

## Mechanisms of tricuspid regurgitation in patients with hypoplastic left heart syndrome undergoing tricuspid valvuloplasty

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**Objectives:** Tricuspid regurgitation (TR) remains a risk factor for morbidity and mortality through staged palliation in patients with hypoplastic left heart syndrome (HLHS). Reports on the mechanisms associated with TR in patients with HLHS are limited. Thus, we sought to describe our experience with tricuspid valve (TV) repair in these patients, focusing on the mechanisms of TR and corresponding surgical techniques.

**Methods:** We performed a retrospective single-center review (January 2000 to December 2012) of patients with HLHS undergoing TV repair and completing Fontan circulation. We evaluated the pre- and postoperative echocardiograms, intraoperative findings, and surgical techniques used.

**Results:** A total of 53 TV repairs were performed in 35 patients with HLHS completing staged palliation. TV repairs were performed at stage II in 15, between stage II and III in 4, at stage III in 27, and after stage III in 7. The surgical techniques for valvuloplasty included annuloplasty (38%), anteroseptal (AS) commissuroplasty (66%), anterior papillary muscle repositioning (11%), multiple commissuroplasties (9%), septal-posterior commissuroplasty (9%), and fenestration closure (4%). The predominant jet of TR emanated along the AS commissure in 68% of the cases. All patients survived the procedure and were discharged. Preoperative echocardiography showed a dilated TV annulus on the lateral dimension, anteroposterior dimension, and area that was significantly reduced after TV repair ( $P < .0001$ ). The preoperative mean TR, as assessed by lateral ( $P = .002$ ) and anteroposterior ( $P = .005$ ) vena contracta, was also significantly reduced after TV repair. TV repair did not significantly affect right ventricular systolic function immediately after surgery ( $P = .17$ ) or at the most recent follow-up visit ( $P = .52$ ). Patients with anterior leaflet prolapse were at increased risk of worse outcomes, including moderate or greater right ventricular dysfunction ( $P = .02$ ). Patients requiring reoperation for TV repair were younger (6.3 vs 28.1 months,  $P < .0001$ ) at the initial operation. One patient died of heart failure. Freedom from TV replacement and transplant-free survival were both 97% at the most recent follow-up point.

**Conclusions:** TR in patients with HLHS commonly emanates from the AS commissure. The associated mechanisms are often annular dilatation and anterior leaflet prolapse. Preoperative anterior leaflet prolapse was associated with worse outcomes. Annuloplasty, closure of the AS commissure, and repositioning of the anterior papillary muscle are effective in addressing TR in the short- and mid-term in this challenging population. (*J Thorac Cardiovasc Surg* 2014;148:832-40)

The outcomes of patients with hypoplastic left heart syndrome (HLHS) have improved dramatically during the past 2 decades.<sup>1-3</sup> However, tricuspid regurgitation (TR) remains a risk factor for adverse outcomes in this challenging population.<sup>4-7</sup> Despite significant TR in 25% of survivors through staged palliation, few data exist on

the underlying pathophysiology of TR in patients with HLHS. The reported mechanisms of TR in HLHS are complex and multifactorial and include structural abnormalities of the tricuspid valve (TV) and functional causes such as right ventricular (RV) dysfunction and dilatation of the TV annulus.<sup>8</sup> Moreover, increasing age, with geometric changes of the annulus, leaflet prolapse, papillary muscle (PM) displacement, and subsequent leaflet tethering have been associated with TR in patients with HLHS.<sup>9,10</sup>

Several reports have suggested that TV repair decreases the severity of TR and could improve the outcomes in patients with HLHS.<sup>11-14</sup> De Vega type annuloplasty, selective annuloplasty, cleft closure, chordal shortening, posterior leaflet obliteration, and commissure closure have been used to repair the TV in this population; however, no standard surgical approach exists. Recent advances in echocardiography and the understanding of the

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### Abbreviations and Acronyms

|      |                                   |
|------|-----------------------------------|
| AP   | = anteroposterior                 |
| AS   | = anteroseptal                    |
| HLHS | = hypoplastic left heart syndrome |
| IQR  | = interquartile range             |
| PM   | = papillary muscle                |
| RV   | = right ventricular               |
| TR   | = tricuspid regurgitation         |
| TV   | = tricuspid valve                 |

mechanisms of RV and TV function in the systemic circulation have made it possible to systematically evaluate the TV preoperatively with the aim of tailoring the surgical technique to specific anatomic abnormalities. Nevertheless, significant discrepancies between the echocardiographic and surgical findings have been reported and have complicated this approach.<sup>11</sup>

The purpose of our study was to evaluate the underlying echocardiographic mechanisms of TR in patients with HLHS completing staged palliation and correlate those with the efficacy and durability of the surgical approach undertaken.

## METHODS

A retrospective analysis was undertaken of all patients with HLHS completing staged palliation through the Fontan procedure at Boston Children's Hospital from January 2000 through December 2012. The cardiac surgery and cardiac intensive care unit databases, hospital medical records, data from the echocardiography laboratory, and information from referring cardiologists were reviewed. All patients were followed up until death, lost to follow-up, or January 2014. Patients with anatomic variants without a morphologic right ventricle as the systemic ventricle were excluded from the present study. It has been consistently reported in published studies that patients showing significant TR before the Norwood procedure are at high risk of morbidity and mortality and thus will not complete staged palliation.<sup>12</sup> Because the main goal of our study was to evaluate the underlying mechanisms of TR through follow-up of patients completing staged palliation and because TV plasty was not routinely performed during the early period of our study, we excluded patients who had undergone TV repair at stage I. The institutional review board approved the present study.

The pre- and postoperative echocardiograms were reviewed, and the parameters of interest were measured by a single experienced echocardiographer (D.W.B.). The existence and degree of TR was assessed with echocardiography using the 2-dimensional lateral and anteroposterior (AP) width of the vena contracta in the orthogonal planes and were categorized as none or trivial (0-0.20 cm), mild (0.20-0.40 cm), moderate (0.40-0.60 cm), or severe (>0.60 cm). In addition, TR was subjectively expressed as 0 (none or trivial), 1 (mild), 2 (moderate), or 3 (severe). The location of the regurgitant jet from the en face views was recorded. The mechanism of regurgitation was ascertained by noting the functional and structural abnormalities, such as valve morphology, leaflet prolapse, leaflet restriction, clefts, and ruptured chord. Prolapse was defined as any leaflet tissue above the plane of the tricuspid annulus in systole. The TV area and AP and lateral diameters were measured, and the Z-scores based on the patients' body surface area were calculated using our own echocardiography laboratory's database.<sup>15</sup> RV function was measured by subjective assessment and categorized as 0 (none or trivial), 1 (mild), 2 (moderate), or 3 (severely depressed).

The operative descriptions of the TV repair were compared with the echocardiographic data. The surgical techniques used were according to surgeon preference. To assess the durability of the TV repair, the echocardiogram at the most recent follow-up visit was reviewed for the degree of TR and assessment of RV function.

Generally, the TV was inspected in patients with HLHS with equal or greater than mild to moderate TR at staged palliation. After opening the right atrium, the surgeon checked the valve morphology and tested the competence with saline. We have previously described different surgical techniques used for TV repair.<sup>13,14</sup> In brief, simple annuloplasty and commissuroplasty were the 2 major techniques used in patients with annular dilatation and/or leaflet prolapse. Partial annuloplasty was performed with a single stitch similar to the De Vega annuloplasty. Less commonly, a prosthetic ring was used. Prolapsing leaflets were resuspended by approximation with interrupted fine polypropylene suture to the adjacent leaflet, thus closing the commissure and improving the linearity of the coaptation area. More recently, we have incorporated repositioning of the anterior PM of the TV toward the septum to improve ventricular sphericity and valve tethering.<sup>13,16</sup> Less commonly used techniques included chordal shortening for chordal elongation and the use of artificial chords for rupture chords.

## Statistical Analysis

If continuous variables were normally distributed, the mean  $\pm$  standard deviation values were used and the median and interquartile range (IQR) if not. The pre- and postoperative changes in the continuous variables, TV dimensions and vena contracta, were compared using a paired *t* test. Associations between dichotomous variables were compared using Fisher's exact test. In the case of the normal assumption not meeting, the analysis of 3 independent variables was performed using the asymptotic Kruskal-Wallis test. For 2 independent variables, the asymptotic Wilcoxon Mann-Whitney rank sum test was used. The Kaplan-Meier curve was used to estimate the freedom of RV dysfunction. All test and graphs were performed using R software.<sup>17</sup>

## RESULTS

From January 2000 through December 2012, 53 TV repairs were performed in 35 patients with HLHS who had completed staged palliation through the Fontan procedure. All patients had mild to moderate or greater TR preoperatively. TV repair was performed at stage II in 15, between stage II and III in 4, at stage III in 27, and after stage III in 7. The characteristics of TV repair are listed in [Table 1](#). The median age at the initial TV repair was 23.20 months (IQR, 6.31-194.0). All patients were alive at discharge without TV reintervention. Seventeen patients required repeat TV repair at a median age of 36.50 months (IQR, 3.81-97.3), with an interval between repairs of 23.30 months (IQR, 18.4-68.7).

## Perioperative Echocardiography

The preoperative echocardiograms showed the predominant jet of TR emanating along the anteroseptal (AS) commissure in 36 (68%), centrally in 12 (23%), and posteriorly in 5 (8%) of the 53 procedures. Preoperative anterior TV leaflet prolapse was identified on the echocardiogram in 31 of the 53 procedures (58%) and was the most common cause of TR. Preoperative echocardiography showed a dilated TV annulus at the lateral dimension, AP dimension,

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