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Surgical repair of "Swiss Cheese" ventricular septal defects with two-patch and right ventricular apex-exclusion technique: mid-term follow-up results

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Abstract

Background "Swiss Cheese" ventricular septal defects represent a serious congenital heart disease with suboptimal clinical outcomes and a lack of consensus regarding its management. This study presents mid-term follow-up results of surgical repairs for "Swiss Cheese" ventricular septal defects, utilizing the two-patch and right ventricle apex-exclusion technique.

Methods A retrospective review was conducted on 13 patients who underwent surgical repair utilizing the two-patch and right ventricle apex-exclusion technique at our institution between May 2014 and October 2021. The procedure involved the closure of defects in the outflow tract ventricular septal and the apex trabecular ventricular septal regions using two patches, with concurrent exclusion of the right ventricular apex from the right ventricular inflow tract.

Results Median follow-up was 4.9 ± 2.1 years (range: 2–9 years). All cases were successful without mortality or major complications. Two years post-surgery, cardiac magnetic resonance revealed median values for left ventricular ejection fraction, right ventricular ejection fraction, left ventricular end-diastolic volume and right ventricular end-diastolic volume of 63.9% ± 1.8% (range: 61–67%), 49.2% ± 2.6% (range: 46–55%), 39.15 ± 2.11 ml (range: 36.2–42.7 ml), 44.55 ± 3.33 ml (range: 38.7–48.6 ml), respectively. No thrombosis occurred. The latest echocardiography results confirmed normal cardiac function in all cases.

Conclusions The surgical repair of "Swiss Cheese" ventricular septal defects utilizing the two-patch and right ventricle apex-exclusion technique is a viable approach with favorable mid-term outcomes. More cases and long-term follow-up results are needed to validate the feasibility and safety of this technique.

Keywords "Swiss Cheese", Ventricular septal defect, Congenital heart disease, Surgical repair

Background

"Swiss Cheese" ventricular septal defects (VSDs) constitute a serious and complex congenital heart disease defined by the presence of four or more muscular VSDs, frequently complicated by severe cardiac dysfunction [1]. Various treatment strategies have been delineated, such as pulmonary artery banding (PAB) [2], transcatheter or intraoperative device closure [3, 4], and surgical repair

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utilizing a variety of approaches, including a right atrial approach, right, left, or bilateral ventriculotomies [5]. There have also been numerous attempts at repair methods, including closure with a single large patch [6] or a composite patch of pericardium and Dacron graft [7], the sandwich technique [8], and the felt sandwich technique [9]. However, the clinical outcomes associated with repairing "Swiss cheese" VSDs are suboptimal, characterized by elevated hospital mortality rates and a heightened propensity for adverse events such as ventricular dysfunction, persistent shunting, and complete heart block [10–12]. Since May 2014, we have implemented a novel surgical technique involving the exclusion of the right ventricular apex (RVA) through the utilization of two fresh autologous pericardium patches [13]. The current study aims to assess the mid-term safety and efficacy of this innovative approach.

Methods

Patients

This study was approved by the institutional review board of the ethics committee in affiliated women and children's hospital of Qingdao university (QFELL-YJ-2023-205) on 31 December 2023. Informed consent was obtained in all cases. Between May 2014 and October 2021, our institution performed surgical repair on 13 patients presenting with "Swiss cheese" VSDs utilizing the RVA exclusion technique. Inclusion criteria necessitated a confirmed diagnosis of "Swiss cheese" VSDs through transthoracic echocardiogram (TTE). Exclusion criteria comprised (1) patients younger than 2 months old, (2) the presence of other associated congenital heart defects requiring palliative treatment, (3) severe pulmonary hypertension with right-to-left shunt of VSDs, and (4) patients whose guardians declined acceptance of this novel technique. Prior to surgery, comprehensive assessments, including physical examination, electrocardiography (ECG), chest radiography, and TTE, were conducted for all cases. The general characteristics of the patients are listed in Table 1.

ECG, electrocardiography; iRBBB, incomplete right bundle brunch block; iLBBB, incomplete left bundle brunch block; cRBBB, complete right bundle brunch block; TTE, transthoracic echocardiogram; LVEF, left ventricular ejection fraction; TR, Tricuspid regurgitation; ASD, Atrial septal defect; DORV, Double-outlet right ventricle; VSD, ventricular septal defect;PDA, Patent ductus arteriosis.

Procedure

Following the initiation of cardiopulmonary bypass (CPB) and the establishment of myocardial protection utilizing the HTK cardioplegic solution, a longitudinal right atriotomy was conducted to expose the VSDs through the tricuspid valve. Subsequently, a meticulous exploration of the "Swiss Cheese" VSDs was performed, involving the precise excision of surplus muscle bundles from the ventricular septum. If deemed necessary, a 10F Foley catheter can be strategically placed, traversing through the ASD or the incision in the atrial septum, mitral valve, the VSD requiring enhanced exposure, and tricuspid valve. By gently pulling the two ends of the Foley catheter, the entire rim of the VSD can be distinctly visualized.

The "Swiss Cheese" VSDs were then divided into two segments by the moderator band: the outflow tract VSDs and the apex trabecular VSDs. The dimensions of these two regions were measured. Subsequently, a fresh

Table 1 Patient characteristics

Patient	Age (mo)	Weight (kg)	Cardiothoracic ratio	ECG	TTE		
					LVEF (%)	TR	Associated cardiac lesions
1	4	4	0.61	–	60	Mild	ASD
2	6	4.6	0.66	–	62	Trivial	DORV
3	3	3.8	0.65	–	65	Trivial	Tetralogy of Fallot
4	2	3.7	0.60	–	57	Moderate	Coarctation of the aorta
5	3	4.1	0.66	iRBBB	62	Trivial	Perimembranous VSD, PDA
6	4	3.8	0.60	iLBBB	60	Mild	ASD
7	13	7	0.72	cRBBB	66	Trivial	Perimembranous VSD
8	5	5.6	0.60	-	62	Mild	ASD
9	6	4.9	0.62	iRBBB	65	Trivial	–
10	3	3.8	0.65	–	58	Mild	–
11	4	5	0.60	–	64	Mild	PDA
12	6	8.5	0.62	–	60	Moderate	–
13	3	6	0.65	–	65	Trivial	Perimembranous VSD, ASD

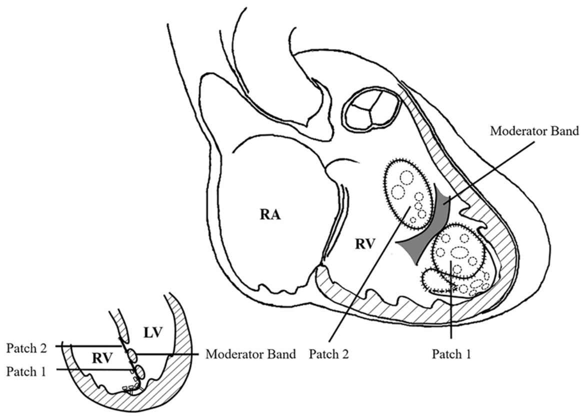

Fig. 1 Two-patch and right ventricular apex-exclusion technique

autologous pericardial patch, matching in size, was harvested for the repair of the apex trabecular VSDs, while a glutaraldehyde-treated autologous pericardial patch or fresh autologous pericardial patch was procured for the outflow tract VSDs. The patches were then sutured along the edges of each VSD segment using 5–0 or 6–0 polypropylene sutures, respectively. Specifically, the apex trabecular patch was sutured between the moderator band and the anterior wall of the right ventricle (Fig. 1). This surgical maneuver aimed to effectively eliminate any existing left-to-right ventricular shunts by excluding the RVA. During the suturing process, three or four interrupted pledgeted sutures were placed between the patch and the trabecular muscles to prevent septal bulging, thereby mitigating the risk of excessive reduction of the right ventricular cavity. Simultaneous correction of combined cardiac lesions was undertaken during the procedure. In cases involving perimembranous VSD, an additional bovine pericardial patch or glutaraldehyde-treated autologous pericardial patch was employed for the repair (Video 1). Transesophageal echocardiography was conducted to identify residual shunts and assess tricuspid regurgitation during the rewarming period.

Postoperative pulmonary hypertension was effectively managed using either treprostinil or bosentan. Follow-up evaluations included ECG, chest radiographs, and TTE at 1 month, 3 months, 6 months, and annually thereafter. Additionally, Cardiac magnetic resonance (CMR) was conducted in all cases two years postoperatively to ensure a thorough assessment of cardiac function and structure.

Results

The procedure was successfully completed in all 13 patients. The median time for CPB and aortic clamping was 93.8 ± 25.8 min (range, 68–167 min) and 66.2 ± 19.3 min (range, 43–122 min), respectively. The median duration of mechanical ventilation was 111.7 ± 76.8 h (range, 32–328 h), and the median postoperative intensive care unit (ICU) stay was 7.2 ± 7.2 days (range, 4–31 days). The median maximum vasoactive-inotropic score (VIS) within the first 24 h post-operation was 9.23 ± 3.64 (range, 6.0–17.5). The detailed posteroperative characteristics are listed in Table 2. Patients were followed up for a median period of 4.9 ± 2.1 years (range, 2–9 years). No deaths or major complications, such as obvious residual shunt (> 2 mm measured by TTE), more than mild tricuspid regurgitation, septal bulging, cardiac dysfunction, or fatal arrhythmias, occurred during the hospitalization or follow-up phases. Two cases exhibited less than minor residual shunts as identified by TTE. These residual shunts were observed to be completely resolved at 10 months and 22 months postoperatively, respectively. The median LVEF, right ventricular ejection fraction (RVEF), left ventricular end-diastolic volume (LVEDV), right ventricular end-diastolic volume (RVEDV), and RVEDV/LVEDV measured by CMR (Fig. 2) two years postoperatively were 63.9% ± 1.8% (range, 61–67%), 49.2% ± 2.6% (range, 46–55%), 39.15 ± 2.11 ml (range, 36.2–42.7 ml), 44.55 ± 3.33 ml (range, 38.7–48.6 ml) and 1.11 ± 0.09 (range, 0.93–1.24), respectively (Table 3). No instances of thrombosis were observed. The most recent ECG showed sinus arrhythmia in all patients, with four exhibiting iRBBB, one with cRBBB, and one with iLBBB. The detailed follow-up data of CMR outcomes are listed in Table 3.

LVEF left ventricular ejection fraction, *RVEF* right ventricular ejection fraction, *LVEDL* left ventricular end-diastolic volume, *RVEDV* right left ventricular end-diastolic volume

Table 2 Posteroperative characteristics

Patient	CPB time (min)	Clamping time (min)	Ventilation time (h)	VIS 24 max	ICU stay (days)
1	86	60	100	8.0	5
2	121	86	159	12.0	7
3	167	122	328	17.5	31
4	105	56	132	14.0	6
5	76	65	99	11.0	5
6	80	58	115	7.0	5
7	83	58	100	6.0	5
8	85	63	150	11.0	7
9	68	43	32	6.0	5
10	91	66	98	8.5	5
11	85	61	44	6.0	4
12	78	55	49	6.0	5
13	94	67	46	7.0	4

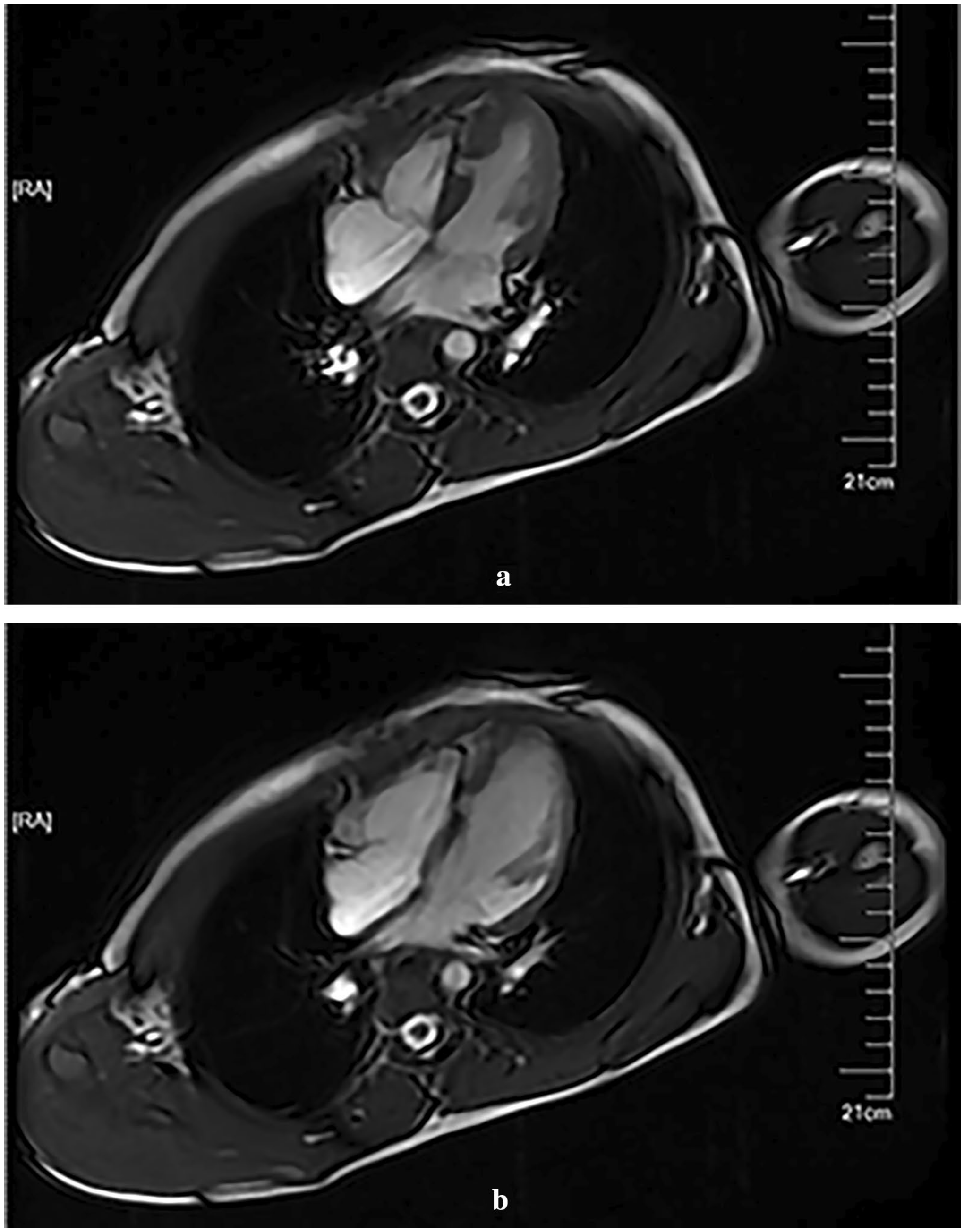

Fig. 2 Representative CMR images at 2 years postoperative follow-up. **a** Systolic Phase; **b** Diastolic Phase

Discussion

"Swiss Cheese" VSDs represent the most critical subtype among muscular VSDs. This subtype exhibits elevated mortality rates, an increased risk of residual shunt and reoperation, as well as heightened susceptibility to ventricular dysfunction and heart block when contrasted with isolated perimembranous defects [14]. Surgical intervention for "Swiss Cheese" VSDs presents a formidable challenge, especially when confronted in neonates and premature infants with low body weight. Currently, there is no consensus regarding the optimal management of "Swiss Cheese" VSDs. Conventional surgical repair for "Swiss Cheese" VSDs yields unsatisfactory outcomes, primarily due to significant intraoperative challenges arising from the obscured boundaries of defects by the moderator band and multiple trabeculations. Closing apparent defects may reveal additional hidden defects, necessitating further intervention [14, 15]. Therefore, in the past

Table 3 Two-year follow-up data of CMR outcomes

Patient	LVEF (%)	RVEF (%)	LVEDV (ml)	RVEDV (ml)	RVEDV/ LVEDV
1	65	50	41.2	42.2	1.02
2	62	47	40.4	44.5	1.1
3	64	46	42.7	48.6	1.14
4	66	49	39.1	45.9	1.17
5	64	55	38.6	48.2	1.25
6	63	51	40.8	47.8	1.17
7	61	50	41.6	38.7	0.93
8	67	48	36.7	40.7	1.11
9	66	47	39.6	42.4	1.07
10	62	51	36.2	44.9	1.24
11	64	46	38.1	40.1	1.05
12	62	52	37.4	41.6	1.11
13	65	48	36.5	40.5	1.11

several decades, various strategies have been described, including device closure, re-endocardialization of the interventricular septum, PAB, as well as diverse surgical approaches and techniques [5, 11, 16].

One classic technique involves repairing all VSDs using a single large autologous pericardial patch, polytetrafluoroethylene patch, or a composite patch composed of Dacron lined with preserved heterologous pericardium [6–8, 17]. This approach has demonstrated satisfactory short- and mid-term outcomes, with specific advantages highlighted in the repair of apex trabecular VSDs [14]. However, it is important to note that an oversized patch may impede the movement of the ventricular septum and limit the development of the right ventricle, potentially leading to late diastolic dysfunction and subsequent complications such as right heart failure, cardiac cirrhosis, and atrial tachyarrhythmias [18, 19]. Brizard et al. [20] introduced a novel approach incorporating intraoperative echocardiography and the utilization of double patches to sandwich the septum. Nevertheless, replicating this technique poses significant challenges, and it has not consistently produced satisfactory outcomes. In their cohort, failure to localize all defects resulted in PAB in two patients, while two other patients required permanent pacemaker insertion. Sakurai et al. [11] reported the successful application of a novel technique for repairing 'Swiss Cheese' ventricular septal defects in a single case, using a combination sandwich patch composed of three polytetrafluoroethylene patches. However, to date, no further studies have been published on this technique.

Since 2014, we have adopted a modified procedure for "Swiss Cheese" VSDs to mitigate the potential drawbacks associated with the "single oversized patch" technique. This approach includes the repair of outflow tract VSDs with one glutaraldehyde-treated or fresh autologous pericardial patch, as well as the repair of apex trabecular VSDs and exclusion of the RVA with another fresh autologous pericardial patch. Mid-term follow-up results were favorable, with no significant complications observed during a median follow-up period of 4.9 years (range: 2–9 years). TTE and CMR revealed satisfactory left and right ventricular systolic and diastolic function, with no discernible reduction in right ventricular volume and no evidence of thrombosis. These findings suggest that the employed technique is safe and may have a lesser impact on the movement of the ventricular septum and fewer constraints on the development of the right ventricle compared to the "single oversized patch" technique.

During the procedure, a Foley balloon catheter can be employed to enhance exposure of the VSDs, as detailed in the preceding procedural section. This approach offers potential advantages in delineating the extent of VSDs and mitigating residual shunting post-operation. The exclusion of the RVA is pivotal to preventing residual shunt. Geva et al. [21] demonstrated that the right ventricular inlet (or right ventricular sinus) constitutes $81 \pm 6\%$ of the combined right ventricular end-diastolic volume and $87 \pm 4\%$ of the combined stroke volume. Therefore, exclusion of the RVA may not significantly influence the right ventricular end-diastolic volume.

For patching, especially in cases involving apex trabecular VSDs, the use of fresh autologous pericardium is recommended due to its reported favorable biological characteristics, including freedom from retraction, stiffness, fibrosis, and calcification [22]. Consequently, it is less likely to adversely impact right ventricular function in the mid- or long-term. During the patching phase, it is advisable to position the suture of the anterior wall of the right ventricle as close as possible to the RVA. Additionally, excising small trabeculae is recommended, and interrupted pledgeted sutures should be strategically placed between the patch and trabecular muscles for intermediate fixation to prevent septal bulging. These maneuvers minimize the volume of the right ventricular cavity separated into the left cardiac system, thereby effectively preventing excessive reduction of the right ventricular cavity and postoperative ventricular dysfunction.

The two-patch and RVA exclusion technique is primarily intended for patients older than 3 months with "Swiss cheese" VSDs, particularly those with muscular VSDs located near the apex. In infants under 2–3 months requiring ventilator support, especially those with complex defects, delicate heart muscles and limited exposure during surgery may negatively impact outcomes of this technique. Therefore, preemptive PAB is recommended to safeguard the pulmonary vascular bed and alleviate symptoms of congestive heart failure [5]. Subsequent definitive repair is typically advised within 3–6 months.

The primary concern centers on the persistent impairment of right heart diastolic function due to the exclusion of the RVA. Additionally, heart block stands out as a potential late complication resulting from the extensive intracardiac sutures. Consequently, patients necessitate continued long-term monitoring, involving regular ECG, TTE, and especially CMR to evaluate right heart functionality.

Conclusions

The two-patch and RVA exclusion technique is a viable approach for the biventricular surgical repair of "Swiss Cheese" VSDs. This method may mitigate the risk of right ventricular diastolic dysfunction attributed to the single oversized patch covering the entire inner right ventricular wall. Thrombosis monitoring and evaluation of right heart function are pivotal aspects of postoperative follow-up, with regular CMR recommended. More extensive case studies and long-term follow-up results are essential to validate the feasibility and safety of this technique.

Abbreviations

ASD	Atrial septal defect
CMR	Cardiac magnetic resonance
CPB	Cardiopulmonary bypass
cRBBB	Complete right bundle brunch block
ECG	Electrocardiography
ICU	Intensive care unit
iLBBB	Incomplete left bundle brunch block
iRBBB	Incomplete right bundle brunch block
LVEDV	Left ventricular end-diastolic volume
LVEF	Left ventricular ejection fraction
PAB	Pulmonary artery banding
RVA	Right ventricular apex
RVEDV	Right ventricular end-diastolic volume
RVEF	Right ventricular ejection fraction
SD	Standard deviation
TTE	Transthoracic echocardiogram
VIS	Vasoactive-inotropic score
VSD	Ventricular septal defect

Supplementary Information

The online version contains supplementary material available at https://doi.org/10.1186/s13019-024-03085-z.

Additional file 1.

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Author contributions

Rui Chen contributed significantly to the article based on the invention of the new approach. Quansheng Xing contributed significantly to the critical revision of the work. Qiteng Xu and Zhen Bing contributed to data collection and manuscript preparation. Bei Lv contributed to preparation of figures. All authors read and approved the final manuscript.

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Availability of data and materials

The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate

All methods involved in this study were carried out in accordance with relevant guidelines and regulations in the declaration of Helsinki. This study was approved by the institutional review board of the ethics committee in Qingdao women and children's hospital (QFELL-YJ-2023-205) on 31 December 2023.

Consent for publication

Informed consents for publication have been obtained from the legal guardians of all subjects.

Competing interests

The authors declare that they have no competing interests.

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