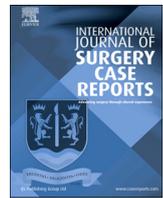




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# Iatrogenic ventricular septal defect: A rare complication of surgical reconstruction of mitral paravalvular dehiscence



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## ABSTRACT

**INTRODUCTION:** Iatrogenic ventricular septal defect is a rare complication after the surgical replacement of cardiac valves. Small defects may have no hemodynamic significance or remain unremarked at the end of the surgical procedure. Understanding of the valvular anatomy alone is not always enough to avoid such complications, especially in the hands of young surgeons.

**PRESENTATION OF CASE:** We present a case of iatrogenic ventricular septal defect that developed early after the surgical closure of a hemodynamically significant mitral paravalvular leak. Although the patient's critical state did not allow surgical intervention and he died, we think the lessons drawn from this case could be helpful to avoid such horrible complications in the future.

**DISCUSSION:** This case documents a rare disastrous complication after imperfect surgical closure of a mitral paravalvular leak. Despite the unfortunate end, in reporting this case we try to direct the light to the possible mechanisms that led to the development of this injury focusing on the embryological and anatomical background.

**CONCLUSION:** Understanding the anatomical and embryological structure of the cardiac fibrotic skeleton should keep cardiac surgeons more vigilant in detecting iatrogenic ventricle septal defects before the development of a devastating hemodynamic state.

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## 1. Introduction

Iatrogenic ventricular septal defect is a rare complication after the surgical replacement of cardiac valves. Small defects may have no hemodynamic significance or remain unremarked at the end of the surgical procedure. Late detection of these defects may lead to serious hemodynamic deterioration, which may render the clinical course irreversible. We present a case of iatrogenic ventricular septal defect that, developed early after the surgical closure of a hemodynamically significant mitral paravalvular leak. Although the patient's critical state did not allow surgical intervention and he died, we think the lessons drawn from this case could be helpful to avoid such horrible complications in the future. In this paper we attempt to study the physioanatomic and embryologic bases that, might have led to the development of this complication.

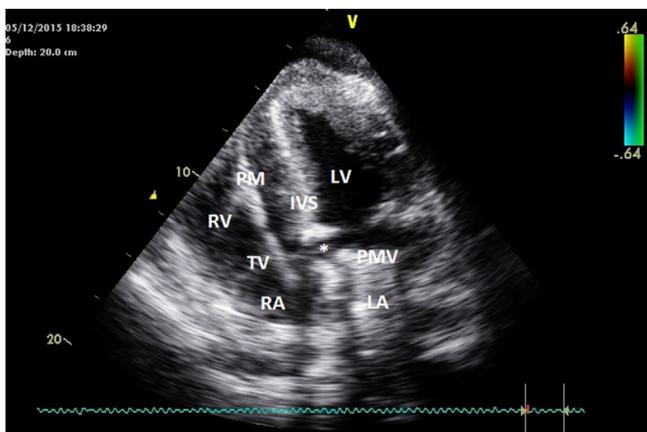
## 2. Presentation of the case

A 61-year-old man was admitted to our cardiac surgical department in December 2015 due to a significant mitral prosthetic

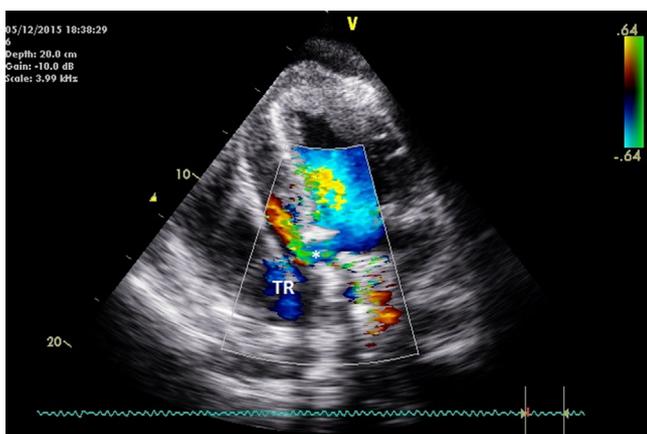
paravalvular leak. The symptoms of congestive heart failure dominated the clinical course. In 2008, the patient was planned to undergo aortic valve replacement due to a calcified stenotic aortic valve. During that procedure, the mitral valve was accidentally injured and the left fibrotic trigone was ruptured, so the mitral valve was replaced too. The patient's past medical history was significant for chronic obstructive pulmonary disease, chronic atrial fibrillation, and previous pacemaker implantation due to third-degree atrioventricular block. The patient was in good health condition until the summer of 2015, when he was admitted to the hospital due to symptoms of dyspnea, fatigue, and weight loss. Transthoracic and transesophageal echocardiography were performed. A significant mitral paravalvular leak was revealed. The hemodynamic function of the aortic mechanic valve was normal. Echocardiography showed diffused hypokinesis and depressed left ventricle function (ejection fraction: 45%) without segmental wall motion disturbance. Based on the tricuspid annular plane systolic excursion (TAPSE) right ventricular dysfunction was also observed (TAPSE: 11 mm). No atrial or ventricle septal flow abnormalities were detected. Coronarography showed no significant coronary stenosis. Laboratory studies performed one week prior to and upon admission showed normal hepatic function, normal renal function, and normal inflammatory biomarkers. An urgent operation was performed. Median sternotomy and a transatrial septal approach were selected. The exposure of the entire mitral valvular ring and

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**Fig. 1.** Two-dimensional Echocardiography image demonstrates iatrogenic ventricle septal defect.



**Fig. 2.** Color wave Echocardiographic image shows left-right shunt through iatrogenic ventricular septal defect.

annulus was only partially feasible. An approximately  $1 \times 1$  cm paravalvular dehiscence was observed in the region near the tricuspid septal annulus. The mitral annulus could not be explored perfectly, due to the pulling effect of the aortic prosthetic ring. In an attempt to reconstruct the dehiscence between the mitral prosthetic ring and the annulus, the surgeon covered the defect by pulling down the lower part of the atrial septum towards the prosthetic ring. Four 2/0 Ethibond-pledgeted sutures were used. Postoperative transesophageal echocardiography showed no paravalvular leak or other abnormal flow patterns. The patient was weaned from the cardiopulmonary bypass. Due to sustaining right ventricle dysfunction epinephrine and norepinephrine was initiated, but gradually stopped by the second postoperative day. Afterward, the patient was weaned from the respirator and extubated. In the fourth postoperative day, he became oliguric, then hypotensive. Central venous pressure (CVP) increased from 12 to 18 cmH<sub>2</sub>O. Circulatory support with high doses of epinephrine, norepinephrine and dopamine was restarted with only minimal beneficial effects. Transthoracic echocardiography showed a 2-cm wide pericardial effusion around the right ventricle, which was drained out through a mini subxiphoid incision. Thereafter, the blood pressure rose from 60 to 90 mmHg and CVP declined to 14 cmH<sub>2</sub>O. Three hours later, as the patient's clinical state showed no further improvement, another echocardiographic examination was performed, which showed signs of severe right ventricle failure (TAPSE: 5 mm) and a 10-mm ventricle septal defect in the region of the membranous septum (Figs. 1 and 2). At this stage, the PCT



**Fig. 3.** Postmortem image shows iatrogenic ventricular septal defect. Signs of endocarditis or myocardial infarction were absent.

was 56 mM/l, and systemic vascular resistance was critically low, mimicking the course of severe sepsis. Further surgical reoperation was considered, but not performed due to the severely critical state of the patient.

Postmortem examination (Fig. 3), in accordance with preoperative echocardiographic images, revealed a ventricle septal defect that was 1 cm in diameter. The defect was identified between the area below the aortic annulus and under the tricuspid septal leaflet. No necrotic signs were seen at the borders of the defect. Postinfarct VSD was precluded as no postmortem signs of early myocardial infarct or coronary occlusion were observed. No signs of active endocarditis were identified.

### 3. Discussion

This case documents a rare disastrous complication after imperfect surgical closure of a mitral paravalvular leak. During the primary operation, the intertrigonal region was accidentally injured and the subsequent mitral valve regurgitation was addressed by the implantation of a mechanical prosthesis. This area was severely scarred, as documented by postmortem examination.

Paravalvular dehiscence might be the result of the earlier trigonal injury, which might have weakened the structure of the fibrotic skeleton in this area. Although iatrogenic ventricular septal defects (VSDs) have been reported after aortic and mitral valve replacement [1], to our knowledge this complication has never been yet described in the English literature. Most of the reported cases dealt with successful surgical or percutaneous closure based on the enormous experience gained in treating congenital VSD [2]. Despite the unfortunate end, in reporting this case we try to direct the light to the possible mechanisms that led to the development of this injury focusing on the embryological and anatomical background. This may make us more vigilant in the future in avoiding complications that originate from structural rupture of the fibrotic skeleton.

#### 3.1. Embryological and anatomophysiological aspects

During the cardiac cycle the movement of the fibrotic skeleton is stationary against the dynamic movement of the atrial and ventricle septum. The implantation of a stented bioprosthesis or mechanical valves in the cardiac skeleton may increase the rigidity of the fibrous zones at the borders of the muscular septum, which may make the septum more vulnerable to stress forces during its dynamic motion. In our case, the Teflon-pledgeted sutures were introduced first to the lower part of the atrial septum, near to the mitral annulus, then through the rigid ring of the mitral mechanical prosthesis. In the setting of high-dose epinephrine-induced tachycardia, the repetitive stress forces on the membranous septum

may profoundly increase, as it is the weakest point of the septum-fibrotic skeleton attachment.

During fetal development, the membranous ventricular septum develops independently from the muscular septum; therefore the membranous portion is not actually a part of the muscular ventricle septum, but rather is part of the aorticopulmonary septum, which fuses during its development with the muscular septum [3]. At the base of the heart, the membranous septum is in relation to the mitral, tricuspid, and aortic annuli through the right fibrotic trigonum, to which the primary atrial septum is attached superiorly [4].

The right fibrotic trigonum is the center of complex anatomical structures, with different embryological origins that, attach and function in relation to each other. These structures are: the primary atrial septum, membranous ventricular septum, aortic annulus (base of non- and right coronary sinus), tricuspid septal annulus, and anteromedial mitral annulus. After aortic and mitral mechanical valve replacement, the dynamics of the right fibrotic trigonum and the related structures may change to adapt to the new stress forces exerted on the structures with dynamic movements around the cardiac cycle. The anterior mitral annulus is not attached to the ventricular musculature, but rather is suspended between the right and left trigone. Therefore, the shape of this part of the annulus dynamically changes through the cardiac cycle. This feature would be lost after implantation of a prosthetic valve in the mitral orifice and the annulus would become immotile and round in shape.

In this setting, the portion of the trigonum underlying the tricuspid septal annulus may be the most vulnerable against tearing forces. The effect of these tearing forces may be more significant in catecholamine-induced tachycardia. This surgical solution, selected to reconstruct the mitral paravalvular leak developed near the right fibrotic trigone, could have been successful in the setting of a native aortic valve, as this may distribute the tearing force exerted through the other two sides of the trigonum, the aortic native annulus, and the tricuspid septal annulus.

### 3.2. Diagnostic considerations

This defect could have been a post-infarction rupture of the ventricular septum. This was precluded due to a negative coronarogram, and the absence of coronary occlusion or evidence of a postinfarct septal defect pathology pattern during postmortem examination [5,6].

Although VSD has been reported as a complication of perianular distribution of prosthetic valve endocarditis [7], in our case endocarditis was not diagnosed. No bacteria or fungi was cultured during the microbiological examination of blood cultures. No vegetations were observed during echocardiography and postmortem examination showed no signs of endocarditis all over the cardiac endocardium.

## 4. Conclusion

Understanding the anatomical and embryological structure of the cardiac fibrotic skeleton should keep cardiac surgeons more vigilant in detecting iatrogenic ventricle septal defects before the development of a devastating hemodynamic state. Other pathological states should be considered and excluded.

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## Conflicts of interest

Authors have no conflicts of interest.

## Sources of funding

Authors have no sponsor or any other external funding to declare.

## Ethical approval

The patient in this case report had, unfortunately, deceased. Due to extreme anonymity, no ethical approval was approved.

## Consent

The patient in this case report had, unfortunately, deceased. His only next of kin is his life's partner with whom he was living for twenty years. No other relatives were found alive. Written informed consent was obtained from her for publication of this case and accompanying images. A copy of the written consent is enclosed.

## Author contribution

Aref Rashed: He decided to summarize the clinical course in a case report for publication. He edited the anatomical and embryological background.

Karoly Gombocz: He prepared the figures and contributed in editing.

Janos Fulop: He edited the surgical part in the report. Nasri Alotti: He revised the report and contributed in editing the conclusion.

## Registration of research studies

One case report, not listed in the research register.

## Guarantor

Aref Rashed.

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