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Review Article

Facing with Limited Options in the Failing Aortic Valve of the Young: Validity of Aortic Cusp Extension Valvuloplasty and Valve Reconstruction

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Review

Available treatment options for Aortic Insufficiency (AI) or Aortic Stenosis (AS) include various techniques of surgical aortic valvuloplasty and Aortic Valve Replacement (AVR) [1-6]. It is uncertain whether AVR is the optimal course of therapy, especially in children and infants in whom Left Ventricular (LV) function has to be preserved for a longer life span. Aortic Valve (AoV) remodeling with Cusp Extension Valvuloplasty (CEV) techniques aim at restoring the morphologic characteristics of the valve where simple techniques of commissuroplasty or isolated cusp repair do not suffice to restore competency of valve apparatus. [7-10].

Growth potential and availability of pulmonary auto graft are major advantages of the Ross procedure, especially in the pediatric population. Despite being technically demanding, CEV may be particularly advantageous in infants and children relative to other alternatives, including freedom from reoperation and need for anticoagulation when mechanical prostheses are used, or complications inherent to the Ross procedure. Indications for AoV surgery are often based on experience with adult patients. In a pediatric patient any delay may be detrimental because of the longer life span. In addition, AI after Balloon Valvuloplasty (BV) can be particularly deleterious during infancy owing to its disruptive nature on both valve planes and cusp integrity [11,12]. Durable CEV techniques may sway the timing for intervention toward earlier surgery [13-15].

Advances in surgical techniques, myocardial protection, and streamlined perioperative management have contributed to a remarkable improvement in early and late mortality after AoV surgery in children. Detailed echocardiographic evaluation of the valve's anatomy and flow kinetics have made repair applicable and safe.

Without precluding future replacement strategies, CEV techniques preserve growth potential of the native tissue with no need for anticoagulation. Excluding lesions (isolated commissural fusion or single cusp prolapsed or tear) for which standard non-CEV techniques have proved durable [16] CEV can be applicable choice in patients with congenital or acquired AI, post-BV AI or/and AS plus AI and any degree of de conditioned bicuspid AoV with worsening AI, cusp or combined annular and cusp structural failure. As recommended [13,14,17] patients selected for CEV have usually an echocardiographic profile consistent with aortic annulus z value of -1.5 or greater without LV or mitral annulus hypoplasia, and several other echocardiographic Doppler derived indices that determine severity of valve dysfunction and timing for ACEV. These include (1) regurgitant jet-to-annulus diameter ratio of 35% or greater or progressive increase LV end-diastolic dimension (LVEDD)z value of -2.5 or greater, (2) peak instantaneous gradient of 40 mm Hg or greater associated with progressive LV hypertrophy, or (3) mixed lesions with a variable degree of AI and AS. Tricuspidization can be added in bicuspid AoV with eccentric opening, in cases in which raphe were well developed, and patients with aortic valve of limited cusp mobility away from the hinge point. Compromised mobility of cusp at the hinge point, extensive and multiple cusp dysplasia, and extensive commissural fibrosis

extending to the coronary ostia were relative contraindications.

The main principles for CEV are summarized here

After median sternotomy autologous pericardium treated with a 0.625% glutaraldehyde solution, bovine pericardium orthin polytetrafluoroethylene membrane for the cusp reconstruction is chosen. Aortobicaval cardiopulmonary bypass with moderate hypothermia (32°C), antegrade and retrograde myocardial preservation is used. After an oblique aortotomyincision each cusp is evaluated as to the extent of tissue deficiency, the shape, and the irregularities of the free edge. Only each cusp's thickened free margin and body are thinned out, leaving its base and unaffected body intact. When tricuspidization is needed, the fused cusp is cut at the raphe precisely to the aortic wall. Any sub commissural fused tissue is released. A pericardial extension is then fashioned to fit the specific architecture of each cusp, but slightly oversized in depth (10% to 15%) and length (up to 25%). Continuous 5-0 or 6-0 polypropylene suture is used. The sutures are placed from the cusp's center toward each commissure. The suture line on the pericardial site is slightly wider than that on the cusp to support a generous mural edge. The depth of each sinus is assessed. Each neo cusp's free edge is leveled with the sinotubular bar at the commissural level but more caudally at the center. The commissural ends are suspended at the level of the sinotubular bar using transmural pledgeted polypropylene sutures. Suspension is tailored to provide optimal coaptation, avoid crowding of the sub commissural triangle, and reestablish normal semi lunar appearance of each neocusp. When severe dilatation of the ventricular–aortic junction is present, a reduction annuloplasty at the sub commissural area between right and left cusps is performed. When the cusp is prolapsed, no attempt is made to excise any portions, but the pericardial extension is sutured to the cusp's free margin and, consequently, suspended to the aortic wall as described (Figure 1).

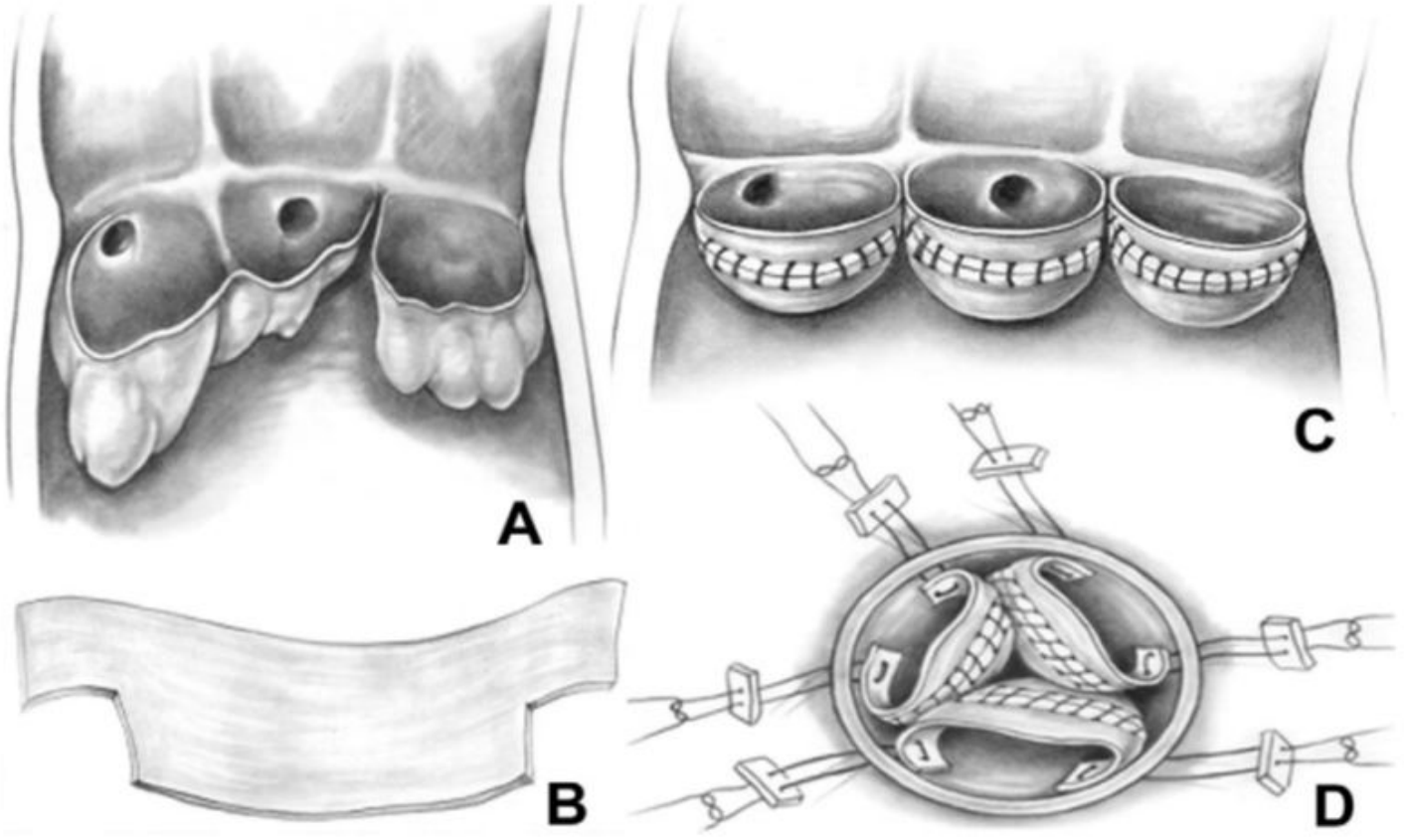

Figure 1: Cusp Extension Valvuloplasty with Tricuspidization.

(A)Each cusp is evaluated as to the extent of tissue deficiency, the shape, and the irregularities of the free edge. Only the thickened free margin and body are thinned out, leaving its base and unaffected body intact. When tricuspidization is needed, the fused cusp is cut at the raphe precisely to the aortic wall.

(B) A pericardial extension is then fashioned to fit the specific architecture of each cusp, but slightly oversized in depth (10% to 15%) and length (up to 25%).

(C) The suture line on the pericardial site is slightly wider than on the cusp to support a generous mural edge. Each neocusp's free edge is leveled with the sinotubular bar at the commissural level.

(D) The commissural ends of the constructed cusps are suspended at the level of the sinotubular bar using transmural pledgeted polypropylene sutures.

As reported [13,18] CEV with selective tricuspidization effectively reduced AI and AS and improved LV wall thickness and LV dimensions in infants and children. Promising early outcomes have been reported in adolescents with congenital or acquired aortic valve disease. Most series are relatively small, or include a combination of simple and complex repairs [7,14]. As shown [13,14,19], this repair strategy allows expeditious LV reverse remodeling even in patients with significant LV dilation or LV wall thickness with satisfactory long-term durability and freedom from AVR. The *z* values of LVEDD, aortic annulus, aortic sinus diameter, sinotubular junction diameter, and LV wall thickness, which improved after ACEV, remained relatively stable in patients who met no AVR criteria at last echocardiographic follow-up after ACEV. Use of a cusp extension as part of an aortic valve repair strategy in patients with rheumatic AI and a structurally normal aortic valve can be fairly durable [20]. Our experience and that of others [13-15,17,19] indicate that CEV provides a comprehensive repair and restores all the important anatomic features of the aortic valve. Most critical among these, in a congenitally abnormal valve, are the attenuated sub commissural triangles, the foreshortening of the cusp's free margin, the shallow sinus (es) of Valsalva, and the eccentric and uneven orifice opening. Cusp extensions counteract the valve's inherent sinus (es) of Valsalva shallowness, reestablish normal depth of the sinuses, secure adequate and longer coaptation surface, and restore the normal "Crown like" appearance of the valve. Cusp resuspension at the level of the sinotubular bar, aiming at a wider sub commissural triangle allows more freedom of the cusp's movement. Tricuspidization ensures a larger central opening and minimizes turbulence. Several substitutes have been used to augment the cusps. These include autologous or bovine pericardium, and polytetrafluoroethylene membrane. Glutaraldehyde-treated autologous pericardium has been most consistently used in the aortic position. Although glutaraldehyde promotes calcification and fibrosis, it is needed for collagen fiber linkage and, hence, tissue strength. The concentration and duration of treatment with glutaraldehyde can vary [13-15,17]. Shorter period of rinsing has been advocated for smaller children and infants [19]. This might promote increased pericardial pliability and minimizes the incidence of excessive stiffness and early calcification of the reconstructed cusps.

Published studies have demonstrated an inherent durability failure component with CEV strategies associated with progressively decreasing freedom from aortic valve re intervention [Table 1]. As expected, moderate or greater recurrent AI or AS at interval follow-up after CEV was an independent predictor of failure [17]. With respect to freedom from AI or AS after CEV, outcomes are often unpredictable. Some patients exhibit rest enosis, whereas others have recurrent AI or both. It is recommended that in isolated AS with AoV annulus *z* value less than 1.5 the Ross procedure should be considered as the preferred choice due to early failure of CEV strategies.

Author	Year	Study Type: (Only CEV included)	Material used /Selective Tricuspidization	Age at Repair (years): Median or Mean	Patients N	Follow-up (years) Median	Early Mortality	Late Mortality	AI at last follow-up[§] N(%)	Freedom from Valve Replacement (years)
Kalangos A et al[(a)][20]	1999	Retrospective (YES)	Autologous pericardium/ NO	11.5±2.7	41	3 (0.4-5)	None	1	1(2)	Not reported
Pretre R et al[(b)][18]	2006	Retrospective (YES)	Bovine pericardium/ YES	18 (10-27)	12	1.1 (0.3-3.9)	None	?	1(8)	Not reported
Alsoufi B et al[13]	2006	Retrospective (YES)	Autologous pericardium/ YES	11.4 (5-17.6)	22	1.7 (0.1-5)	None	None	7(31)	80±18% (at 2.5)
McMullan DM et al[(c)][15]	2007	Retrospective case-control (YES)	Autologous pericardium/ YES	12.6 (2.6-18)	21	3.1 (0.2-7.4)	None	None	6(28)	90% (at 3)
Bacha EA et al[(d)][14]	2008	Retrospective (NO)	Autologous pericardium/ NO	8.8 (0.1-18)[1]	65(81)[2]	4.5 (0.1-16)	1	2	21(26)	54±9% (at 7.5)
Polimenakos AC et al[17]	2010	Retrospective (YES)	Autologous pericardium/ YES	9.3±2.7	139	14.4 (0.1-21.4)	None	None	44(31)	71.8±5.1% (at 10)

Table 1: Studies in children and adolescents with aortic valve repair where cusp extension valvuloplasty was applied

[(a)]Only patients with rheumatic valve disease

[(b)]Only patients with bicuspid aortic valve

[(c)]Case control study comparing CEV (21 patients) vs Ross operation (25 patients)

[(d)]Combined study the CEV (65) and non-CEV (16) patients

[1]Patients who underwent CEV

[2]In parenthesis are all patients; 65 only had CEV

[§]Moderate or Severe aortic insufficiency

Considering that (1) despite satisfactory long-term outcomes [3-5,21],the Ross procedure requires valves-conduit replacement in the right ventricular outflow and is associated with increasing evidence of progressive neo aortic root dilation or auto graft failure; (2) small valve prostheses, especially in infants and children, can be associated with patient–prosthesis mismatch as the child outgrows the valve, morbidity related to long-term anticoagulation, and the need for future valve replacement [5,22], CEV with selective tricuspidization provides an alternative that can neutralize many of the drawbacks of early valve replacement. It has extremely low operative early or long-term mortality, can be associated with reproducible results, and arrests the disease process until such an age that valve replacement becomes more advantageous. As a result, with improved patient selection and refinement of cusp extension valvuloplasty techniques, the durability of CEV should continue to improve. Thus, CEV might represent an attractive option in infancy and early childhood and the platform as bridge-to-AVR later in childhood and adolescence.

In conclusion, results with CEV in children and adolescents with complex congenital or acquired aortic valve disease encourage earlier and more aggressive management considering that LV has to preserve function for a longer life span. The aortic annulus *z* value, transvalvular pressure gradient, LV hypertrophy progression, LV dimensions (such as LVEDD *z* value), and regurgitant jet-to-annulus diameter ratio may guide optimal timing and strategy. When certain anatomic and functional valve criteria are met, CEV with selective use of tricuspidization is a safe, effective, and reproducible surgical choice. It allows expeditious LV reverse remodeling with satisfactory long-term durability and freedom from AVR. Thus, AVR with prosthesis or autologous pulmonary valve can be reserved for patients not suitable for CEV or as a late resort after CEV failure in late childhood or adolescence.

Multi-center studies comparing CEV-related repair techniques with AVR are needed to refine selection criteria and determine patients' suitability for each strategy.

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