A successful repair of pentacuspid aortic valve

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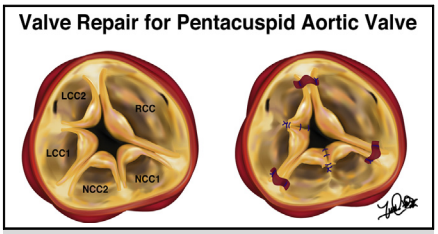


Repair of pentacuspid aortic valve.

CENTRAL MESSAGE

Complex repair of a pentacuspid aortic valve by tricuspidization.

See Commentary on page 73.

Video clip is available online.

Pentacuspid aortic valve (PAV) is an extremely rare congenital cardiac anomaly.[1] Simonds[2] first described PAV in 1923; subsequently, 8 cases, including 5 treated with aortic valve replacement, have been reported. Aortic valve replacement is the standard treatment for PAV-related aortic regurgitation (AR); there have been no reports of aortic valve repair in PAV.

We present new insights into PAV repair using tricuspidization involving resection of the rudimentary commissure, and double plication of the annuloventricular junction (AVJ) and sinotubular junction (STJ).

CASE REPORT

A 52-year-old woman was admitted because of AR and left ventricular dilation. Transthoracic echocardiography revealed severe AR. The left ventricular diastolic diameter was 66 mm and ejection fraction was 62%. Transesophageal echocardiography and computed tomography revealed PAV (Figure 1). The diameters of the AVJ, sinus of Valsalva, and STJ were 24 (index: 15.3 mm/m^2), 30, and 29 mm, respectively. The mean and peak pressure gradients were 11 and 20 mm Hg. The patient's body surface area was 1.57 m^2.

OPERATIVE PROCEDURE

After cardiac arrest, the aorta was divided 2 cm above the STJ. The aortic valve had 5 leaflets: 4 equal-sized cusps and 1 large cusp (Figure 2, *A*). Rudimentary commissures were found in the left coronary cusp (LCC) and noncoronary cups (NCC), separating the LCC and NCC into 2 equal cusps (LCC1, LCC2, NCC1, and NCC2). The heights of

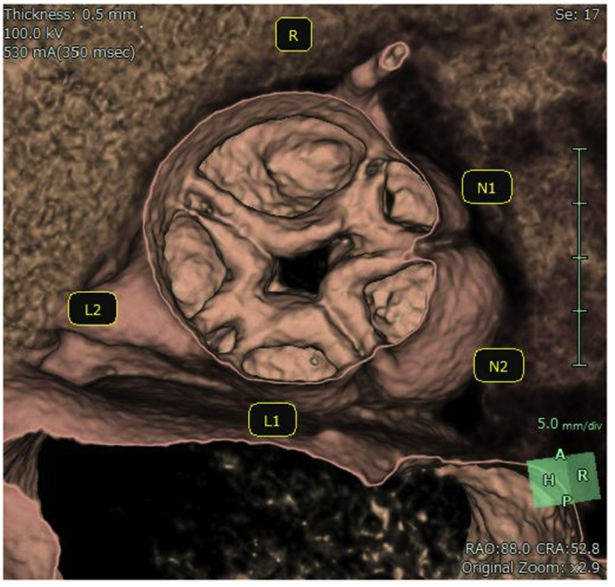


FIGURE 1. Preoperative computed tomography. Aortic valve consisted of 5 leaflets: 2 small left coronary cusps, 2 small noncoronary cusps, and 1 relatively large right coronary cusp. *L1/L2*, Left coronary cusp 1 or 2; *N1/N2*, noncoronary cusp 1 or 2; *R*, right coronary cusp.

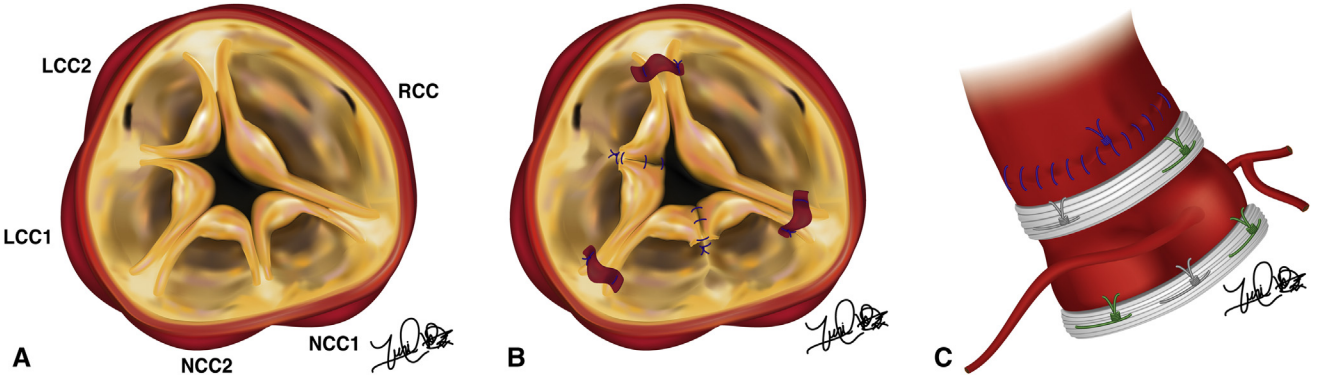
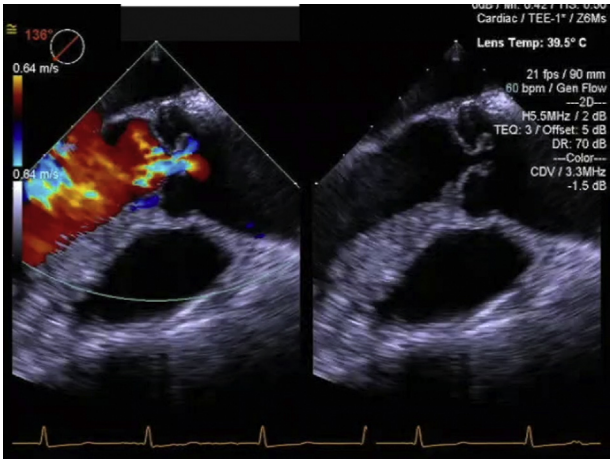


FIGURE 2. Operative schema. A, A rudimentary commissure was found in the left coronary cusps (*LCC*) and noncoronary cusps (*NCC*) that separated LCC and NCC into 2 equal cusps (LCC1/LCC2 and NCC1/NCC2). B, The rudimentary commissures were removed and translocated to a position lower in the nadir level for each cusp by suturing 2 small cusps. C, Aortoventricular junction (AVJ) plication and sinotubular junction (STJ) plication using external bands made of Woven graft. AVJ diameter of 20 mm was calculated by reported recommended AVJ index of 13.3 mm/m^2 and body surface area of 1.57 m^2; 13.3 × 1.57 = 20.9 mm. STJ diameter of 24 mm was calculated by AVJ; AVJ × 1.2 (assuming normal ratio of AVJ: STJ). *LCC1/LCC2*, Left coronary cusp 1 or 2; *NCC1/NCC2*, noncoronary cusp 1 or 2; *RCC*, right coronary cusp.

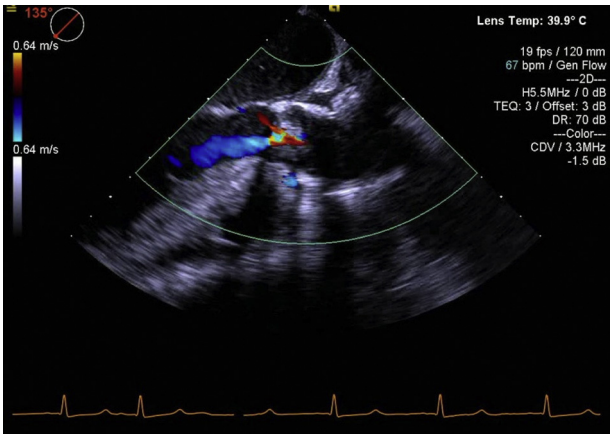
all commissures were similar, although the cusp nadir of LCC1, LCC2, NCC1, and NCC2 was shallower than that of the right coronary cusp (RCC). The 4 small cusps had mildly thickened and calcified free margins. The geometric heights of the RCC, NCC1, NCC 2, LCC1, and LCC2 were 19, 20, 15, 20, and 15 mm. Further, the AVJ and STJ were 24 and 26 mm, respectively.

The aortic root was dissected to the AVJ and secured with an external 5-mm-wide woven graft band (Japan Lifeline Co Ltd). The band was reinforced with five 3-0 Nespolene pledgeted sutures (Alfresa Pharma, Co) in the AVJ on the horizontal plane of each commissure. The AVJ was plicated to 20 mm. To create 3 symmetric cusps, the rudimentary commissures (LCC1/LCC2 and NCC1/NCC2) were incised, and the leaflet was translocated to a lower position at the nadir of each cusp using interrupted sutures (LCC1 + LCC2 and NCC1 + NCC2) (Figure 2, *B*). Central plication was added for prolapse of the neo-NCC using two 6-0 polypropylene interrupted sutures. Reinforcement with an autologous pericardium strip using a 5-0 polypropylene mattress stitch was applied for each commissure.

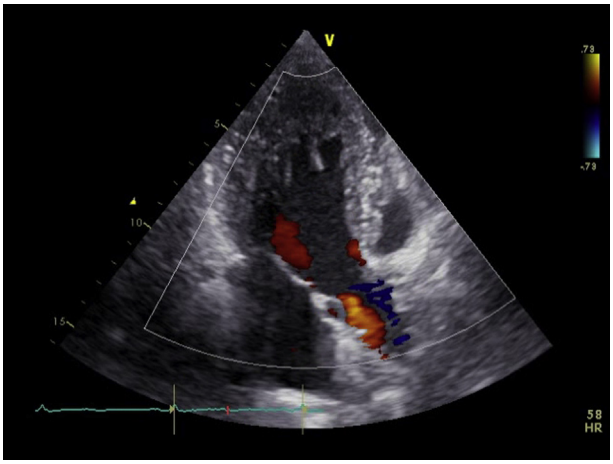
STJ plication was also performed using an external 7-mm-wide woven graft band. The band was sutured using three 3-0 Nespolene spaghetti sutures at the top of each commissure and another suture at the top of LCC1/LCC2 commissure. The STJ was then plicated to 24 mm. Distal anastomosis was completed after confirming the competency of the aortic valve (Figure 2, *C*). Intraoperative



VIDEO 1. Under cardiac arrest, the aortoventricular junction plication was performed using an external band with 5 reinforced 3-0 Nespolene pledgeted sutures. The rudimentary commissure was detached and translocated to a position lower in the nadir level for each cusp. Then, central plication and commissural plication using autologous pericardium strip were added. After confirming the competency of the aortic cusp, sinotubular junction plication and distal anastomosis were completed. Video available at: https://www.jtcvs.org/article/S2666-2507(22)00220-6/fulltext.



VIDEO 2. Transesophageal echocardiography at discharge. Video available at: https://www.jtcvs.org/article/S2666-2507(22)00220-6/fulltext.



VIDEO 3. Transthoracic echocardiography at 3 years. Video available at: https://www.jtcvs.org/article/S2666-2507(22)00220-6/fulltext.

transesophageal echocardiography demonstrated central mild residual AR and an adequate aortic valve area of 1.5 cm^2 (Video 1). The patient's postoperative course was uneventful, and annual transthoracic echocardiography follow-up showed mild central AR at 3 years (Figure E1, Videos 2 and 3).

DISCUSSION

The morphologic variations of PAV remain unclear. Kuroki and colleagues[3] reported different valve configurations with a relatively large LCC and RCC and 3 small divided NCCs. In the present case, the PAV comprised 2 small LCCs, 2 small NCCs, and 1 relatively large RCC.

Recently, repair of regurgitant uni/bicuspid aortic valves has become an increasingly used alternative to replacement in the treatment of severe AR.[4] Prosthetic aortic valve replacement was suboptimal in this younger patient, considering the need for continuous anticoagulation and lifelong risk of bleeding and thromboembolism with a mechanical valve or expected structural valve deterioration with a bioprosthetic valve. We believe aortic valve repair was justified in this patient as it showed acceptable early- and mid-term outcomes.

The preoperative AVJ index was large relative to the restricted supernumerary cusp as reported by de Kerchove and colleagues,[5] who recommended annuloplasty for AVJ of $\geq$28 mm (index: 13.3 mm/m^2). Regarding the surgical approach for tricuspidization, the angles of the three fashioned commissures should be considered with the depth and height of the neoleaflets relative to the annulus. In this case, 2 relatively small LCCs and NCCs could be plicated into a neo-LCC and NCC, similar to the normally functioning tricuspid aortic valve. Aortic valve repair was a potentially more attractive option, given that it can provide sufficient durability and/or a favorable bridge to prosthetic valve replacement. This study was reviewed and approved by the institutional review board (institutional review board/ethical review board number [1144] and date [October 13, 2021]). Informed written consent was granted.

CONCLUSIONS

PAV repair provided a satisfactory outcome. Tricuspidization of PAV may be feasible in younger patients.

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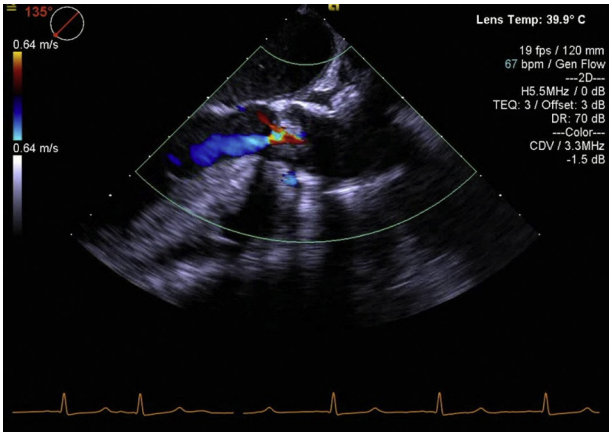

FIGURE E1. Preoperative transthoracic echocardiography (*TTE*), TTE at discharge, and TTE at 3 years. Left ventricular diastolic diameter (*LVDd*): preoperative 66 mm, at discharge 52 mm, and at 3 years 52 mm. Left ventricular ejection fraction (*LVEF*): Preoperative 62%, at discharge 49%, and at 3 years 72%, Mean pressure gradient: preoperative 11 mm Hg, at discharge 17 mm Hg, and 3 years 16 mm Hg, Peak pressure gradient: preoperative 20 mm Hg, at discharge 29 mm Hg, and 3 years 30 mm Hg.