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Surgery for Congenital Heart Disease

The cone reconstruction of the tricuspid valve in Ebstein’s anomaly. The operation: early and midterm results

José Pedro da Silva, MD, José Francisco Baumgрат, MD, Luciana da Fonseca, MD, Sônia Meiken Franchi, MD, Lilian Maria Lopes, MD, Gláucia Maria P. Tavares, MD, Andressa Mussi Soares, MD, Luiz Felipe Moreira, MD, and Miguel Barbero-Marcial, MD

Objectives: We sought to describe a new technique for tricuspid valve repair in Ebstein’s anomaly and to report early echocardiographic results, as well as early and midterm clinical outcomes.

Methods: From November 1993 through August 2005, 40 consecutive patients with Ebstein’s anomaly (mean age, 16.8 ± 12.3 years) underwent a new surgical repair modified from Carpentier’s procedure, the principal details of which are as follows. The anterior and posterior tricuspid valve leaflets are mobilized from their anomalous attachments in the right ventricle, and the free edge of this complex is rotated clockwise to be sutured to the septal border of the anterior leaflet, thus creating a cone the vertex of which remains fixed at the right ventricular apex and the base of which is sutured to the true tricuspid valve annulus level. Additionally, the septal leaflet is incorporated into the cone wall whenever possible, and the atrial septal defect is closed in a valved fashion.

Results: There was 1 (2.5%) hospital death and 1 late death. Early postoperative echocardiograms have shown good right ventricular morphology and reduction in tricuspid regurgitation grade from 3.6 ± 0.5 to 1.2 ± 0.5 (P < .0001). After mean follow-up of 4 years, the functional class (New York Heart Association) improved from 2.6 ± 0.7 to 1.2 ± 0.4 (P < .0001). Two patients required late tricuspid valve re-repair, and there was neither atrioventricular block nor tricuspid valve replacement at any time.

Conclusions: This surgical technique for Ebstein’s anomaly can be performed with low mortality and morbidity. Early echocardiograms showed significant reduction of tricuspid insufficiency, and the follow-up showed improvement in patients’ clinical status and low incidence of reoperation.

Ebstein’s anomaly of the tricuspid valve (TV) constitutes approximately 1% of congenital heart defects and has presented an ongoing challenge since the initial repair attempts in 1958.1,2 Danielson and associates’ approach3 has accumulated a large experience in repair of the TV at the level at which it exists in the right ventricle (RV). However, the goal of eliminating tricuspid regurgitation has limited the procedure to a certain subset of anatomic variations, and TV replacement (TVR) has been necessary in 36% to 65% of cases.3–5 Carpentier and colleagues6 carefully described and reported a new technique in 1988. In contrast to the transverse plication of the atrialized chamber described by...
Danielson and associates, they plicated the RV longitudinally and also returned the TV to the anatomically correct level, thus obtaining good right ventricular morphology. The tricuspid annulus was remodeled and reinforced with a prosthetic ring. Carpentier was able to apply his procedure almost to all anatomical presentations of the disease, but hospital mortality was high (14%) in his initial series, and late complications were also important. Quaegebeur and coworkers performed a slightly modified operation without the use of a prosthetic ring. They reported no early deaths but still observed a high incidence of moderate and severe TV regurgitation. Beginning in 1989, our group developed and subsequently began to routinely perform a new surgical technique, termed cone reconstruction. It uses some principles of the Carpentier technique but reconstructs the TV in a markedly different manner: the cone-shaped valve opens to a central blood flow and closes with full coaptation of leaflets. The objective of this study is to relate in detail the technical aspects of this operation, as well as to assess the early and late clinical outcomes.

Patients and Methods
Between November 1993 and August 2005, 40 consecutive patients with Ebstein’s anomaly of the TV underwent the surgical repair described herein after informed consent was obtained from each patient and study project approval was given by the hospital review board. The patient characteristics are depicted in Table 1. The operation was indicated for the following patients: (1) those with symptomatic New York Heart Association (NYHA) functional class III or IV; (2) those with NYHA functional class I or II with cardiomegaly and a cardiothoracic ratio of 0.65 or greater; (3) those with significant cyanosis and polycythemia; (4) those with paradoxical embolism; and (5) those with tachycardia and accessory atrioventricular (AV) bundle. Five symptom-free patients did not undergo operations because they did not fulfill the above criteria. In addition, the newborn patients were excluded because they were not managed with this technique. No other patient with Ebstein’s anomaly was excluded for any reason.

Surgical Technique
After installing extracorporeal circulation with bicaval cannulation and infusing blood cardioplegia for myocardial protection, an oblique right atriotomy is performed (Figure 1, A). An incision is then initiated near the point of divergence of the displaced attachment of the anterior tricuspid leaflet from the plane of the normal annulus. This incision is then prolonged posteriorly, detaching the anterior and posterior tricuspid leaflets from their anomalous attachments in the RV as a single piece.

Next, the abnormal papillary muscles and other tissues between the individual leaflets and the corresponding right ventricular wall areas are divided, taking particular care to preserve the right ventricular apex attachments. This gives access to the subvalvular apparatus while retaining support to the eventual valve. The resulting excellent exposure permits division of fused papillary muscles, fenestration of obliterated interchordal spaces, and triangular resections on the leaflet distal third as necessary to improve the right ventricular blood inflow afterward.

Then the septal edge of the anterior leaflet is carefully mobilized by freeing its connections to the interventricular septum. Therefore only the normal attachment of the anterior leaflet to the true tricuspid annulus and the proper subvalvular apparatus are left in place (Figure 1, B). Then the free edge of the posterior leaflet is rotated clockwise and sutured to the anterior leaflet septal edge, forming a new TV resembling a cone (Figure 1, C).

An important technical consideration is management of the septal leaflet. This leaflet is hypoplastic and displaced downward in Ebstein’s anomaly. However, in 22 patients it was sufficiently developed to take part in the newly constructed TV. Therefore it was extensively mobilized by taking down its proximal edge and releasing the adhesions from the ventricular septum. When it is too short to reach the true tricuspid annulus, longitudinal elongation is done by plicating its proximal edge toward the center.

On completion of these preparations, the anterior edge of the septal leaflet is sutured to the septal edge of the anterior leaflet (Figure 2, A). Anchoring the free edge of the posterior leaflet to the other side creates a wider cone (Figure 2, B). In some cases, a short septal leaflet can be completed with the posterior leaflet tissues, which, on rotation, will form its proximal portion. In a few cases, the septal leaflet is represented by only a fibrous tissue ridge that goes from the membranous septum to the right ventricular apex. It still can be useful as an attachment for the posterior TV leaflet.

### Table 1. Preoperative patient characteristics

<table>
<thead>
<tr>
<th>Variable</th>
<th>Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age, y (range; mean)</td>
<td>1.49; 16.76 ± 12.27</td>
</tr>
<tr>
<td>Sex</td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>24 (60%)</td>
</tr>
<tr>
<td>Female</td>
<td>16 (40%)</td>
</tr>
<tr>
<td>Cyanosis</td>
<td>21 (52.5%)</td>
</tr>
<tr>
<td>Edema</td>
<td>2 (5.0%)</td>
</tr>
<tr>
<td>Left ventricular cardiomyopathy</td>
<td>2 (5.0%)</td>
</tr>
<tr>
<td>Cardiothoracic ratio (%)</td>
<td>64 ± 6.01</td>
</tr>
</tbody>
</table>

Associated heart defect

| ASD-oval foramen                        | 35 (87.5%) |
| Accessory conduction pathway            | 9 (22.5%) |
| VSD                                     | 1 (2.5%) |
| Pulmonary stenosis                      | 3 (7.5%) |
| Anomalous pulmonary venous connection   | 1 (2.5%) |

ASD, Atrial septal defect; VSD, ventricular septal defect.

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**Abbreviations and Acronyms**

- ASD = atrial septal defect
- AV = atrioventricular
- NYHA = New York Heart Association
- RV = right ventricle
- TV = tricuspid valve
- TVR = tricuspid valve replacement

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**TABLE**
After this, the atrialized RV is longitudinally plicated to exclude its thin part (used in about 80% of cases). Endocardial placement of this suture avoids damage to the coronary arteries.

The new valvular annulus is constructed at the anatomically correct level by means of plication of the true tricuspid annulus to match the proximal circumference of the already constructed cone-shaped valve (Figure 1, C), which is then attached to it with care when suturing near the AV node, where only very superficial bites should be taken to avoid heart block (Figure 1, D, and Figure 2, C).

Finally, regarding the issue of atrial septal defect (ASD) closure, the capacity to shunt right to left must be preserved postop-
eratively. The foramen ovale is thus closed in a valved fashion, usually with a single stitch (Figure 1, C and D), but the fossa ovale must be opened along its superior oblique margin if the atrial septum is found to be intact during the operation. An ostium secundum ASD, if present, is closed with a valved patch technique.

Additional surgical procedures were performed to repair the associated heart anomalies: 1 ventricular septal defect closure, 1 mitral valve repair, 3 enlargements of the right ventricular outflow tract with a monocuspid bovine pericardial graft, and 9 cases of surgical section of accessory conduction pathways.

**Echocardiography**

The echocardiograms of all patients obtained preoperatively and at hospital discharge were reviewed. The anteroposterior diameter of the true tricuspid annulus, defined as the junction between the right atrium and the RV, was measured in a 4-chamber view. The TV performance was revised for stenosis, insufficiency, or both, with the insufficiency grade classified as 1 to 4 according to the method of Suzuki and coworkers.

**Follow-up Clinical Assessment**

All patients were followed over a period of 3 to 143 months (mean, 49 months). The recent clinical condition of all patients was obtained by means of outpatient evaluation or telephone interview. The heart failure functional class, assessed by using the NYHA scale, and important medical events were recorded.

**Statistics**

The numeric data are expressed as means and standard deviations. Statistical analysis was performed with the Graphpad Prism software (version 4.0; Graphpad Software, Inc, San Diego, Calif). Preoperative and postoperative changes in functional class and grades of tricuspid insufficiency were analyzed by using the Wilcoxon signed-rank test, and the change in TV annulus size was analyzed by using the Student paired t test.

**Results**

The cardiopulmonary bypass time varied from 45 to 180 minutes (mean, 104 ± 33 minutes), and the aortic cross-clamp time went from 25 to 115 minutes (mean, 70 ± 21 minutes). The single hospital death (2.5%) in this series occurred as a result of biventricular hypoxic cardiomyopathy. Although the TV repair was effective, low cardiac output was the cause of death on the fourth postoperative day.

The preoperative and early postoperative echocardiographic results, as well as the preoperative and recent functional class, are shown in Table 2. There was a reduction in tricuspid annulus diameter postoperatively because of surgical plication, which did not result in definitive TV stenosis. The 2 patients who presented with early transvalvular peak gradients of 8.0 and 11.7 mm Hg had these gradients decreased to 5.0 and 7.5 mm Hg, respectively, on late echocardiographic studies. The echocardiogram of patient 32 (Figure 3), a 4-year-old girl, besides indicating good positioning of the TV and restoration of the normal right ventricular morphology, also shows some growth of the tricuspid annulus, the anteroposterior diameter of which went from 14.9 to 17.4 mm in 1 year.

There was 1 late death caused by TV bacterial endocarditis after dental infection. Reoperation was required in 2 patients (the first and twelfth patients) because of severe tricuspid regurgitation: both underwent repeat valve repair at the fourth and fifth postoperative years, respectively. The first patient is currently in excellent clinical condition, and the other still has grade 3 TV regurgitation and moderate right ventricular dysfunction. One patient with severe biventricular cardiomyopathy had a stroke in the sixth postoperative year. He is presently in the seventh postoperative year, and despite taking anticoagulants and optimized medical therapy for heart failure, he persists in functional class IV. The remaining patients are event free to date, and 1 patient even gave birth to a healthy baby after an uneventful pregnancy 2 years after the operation. There was neither TVR nor pacemaker implantation in any patient from this series.

Of the 9 patients with Wolff–Parkinson–White syndrome who had surgical division of the accessory AV conduction pathway, 8 were successful. The failed case (in the twelfth patient) was submitted to repeat catheter ablation attempts afterward. Eventually, the patient underwent reoperation because she also had TV regurgitation.

**Discussion**

This technique was conceived while bearing in mind Carpenter’s concepts of bringing the TV leaflets to the true tricuspid annulus level and longitudinal plication of the atrialized RV as necessary to restore the right ventricular volume and morphology in Ebstein’s anomaly repair. Nev-

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**Table 2. Early postoperative echocardiographic results and recent functional class of heart failure compared with preoperative data**

<table>
<thead>
<tr>
<th></th>
<th>Preoperative</th>
<th>Postoperative</th>
<th>P value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Tricuspid insufficiency, n</td>
<td></td>
<td></td>
<td>&lt;.0001</td>
</tr>
<tr>
<td>Grade 1</td>
<td>0</td>
<td>31</td>
<td></td>
</tr>
<tr>
<td>Grade 2</td>
<td>1</td>
<td>5</td>
<td></td>
</tr>
<tr>
<td>Grade 3</td>
<td>15</td>
<td>3</td>
<td></td>
</tr>
<tr>
<td>Grade 4</td>
<td>24</td>
<td>0</td>
<td></td>
</tr>
<tr>
<td>TTA AP diameter, mm</td>
<td>34.3 ± 6.5</td>
<td>15.1 ± 3.7</td>
<td>&lt;.0001</td>
</tr>
<tr>
<td>Functional class (NYHA), n</td>
<td></td>
<td></td>
<td>&lt;.0001</td>
</tr>
<tr>
<td>Class I</td>
<td>5</td>
<td>32</td>
<td></td>
</tr>
<tr>
<td>Class II</td>
<td>8</td>
<td>5</td>
<td></td>
</tr>
<tr>
<td>Class III</td>
<td>25</td>
<td>0</td>
<td></td>
</tr>
<tr>
<td>Class IV</td>
<td>2</td>
<td>1</td>
<td></td>
</tr>
</tbody>
</table>

**NYHA, New York Heart Association; TTA, true tricuspid annulus; AP, anteroposterior.**
Nevertheless, it differs markedly in terms of surgical details, as well as the resulting TV geometry and function. Carpentier and colleagues detach the dislocated TV from the interior of the RV, bring it to the normal level of the tricuspid annulus, and rotate it partially only to reach the septal tricuspid region. Therefore reimplantation is to proceed in a monocuspid format, thus generating an off-center diastolic blood flow. In contrast, the cone reconstruction proceeds...
through a complete clockwise rotation of the lateral margin of the TV megaleaflet while preserving its right ventricular apex attachments. Furthermore, the entire circumference of this cone base (which might or might not include a contribution from the septal leaflet, depending on its state of development) is sutured to the normal level of the tricuspid annulus. Unlike with the method of Carpentier and colleagues, this includes the septal region, resulting in central diastolic blood flow and restoration of the septal leaflet rule on the TV closure mechanism. AV block, a complication not observed in this series of patients, is avoided by the use of very superficial sutures near the AV node area.

Of note, this technique might reduce the tricuspid annulus to a smaller size than the mitral annulus, raising concerns about TV stenosis; however, to date, that problem has not existed. Although mild early TV stenosis occurred in 2 patients, it reversed on follow-up echocardiographic studies. The inclusion of the septal leaflet to enlarge the newly reconstructed TV seems to add in the TV stenosis prevention, particularly in adult patients. The rotation of cordless posterior leaflet tissue to be fitted to the hypoplastic septal leaflet, completing its proximal aspect, is a helpful maneuver to obtain a bigger TV.

As in the series of Quaegebeur and coworkers, we did not use an annuloplasty ring device. That seems to be important in children because a permanent fixation of an already diminished tricuspid annulus could result in late stenosis. Figure 3 shows an example of TV annulus growth while keeping a good coaptation of TV leaflets and, consequently, no regurgitation. Additionally, systolic constriction of the tricuspid annulus was seen at the same echocardiogram. This annular flexibility, which might contribute to the valve opening and closing mechanism, was observed thanks to echocardiographic markers from interrupted suture knots placed at the true tricuspid annulus. The possibility of tricuspid annulus growth and flexibility, good midterm clinical outcomes, and few reoperations in this series of patients stand against routine use of the tricuspid ring with this procedure but do not rule out its need in older patients, especially in those with friable valvular tissues.

In operations described by Carpentier and colleagues and Quaegebeur and associates, a monocusp mechanism of closure is the result, and their follow-up studies documented a substantial incidence of TV regurgitation. It is hoped that the full circumferential attachment to the TV annulus described in this procedure, providing a leaflet-to-leaflet coaptation, will reduce that incidence of regurgitation, but it has to be confirmed by late anatomic and functional studies.

It is reassuring that no AV block occurred in any of the patients. This coincides with the results of Wu and Huang’s recent series. They used fresh autologous pericardium in 80% of their patients to reconstruct the septal portion of the TV, which they suture a little below the septal annulus level. As in this series, no occurrence of AV block was observed after suturing at the septal portion, although they also touched on the importance of placing superficial sutures in this area.

Ullmann and associates reported a technique using the septal tricuspid leaflet, which had its proximal edge mobilized and sutured to the septal tricuspid annulus area. They also did not have any cases of heart block. It should be emphasized, however, that their approach did not include plication of the right ventricular atrialized portion or reduction of the tricuspid annulus, and as they themselves have pointed out, that technique is limited to patients with more favorable anatomy, with replacement of the TV being necessary in 5 of 29 patients. Besides being different, this operation was not restricted to only patients with well-developed septal leaflets, which represent a small portion of the wide anatomic spectrum of Ebstein’s anomaly. Actually, it was applied to all anatomic variations presented in 40 consecutive patients with Ebstein’s anomaly. In 22 of them, the septal leaflet was incorporated in the reconstructed valve, and in 18 patients the septal leaflet was disregarded, but the idea of making a cone was kept in all patients.

In general, the surgical technique to approach the ASD in Ebstein’s anomaly has not been a major concern to authors performing only routine closure. In this cohort a valved interatrial communication was always provided, allowing only right-to-left blood shunting when the atrial pressure becomes greater in the right side. This has proved useful in the early postoperative course of 5 patients with important right ventricular dysfunction, who were nevertheless able to maintain good systemic cardiac output, albeit with mild oxygen desaturation that got better once the RV improved. Chauvaud has already reported improvement in results with the use of a bidirectional cavopulmonary shunt used as an adjunctive procedure to Carpentier’s operation in patients with severe right ventricular dysfunction. Lately, Chauvaud and colleagues have used that technique in 36% of patients, with the goal of decreasing the right ventricular preload in cases of severely depressed right ventricular contractility. With that procedure, they had a significant reduction in mortality caused by right ventricular failure. Alternatively, in this series a unidirectional ASD was left, aiming to decompress the RV and to increase left ventricular preload by means of temporary right-to-left shunting. Volume, inotropic drugs, and vasodilators were used to improve the right ventricular performance and consequently increase the pulmonary perfusion. The cavopulmonary shunt was a strategy reserved only as a next-step procedure for cases of low right-sided cardiac output insufficient to maintain reasonable systemic oxygen saturation, which did not happen in any patient. It is necessary to point out, though, that the 2 series might not be comparable because in
this series the patients were younger and presented with lower mean cardiothoracic ratios than in the series of Chauvaud and colleagues.15

The Danielson operation, despite some technical modifications, remains highly associated with the need for TVR. Kiziltan and coworkers4 reviewed their series of 323 patients with Ebstein’s anomaly, with TVR performed in 158 (48.9%) patients. In regard to long-term results, they found that the freedom from bioprosthesis replacement was 97.5% ± 1.9% after 5 years and 80.6% ± 7.6% after 10 years. They also found no statistically significant difference at 10 and 12 years in freedom from reoperation after TVR compared with freedom from reoperation after TV repair. These good results, according to the authors, might be related to the large size of bioprosthesis that can be implanted relative to patient somatic size and to the normally low right ventricular systolic pressure in patients after Ebstein’s anomaly repair. However, these results are for a limited period and do not rule out the ultimate need for tricuspid prosthesis replacement and therefore do not decrease the importance of creating an efficient and durable TV repair operation.

The indications for surgical intervention in patients with Ebstein’s anomaly remain controversial in asymptomatic patients, although the natural history of the disease is a relentless progression to congestive heart failure, arrhythmias, or both16,17 in the majority of patients not undergoing operations. Mortality for these late-stage complications is high.18,19 It seems also to be true that surgical treatment at late stages has less chance of reversing the ravages of the disease completely. That might be the case in 1 early death and another late progression to heart failure in this series. These events seem to be related to the state of the 2 patients who had left ventricular myocardiopathy preoperatively rather than to the surgical technique. This further reinforces the notion that surgical intervention should come earlier, before deterioration of right and left ventricular function.

In conclusion, this surgical technique that reconstructs the TV in a cone shape, which results in a central flow through the tricuspid orifice and a full coaptation of the leaflets, can be performed with low mortality and morbidity. Early echocardiography showed significant reduction in tricuspid insufficiency, and the follow-up showed clinical improvement in the majority of patients, low incidence of reoperations, and no need for TVR. Further studies and longer follow-up are required to evaluate the behavior of the TV and RV after this procedure.

References

Discussion
Dr Jan M. Quaegebeur (New York, NY). Dr da Silva, I would like to congratulate you on the presentation of an ingenious technique. It is interesting to note that although Ebstein’s anomaly is a very rare condition, there have been 3 presentations dedicated to this anomaly in this meeting. Therefore the quest for a better repair of Ebstein’s anomaly continues. You have described your experience over 10 to 12 years with an innovative technique, which tries to improve TV function by realizing a complete coaptation between valvular tissue in comparison or in contrast with the previous repairs by all of us that relied on a monocuspid or bicuspid valve where the coaptation is between the valve leaflets, namely the anterior and posterior leaflets, and the ventricular septum. Forty patients were presented with a low early mortality rate and only 1 late death, which are very good results.
I am not entirely clear about the age of your patients. You said that the mean age was 16.8 years, but I would like it if you could answer briefly how many patients were less than 10 years of age, for instance.

Dr da Silva. Well, the youngest in this series was 22 months old, and the oldest, I think, was 49 years old. Afterward, we had more patients, and among them there was a 3-month-old girl who underwent repair with this technique.

I will say that we have 16 patients younger than 13 years. Therefore 24 patients were older than that age. But I think it is better to use this technique in younger patients, especially those less than 18 years of age.

Dr Quaegebeur. You have provided me with the article and beautiful illustrations, I must say. It happens that last week I had a patient on the schedule with Ebstein’s anomaly, and therefore I said, well, we will see how we can maybe improve the technique because, theoretically, it would be better to have coaptation between leaflet tissue than with leaflet tissue and septal tissue. Invariably, in our experience the residual tricuspid incompetence is at the level at which you have absent leaflet tissue. Therefore the concept, I think, is very sound.

This patient had muscularization of the posterior leaflets, which happens quite often. There was absolutely no leaflet tissue at the level of the septum. You have described the attachment of the anterior leaflet at the level of the anterosetal commissure like a bit of fibrous continuity before it reaches the papillary muscle of the anterior leaflet. This patient did not have such an attachment. In my experience, I could not perform this technique.

There is a great variability in the morphology of the valve in Ebstein’s malformation. My question is this: Are you always able to perform this technique, or are there patients in whom you do something different?

Dr da Silva. Well, in the type D of Carpentier’s classification, I think we have to go to valve replacement. I also think that in older patients, if you have difficulty finding tissue enough to construct a good tricuspid valve, it is good to indicate valve replacement or the Carpentier or other repair technique if possible. But in my experience, all cases were done with this technique. If you push to it, you can do a lot of maneuvers that can compensate for having little tissue. One of them is to take down the posterior leaflet from its attachment without support and then get the support for that leaflet at the septal tissues. You can have some residual, malformed septal leaflet that can be useful as a subvalvular support for the posterior leaflet. And with that I think you can reconstruct the cone the way I did many times. Actually, I have movies on that concept, I think, is very sound.

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important adjunct to the operation that I have started to do was
introduced by Dr Sano. This involves resecting a portion of the RV
to reduce its size. I now resect the inferior wall of the RV between
the acute margin and the posterior descending coronary artery and
parallel to the right coronary arterial branches, and therefore cor-

onary artery compromise is eliminated or minimized. I think
anything to reduce the size of the large atrialized RV on the other
side of the tricuspid repair will help in the long term. I congratulate
you and the authors and look forward to the opportunity to apply
some of your techniques.

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