Are bicuspid aortic valves a limitation for aortic valve repair?*

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Abstract

**Objective:** To compare the mid-term results after aortic valve (AV) repair in bicuspid AVs with those in tricuspid AVs. **Methods:** Between 2000 and 2010, 100 patients (mean age 47.2 years) underwent AV repair procedures for insufficient bicuspid AV (n = 43) and tricuspid AV (n = 57). Aortic regurgitation (AR) more than moderate was present in 31/43 and 21/57 patients in the bicuspid AV and the tricuspid AV group, respectively. Concomitant root replacement by either the reimplantation or the remodeling technique was performed in 42 patients (bicuspid AV 17/43, tricuspid AV 25/57). All patients were prospectively studied with postoperative and further annual clinical assessment and echocardiography. Follow-up was 99% complete with a mean follow-up time of 22 months. **Results:** Three patients died during the initial hospitalization, all due to postoperative cardiac failure. Overall actuarial 3 years’ survival was 93 ± 4.2% without significant differences between the two groups. Overall actuarial 3 years’ freedom from AV-related reoperation was 86 ± 5.1% without significant differences between the groups (85 ± 9.7% for bicuspid AV, 86 ± 6.0% for tricuspid AV; log-rank test: p = 0.98). Overall actuarial 3 years’ freedom from recurrent AR ≥ moderate was 100% and AR > trace was 71.3 ± 8.2% without significant differences between the groups (76.5 ± 11.7% for bicuspid AV, 71.4 ± 9.4 for tricuspid AV; log-rank test: p = 0.97). **Conclusions:** The mid-term outcome in terms of survival, freedom from reoperation or recurrent AR is similar for both groups of patients after AV repair procedures. Therefore, we advocate valve repair also in patients presenting with an insufficient bicuspid AV.

Keywords: Valve lesions; Aortic valve; Valve repair

1. Introduction

The bicuspid aortic valve (BAV) is a congenital cardiac malformation with an incidence of up to 2% of live births in the general population, and may be associated with diseases of the ascending aorta, such as aneurysm or dissection [1]. The presence of a BAV may be complicated mostly by the progression to aortic valve (AV) stenosis and subsequent valve replacement [2]. By contrast, up to 20% of all BAV patients develop a pure AV regurgitation [3,4]. These patients present to valve operation often during the fourth decade of life [5,6], which is a reason why a possible valve sparing would be a benefit over valve replacement with the prosthesis related limitations and complications [7,8].

Although repair techniques evolved over the past two decades, concerns remain on recurrent aortic valve regurgitation (AVR), especially for BAVs. The aim of our study was to examine the influence of BAVs on the recurrence of regurgitation, the reoperation, and the survival rates in the mid-term after AV repair and compare this data with those from patients presenting with tricuspid AVs (TAVs).

2. Patients and methods

2.1. Patient selection and data collection

This study was approved by the Ethics Committee of the Technische Universitaet Muenchen, Munich. A total of 225 patients, who were scheduled for AV-sparing operations between April 2000 and March 2010 at the German Heart Centre Munich, were included in our valve-sparing database. Patients who underwent only root replacement by the root reimplantation or the root remodeling technique, without repairing the AV directly, were excluded (n = 117). Furthermore, another eight patients in whom a repair of the AV was...
converted to a valve replacement or Bentall procedure during the operation were excluded from the study cohort. These patients were scheduled for an AV-sparing operation, due to the diagnosed AV regurgitation with or without root dilatation. During the procedure, advanced calcification of the AV was detected, and the surgeon decided to implant an AV prosthesis. Accordingly, a repair of the AV using one or more different techniques was performed in 100 patients. The assessment of the pre- and postoperative aortic regurgitation (AR) was determined by transthoracic or transesophageal echocardiography. Regurgitation of the AV was graded as none, trace, mild, moderate, or severe. The ventricular function was graded using two-dimensional (2D) echocardiography and evaluated as normal (ejection fraction $\geq 50\%$), moderately impaired (ejection fraction 30–49%), or severely impaired (ejection fraction $< 30\%$). All patients were prospectively studied with annual clinical assessment and echocardiography.

2.2. Study group

There were 80 male and 20 female patients, with a mean age of $47.2 \pm 18.6$ years (range: 1–83.7 years). The patients were assigned to two different groups according to the AV anatomy: BAV ($n = 43$) and TAV ($n = 57$). Table 1 shows the demographic data of all patients according to the AV anatomy.

Three patients presented with Marfan’s syndrome, which was diagnosed according to the Ghent criteria or by genetically test preoperatively. Five patients were less than 18 years of age at time of the operation. Three received an emergency operation on the day of admission to our hospital. Ten patients (10%) had undergone one or more previous cardiac interventions such as Ross operation ($n = 1$), CoA resection ($n = 5$), persistent ductus arteriosus (PDA) closure ($n = 2$), ventricular septal defect (VSD) closure ($n = 3$), double-outlet right ventricle correction ($n = 1$), and AV repair by means of a Trusler-plasty ($n = 1$).

Preoperative echocardiographic data were available in all patients. A BAV was present in 43/100 (43%) patients. For these patients, the AR was judged as less than moderate in 12/43 (28%) patients and more than or equal to moderate in 31/43 (72%) patients. A TAV was present in 57/100 (57%) patients. For the patients with a TAV, the regurgitation was measured as less than moderate in 36/57 (63%) patients and more than or equal to moderate in 21/57 (37%) patients.

Ninety-four out of 100 patients (94%) presented with sinus rhythm, four with atrial fibrillation, and two had a preoperatively implanted pacemaker. The list of patients summarized in Table 1 with congenital heart disease, other than BAV or Marfan’s syndrome, presented at the operation additionally to the AV regurgitation with atrial septal defect or patent foramen ovale ($n = 3$), PDA ($n = 1$), and VSD with coronary anomaly ($n = 3$).

2.3. Valve morphology and surgical technique

The AV morphology and the identification of all pathological details is the essential prerequisite for a successful AV repair. There are different etiologies for AVR, some, which are independent from the number of cusps, and others, which are specific to BAVs.

Ring dilatation is present in the majority of regurgitant AVs and may be independent or in combination with aortic root dilatation. In the first case, where aortic root replacement was not necessary, subcommissural plasty was

<table>
<thead>
<tr>
<th>Bicuspid AV</th>
<th>Tricuspid AV</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number of patients</td>
<td>43</td>
<td>57</td>
</tr>
<tr>
<td>Mean age ± SD</td>
<td>39.5 ± 14</td>
<td>52.9 ± 19.6</td>
</tr>
<tr>
<td>Gender</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>42 (97.7%)</td>
<td>38 (67.7%)</td>
</tr>
<tr>
<td>Female</td>
<td>1 (2.3%)</td>
<td>19 (33.3%)</td>
</tr>
<tr>
<td>Left ventricular ejection fraction (96/100)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>≥50%</td>
<td>28 (68.3%)</td>
<td>41 (74.5%)</td>
</tr>
<tr>
<td>30–50%</td>
<td>12 (29.3%)</td>
<td>13 (23.6%)</td>
</tr>
<tr>
<td>&lt;30%</td>
<td>1 (2.4%)</td>
<td>1 (1.8%)</td>
</tr>
<tr>
<td>NYHA functional class (80/100)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>I</td>
<td>18 (58.1%)</td>
<td>14 (28.6%)</td>
</tr>
<tr>
<td>II</td>
<td>10 (32.3%)</td>
<td>15 (30.6%)</td>
</tr>
<tr>
<td>III</td>
<td>3 (9.7%)</td>
<td>17 (34.7%)</td>
</tr>
<tr>
<td>IV</td>
<td>0</td>
<td>3 (6.1%)</td>
</tr>
<tr>
<td>AV pathology without aortic root or ascending aorta aneurysm</td>
<td>13 (30.2%)</td>
<td>19 (33.3%)</td>
</tr>
<tr>
<td>Sinus of Valsalva or ascending aortic aneurysm</td>
<td>30 (69.8%)</td>
<td>35 (61.4%)</td>
</tr>
<tr>
<td>Type A dissection</td>
<td>0</td>
<td>3 (5.3%)</td>
</tr>
<tr>
<td>Marfan’s syndrome</td>
<td>0</td>
<td>3 (5.3%)</td>
</tr>
<tr>
<td>Coronary artery disease</td>
<td>3 (7.0%)</td>
<td>12 (21.4%)</td>
</tr>
<tr>
<td>Mitral valve disease</td>
<td>11 (25.6%)</td>
<td>32 (56.1%)</td>
</tr>
<tr>
<td>Tricuspid valve disease</td>
<td>5 (11.6%)</td>
<td>19 (33.3%)</td>
</tr>
<tr>
<td>Hypertension</td>
<td>18 (41.9%)</td>
<td>37 (66.1%)</td>
</tr>
<tr>
<td>Hyperlipidemia</td>
<td>10 (19.2%)</td>
<td>24 (24.6%)</td>
</tr>
<tr>
<td>Diabetes</td>
<td>0</td>
<td>3 (5.3%)</td>
</tr>
<tr>
<td>Congenital heart disease</td>
<td>3 (7%)</td>
<td>12 (21.4%)</td>
</tr>
<tr>
<td>Renal insufficiency</td>
<td>2 (4.7%)</td>
<td>9 (15.8%)</td>
</tr>
</tbody>
</table>


* When frequency was less than 5 in one or both groups — Fisher’s exact test.
performed, by placement of one pledgeted 3/0 polyester suture for each commissure, parallel to the valve annulus as described by Cabrol and colleagues [9]. Thus, the commissural angle was narrowed, reducing the valve annulus and increasing the coaptation area of the cusps. In order to avoid any excessive valve area reduction, a dilator was used during the suture-tying maneuver. If root dilatation was present (>45 mm in the BAV group and >50 mm in the TAV group), an annular reinforcement was performed using the AV reimplantation technique. After resection of the aortic sinuses, the AV annulus and subcommissural triangles of the noncoronary cusp were implanted inside a vascular graft using two proximal suture lines, one subannular mattress suture line and one hemostatic continuous suture line within the graft as described by David and Feindel [10].

In the present cohort, a ring dilatation, as described during the operation, concomitant or not to other cusp pathologies was present in 26/43 (61%) and 41/57 (72%) patients with bicuspid and TAVs, respectively. In the BAV group, 26/43 (61%) patients underwent a subcommissural plasty and 16 (37%) patients underwent a root replacement using the AV reimplantation technique. In the TAV group, a subcommissural plasty was performed in 25/57 (44%) patients and an annular reinforcement using the AV reimplantation technique was performed in 21/57 (37%) patients.

Cusp prolapse is a condition, which may be present in both types of AVs, tricuspid and bicuspid. It occurs in cusps with an excessive length of the free edge, which lead to a displacement of the free edge and the coaptation area of the cusp toward the left ventricular outflow tract. In TAVs, the regurgitation may be due to the prolapse of one or more cusps, with elongation of the free edge and in some cases with thinning and fenestration of the cusp. In our cohort, a prolapse of one or more cusps was present in 38 out of 57 (67%) patients with TAVs To resuspend the cusp, in the majority of cases, a triangular plication (32/57; 56%) of the free edge was performed with a polypropylene suture near the commissure as described by Trusler or in the central portion of the free edge of the cusp as described by El Khoury [11,12]. In two patients, a triangular resection of the prolapsing cusp was necessary. In BAVs, the regurgitation due to prolapsing cusps may have two different etiologies. First, the cusps may be symmetric and one or both may be elongated and prolapsing. In this case, free edge plication or free edge reinforcement, as described by David and colleagues [13], was the used technique for cusp correction. A triangular plication was performed in 15/43 (35%) and 20/43 (47%) patients presenting with a BAV, for the conjoint and the nonconjoint cusp, respectively. Second, the valve pathology is influenced by the presence of the conjoint cusp, which emerges from the congenital fusion of two cusps. In our cohort, the conjoint cusp emerged in 38 patients, two and one from the fusion of the left coronary and right coronary cusps, the aconary and the right coronary cusps, and the aconary and left coronary cusps, respectively. This conjoint cusp mostly presents with a raphe (26/43; 61%), a fibrous ridge, which can be short or tall, stiff and solid, or flexible and soft with or without fenestrations. The prolapse of this conjoint cusp may develop due to excessive cusp tissue, with or without a tall raphe. If this raphe was flexible and soft, the correction was performed by free edge triangular plication of the conjoint cusp. In case of a stiff and eventually calcified raphe, a triangular resection was performed and the free margins of the conjoint cusp were reapproximated as described by El Khoury and colleagues [12,14]. If the nonconjoint cusp was also elongated or was prolapsing after the repair of the conjoint cusp, this had to be adjusted by triangular plication. A triangular resection of the raphe was necessary in 21/43 (49%) patients presenting with a BAV.

Cusp fenestrations where closed in both tricuspid (6/57; 11%) and bicuspid (1/43; 2%) AVs For extended defects, a glutaraldehyde-fixed patch of autologous pericardium was prepared, which was larger than the cusp defect in order to avoid cusp restriction. The patch was then sutured into the defect using a running 6/0 polypropylene suture. For small fenestrations, a direct closure with a 6/0 polypropylene suture was performed.

2.4. Follow-up

Follow-up was conducted annually after the operation by mail questionnaire and telephone contact with the patient and the referring cardiologist. The clinical assessment and the echocardiography were accomplished by the referring cardiologist or in our clinic. The latest follow-up examinations included electrocardiography (ECG), Doppler echocardiography, and physical examination. All data were entered into our database. Follow-up was 99% complete with a mean follow-up time of 22 months (range: 1 month to 8.6 years), and a cumulative follow-up of 167 patient years. The functional status was determined according to the New York Heart Association (NYHA) class. The cause of death was determined from hospital records.

2.5. Statistical analysis

Categorical variables were reported using the number and percentage of observations. Continuous variables were reported as means ± standard deviation or median with ranges. Categorical variables were compared between the groups using the chi-square test (and, when appropriate, Fisher’s exact test), and continuous variables were compared using a two-sample t-test (and, when appropriate, Mann–Whitney test). The outcome parameters were defined as time from the primary AV operation to death, to AV-related reoperation or to a recurrent AVR. The probability of freedom from events was estimated according to the Kaplan–Meier method. Freedom-from-events curves were compared using the log-rank test. A p-value <0.05 was considered to indicate statistical significance.

According to Concato and colleagues [15], multivariate analysis could not be performed, since the ratio of events per variable was too small. Analyses were performed with SPSS PASW Statistics 18 for Windows.

3. Results

3.1. Operative characteristics

A root replacement using the reimplantation technique was performed in 16 out of 43 (37%) and 21 out of 57 (37%) patients diagnosed with a BAV and a TAV, respectively. A root
replacement using the remodeling technique was performed in four out of 43 (9%) and three out of 57 (5%) patients diagnosed with a BAV and a TAV, respectively. Operative data according to the AV cusp number are shown in Table 2.

### 3.2. Survival

There were no operative deaths in this study cohort. Three patients died during the initial hospitalization, all due to postoperative cardiac failure. The first patient was preoperatively diagnosed with mild regurgitation of a TAV, type A aortic dissection, and normal left ventricular ejection fraction (LV-EF). He underwent an emergency operation with supracoronary replacement of the aorta ascendente, hemiarch replacement, descending aortic stent implantation, and AV repair. The patient died 10 weeks after the operation after pulmonary and gastrointestinal complications. The second patient was preoperatively diagnosed with mild regurgitation of a TAV, severe regurgitation of the mitral valve, dilatative cardiomyopathy with an LVEF of 25%, and pulmonary hypertension. He underwent an aortic and mitral valve repair and died 14 days after the operation. This patient required re-exploration twice for surgical bleeding. The third patient was preoperatively diagnosed with moderate TAV regurgitation and a sinus of Valsalva aneurysm. He underwent an AV repair with replacement of the sinus of Valsalva and died 2 days after the operation.

One more patient died during the follow-up period, 2.5 years after the primary operation due to a myocardial infarction and was preoperatively diagnosed with severe regurgitation of a BAV and aneurysm of the ascending aorta. A coronary heart disease was not diagnosed at time of the operation.

Overall survival was not statistically significantly different between the groups (p = 0.59) with an estimated 3-year survival probability of 93 ± 4.2% in the total population, 80 ± 18% in the BAV group, and 94 ± 3.2% in TAV patients (Fig. 1).

Marfan’s syndrome, preoperative comorbidities, and several concomitant procedures were not significant predictors for death in univariate analysis, neither for the BAV nor for the TAV group.

### 3.3. Reoperations

After hospital discharge, eight patients underwent a reoperation for AV-related disorders. Five out of the eight patients presented before the reoperation with a TAV with a more or equal to moderate AR. Two out of the eight patients presented before the primary operation with a BAV with a more than moderate AR, and one presented with a BAV with stenosis having a mean pressure gradient upon the valve of 60 mmHg. One out of the eight reoperated patients presented with Marfan’s syndrome and he underwent a remodeling of the aortic root procedure without a reinforcement of the AV anulus. He developed a severe recurrent AR 1 year after the primary operation. At time of the reoperation, an AV replacement was performed in seven out of eight reoperated patients and a Ross operation in one patient. All eight patients are alive at time of the latest follow-up. The data of the reoperated patients are shown in Table 3.

Actuarial overall freedom from AV-related reoperation was not statistically significantly different between the two groups (p = 0.98). The estimated 3-year freedom from

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**Table 2. The operative data according to the number of aortic valve cusps.**

<table>
<thead>
<tr>
<th></th>
<th>Bicuspid AV</th>
<th>Tricuspid AV</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number of patients</td>
<td>43</td>
<td>57</td>
<td></td>
</tr>
<tr>
<td>Concomitant aortic operation</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Remodeling of the aortic root</td>
<td>4 (9.3%)</td>
<td>3 (5.3%)</td>
<td>0.45</td>
</tr>
<tr>
<td>Reimplantation of the aortic valve</td>
<td>16 (37.2%)</td>
<td>21 (36.8%)</td>
<td>0.78</td>
</tr>
<tr>
<td>Supracoronary replacement of the ascending aorta</td>
<td>9 (20.9%)</td>
<td>8 (14%)</td>
<td>0.58</td>
</tr>
<tr>
<td>Urgent operation</td>
<td>0</td>
<td>3 (5.3%)</td>
<td>0.12</td>
</tr>
<tr>
<td>Aortic cross clamp time (min)</td>
<td>93.9 ± 41.5</td>
<td>108 ± 41.1</td>
<td>0.53</td>
</tr>
<tr>
<td>median (range)</td>
<td>77 (11–162)</td>
<td>98.5 (27–206)</td>
<td></td>
</tr>
<tr>
<td>CABG</td>
<td>2 (4.7%)</td>
<td>8 (14.0%)</td>
<td>0.18</td>
</tr>
<tr>
<td>Aortic arch or hemiarch replacement</td>
<td>1 (2.3%)</td>
<td>5 (8.8%)</td>
<td>0.23</td>
</tr>
<tr>
<td>MV-repair/replacement</td>
<td>1 (2.3%)</td>
<td>13 (22.8%)</td>
<td>0.03</td>
</tr>
<tr>
<td>TV-repair</td>
<td>0</td>
<td>6 (10.5%)</td>
<td>0.03</td>
</tr>
<tr>
<td>Other congenital (PDA, PFO, ASD, VSD)</td>
<td>3 (7.0%)</td>
<td>7 (12.3%)</td>
<td>0.5</td>
</tr>
</tbody>
</table>

AV: aortic valve; ASD: atrial septal defect; CABG: coronary artery bypass grafting; MV: mitral valve; PDA: persistent ductus arteriosus; PFO: persistent foramen ovale; TV: tricuspid valve; and VSD: ventricular septal defect.

- When frequency was less than 5 in one or both groups — Fisher’s exact test.
- * Cross clamp time not normally distributed — group means compared by Mann–Whitney test.
reoperation probability was 86 ± 5.1% for the entire study population (Fig. 2), 85 ± 9.7% for the BAV, and 86 ± 6% for the TAV group (Fig. 3).

Marfan’s syndrome, preoperative comorbidities, and several concomitant procedures were not significant predictors for reoperation in univariate analysis, neither for the BAV nor for the TAV group. The only exception was the group of patients with a TAV repair and concomitant root replacement using the AV reimplantation technique. They had a significantly lower reoperation rate compared to those without root replacement (p = 0.045). Five out of 36 patients who presented with a regurgitant TAV at primary operation and underwent valve repair without root replacement developed a recurrent AR and were reoperated during the follow-up period. By contrast, none of the 21 patients with a regurgitant TAV and valve repair concomitant to David operation underwent reoperation on the AV.

### 3.4. Left ventricular function

Left ventricular function at final follow-up was documented in 79/88 patients, who were alive and not reoperated, and graded as normal (≥50%) in 52/79 (66%) and moderately impaired (30–49%) in 27/79 (34%).

### 3.5. AV function

Eighty-seven patients had no further reoperation and are alive and not lost until the final follow-up. In all these patients, AV function was assessed by echocardiography at the final follow-up examination. At final follow-up, AR was documented as none or trace in 73/87 (84%) patients and as mild AR in 14/87 (16%). Except the reoperated patients, no patient presented at the time of the final follow-up with a more or equal to moderate AR.
4. Discussion

The congenital cardiac malformation represented by the presence of a BAV may be asymptomatic up to the seventh decade of life [16], and may be associated with aneurysm or dissection of the ascending aorta and furthermore, complicated by a progressive dysfunction of the bicuspid valve. Pure BAV stenosis is described in the literature to be the most commonly reported dysfunctional state, affecting about 72% of the patients with a BAV pathology [6]. In this case, the indication for valve replacement is clear and is also supported by the reported mean age of more than 63 years for the patients presenting with stenotic BAVs [6,17] By contrast, the decision of whether to repair or replace the valve in case of pure AVR, which occurs in about 20% of the symptomatic BAV patients, is still controversial.

In a cohort of 100 patients following AV repair with or without root replacement at our institution, the BAV was not identified to be a significant predictor for mortality, reoperation, or recurrent AVR. Furthermore, the freedom from recurrent moderate or higher valve regurgitation was similar for patients presenting with a tricuspid or a BAV. Hence, we advocate AV repair also in patients presenting with an insufficient BAV.

Patients presenting with a BAV with pure valve regurgitation tend to become symptomatic in the third and fourth decade of live [5,6,18]. Thus, the possibility of valve repair should be considered in this group of patients in order to avoid the valve-related events and premature degeneration, which may occur after valve replacement with mechanical or biological prosthesis [7,8].

Reported data of several studies [6,18–20] for BAVs demonstrate that the different etiologies of bicuspid valve regurgitation, such as ring dilatation, prolapse or retraction of one or both cusps, and fenestrations of the cusps, may emerge isolated or in combination, leading to a complex valve pathology. First, a ring dilatation is described to be present in 39–48% of the patients diagnosed with a regurgitant BAV [6,19]. In the present study, 61% of the patients with a BAV presented a dilated AV annulus at the time of primary operation. It may occur in combination with a dilated aortic root or isolated. Nevertheless, as described by El Khoury and colleagues [12], if a BAV repair is to be attempted, an aggressive stance should be taken toward root replacement. The authors report that a root replacement was found to be protective against a recurrent AVR, and advocate root replacement also in BAV patients with root diameters lower than 4.5 cm if the aortic wall tissue is thin and fragile. In another study performed by Schafers and colleagues, the authors recommend to replace the dilated part of the aortic root if enlargement is already present at the time of surgical intervention on the AV. The cutoff point for root replacement chosen by the authors was a sinotubular junction diameter of 3.3–3.5 cm [21]. In the present study, we performed root replacement only if the diameter of the aortic root was at least 4.5 cm for the patients with a BAV and 5 cm for the patients with a TAV. Hence, for isolated dilated aortic annulus without a dilated root, subcommissural plasty was the only used technique for annular reinforcement. Until now, the omission of root replacement has not revealed to be a predictor for recurrent BAV regurgitation in the present
group. In contrast to the BAV group, 5 out of 36 patients who presented with a regurgitant TAV at primary operation and underwent valve repair without root replacement developed a recurrent AR and were reoperated during the follow-up period. The freedom from reoperation at final follow-up was significantly lower in this patients compared to those TAV patients with annular reinforcement by means of the David root replacement. Nevertheless, on examining the data of this five reoperated patients in detail, it is obvious that the missing reinforcement of the annulus by the reimplantation technique is not the reason for reoperation, but, more precisely, the missing annular reinforcement in general. Two out of the five patients underwent a Yacoub procedure without annular reinforcement, other two underwent cusp plication but without subcommissural plasty, and the fifth underwent an incomplete annular plasty by subcommissural stitches only in the acornary to right coronary commissure. Hence, we advocate annular reinforcement in regurgitant TAVs not necessarily by root replacement using the reimplantation technique, but definitely by subcommissural plasty for all three commissures. However, only further long-term observations will be able to determine if combined procedures using both, the repair techniques for the pathological cusps and the root replacement, offer a real lower incidence of AV-related reoperation in both regurgitant BAVs and TAVs.

Another most commonly reported mechanism leading to regurgitation among congenital BAVs is cusp prolapse or retraction of the cusps. This prolapse, which usually affects the conjoint cusp, results from the combined effects of the myxomatous degeneration and stretching of the cusp and an inadequate support by a flexible and soft raphe [6]. These data are similar with those from our study cohort with 29/43 (64%) and 12/43 (28%) patients with a prolapsing conjoint, and nonconjoint cusp, respectively. The preoperative echocardiography assessment and intraoperative morphopathological identification of these different etiologies represent the basic condition for a successful performance of AV repair.

Following these steps, El Khoury and colleagues reported a series of 122 patients with regurgitant BAV due to one or more of the above-described etiologies with a mean follow-up time of 61 months. The authors reported an overall survival and freedom from AV-related reoperation at 8 years of 97 ± 3% and 83 ± 5%, respectively. Furthermore, the freedom from recurrent AR >2+ was 94 ± 3% at 5 years [12]. Cosgrove and colleagues report a series of 94 patients with regurgitant BAV, who underwent valve repair by means of triangular resection or mid-leaflet plication of the prolapsing leaflet without subcommissural plasty [5]. The freedom from AV-related reoperation was 87% and 84% at 5 and 7 years, respectively. The authors conclude after evaluating their data, that any residual AR jeopardizes repair durability. In another series of Danielson and colleagues, the authors reported a freedom from reoperation of 91% at 5 years for the patients presenting with a regurgitant BAV at primary operation. They conclude that the risk of reoperation is acceptable in young patients, particularly when compared with the anticipated risk of structural valve degeneration of heterograft valves [19], Schafers and colleagues report a series of 173 BAV patients, who were assigned to three groups according to the operations technique, isolated valve repair, valve repair and supracommissural aortic replacement, and remodeling of the aortic root. The freedom from AR ≥ 2 at 5 years varied in their cohort between 91% and 96%, with the highest reoperation rate in the group with supracommissural aortic replacement. The authors conclude that the reduction of the sinotubular diameter may induce symmetric cusp prolapse, which has to be corrected in order to achieve valve stability in the long term [20].

The data from the studies mentioned above are in line with the results from our cohort, with a 3-year freedom from AV-related reoperation, recurrent a more or equal to moderate AR, and recurrent AR > trace of 84 ± 9.7%, 100% and 76.5 ± 11.7%, respectively. Compared to the patients from the TAV subgroup, the patients with a BAV presented a significantly lower incidence for comorbidities in the present study, and were in a better clinical state at the time of the primary operation. This fact may be generated by the significantly younger age of the BAV patients at the time of primary operation.

The surgical techniques performed by the authors of the present study were in line with the techniques described by El Khoury or Danielson and based on the preservation of the bicuspid shape of the AV. By contrast, in a biomechanical study reported by Conti and colleagues, the authors suggest that a BAV repair without tricuspidalization is insufficient, because a bicuspid valve, although normally coapting, is overloaded with stress and may therefore be prone to accelerated degeneration or tearing [22].

Additional observation is necessary to determine whether the strategy of BAV repair is superior in the long term over early replacement with biological or mechanical devices. Having demonstrated that the necessity for reoperation is low in the mid-term, in our opinion, the benefit of BAV repair outweighs the risk of reoperation; therefore, repair should be considered whenever technically possible.

5. Study limitations

The main limitation of our study was the small number of patients in both subgroups, which precluded a subanalysis regarding detailed valve pathology or valve repair techniques. Furthermore, the reduced follow-up time may also lead to a misestimation of the calculated results.

6. Conclusion

There is no statistically significant difference in terms of early or mid-term complications, mortality, valve-related reoperation, or recurrent AV regurgitation between patients who presented with bicuspid or tricuspid AVs. Further observations will be necessary in order to determine whether BAV repair is superior over valve replacement, and whether one of the used repair techniques is a significant predictor for recurrent AV regurgitation. Nevertheless, we advocate for BAV repair in pure regurgitant valves, with concomitant annular reinforcement and, if necessary, root replacement.
References


Appendix A. Conference discussion

Dr. A. Franco-Cereceda (Stockholm, Sweden): I have a couple of questions. The first one relates to the size of the aortic root. You state in your manuscript that you have a cutoff level of 50 mm in both bicuspid and tricuspid aortic valves. What do you think is the rationale for having the same cutoff level? Usually we go somewhat lower with a bicuspid valve, considering that there might be an inherent weakness of the aortic wall in these patients. And in conjunction with that, do you have any follow-up data on possible continuous progression of root dilatation in these patients, which I think is very important to know? You operate on fairly young patients with bicuspid aortic valves, and it is certainly very important to know if they continue to dilate if you have not already changed the aortic root at the primary surgery.

Secondly, in your presentation last year you stated that there was a tendency for the degree of aortic valve regurgitation to be a predictor for poor outcome postoperatively. Last year you had 102 patients; now you have an additional 100 patients. Combining these, can you confirm what you thought last year or should we disregard that? And if you can confirm it, does this mean that we are actually operating on these patients too late?

My final question relates to the end-diastolic diameters. There are some indications that a large ventricle may be predictive for recurrence of late aortic regurgitation in these patients, and I wonder if you have any comments on that, too?

Dr. Badiu: Concerning your first question about the limit of 50 mm, we look at the tissue consistency in every patient. The 50 mm is just a border that we use in order to have a dimension, but the most important criterion is the tissue quality. If we see that the tissue is very thin, if it is less than 50, we go for a replacement of the root also, not only in patients with a bicuspid valve, but also in patients with a tricuspid aortic valve, but if the tissue quality is normal, the policy is to follow the 50 mm rule.

In terms of progression, we don’t have long-term follow-up. We examine these patients 6 months and 1 year after the operation, and we don’t see any further dilatation at that stage, but I think this may occur in the long-term. To date we don’t have an answer, because the follow-up is short.

Regarding your second question, you are right, we presented patients from our aortic valve-sparing database last year, but some of those were patients without valve repair. They were patients with root replacement with or without valve repair. In that group we saw that severe AR previous to the operation seems to be a predictor in the long term for recurrent regurgitation. In the present cohort, first of all, we didn’t have so many patients with severe aortic regurgitation in the bicuspid valve group, and the regurgitation degree previous to the primary operation was not a predictor for recurrent AR. To date we do not have further data for the cohort presented last year.

Concerning your third question, I think it was about the dimension of the ventricle. We just compared the ventricles by their function and not by their dimensions. The patients with severe reduced ventricular function were those who also had problems directly postoperatively. Unfortunately we do not have a comparison between the dimensions of the ventricle and the postoperative outcome.

Dr. G. El Khoury (Brussels, Belgium): If I can comment on the last question, in our experience, we found, and we don’t know why, that the patient with a large ventricle with really very bad function has a really very bad prognosis. They come early for reoperation. When we have a really big ventricle, severely depressed left ventricle, in our experience, we tend to replace the valve rather than repair it.