Unicuspid aortic valve (UAV) anatomy is occasionally encountered in adolescents or young adults and not infrequently associated with aneurysm of the ascending aorta and aortic root. To manage both defects without aortic valve replacement we propose a combination of remodeling of the aortic root combined with bicuspidization of the UAV.

Methods. Between 1 December 2007 and November 2013, 25 patients (23 males; mean age, 38 ± 12 years; range, 21 to 65 years) with aortic regurgitation as a result of UAV and aortic root dilatation underwent remodeling of the aortic root and bicuspidization of the UAV. The dilated aortic root tissue was resected, leaving the wall adjacent to the normal commissure and at 180 degrees orientation and similar height for the new commissure. The graft was configured to create two symmetric tongues of graft and sutured to the remnants of the aortic root wall. The dysplastic right coronary cusp was resected, and autologous pericardial patches augmented the deficiency of cusp tissue between the left and noncoronary cusps. A suture annuloplasty was used in 20 cases. All patients were followed clinically and echocardiographically at 3, 6, and 12 months and at yearly intervals. Cumulative follow-up was 677 months (mean, 27 ± 18 months).

Results. No early or late death occurred. Intraoperative echocardiography revealed minimal or no aortic regurgitation in all patients; at discharge, systolic mean gradient was 6 ± 3 mm Hg. There was no bleeding or thromboembolic event during the follow-up. One patient exhibited endocarditis and underwent reoperation. Two patients experienced relevant recurrent aortic regurgitation for limited suture dehiscence between the patch and the cusp and were reoperated on between 16 and 32 months postoperatively. One patient underwent biologic valve replacement, and two valves were re-repaired. At 5 years, freedom from reoperation and aortic valve replacement was 81% and 91%, respectively.

Conclusions. In the presence of UAV and aortic root dilatation, the concept of valve bicuspidization and root remodeling can be applied with satisfactory hemodynamic results. The hemodynamic function of an aortic valve preserved by this concept is good. If sufficient stability can be achieved, aortic valve replacement can be avoided in young patients with aortic regurgitation caused by UAV and root aneurysm.

controversially in the presence of annular and aortic dilatation [10, 11]. Valve-preserving root replacement has become an increasingly accepted alternative to composite valve replacement in tricuspid and also bicuspid aortic valve anatomy. We have also previously developed a bicuspidizing reconstructive approach to the unicuspid aortic valve, preserving normal cusp tissue, creating a second commissure of normal height, and augmenting the cusp tissue with pericardium [12]. We found that a symmetric design achieves near-normal hemodynamics [13].

To avoid the disadvantages of the autograft or anticoagulation, using a reconstructive approach that normalizes valve function is desirable. We hypothesized that a combination of root remodeling and bicuspidization of the UAV should achieve this goal, normalization of valve function and elimination of aortic dilatation. The objective of this analysis was to describe the technique and review the early results of the first consecutive cases as a pilot series.

Patients and Methods

Patients

Between December 2007 and November 2013, we treated 25 patients surgically for aortic dilatation involving the root in the presence of a UAV. The majority (n = 23) were male, the age ranged from 21 to 65 years with a mean age of 38 ± 12 years. One patient had previously undergone balloon valvuloplasty and a previous commissurotomy for congenital aortic stenosis. Aortic regurgitation (AR) was predominant in 24 individuals; 1 had predominant stenosis. The primary indication for surgery in 21 patients was symptomatic aortic valve disease, in 4 it was aortic aneurysm (>5 cm). Aortic root size varied from 42 to 55 mm (mean, 45 ± 5 mm). The mean peak systolic gradient was 6 to 74 mm Hg and the mean systolic gradients ranged from 3 to 48 mm Hg; AR was mild in 1 patient, moderate in 5 patients, and severe in 19 patients. Aortic aneurysm with a maximum aortic diameter exceeding 5 cm was present in all patients. Further patient characteristics are summarized in Table 1. All patients were treated in identical fashion by bicuspidization the UAV and remodeling the aortic root. All patients were followed up clinically and echocardiographically; follow-up was complete, ranging from 7 to 68 months (mean, 27 ± 18 months; cumulative, 677 months).

Statistical Analysis

Kaplan-Meier curves were calculated for freedom from reoperation and freedom from valve replacement using a commercially available software package (Prism; GraphPad, Inc, San Diego, CA). The curves were compared between the groups using the Mantel-Haenszel log rank test. A probability value of less than 0.05 was considered as statistically significant.

| Table 1. Preoperative Patient Characteristics |
| Characteristic | Range | Mean | Median |
| Sex (M/F) | 23/2 | | |
| Age (y) | 21–65 | 38 ± 12 | 34 |
| AR (grade) | 2.5–3.5 | 2.9 ± 0.3 | 3 |
| Preoperative peak gradient (mm Hg) | 6–74 | 21.4 ± 17 | 20 |
| Preoperative mean gradient (mm Hg) | 3–48 | 11.5 ± 10 | 10 |
| Maximal diameter ascending aorta (mm) | 50–64 | 51 ± 4 | 50 |
| LVEDD (mm) | 46–70 | 59 ± 16 | 55 |
| LVESD (mm) | 25–50 | 43 ± 6 | 40 |
| AVJ diameter (mm) | 28–37 | 32 ± 3 | 31 |
| Sinus diameter (mm) | 42–55 | 45 ± 5 | 43 |
| STJ diameter (mm) | 27–45 | 39 ± 4.5 | 40 |
| EF | 0.48–0.70 | 0.59 ± 0.05 | 0.60 |

AR = aortic regurgitation; AVJ = aortoventricular junction; EF = ejection fraction; LVEDD = left ventricular end-diastolic diameter; LVESD = left ventricular end-systolic diameter; STJ = sinotubular junction.
keeping the suture line close to the existing or anticipated cusp insertion lines (Fig 2).

Autologous pericardium was then harvested and pretreated in 1.5% glutaraldehyde for 3 minutes and rinsed for 3 minutes in normal saline solution. For each of the two cusps a patch was tailored to bridge the gap between the existing cusp and the new commissure. The pericardium was then sutured (polypropylene 4-0) into the planned insertion line, ie, close to the suture between the Dacron tongue and the aortic wall. The patch was trimmed to final size, and using the same suture material the patch was then connected to the left and the noncoronary cusps (Fig 3). Finally effective height was measured [15]. If effective height was less than 10 mm, the free margin of the pericardial patch was shortened with plicating sutures.

In the last 20 patients an annuloplasty suture was used if basal diameter exceeded 25 mm. A two-braided poly-ester (Ethibond, Ethicon, Hamburg, Germany; n = 3) or polytetrafluoroethylene (PTFE; Gore-Tex CV-0, Gore, Munich, Germany; n = 17) suture was passed around the root at the level of the basal ring; it was then tied around a Hegar dilator chosen according to body surface area [16]. Concomitant coronary bypass grafting was necessary in 1 patient.

Results

In all patients the planned procedure was completed without intraoperative conversion to replacement. Mean ischemic time was 87 ± 12 minutes (range, 60 to 113 minutes). Intraoperative transesophageal echocardiography revealed minimal or no AR in all patients. No early or late death occurred, and no patient experienced atrioventricular conduction disturbance. At discharge transthoracic echocardiography documented stable diastolic valve function. Peak systolic gradient was 12 ± 5 mm Hg (range, 4 to 22 mm Hg), and the mean systolic mean gradient was 6 ± 2.5 mm Hg (range, 3 to 12 mm Hg). During the follow-up there were no bleeding or thromboembolic events. One patient experienced endocarditis;
he could be stabilized by antibiotic treatment and underwent reoperation for recurrent regurgitation within the first postoperative year.

In total, 3 patients experienced relevant recurrent AR and were reoperated on between 16 and 32 months postoperatively. The cause of recurrent AR was a limited suture dehiscence between the patch and cusp (n = 2) or healed endocarditis (n = 1). One patient underwent biologic valve replacement, 2 underwent re-repair. At 5 years, freedom from reoperation was 81% (Fig 4). Because 2 patients could be re-repaired, freedom from aortic valve replacement was 91% at 5 years (Fig 5).

Until now, aortic valve competence has remained stable in all patients. At latest follow-up, peak systolic gradient was 11 ± 7 mm Hg and mean systolic gradient was 5 ± 2 mm Hg. Left ventricular dilatation was regressive in all patients (at latest follow-up, mean end-diastolic diameter was 52 ± 6 mm and mean end-systolic diameter was 38 ± 5 mm). Transthoracic echocardiography has not shown evidence of calcification or degeneration of the cusp tissue or the pericardial patch.

Of 5 patients without annular support, 2 underwent reoperation (40%) versus 1 of 20 (5%) who were treated by suture annuloplasty. Of these, 3 had undergone annuloplasty with a polyester suture and 17 with a PTFE suture. So far none of the patients with PTFE suture annuloplasty has exhibited recurrent AR.

Comment

Unicuspid aortic valve morphology has been a frequent cause of aortic valve dysfunction in our experience; it has been found in 60% of individuals requiring aortic valve or root surgery in our experience (unpublished data). The typical patient is younger than those with bicuspid aortic valve morphology. Thus, valve dysfunction in a UAV to the point of requiring surgical treatment seems to occur 10 to 20 years earlier than with bicuspid valves [2]. With a mechanical composite, the linearized risk of thromboembolic complications and anticoagulation-related hemorrhage is low [17], but the cumulative risk may be substantial because of the long life expectancy. Valve replacement with a pulmonary homograft is discussed controversially in the presence of annular and aortic dilatation. Standard biologic xenografts have suboptimal durability for this age range [18]. In addition, if root replacement is required, the reoperation may be rather complex. These considerations, and the good hemodynamic function achieved with the bicuspidizing approach to the unicuspid valve stimulated us to explore repair rather than replacement also for patients with unicuspid anatomy and concomitant root dilatation.

Originally root remodeling was developed for tricuspid aortic valves [19]. We extended this approach to bicuspid aortic valves with good long-term results [20, 21]. In fact, the durability of combined bicuspid aortic valve repair and concomitant root remodeling has exceeded that of isolated valve repair [21]. Consequently we decided to combined root remodeling with our previously published repair technique of bicuspidization of the UAV [12], modifying the remodeling technique to the anatomic characteristics of this setting. In the current analysis we describe the technical details and assess early valve function of this concept.

So far the reproducibility of the technique has been good. We have been able to apply the concept to all patients with unicuspid valves as long as cusp fibrosis or calcification was absent or limited to the right coronary cusp. The early echocardiographic results confirm the good hemodynamic function we have observed previously [13]. In fact, postoperative systolic gradients at rest and during exercise were similar to those of an age-matched patient population with a tricuspid aortic valve [13]. Thus, the current results can be interpreted as proof of concept.

The next important question will be the durability of this repair. Similar to our observations in bicuspid valve repair, the current results imply that repair...
stability may also be related to the presence of annular dilatation [21]. We have focused on this detail in the past few years. First results have shown a lower incidence of postoperative residual regurgitation using a basal suture annuloplasty [16].

Especially when using PTFE material for suture annuloplasty, reoperations have become rare. Even though the current numbers are small and the results far from statistically significant, they seem to confirm the observation made in bicuspid repair [16].

Contrary to many other cusp repair maneuvers in the context of valve-preserving surgery, this valve morphology requires the use of a relatively large patch. The durability of the patch material can be expected to primarily influence the stability of our technique. Pericardial patch insertion for aortic valve repair was first used by Ross in 1963 [22]. We use pericardium also to stabilize aortic cusps that prolapse in the presence of congenital fenestrations, and for correction of defects after excision of a calcified raphe in prolapsing bicuspid valves [23]. Unfortunately, its use in bicuspid valve repair has been associated with an increased probability of reoperation.

Others used pericardium for cusp replacement or extension in congenital aortic valve disease [24–26]. Alsoufi and colleagues [24] used cusp extension with pericardium for congenital aortic valve disease and reported a 5-year freedom from valve replacement of 75% with a median follow up of 1.7 years. Prêtre and associates [25] used pericardium for tricuspidization of regurgitant bicuspid aortic valves with a follow-up of less than 4 years. Interestingly, the repair concept including pericardial extension was only used in 21% of pediatric aortic valve operations. Tolan and coworkers [26] also used pericardium for tricuspidization in six stenotic bicuspid aortic valves with a median follow up of 34 months. With our limited follow-up—68 months for the longest follow-up and a mean of 27 ± 18 months—our results are similarly good with a freedom from reoperation at 5 years of 81%. Further follow-up will determine the role of pericardium in aortic valve reconstruction and in this setting in particular.

Limitations

There are several limitations to generalizing the current results. The number of patients is very limited, and so is the follow-up. At this point a 5-year freedom from reoperation of 81% appears suboptimal compared with mechanical composite replacement or pulmonary autograft. On the other hand, interference with right ventricular function is avoided [27] and so is the need for anti-coagulation. In addition it must be considered that the current data include or represent the learning curve with this technique. It can be expected that the patients will need a reoperation at some point in the future. Because the root has been replaced at the first procedure, this can be expected to be a simple aortic valve replacement.

A limitation also applies to our changing approach to the aortoventricular junction. Before 2009 no stabilization was used for the basal ring. Based on our own observations we have subsequently used suture annuloplasty whenever basal diameter exceeded 27 mm. Only one of the patients who had aortoventricular stabilization had to be reoperated on, and he exhibited regurgitation after an episode of endocarditis. The cohort is also heterogeneous regarding the material of suture annuloplasty; our current observation implies that the results will improve with more patients treated by PTFE annuloplasty [16]. Nevertheless, the stability of comitant unicuspid repair and root remodeling seem to be superior to the stability of isolated unicuspid repair, underlining the importance of stabilizing the aortic root. Definitely, a longer follow-up and a bigger patient population will be necessary to confirm the present results.

Conclusions

Unicuspid aortic valve is an anatomic variant that can lead the patient to surgery at a young age and requires surgery because of valve dysfunction or aortic dilatation. Reconstructive surgery, consisting of bicuspidization of the UAV combined with root remodeling and stabilization of the aortoventricular junction, provides a promising alternative to valve replacement. First results are satisfactory, but a longer follow-up is mandatory to confirm the stability of the procedure.

References

DISCUSSION

DR YVES D’UDEKEM (Victoria, Australia): Most of the teams who have used the Yacoub technique of the root replacement have shown that the technique is doomed to fail because of the dilatation of the aortoventricular junction.

So, you’re stating that the annuloplasty you’re using is stabilizing the root. I believe that in such case of unicuspid aortic valve, you just need a partial stabilization of the root, the bit that is in front of the calcified area or the fused commissure of the unicuspid valve is usually stable and will not dilate, I believe.

In your follow-up, you have 81% freedom from aortic regurgitation at 5 years, but you had only 3 patients left at 5 years, so it’s likely to be a shorter follow-up than that. Can you tell me if in other indications you have a longer follow-up on this type of annuloplasty?

DR AICHER: To the first question, I disagree that the remodeling technique is doomed to failure because of dilatation of the aortoventricular junction—the reason is more likely a cusp problem.

To the second question, I agree that we need a longer follow-up for that operation. We have a longer follow-up for the annuloplasty, we started with 2008.

DR U’DEKEM: Specifically for the annuloplasty?

DR AICHER: We published the first data last year for that annuloplasty in about 190 patients with a bicuspid aortic valve. We found that the results were better with annular stabilization, especially in isolated valve repair.

DR ERIC E. ROSELLI (Cleveland, OH): First of all, congratulations on a very nice experience. Once again, you guys are teaching us about this disease and that is appreciated.

My approach has been to use a reimplantation instead of remodeling technique, but I think the idea is the same: to stabilize the aortoventricular junction. My question refers to the use of the autologous pericardium. In recent patients where I’ve used a similar strategy where we bicuspidize the valve and reimplant the valve, I have used the CorMatrix tissue matrix for the reconstruction. Do you have any experience with that and can you comment on it?

DR AICHER: Sorry. I think I didn’t get your question.

DR ROSELLI: My concern about the autologous pericardium is that it will calcify later on, and so the tissue matrix technology has shown that there may be the potential for tissue remodeling and regeneration. I’ve used that with some success. Do you have any experience using anything besides autologous pericardium?

DR AICHER: We used autologous pericardium in most patients and heterologous (equine) pericardium only in a few, until now I saw one pericardial patch calcifying—all the others are still doing well. I have no experience with other material than the autologous or heterologous pericardium.

DR ROSELLI: Thank you.

DR DOMENICO MAZZITELLI (Munich, Germany): Thank you.

I really enjoyed your presentation. My question is related to the two cases you had to reoperate on because of the dehiscence between the pericardial patch and native tissue. And I know previous work of your group, which also showed, among different repair techniques, that the pericardial patches were doing worse.

So my simple question is, have you never taught to resect also the so-called healthy part of the valve and put in two new patches of pericardium?

DR AICHER: We learned from our results in bicuspid aortic valve repair that the bigger the pericardial patch, the higher the risk of reoperation. That’s the reason why we only do that operation in patients where we have more than 50% of normal tissue left. We are afraid having a complete cusp out of pericardium—the risk of reoperation is probably too high.
DR PRANAVA SINHA (Washington, DC): If I heard you correctly, you treated your pericardial patch with glutaraldehyde for 2 minutes?

DR AICHER: Yes.

DR SINHA: How did you decide on that time?

DR AICHER: That’s a good question, but I don’t really have an answer for it. The reason for pretreatment with glutaraldehyde is an easier processing and prevention of shrinkage after sewing.

DR SINHA: I did some study in our lab, and the data was presented at the Western Thoracic meeting 2 years ago, and is published in the JTCVS (Journal of Thoracic and Cardiovascular Surgery). We found that if you treat the pericardium for more than 30 minutes, the level of calcification is the similar as untreated pericardium. If you treat it less than 30 minutes, you see more calcification. And that’s because if you treat it inadequately, you have partial crosslinking of the aldehyde with the protein, which is where the site for calcium binding and chelation is. So our preference if you’re going to use treated pericardium, to treat it for at least 30 minutes.

DR EZZELDIN A. MOSTAFA (Cairo, Egypt): My questions actually are about the circular annuloplasty. I assume there are three differences in our technique; namely, you are using many polyfilament (PTFE), and we use three monofilament (propylene) sutures, and we tie on beating heart according to the TEE (transesophageal echocardiography) - guided maneuvers. What do you think of these differences?

DR AICHER: We also started with an Ethibond suture, but we had some problems like erosion of the ventricular septum, and also the circumflex artery. That was the reason why we changed the material to a Gore-Tex suture. With that material we did not have any of those problems.

DR MOSTAFA: What about tying on a beating heart?

DR AICHER: We never tried this. We knot the suture before we open the aortic clamp.

DR MOSTAFA: I see. Thank you.