Original Article

Biventricular remodeling after cone repair in Ebstein anomaly: magnetic resonance imaging data analysis

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Background: Various surgical techniques have been reported for repairing Ebstein anomaly. Cone repair provides nearly anatomical tricuspid valve (TV) reconstruction with promising outcomes. We reviewed our experience with cone repair to evaluate biventricular remodeling and the outcomes of the annular support procedure.

Methods: Between January 2008 and December 2021, cone repair was performed in 33 consecutive patients with Ebstein anomaly. Mean age was 32.0±16.8 years (range, 1.1–66.8 years). Previous TV repair had been performed in two patients with the Hetzer and the Carpentier techniques (6%). Severe tricuspid regurgitation (TR) was observed in 32 patients (97%). Twenty patients had preoperative magnetic resonance imaging (MRI) data; 15 patients had postoperative MRI data.

Results: Modifications included the addition of an annuloplasty band (21 patients with pericardial strips, two patients with prosthetic rings) in 23 patients (69.7%) and papillary muscle repositioning in one patient (3%). Bidirectional cavopulmonary anastomosis was performed in two patients (6%), with one of them undergoing a Fontan operation in the third postoperative years. No mortality was observed. The mean follow-up duration was 7.5±4.6 years. Two patients (6%) required late TV re-repair in the first and sixth postoperative years. At follow-up, five patients (16.1%) reported no or trivial TR, 16 (51.6%) had mild TR, and 5 (16.1%) had mild to moderate TR. Freedom from late TV reoperation was 78.8%±13.4% at 5 years. The TV reoperation rate was significantly low in the patients who underwent tricuspid annuloplasty with a band (P=0.02). Preoperative and postoperative MRI data demonstrated a significant right ventricular (RV) volume decrease after cone repair [RV end-diastolic volume index (mL/m2): preoperative/postoperative =207.4±40.2/105.5±41.3, P=0.001]. Left ventricular ejection fraction (LVEF) remained unchanged after cone repair, while left ventricular stroke volume (LVSV) significantly increased [LVEF (%): preoperative/postoperative =60.8±5.3/61.2±5.4, P=0.10; LVSV (mL): preoperative/postoperative =64.0±1.8/71.4±12.7, P=0.041].

Conclusions: Cone repair for Ebstein anomaly has low mortality and morbidity rates. The addition of an annuloplasty band was associated with a low incidence of the TV reoperation. Moreover, cone repair might impact left ventricular function due to ventricular interdependency. Longer follow-up is essential to determine the late durability of cone repair and both ventricular functional changes.

Keywords: Congenital heart disease; Ebstein anomaly; tricuspid regurgitation (TR); heart valve surgery

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Introduction

Ebstein anomaly is a rare disease, accounting for 1% of congenital heart diseases (1). The malformation is right ventricular (RV) myopathy with failure of tricuspid delamination and highly variable tricuspid valve (TV) morphology. Ebstein anomaly is characterized by downward displacement of the hinge point of the septal leaflets towards the apex of the RV, dilatation of the tricuspid annulus, and atrialized RV (2).

This anomaly has a broad clinical presentation ranging from a highly symptomatic neonate to an asymptomatic adult. Most patients with Ebstein anomaly require surgical management at some point in their life (3).

Surgical management of Ebstein anomaly has evolved, with cone repair providing a near anatomic correction, advancing from earlier repairs based on anterior leaflet monocusp coaptation with the ventricular septum (4-6).

Some studies have reported magnetic resonance imaging (MRI) data-based biventricular function and volume changes after cone repair (7-9). This study aimed to analyze our experience with cone repair in patients with Ebstein anomaly and late biventricular remodeling. We present this article in accordance with the STROBE reporting checklist (available at https://jtd.amegroups.com/article/view/10.21037/jtd-2024-2185/rc).

Highlight box

Key findings

- Cone repair for Ebstein anomaly shows low mortality and morbidity, with significant biventricular remodeling. Adding an annuloplasty band reduces tricuspid valve (TV) reoperation rates (P=0.02). Right ventricular volume decreased significantly post-repair (P=0.001), and left ventricular stroke volume improved (P=0.041).

What is known and what is new?

- Cone repair reconstructs the TV with favorable outcomes.
- This study highlights improved biventricular remodeling, reduced TV reoperation with annuloplasty bands, and detailed magnetic resonance imaging (MRI)-based functional insights.

What is the implication, and what should change now?

- Cone repair is durable and enhances ventricular function. Longer follow-up is needed to assess late durability. Annuloplasty bands should be standard, and routine MRI can improve surgical planning and outcomes.

Methods

Patients

The study was conducted in accordance with the Declaration of Helsinki and its subsequent amendments. The study was approved by the Institutional Review Board of Bucheon Sejong General Hospital (No. 2023-12-007-004) and individual consent for this retrospective analysis was waived. Between January 2008 and December 2021, 33 consecutive patients with Ebstein anomaly of the TV underwent cone repair.

Indications for surgery included symptoms of fatigue, cyanosis, shortness of breath, or decreased exercise tolerance as a symptom of heart failure. Progressive RV enlargement, RV dysfunction, or atrial tachyarrhythmia supported the need for surgery in the absence of symptoms. All cases referred for surgery were discussed at a multidisciplinary team conference for surgical considerations and final decision-making.

Surgical procedure

The cone reconstruction technique was performed as described in previous reports (4-6). The principle of cone repair involves complete surgical delamination and mobilization of all available leaflet tissues, which are reattached to the true right atrioventricular junction, creating a 360-degree leaflet cone (*Figure 1*). Briefly, surgical circumferential leaflet delamination was performed from the underlying RV. Mobilized leaflets (anterior, diminutive inferior, and septal) were joined to side to side to create a circumferential cone of leaflet tissue. Internal RV plication was performed on smooth, non-trabeculated, inferior RV wall endocardium extending from apex to annulus. Plication sutures were carefully placed to avoid the epicardial branches of the right coronary artery. Plication typically crosses the true annulus to partially reduce the size of the dilated annulus.

Annuloplasty was implemented in subsequent patient series to stabilize the annulus and to prevent postoperative dehiscence of the valve leaflets. The indication for annuloplasty was the anatomic annulus size and fragility. Annuloplasty was performed when intraoperative findings indicated a significantly dilated anatomic annulus with fragile tissue, suggesting a high risk of dehiscence. Fresh auto-pericardium, fixed auto-pericardium, bovine

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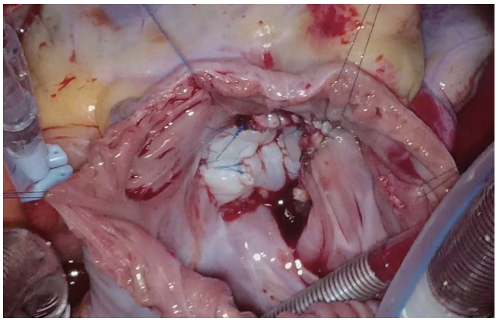

Figure 1 Intraoperative findings of cone procedure.

pericardium and commercial annuloplasty ring was used for annuloplasty. The materials for annuloplasty were determined based on the surgeon's preference. 'Band annuloplasty' means partial annuloplasty. We used the term 'band' to describe a non-commercial ring annuloplasty. Pediatric patients underwent partial annuloplasty. The main purpose of annuloplasty was annular reinforcement to prevent dehiscence.

The sutures for valve reattachment were shifted a few millimeters inferior from the vein of D to avoid an iatrogenic heart block (10). Moreover, a bidirectional cavopulmonary shunt (BCPS) has been applied selectively, generally for severe RV dysfunction (11-13).

Echocardiography

Echocardiography was routinely performed preoperatively, intraoperatively and at discharge. Subsequent, outpatient department-based follow-up echocardiogram was performed at Bucheon Sejong General Hospital (mean interval after operation: 5.9±4.5 years) postoperatively. The anteroposterior diameter of the true annulus, defined as the junction between the right atrium and the RV, was measured in a four-chamber view. The degree of tricuspiand hepatic venous flow change). Additionally, Tricuspid regurgitation (TR) was graded as follows: 0, none; 1, trivial; 1.5 trivial to mild; 2, mild; 2.5, mild to moderate, 3, moderate; 3.5, moderate to severe; and 4, severe. To assess late TV function, data of the last available echocardiography were analyzed.

Conventional echocardiographic criteria were employed to evaluate RV dysfunction including fractional area change, tricuspid annular plane systolic excursion, and qualitative assessment. Significant RV dysfunction was defined as moderate or severe ventricular dysfunction on discharge echocardiography.

Cardiac magnetic resonance

Cardiac MRI was performed preoperatively and postoperatively using a Philips INGENIA 3.5T (Philips medical system, Netherlands). Subsequent, outpatient department-based follow-up cardiac MRI was performed at Bucheon Sejong General Hospital (mean interval after operation: 5.2 years) postoperatively. The atrialized ventricle included for RV volume measurements. Ventricular function and volumetric data were quantified using the following markers, indexed for body surface area as appropriate: RV and left ventricular (LV) ejection fraction (LVEF) (in percentage), RV and LV cardiac index (CI) (in liters per minute per meters^2), RV and LV stroke index (in milliliters per meters^2). Biventricular remodeling was assessed by measuring planimetered area: anatomic RV, LV stroke volume (SV), and body surface area-indexed ventricular volumes (in milliliters per meters^2): RV and LV end-diastolic volume index (EDVi), RV and LV end-systolic volume index (ESVi).

Statistical analysis

Preoperative and postoperative data were collected from the patients' medical records. The characteristics of the study population were expressed as frequencies, medians with ranges, or means with standard deviations, as appropriate. Paired *t*-test analysis was used to compare the preoperative and postoperative TR, TV anteroposterior diameter. Overall survival and freedom from TV reoperation were analyzed using Kaplan-Meier survival analysis with a log-rank test to compare the factors. One patient who underwent TV replacement 2 days after cone repair was excluded from the analysis for freedom from late TV reoperation and comparison of echocardiographic evaluation before and after surgery. The SPSS version 21.0 software (SPSS, Inc, Chicago, IL, USA) was used for all statistical analyses, and a P value <0.05 was considered statistically significant.

Results

Baseline characteristics

The mean age was 32.0±16.8 years (range, 1.1 to 66.8 years),

Table 1 Preoperative patient characteristics

Variables	Value (n=33)
Age, years	29.9 (18.1–45.4)
Weight, kg	56.9 (49.0–68.5)
Body surface area, m^2	1.6±0.4
Gender	
Female	22 (66.7)
Male	11 (33.3)
Symptoms	
Exercise intolerance	13 (39.4)
Fatigue	4 (12.1)
Cyanosis	3 (9.1)
Palpitations/prior syncope	7 (21.2)
LV dysfunction (EF <50%)	2 (6.1)
Associated heart defect	
ASD	23 (69.7)
Atrial arrhythmia	9 (27.3)
WPW syndrome	6 (18.2)
Atrial fibrillation	1 (3.0)
Atrial flutter	3 (9.1)
Supraventricular tachycardia	5 (15.2)
Previous catheter ablation	6 (18.2)
Previous cardiac operation	
TV repair	2 (6.1)

Data are presented as n (%), mean ± SD or median (IQR). ASD, atrial septal defect; EF, ejection fraction; IQR, interquartile range; LV, left ventricle; SD, standard deviation; TV, tricuspid valve; WPW, Wolff-Parkinson-White.

with 8 (24.2%) individuals being children (less than 18 years of age). The mean length of follow-up was 7.5±4.6 years. Patient demographic characteristics are presented in *Table 1*.

Operative data

Technical modifications of cone repair are shown in *Table 2*. TV annulus support procedure was performed in 23 patients, a pericardial strip in 21, and a prosthetic ring in two. Papillary muscle repositioning was performed on one patient. Previous TV repair had been performed in two patients with Hetzer and Carpentier techniques (6%). The concomitant procedures included RV plication in 29 cases, atrial septal defect closure in 22 cases, right atrial reduction in 9 patients, BCPS in 2 patients, RV outflow track widening in 1 patient, and septal myectomy via aortotomy in 1 patient. Antiarrhythmic procedures were performed in nine patients (four right atrial maze procedures, four cavotricuspid isthmus ablations, and one biatrial maze procedure). The associated procedures are listed in *Table 2*.

Clinical outcomes

No early or late mortalities were reported. A 2 years-old-girl who initially underwent cone repair and BCPS developed progressive RV dysfunction and underwent extracardiac conduit Fontan operation 3 years after first operation. Furthermore, early TV reoperation was performed. A 26-years-old man who underwent cone repair and right-sided maze procedure for severe TR and atrial flutter developed acute severe TR due to TV detachment. Subsequently, he underwent TV replacement 2 days after initial operation. Two cases of late TV reoperation were identified. One case was of a 1-year-old boy who underwent cone repair with BCPS for severe TR and RV dysfunction. He developed RV dilatation and moderate to severe TR and underwent RV reductionplasty and TV re-repair 6 years after first operation. A 54-year-old man initially underwent cone repair for severe TR. One year after the operation, the patient underwent TV ring annuloplasty for severe TR due to TV detachment. At five years, freedom from TV reoperation was 78.8%±13.4% (*Figure 2*). The TV reoperation rate was significantly lower in patients who underwent tricuspid annuloplasty. The log-rank test yielded a P value of 0.02, while the additional Chi-square test showed a similar result, with a P value of 0.02, confirming the protective effect of annuloplasty (*Figure 3*). TV reoperation rate was not different between patients under 18 years old and those over 18 years old (P>0.99).

Echocardiography for TV function

The preoperative, predischarged, and last echocardiographic data are displayed in *Table 3*. No clinically significant TV stenosis was observed during the immediate postoperative period. One patient exhibited a moderate degree of tricuspid stenosis on the latest echocardiogram obtained 5.9 years after surgery.

Before discharge, except for patients who underwent

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Table 2 Procedural data and clinical outcomes

Variables	<18 years (n=8)	≥18 years (n=25)	Total (n=33, %)
Concomitant procedure			
Right ventricle plication	8	21	29 (87.9)
ASD closure	6	16	22 (66.7)
Right atrial reduction	3	6	9 (27.3)
Bidirectional cavopulmonary shunt	2	0	2 (6.1)
Right ventricular out track widening (PV valvotomy, MPA widening)	1	0	1 (3)
Septal myectomy	0	1	1 (3)
Antiarrhythmic procedure			
Right atrial maze procedure	0	4	4 (12.1)
Biatrial maze procedure	0	1	1 (3)
Isolated cavotricuspid isthmus ablation	0	4	4 (12.1)
Technical modification of cone repair			
TV annulus support procedure	3	20	23 (69.7)
Pericardial strip	3	18	21 (63.6)
Prosthetic ring	0	2	2 (6.1)
Papillary muscle reposition	0	1	1 (3)
Cardiopulmonary bypass time, minutes	215.4±59.8	202.9±57.1	205.9±57.1
Cross clamp time, minutes	152.0±28.8	148.8±48.8	149.7±44.4
TV reoperation	1	2	3 (9.1)

Data are presented as n (%), mean ± SD or number. ASD, atrial septal defect; MPA, main pulmonary artery; PV, pulmonary valve; SD, standard deviation; TV, tricuspid valve.

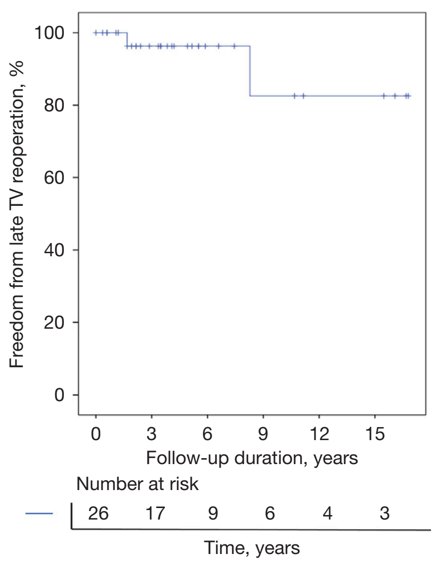

Figure 2 Overall freedom from tricuspid valve reoperation. TV, tricuspid valve.

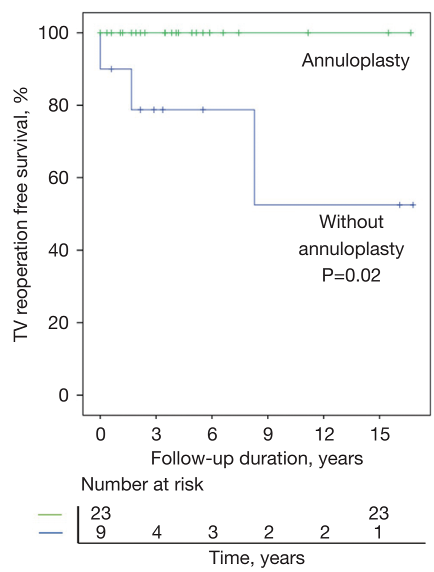

Figure 3 Freedom from TV reoperation (with or without annuloplasty). TV, tricuspid valve.

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Table 3 The preoperative, predischarged, and last echocardiographic results

Variables	Pre (n=33)	Predischarged (n=31)	Last echo (>6 months, n=31)
Interval after operation (years)			5.9±4.5
TR grade	3.9±0.3	1.5±0.8	1.9±0.7
No	0	5 (16.1)	1 (3.2)
Trivial	0	7 (22.6)	4 (12.9)
Trivial to mild	0	1 (3.2)	1 (3.2)
Mild	0	15 (48.4)	16 (51.6)
Mild to moderate	0	3 (9.7)	5 (16.1)
Moderate	1 (3.0)	0	4 (12.9)
Severe	32 (97.0)	0	0
TS (≥ moderate)	0	0	1 (3.2)
LV dysfunction	2 (6.0)	0	
RV dysfunction	16 (48.5)	18 (58.0)	

Data are presented as n (%) or mean ± SD. LV, left ventricle; RV, right ventricle; SD, standard deviation; TR, tricuspid regurgitation; TS, tricuspid stenosis.

TV replacement, 28 patients (90.3%) demonstrated mild or lesser degree of TR. None of the patients exhibited moderate degree or more degree of TR. A total of 31 patients underwent the last follow-up echocardiography more than 6 months after cone repair (mean interval: 5.9±4.5 years). Among them, 22 patients (71.0%) exhibited mild or lesser degree of TR, and four (12.9%) patients had moderate TR.

Preoperative and postoperative cardiac MRI data

Of the 33 patients included in this study, 25 underwent cardiac MRI (20 preoperatively and 15 postoperatively). Among them, 11 patients underwent both preoperative and postoperative cardiac MRI, with a median follow-up duration of 5.2 years (interquartile range: 2.7–7.9 years). The preoperative and postoperative MRI data for these 11 patients demonstrated a significant reduction in RV volume after cone repair [RVEDVi (mL/m^2): preoperative/postoperative =207.4±40.2/105.5±41.3, P=0.001; RVESVi (mL/m^2): preoperative/postoperative =104.5±21.3/61.2±38.5, P=0.047]. Although LVEF did not show a significant difference after cone repair [LVEF (%): preoperative/postoperative =60.8±5.3/61.2±5.4, P=0.10], LVSV significantly increased [LVSV (mL): preoperative/postoperative =64.0±1.8/71.4±12.7, P=0.041]. The preoperative and postoperative cardiac MRI data are presented in *Table 4*.

Discussion

Excellent clinical results of cone repair have been reported in several studies (3,5,9,14). The major findings of our study indicate that following cone repair, (I) the long-term mortality and morbidity were also excellent, consistent with the findings of the other studies; (II) the degree of TR was reduced to no more than mild degree in 87.9% of patients; (III) utilization of an additional annuloplasty band lowered the reoperation rate; (IV) RV volume was markedly reduced based on MRI data; (V) LV stroke volume was increased after cone repair (preoperative MRI: 64.0±1.8/71.4±12.7 mL, P=0.041).

Annular reinforcement after cone repair is considered an important procedure as the inferior annulus is particularly susceptible to annular dilatation. This is also the location of the suture line for plication of the atrialized RV. Moreover, tension occurs in this area due to RV contraction and relaxation (10,15). This study identifies the annular reinforcement procedure as having a protective effect on the reoperation rate following cone repair.

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Table 4 MRI data (n=11)

Parameters	Preoperative MRI data (n=11)	Postoperative MRI data (n=11)	P value
RVEDVi (mL/m^2)	207.4±40.2	105.5±41.3	0.001
RVESVi (mL/m^2)	104.5±21.3	61.2±38.5	0.047
RVEF (%)	46.4±9.6	45.2±10.5	0.57
RVSV (mL)	144.4±47.6	75.0±16.7	0.02
RVCO (L/min)	9.7±3.0	4.9±2.0	0.02
RVSI (mL/m^2)	90.2±30.5	46.4±11.3	0.008
RVCI (L/min/m^2)	6.1±2.0	3.0±1.2	0.01
LVEDVi (mL/m^2)	80.6±34.6	69.6±11.0	0.61
LVESVi (mL/m^2)	32.8±16.1	27.4±7.2	0.53
LVEF (%)	60.8±5.3	61.2±5.4	0.10
LVSV (mL)	64.0±1.8	71.4±12.7	0.04
LVCO (L/min)	4.4±1.0	4.5±1.2	0.82
LVSI (mL/m^2)	39.7±6.3	42.3±5.7	0.31
LVCI (L/min/m^2)	2.7±0.6	2.7±0.5	0.41

Data are presented as mean ± standard deviation. LVEDVi, left ventricle end-diastolic volume index; LVESVi, left ventricle end-systolic volume index; LVEF, left ventricle ejection fraction; LVSV, left ventricle stroke volume; LVCO, left ventricle cardiac output; LVSI, left ventricle systolic index; LVCI, left ventricle cardiac index; MRI, magnetic resonance imaging; RVEDVi, right ventricle end-diastolic volume index; RVESVi, right ventricle end-systolic volume index; RVEF, right ventricle ejection fraction; RVSV, right ventricle stroke volume; RVCO, right ventricle cardiac output; RVSI, right ventricle systolic index; RVCI, right ventricle cardiac index.

Our MRI data demonstrated a significant reduction in RV volume following cone repair. We measured the anatomic RV volume (atrialized RV + functional RV) since measuring functional RV volume alone does not fully capture the volume changes after cone repair. Qureshi *et al.* recommend that cardiac MRI RV volumes include the complete anatomic RV, which consists of both atrialized and functional RV preoperatively (16).

Before cone repair, an increase in RV preload improves myocardial contraction based on the Frank Starling mechanism but simultaneously reduces effective cardiac output due to reduced forward flow attributed to significant TR (17,18). After successful cone repair, the reduction in TR and the inclusion of atrialized RV into the functional RV may expose the RV to sudden decreases in preload and relatively increased afterload. However, effective forward flow from the RV is increased.

Some studies have demonstrated that the forward flow from the RV is increased, resulting in improved LV filling and higher LVSV, as demonstrated by conventional echocardiographic parameters and MRI (19,20). The reduction in RV volume after cone repair allows the septum to return to a more physiological position. This improved septal configuration and synchrony enhances left ventricular end-diastolic filling, leading to increased preload and ultimately a rise in LVSV (8,18,19). These optimized preload conditions post-repair could enhance stroke volume through the Frank-Starling mechanism. Before repair, excessive TR leads to ineffective forward flow, reducing left ventricular preload. Following cone repair, the improved RV function contributes to more efficient pulmonary circulation and left atrial filling, which enhances left ventricular stroke volume (20). Therefore, while other left ventricular parameters remain stable, the increase in LVSV reflects a functional improvement in left ventricular performance following cone repair, supporting the broader benefits of the procedure beyond RV remodeling. These findings can elucidate our MRI results after cone repair, indicating an increased LVSV.

The timing of operation for Ebstein anomaly remains controversial, particularly in asymptomatic patient with normal exercise capacity. If the results of cone repair are

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proven durable and if both ventricular functions improve, a paradigm shift toward earlier operation may occur. In particular, our study demonstrated that LVSV increased after cone repair. This is evidence of biventricular reverse remodeling after cone repair.

Limitations

This study has some limitations. First, this was a retrospective, nonrandomized, single-center study. Second, preoperative and postoperative MRI scans were not performed on all patients. Third, the study population was small because Ebstein anomaly is a rare disease.

Conclusions

Cone repair for Ebstein anomaly demonstrates low mortality and morbidity rates. Despite the excellent surgical results in most patients with improved clinical outcomes, ventricular function, and biventricular reverse remodeling need to be assessed on a long-term basis, and the assessment tools also are not standardized. Our data demonstrates the potential of LV reverse remodeling based on MRI results. The long-term ventricular function and clinical symptoms were preserved. The addition of an annuloplasty band was associated with a reduced incidence of TV reoperation. In addition, cone repair might affect LV function due to ventricular interdependency. Prolonged follow-up is essential to determine the late durability of cone repair and both ventricular functional changes.

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Footnote

Reporting Checklist: The authors have completed the STROBE reporting checklist. Available at https://jtd.amegroups.com/article/view/10.21037/jtd-2024-2185/rc

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Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. The study was conducted in accordance with the Declaration of Helsinki and its subsequent amendments. The study was approved by the Institutional Review Board of Bucheon Sejong General Hospital (No. 2023-12-007-004) and individual consent for this retrospective analysis was waived.

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