unable to control breathing or have arrhythmia. In this case, we had also performed the full-volume 3D and 3D color echocardiography that reconstructed the imaging from four interrupted cardiac cycles during seven consecutive beats. The analysis of full-volume data sets showed results similar to the real-time imaging, however, with reduced resolution for larger scanning depth. The 3D color imaging, also obtained from reconstruction, was influenced by the limitation of Doppler technique in interrogation angles between ultrasound beam and flow direction. Copious color signals in the aorta and cardiac chambers hindered us from clearly demonstrating the blood flow from the fistula like in those images shown by TEE. Thirdly, the off-line analysis can provide precise anatomical information and allow the examiner to delineate the pathologic structure with appropriate planes of view which may be impossible or difficult to obtain from the images acquisition in a conventional 2-dimensional echocardiography. Thereafter and finally, the imaging can be acquired from the projection, which may be different from the views in analysis, with least acoustic attenuation to increase the signal acquisition and be suitable for the examiners' preferences.

In this case, the preoperative 3D echocardiography was demonstrated to provide a precise localization of fistula and a complete assessment of the extent of infection. 3D echocardiography clearly showed the aortocardiac fistula that penetrated the aortic annulus near the NCC and spared the tricuspid valve. The vegetation could be viewed with clear demarcation after establishing the anatomical relationship by 3D imaging. The fistula, difficult to be completely trace in conventionaly echocardiography, could be delineated with the entire tract by appropriate projection after post-processing. The vegetation detected in the RA was shown to be perivalvular extension from the aortocardiac fistula, not coexisting on the tricuspid valve. Therefore, the preoperative planning included surgical debridement, repair of aortocardiac fistula and replacement of the aortic valve without intervening the tricuspid valve. 3D echocardiography is a valuable diagnostic tool in complicated cases, providing refinements in the intervention strategy. Accurate preoperative diagnosis made the emergent surgery go smoothly.

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三維實時超音波診斷主動脈瓣心內膜炎併發主動脈右心房瘻管 — 病例報告

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儘管診斷與治療方法日新月異，感染性心內膜炎仍造成相當的罹病率與死亡率。發生於主動脈瓣的感染性心內膜炎，可侵犯瓣輪部位，造成嚴重的瓣膜逆流，併發心臟傳導異常及較少見的心臟瘻管。我們報告一位罹患 *Staphylococcus capitis* 感染性主動脈瓣心內膜炎的患者，經心臟超音波診斷出嚴重的主動脈瓣逆流併發侵犯瓣輪部，進而形成主動脈和右心房間的瘻管。雖然起初血行動力學穩定，完成抗生素四週的療程。但於瓣膜置換手術前一日,因突發加劇的完全主動脈瓣逆流造成急性心衰竭，必須進行緊急手術，幸而術前正確的三維實時心臟超音波診斷，提供施術者完整的病灶及併發症解剖資訊，使得緊急外科手術治療順利完成。

關鍵詞：感染性心內膜炎、主動脈右心房瘻管、三維實時心臟超音波。