MINI-FOCUS ISSUE: TRANSCATHETER INTERVENTIONS

CASE REPORT: CLINICAL CASE

Caged-Ball Mitral Prosthesis Explanted After 50 Years

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ABSTRACT

We report a unique case of a Starr-Edwards prosthesis (model 6310, cloth covered) implanted in the mitral position by Christian Barnard that was successfully explanted and replaced after 50 years, the longest period free from valve dysfunction ever reported. Reoperation also included replacement of the native aortic valve combined with tricuspid valve annuloplasty. (Level of Difficulty: Beginner.) (J Am Coll Cardiol Case Rep 2021;3:884–7) © 2021 The Authors. Published by Elsevier on behalf of the American College of Cardiology Foundation. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

HISTORY OF PRESENTATION

A 60-year-old white woman, in good clinical condition during the past few years, was admitted for the onset of exertional dyspnea and palpitations. She had been febrile (38.2°C) for 2 days. Physical examination revealed an arrhythmic pulse, a mean heart rate of 118 beats/min, regular arterial pressure (systolic blood pressure 110 mm Hg, diastolic blood pressure 70 mm Hg), and normal oxygen saturation. At cardiac auscultation, a holosystolic, rumbling, Levine grade 3 murmur radiating to the left armpit and not described previously was heard. A diastolic, rolling, Levine grade 2 murmur was also detectable at the apex. Thoracic auscultation revealed basal bilateral pulmonary rales. As a caged-ball Starr-Edwards mitral prosthesis holder, she was on oral anticoagulation therapy. Notably, the international normalized ratio (INR) level was 2.7 on arrival, and the patient maintained good adherence to warfarin. Atrial fibrillation was documented by the electrocardiogram. Echocardiography revealed a normal ejection fraction, no significant variation in the mitral transprosthetic gradient, and a perivalvular leak originating from the posteromedial region of the prosthetic ring.

LEARNING OBJECTIVES

- To be able to correctly handle follow-up of patients with prosthetic valves characterized by both high durability and non-negligible rates of various complication.
- To understand the role of echocardiography in the prompt identification of complications and directing the surgical strategy.

PAST MEDICAL HISTORY

The woman’s past medical history included rheumatic heart disease, diagnosed at the age of 5 years and resulting in the development of severe mitral
stenosis. The 1960s were pioneering for cardiac surgery in Europe, and access to cardiac surgical treatment was very difficult in Italy. Her brother, a navy captain who frequented the routes to Africa, had heard about cardiac surgery in South Africa and of Christian Barnard, who became famous at that time for performing the first heart transplantation. After consult with family members, she conducted a long journey, together with her brother, and was addressed to Barnard’s management. She underwent mitral valve replacement with a caged-ball Starr-Edwards mitral prosthesis at the Department of Surgery of the University of Cape Town in South Africa when she was 10 years old. Afterward, the patient enjoyed good health, got married when she was 24 years old, and completed 2 pregnancies. Her subsequent medical history was unremarkable, and she did not even experience one of the most common inconveniences of this type of valve, the noise. The patient scrupulously followed oral anticoagulation therapy, keeping her INR between 2.5 and 3.5.

DIFFERENTIAL DIAGNOSIS

Taking into account the patient’s clinical history and presentation, the following conditions should be considered in the differential diagnosis.

PROSTHETIC VALVE THROMBOSIS. Inadequate anticoagulation or an acquired lupus anticoagulant may have led to mechanical mitral prostheses thrombosis. The therapeutic INR level at admission, her good adherence to oral anticoagulation therapy, and the normal echocardiographic transvalvular gradient allowed us to exclude this condition.

PANNUS. Fibrous tissue overgrowth around the prosthetic valve can lead to pannus formation. Increased transprosthetic gradients and the echocardiographic identification of masses, together with the long time elapsed from her surgery, are consistent with the possible formation of a pannus.

PROSTHETIC VALVE INFECTIOUS ENDOCARDITIS. An episode of fever reinforced the suspicion of prosthetic valve infective endocarditis. Serial blood cultures, and transthoracic and transesophageal echocardiography to identify vegetations should be performed.

PROSTHETIC VALVE DEHISCENCE. As 50 years had passed since implantation of the prosthetic mitral valve, and considering the valve structure, which consisted of the housing system, caged ball, and bulky sewing ring, prosthetic valve dehiscence should be considered. Having found a perivalvular leak originating from the postero-medial region of the prosthetic ring, a transesophageal echocardiographic examination should be done to better clarify the diagnosis.

INVESTIGATION

Echocardiography revealed a perivalvular leak originating from the postero-medial region of the ring. Left ventricular dimensions and ejection fraction were normal. The left atrium was moderately dilated. The right ventricle was enlarged, with a mild reduction of systolic function. The tricuspid annulus was dilated, and severe functional tricuspid regurgitation was detected. Pulmonary artery systolic pressure, estimated at 44 mm Hg, was slightly increased. Aortic valve deterioration was appreciated due to diffuse calcification, and cusp remodeling inducing moderate aortic regurgitation. Transesophageal echocardiography revealed a heavily calcified mitral annulus and confirmed the presence of partial posterior prosthetic ring detachment with severe perivalvular leak. Normal ball excursion within the cage was observed; transprosthetic mean gradient was 5.8 mm Hg. No vegetations or pannus were appreciated.

Unfortunately, the ultrasound machine on which the images were stored was recruited and formatted for use in the coronavirus disease 2019 intensive care unit, and echocardiographic images could not be retrieved.

At the radioscopic evaluation, the Starr-Edwards valve showed normal ball excursion (Figures 1A and 1B, Video 1). Significant coronary artery disease was ruled out by coronary angiography. Left ventriculography was not performed. Blood cultures and markers of inflammation were negative.

MANAGEMENT

The patient underwent double-valve replacement and tricuspid annuloplasty through a standard median sternotomy and cardiopulmonary bypass at a flow rate of >2.4 L/m² under moderate hypothermia (32°C). Extracorporeal circulation time and aortic clamping time were 180 and 155 min, respectively. The Starr-Edwards caged-ball prosthesis (model 6310) was explanted, and the mitral annulus was decalcified. It was replaced with an On-X 25/33-mm valve prosthesis (CryoLife). In addition, the aortic valve was replaced with a bileaflet prosthesis (LivaNova Bicarbon Fitline, 19 mm) and tricuspid annuloplasty was performed with a 28-mm Physio Ring (Edwards Lifesciences). The post-operative course was uneventful.

At macroscopic analysis, no impairment of ball excursion of the mitral prosthesis was observed.
The surfaces of both the sewing ring and the valve cage were partially and asymmetrically endothelialized. Moreover, sewing ring deformation due to heavy calcification at the point of detachment was found. Surgical inspection confirmed the absence of pannus. Pre-discharge echocardiography confirmed normal gradients and the absence of perivalvular leaks of both the mitral and aortic prostheses. The patient was discharged in good clinical condition.

DISCUSSION

We report a unique case of a Starr-Edwards ball valve implanted in the mitral position in 1969 by Christian Barnard that required replacement for partial prosthesis dehiscence and cardiac failure in the context of moderate aortic regurgitation and severe tricuspid regurgitation with tricuspid annulus dilation (1).

There are 3 main peculiarities of the present case:

1. The Starr-Edwards ball valve lasted, without complications, for 50 years, the longest period free from valve dysfunction ever reported.
2. The caged ball was made of steel, and its movement into the cage was normal. Dysfunction occurred due to mitral prosthetic valve dehiscence.
3. Reoperation occurred in a complex context that included not only replacement of the prosthetic valve in the mitral position, but also aortic valve replacement and tricuspid valve annuloplasty.

Structural damage to the explanted valve was due only to heavy calcification of the sewing ring.

The Starr-Edwards valve was the first mechanical valve prosthesis successfully implanted in the mitral position in humans. The ball was built using both steel and silicone. Despite considerable steric hindrance leading to higher than physiological transvalvular gradients, these valves ensured a sufficient hemodynamic profile and safe durability. However, a non-negligible complication rate was reported,
depending on the time of implantation. The most common causes of valve dysfunction are lipid absorption into the ball in early silicone rubber balls and pannus tissue overgrowth. Moreover, changes in the valve profile determined forced lateral flow and further increments of transvalvular gradients. These morphological and functional changes lead to increased hemolysis (2). Pannus tissue overgrowth is another frequent complication resulting in prosthetic dysfunction due to reduced ball excursion or detachment of the sewing ring.

In the present case, the steel composition of the caged ball allowed preservation of the morphological profile and normal movement into the cage. Furthermore, Starr-Edwards valve complications were related to endocarditis, aortic wall injury, or left ventricular posterior wall damage due to implantation position. Fracture or systemic embolization have rarely been reported. The incidence of the aforementioned problems remained persistent, although several engineering modifications were made over the years to reduce or prevent complications. Tilting disc and bileaflet mechanical prostheses with newer designs and better performance were subsequently introduced. Notwithstanding this, Starr-Edwards valves were largely used, showing good durability. Survival rates of patients undergoing mitral valve replacement were reported to range between 51.0% and 75.0%, 23.0% and 78.9% and 87.0% at 10, 20, and 30 years, respectively (3, 4). Freedom of patients from reoperation for mitral valve replacement was reported to be 61.0% and 8.0% and 33% at 10, 20, and 30 years, respectively (5).

In the present case, the reason for reoperation was the onset of signs and symptoms of heart failure along with echocardiographic evidence of new paravalvular leak. Intraoperative evaluation confirmed heavy calcification involving the sewing ring and leading to partial mitral prosthetic valve dehiscence. We hypothesize that late onset of dehiscence is related to the slow development of the bulky calcified lesion.

In this context, echocardiography remains the first-line imaging tool for the assessment of prosthetic valves. If valve dysfunction is detected, closer clinical and echocardiographic follow-up is indicated.

**FOLLOW-UP**

The 6-month follow-up was uneventful. The patient was in atrial fibrillation with good heart rate control under beta-blocker therapy and reported improved exercise tolerance. Transthoracic echocardiography revealed a normal left ventricular ejection fraction. No paravalvular leaks were detected in both the mitral and aortic prosthetic valves. Transprosthetic gradients were within the normal range. No signs of pulmonary hypertension were detected.

**CONCLUSIONS**

To the best of our knowledge, we describe the latest case ever reported of explantation of a Starr-Edwards valve prosthesis in the mitral position before the onset of complications. Of note, despite its 50-year duration, no structural damage to the cage and ball mechanism was detected, confirming the exceptional durability of this “ancient” mechanical prosthetic valve.

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**KEY WORDS** aortic valve, echocardiography, mitral valve

**APPENDIX** For a supplemental video, please see the online version of this paper.