Cyclic Intermittent Aortic Regurgitation of a Mechanical Bileaflet Aortic Valve Prosthesis: Diagnosis and Clinical Implications

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We describe a rare case of intermittent dysfunction of a bileaflet mechanical aortic valve prosthesis. Subvalvular pannus overgrowth on the inflow aspect of the prosthesis impeded normal closure of one leaflet causing severe aortic regurgitation, which occurred intermittently. The complementary role of transthoracic and transesophageal echocardiography and cine-fluoroscopy in the diagnosis of this case is described. (J Am Soc Echocardiogr 2007; xx:xxx.)

CASE REPORT

A 47-year-old woman was admitted to our hospital because of mild dyspnea on effort. She had undergone aortic valve replacement with a 21-mm Sorin Bicarbon (Saluggia, Vercelli, Italy) 14 years before because of aortic valve stenosis and regurgitation caused by rheumatic valve disease. Since then she had received adequate anticoagulant therapy even though the last international normalized ratio values were slightly low (few values between 1.5 and 2). Her clinical condition had been fairly good until she noticed exertional dyspnea 3 months before (New York Heart Association functional class II). Her blood pressure was normal (130/70 mm Hg) and pulse rate was 70/min (in sinus rhythm). Auscultation revealed a grade 2/6 systolic murmur consistent with aortic stenosis and normal prosthetic valvular clicks. No signs of heart failure were present.

Transthoracic echocardiography (TTE) was performed and showed apparent normal leaflet motion (however, quantitative assessment was not achieved). No distinct mass of abnormal echoes attached to the prosthesis was identified whereas a suggestion of an abnormal fibrous structure at the level of the left ventricular outflow tract was posed. The left ventricle was slightly enlarged with a normal ejection fraction (55%). On Doppler examination mean (44 mm Hg) and peak (77 mm Hg) aortic gradients were higher than previous values recorded 1 year before and aortic valve area was 0.8 cmq (0.5 cm2/m2; calculated with the continuity equation). The right ventricle and systolic pulmonary pressure (29 mm Hg) were normal. Physiologic diastolic prosthetic jets were identified inside the prosthesis and no paraprosthetic jet was recorded. However, severe aortic regurgitation occurred intermittently every 5 to 8 beats. Multiplane transesophageal echocardiography (TEE) was, therefore, performed and confirmed TTE data showing normal systolic motion of two aortic leaflets whereas diastolic motion of the leaflets and closing angles were not defined. Severe aortic regurgitation occurred intermittently as from the transthoracic approach and TEE allowed clear visualization of a dense mass of echoes located immediately below the aortic prosthesis in the outflow tract. This structure appeared to be circular in the different TEE views without direct contact with the prosthetic leaflets. No masses inside the prosthetic valve (indicative of thrombus formation or pannus inside the valve) or mobile echoes or vegetations attached to the prosthetic disks or ring were visualized. Because both TTE and TEE failed to identify opening and closing angles of the two leaflets, cine-fluoroscopy (CF) was performed. This examination allowed proper visualization of the disks showing normal systolic motion throughout the examination (constant complete opening angle of 18 degrees) and intermittent incomplete closure of one of the two every 5 to 8 beats (130- vs 140-degree diastolic closure angle). The final diagnosis was pannus formation causing moderate aortic stenosis (as a result of subvalvular obstruction without interference with systolic disk motion) and severe intermittent aortic regurgitation (as a result of intermittent pannus interference with disk closure of one disk) and based on these data we referred the patient for surgery. At surgery, inspection and visualization of the structure with a cardiac optical fiber device confirmed echocardiographic data showing a circular pannus formation below the aortic prosthesis. The prosthetic valve leaflets were not involved and the disk motion appeared unrestricted. Figures 1, 2, and 3 and Videos 1, 2, and 3 show the images of TTE, TEE, and CF demonstrating the

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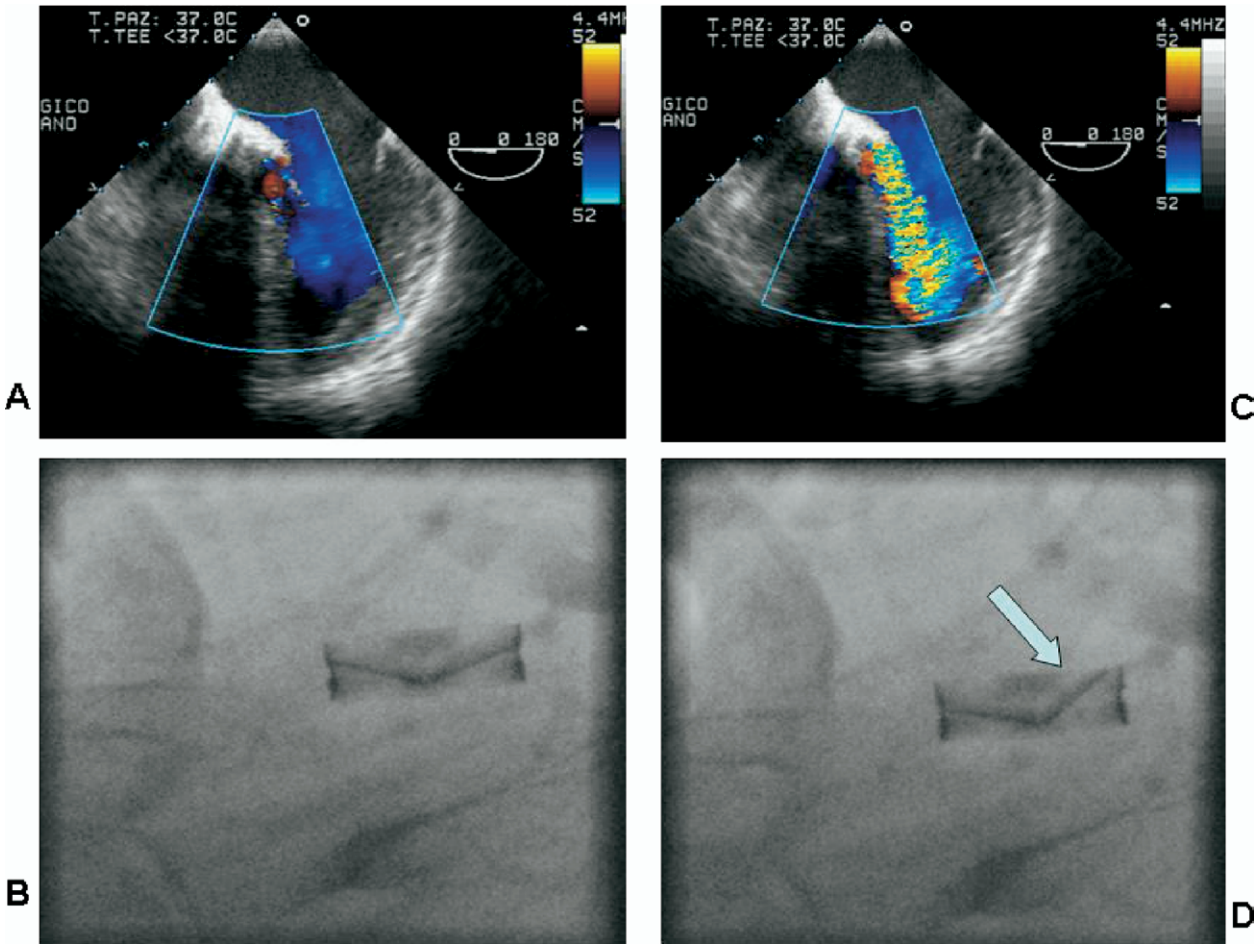

Figure 1 Apical 4-chamber, transesophageal echocardiogram showing intermittent severe aortic regurgitation associated with CF demonstrating intermittent diastolic of one leaflet: Doppler color shows trivial aortic prosthetic regurgitation (**A**), intermittent severe aortic prosthetic regurgitation (**C**). CF: normal diastolic closure of both prosthetic leaflets (140°) (**B**), intermittent incomplete closure (130°) of one of the two leaflets (arrow) (**D**).

complementary role of these techniques in this uncommon diagnosis.

Pannus was fully excised and the aortic valve was replaced with a Carpentier biological valve. Histologic examination revealed a structure of fibroconnective tissue consistent with pannus formation.

DISCUSSION

Mechanical prosthetic valve dysfunction may be the result of thrombosis or pannus formation, and even though the most common underlying pathology is thrombus formation, pannus alone or pannus in combination with thrombosis are not rare conditions.[1,2] The distinction between thrombus formation and pannus as the underlying cause of valve is essential[3] and has important clinical, prognostic, and therapeutic implications because thrombolytic therapy has emerged as an alternative to reoperation.[4-6] In this regard clinical presentation may vary between an insidious illness with few and nonspecific symptoms and a severely impaired hemodynamic condition rapidly progressing to shock and death. Therefore, a quick, effective, and easy noninvasive diagnostic method is mandatory. TTE, TEE, and CF are complementary methods to make a rapid, complete, and correct diagnosis including valve morphology, presence of thrombus or pannus, characteristics of prosthetic flow (gradients, valve area, intraprosthetic or periprosthetic regurgitation), and leaflet motion.[7-9]

To our knowledge this is the first report concerning prosthetic aortic valve obstruction without systolic alteration of leaflet motion associated with intermittent severe aortic regurgitation related to perivalvular pannus ingrowth. Pannus did not extend to the tilting disk or the hinge and no interfer-

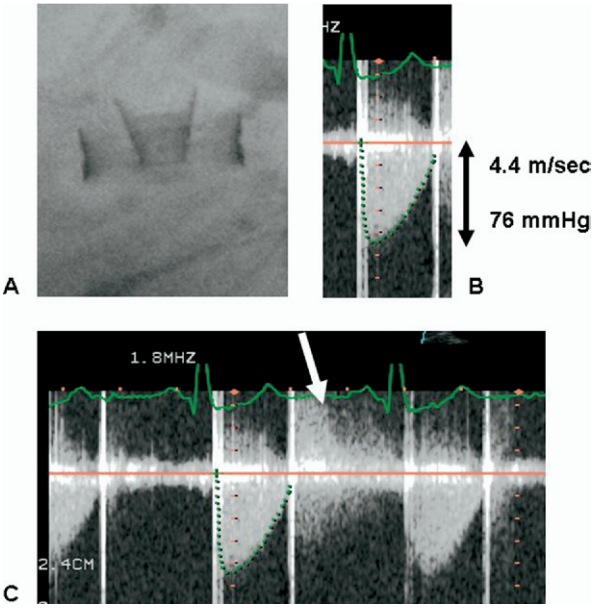

Figure 2 CF shows normal opening prosthetic leaflets (**A**); transthoracic echocardiogram continuous wave Doppler showing elevated systolic transprosthetic velocity, corresponding to an elevated gradient (**B**), and intermittent diastolic insufficiency (arrow) (**C**).

ence with systolic motion of the two leaflets occurred. Thus, continuous Doppler showed a high transprosthetic gradient even though the two mechanical leaflets opened appropriately as documented qualitatively by echocardiography and quantitatively by CF. On the contrary the diastolic motion of one of the two leaflets was markedly impaired intermittently causing severe aortic regurgitation as shown by color-coded echocardiography. In this regard even though 2-dimensional echocardiography failed to visualize closing angles of the prosthetic valve, CF clearly demonstrated prosthetic dysfunction (intermittent incomplete closure of one of the two leaflets). This is in complete agreement with recent reports showing feasibility and diagnostic accuracy of quantitative assessment of mechanical leaflet motion by TTE and TEE in comparison with CF. TTE and TEE have, in fact, high feasibility in the identification of opening and closing angles for mitral prosthesis while they correctly identify aortic leaflet motion (particularly in bileaflet valves) in a minority of cases.[10] In our case, therefore, CF was very useful in detecting abnormal closing angles and 2-dimensional echocardiography (particularly TEE) was able to visualize pannus ingrowth below the valve, thus, posing indication for surgery.

Intermittent closure alteration of mechanical disks in mitral prosthesis is a well-known phenomenon[11] as a result of varying degrees of intermittent obstruction to the disk opening (caused both by pannus or thrombus) related to cyclic changes in atrial-ventricular gradients. On the contrary intermittent aortic regurgitation in mechanical aortic valves is a very rare condition[12] and cyclic restriction of leaflet motion may not be easily explained by hemodynamic cyclic changes while it could be related to pannus interference that impedes normal closure of disks in an intermittent way. This may be related to pannus, which had grown at a very short distance from the valve and narrowed the left ventricular outflow tract.

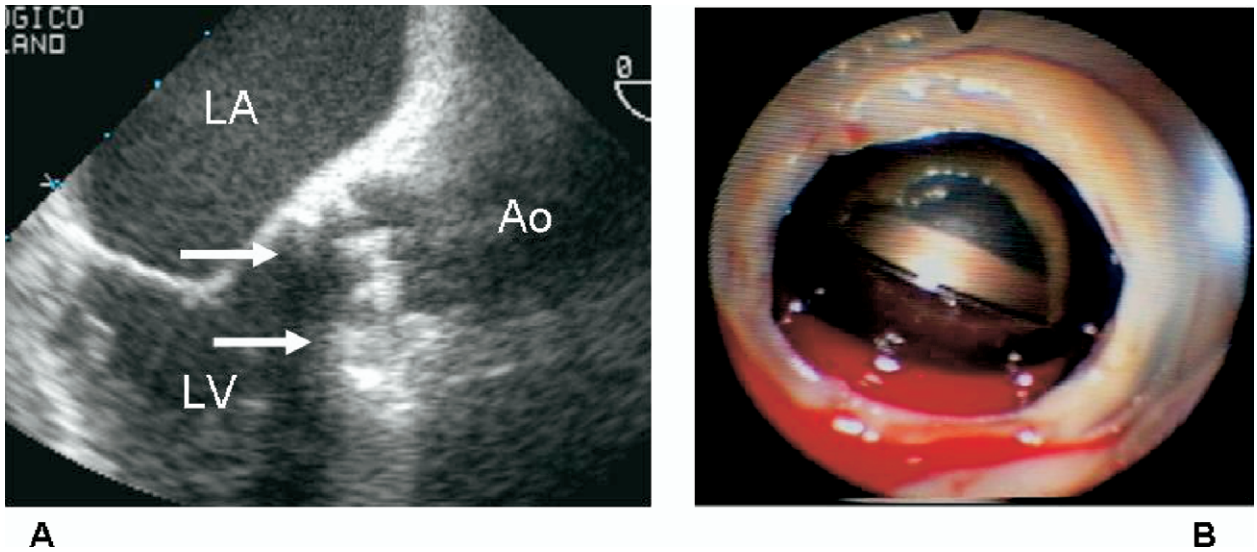

Figure 3 Echocardiographic and anatomical correspondence of prosthetic pannus. Transesophageal echocardiography showing dense mass of echos located immediately below the aortic prosthesis in the outflow tract (arrows) (**A**). Pannus visualized by an intracardiac fibroscope at surgery (**B**).

Conclusion

Aortic prosthetic valve obstruction with normal systolic motion of the tilting disk associated with intermittent diastolic regurgitation as a result of subvalvular pannus interference with the closing position of the leaflets is a rare cause of prosthetic dysfunction in the late postoperative period. TTE, TEE, and CF are quick, effective, and complementary diagnostic tools to diagnose this rare condition.

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