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Case Report

Novel Treatment for Atrial Septal Defect without an Inferior Septal Rim and Partial Anomalous Right Lower Pulmonary Venous Connection

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ABSTRACT: There is no consensus regarding the ideal surgical procedure for treatment of combined atrial septal defect without an inferior septal rim and partial anomalous right lower pulmonary venous connection. A 41 year-old woman had previously undergone ineffective surgical closure of an ostium secundum-type atrial septal defect. Intraoperatively, we ascertained the correct diagnosis of partial anomalous right lower pulmonary venous connection combined with a residual defect. Incision of the atrial septal wall up to the superior margin along the previous suture line yielded a flap adequate for the atrial septal wall, and this flap was slid again into the entire opening. The spatially complex intracardiac lesion was repaired by using only autologous tissue.

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KEYWORDS: atrial septal defect without an inferior septal rim, partial anomalous right lower pulmonary venous connection, autologous tissue

Introduction

Only a few surgical methods are available to treat the combination of atrial septal defect (ASD) and partial anomalous pulmonary venous connection (PAPVC); however, no consensus has been reached regarding the ideal treatment and baffle material. In cases of an ASD without an inferior rim and anomalous right lower pulmonary venous connection to the right atrium, adequate baffling of the pulmonary vein to the left atrium (LA) in right atrium (RA) can avoid pulmonary vein obstruction but might result in stenosis of the inferior vena cava (IVC). Here, we describe a novel treatment for these anomalies that uses only

autologous tissue. Incision of the atrial septal wall up to the superior margin yields a flap adequate for the atrial septal wall, and this flap is slid again into the entire opening. This procedure reduces the risks of stenosis and thrombosis in the left and right heart.

Case

A 41 year-old woman referred to our department for assessment of fatigue had undergone surgical closure of an ostium secundum-type ASD at another hospital at age 5 years. Regarding blood data, all but serum BNP levels (72.3 pg/mL) were within the normal range. Chest X-ray revealed only the mild enhancement of pulmonary vascular

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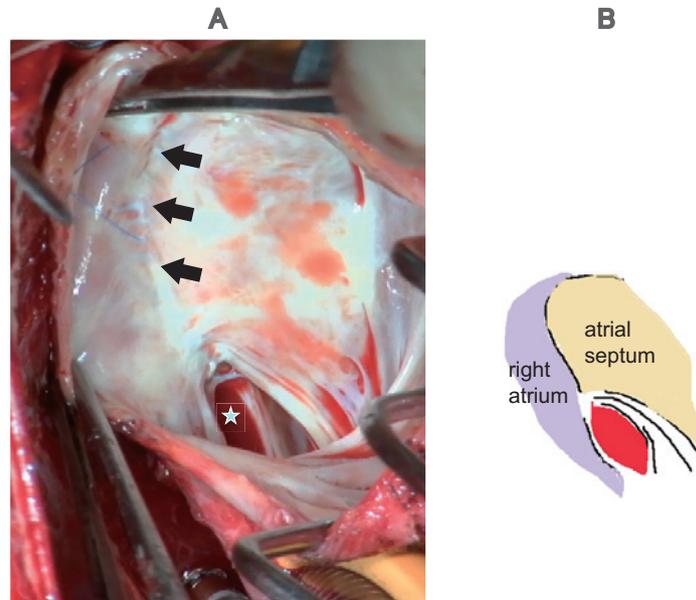


Fig. 1

- A) Intraoperative photo showing the inside of the right atrium, the fossa ovalis ASD, the previous suture covered with endothelial cells, and the opening of the right lower PV with a posterior limb.
 Arrows: the previous suture covered with endothelial cells; star: the orifice of the right lower PV
- B) Schema shows photo dissection (right atrium, atrial septum)

shadow. Transthoracic echocardiography revealed a small ASD, severe tricuspid valve insufficiency, and volume overload of the right heart. Right pulmonary angiography showed the IVC in the pulmonary vein (PV) phase. A catheter was easily inserted from the IVC to the LA and significantly increased oxygen saturation in the RA. The ratio of pulmonary blood flow to systemic blood flow (Q_p/Q_s) was 1.6. On the basis of these findings, we diagnosed unprogrammed significant residual shunt and scheduled definitive repair.

After re-sternotomy and adhesiotomy, elective cardiopulmonary bypass (CPB) was started with the ascending aorta and bicaval cannulation. Cardiac standstill was achieved after administration of a cardioplegic solution. Oblique right atriotomy revealed a residual secundum-type ASD without an inferior rim. The previous suture was located at the cranial position of the ASD and was covered by endothelial tissue (Fig. 1). To determine the anatomy of the LA and other pulmonary venous connections, an incision was made in the atrial septal wall along the previous suture line, up to the superior margin of the fossa ovalis (Fig. 2). The posterior limb of the right lower PV was located in the RA, and no other anomalous pulmo-

nary venous connections were found. Thus, we diagnosed the partial anomalous right lower pulmonary venous connection intraoperatively. Although we saw no evidence of the initial anatomy, we concluded that the orifice of the right lower PV was mistaken for that of the IVC, which led to inappropriate closure and the patient's present condition. After enlarging the orifice through the right lower PV, we confirmed that the flap of the atrial septal wall was sufficiently large to be slid into the entire opening (Fig. 3-A). After creating an inferior rim for the true ASD, the flap and rim were secured with running sutures. The line was designed to connect the superior margin of the ASD on the anterior side with the anterior limb of the right lower PV orifice. Close suturing around the anterior limb was used to avoid obstructing the PV. Immediately after completion of ASD closure (Fig. 3-B), we performed tricuspid valve annuloplasty with an artificial ring.

Following positive weaning from CPB, transesophageal echocardiography showed no residual ASD and negligible tricuspid regurgitation without the stenosis of rerouting from right lower PV to LA and from IVC to RA.

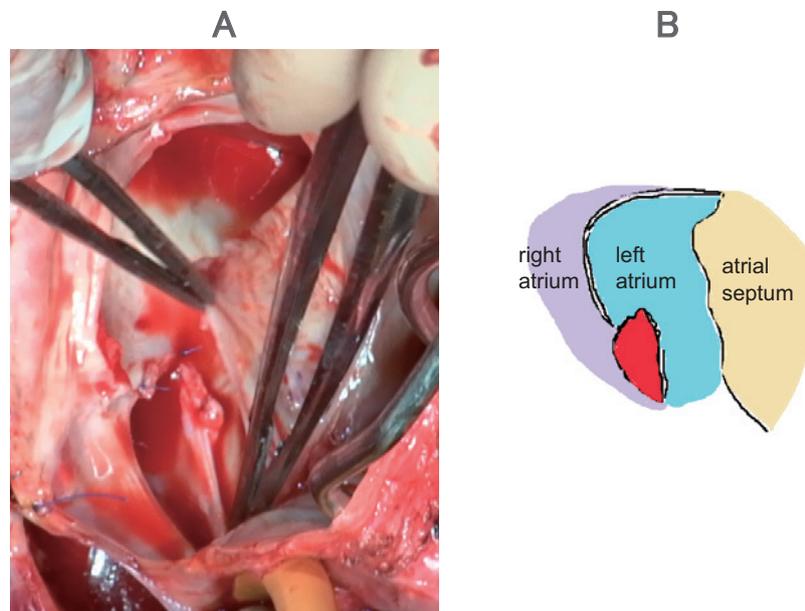


Fig. 2

A) Intraoperative photo showing the incision made in the atrial septal wall along the previous suture line, up to the superior margin of the fossa ovalis, and the enlarged orifice through the right lower PV.

Arrow: posterior limb of the right lower PV

B) Schema shows photo dissection (right atrium, left atrium, atrial septum)

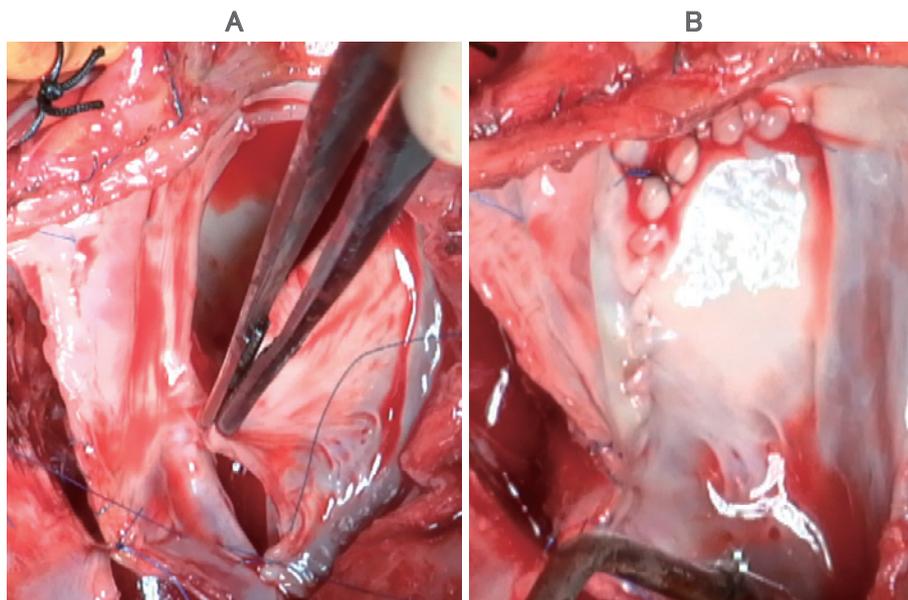


Fig. 3

A) Intraoperative photo showing that the atrial septal flap is large enough to close the ASD and right lower PV.

B) Intraoperative photo showing the inside of the right atrium after completion of ASD closure.

Discussion

It is unusual to pair a single right lower venous connection to the RA with an ASD. The change in the plane of the right PV was minimal. Because a posterior limb is present in this anomaly, the vein is anomalously connected to the RA. Thus, the combination of this anomaly and an ASD without an inferior septal rim is very rare.¹⁾ The surgical considerations for this case are similar to those for scimitar syndrome, i.e., determining the best way connect from the PV to the LA and the optimal baffle material (pericardium, polytetrafluoroethylene, or right atrial wall). We assumed that fresh autologous pericardium would be the best material, which has the advantage of antithrombogenicity; however, because of its distensibility, it bulges outward into the RA and disturbs IVC flow.²⁻⁵⁾ Although polytetrafluoroethylene is a common artificial material in cardiac surgery, particularly in congenital heart surgery, it requires antiplatelet therapy.²⁾³⁾ Intra-atrial rerouting, such as the Senning procedure, is believed to be suitable in terms of structure, but this complex procedure is not often performed, except for the double-switch operation for the corrected transposition of the great arteries, because of reproductibility and arrhythmia issues.⁵⁾ The procedures that use the atrial free wall flap for PAPVC to superior vena cava are impressive,⁶⁻⁸⁾ but in this case, these may not retain sufficient space in the IVC junction with the RA.

From the previous clinical records explaining the intraoperative IVC injury, misjudgment of the right lower PV orifice for the IVC opening was presumed. After enlarging the previous suture, we were able to clearly recognize the altitudinal gap between the upper and lower PVs. This determination was consistent with the results of the preoperative catheter examination.

The number of adult congenital heart disease (ACHD) cases continues to increase because of improved surgical outcomes and medical management. Surgery for ACHD patients is safe, beneficial, and low risk; however, procedures must be individualized to achieve the best results.⁹⁾ This sliding procedure, which uses the autologous tissue only, is reproducible and yields better results in relation to antithrombogenicity; hence, this deserves to be a novel way. However, this novel procedure was made possible by

the enlarged atrial and septal walls, which were attributable to the long period of illness and re-do operation. Although the indications may be limited, this technique is effective in reducing the risks of stenosis and thrombosis in the left and right heart.

Conflicts of interest: None declared

Consent for publication: Informed consent was obtained from the patient and her family for the publication of this case report and accompanying images.

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