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Modified Technique for Repairing Recurrent Pulmonary Venous Obstruction

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Postoperative pulmonary venous obstruction after the repair of total anomalous pulmonary venous connection is a challenging complication, especially when it occurs in the upstream pulmonary vein outside the pericardial sac. Here, we report such a case that was resolved successfully by a novel, modified, sutureless technique that repositioned the pericardial reflection to expand the pericardial sac.

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The incidence of pulmonary venous obstruction (PVO) after repair of total anomalous pulmonary venous connection (TAPVC) ranges from 5% to 18%.[1] With a high recurrence risk, PVO is often refractory to treatment, and in malignant cases extending to the upstream pulmonary vein (PV), poor prognoses are common.[2-4] Moreover, although there is no well-established treatment of such recurrent, upstream PVO, here we report a successful repair by a modified, sutureless technique with autologous tissue.

A 3098-g male neonate was delivered at 38 weeks and 2 days of gestation by induced labor with an Apgar score of 8/8. After delivery, cyanosis was noticed, and echocardiography revealed TAPVC type Ia with vertical vein stenosis, heterotaxy (asplenia), single ventricle, severe pulmonary stenosis, and dextrocardia. A sutureless TAPVC repair and modified Blalock-Taussig shunt were performed at day 0. At 3 months, a right PVO with massive collateral venous formation was observed, and repair was performed with intracardiac resection of the occluded lesion. At 9 months, a recurrent right PVO was observed.

Contrast-enhanced computed tomography showed the recurrent right PVO lesion extending from the PV-atrial junction to the upstream PV (Figure 1). Angiography revealed a mean pulmonary artery pressure of 17 mm Hg, a pulmonary artery index of 208, and a pulmonary vascular resistance of 1.2 Wood units. Aortopulmonary collaterals were occluded by coil embolization. An oxygen saturation in the upper 80s was maintained even after the collateral embolization.

PVO repair and bidirectional Glenn anastomosis were performed. Under cardiopulmonary bypass support, the right PV, surrounding pericardium, and common atrium were dissected out, then the occluded right PV lesion was confirmed as extending to the upstream PV beyond the pericardium. A pericardial incision was made just above the right PV, avoiding phrenic nerve injury, before the pericardium edge was resutured to the distal pulmonary pleura and surrounding tissues beyond the site of the PVO to relocate the pericardial reflection to the chest cavity and to extend the intrapericardial area beyond the occluded PV lesion. After cardioplegic arrest, an atrial incision allowed inspection of the right PV orifice, which was occluded, and a dent was noticed. With the dent guiding, the atrial wall adjacent to the right PV orifice was resected to make a window, and a longitudinal incision was made on the right PV anterior wall beyond the obstructed lesion. Subsequently, the pericardium was sewn to the atrial wall as a baffle to return right PV flow to the atrium (Figure 2).

Total bypass and cross-clamp times were 262 minutes and 48 minutes, respectively. Glenn pressure was steady at 12 to 15 mm H_2O. The postoperative course was uneventful, except for right phrenic nerve palsy, and diaphragmatic plication was performed later. The patient has

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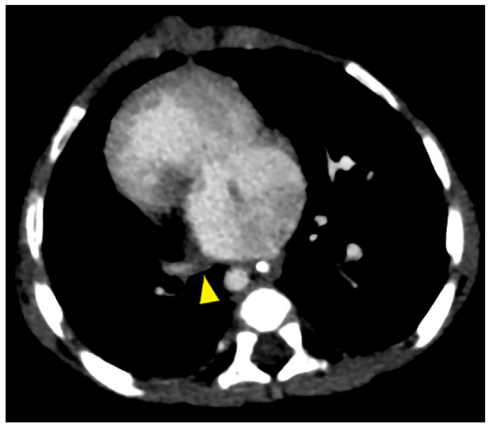

FIGURE 1 Preoperative computed tomography image. The arrowhead shows an occluded lesion of right pulmonary vein.

been followed up postoperatively for 1 year without any signs of recurrent PVO (Figure 3; Supplemental Figure 1).

COMMENT

Patients with TAPVC, heterotaxy, and single ventricle have a poor prognosis. Khan and coworkers[5] conducted a study on infants with a diagnosis of heterotaxy syndrome undergoing TAPVC repair, reporting an in-hospital mortality of patients with single ventricle of 43%, but 30% without single ventricle. In addition, Nakayama and colleagues[6] reported that for functional single-ventricle patients undergoing TAPVC repair, the 5-year survival rate with asplenia was 44.8% vs 87.5% without.

PVO complications after TAPVC repair greatly worsen prognoses. Although patients with functionally univentricular circulation or atrial isomerism were excluded, Seale and coworkers[1] reported estimates of 1-year survival for all patients with postoperative PVO as 62.0% (49.7%-72.1%), but 91.2% (87.6%-93.8%) for those without postoperative PVO. In addition, recurrent PVO further complicates repair, especially in cases in

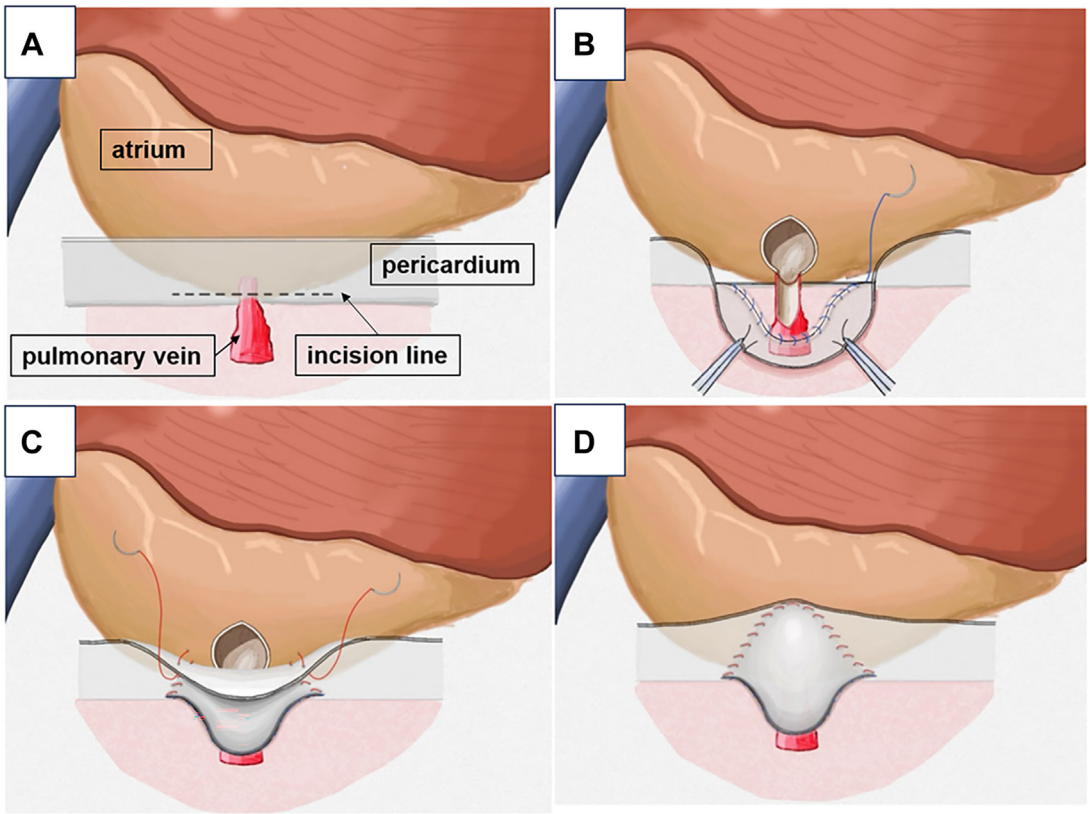

FIGURE 2 (A) An incision was made on the pericardium just above the right pulmonary vein (PV). (B) The pericardium edge was resutured on distal pulmonary pleura and surrounding tissues beyond the site of the PV obstruction. The atrial wall adjacent to the right PV orifice was then cut open to make a window, and anterior wall of right PV was resected longitudinally beyond the obstructed lesion. (C, D) The pericardium was sewn on the atrial wall as a baffle.

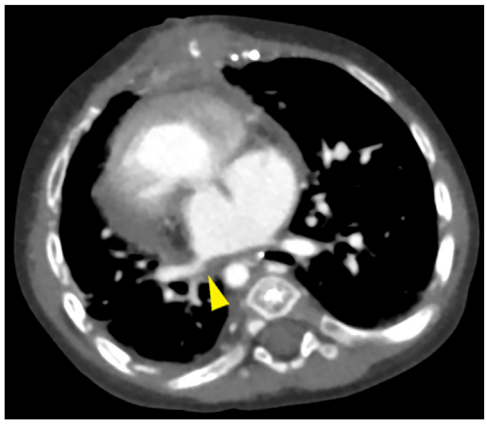

FIGURE 3 Postoperative computed tomography image. The arrowhead indicates repaired pulmonary vein obstruction site.

which it extends beyond the PV-atrial junction to the upstream PV outside the pericardial sac.

Whereas diverse reports have described PVO repair inside the pericardial sac, the sutureless technique is most commonly used and expected to decrease PVO recurrence. In contrast, scarce reports on PVO repair outside the pericardial sac exist because transcatheter interventions are often effective for short-term PVO relief, but occlusion tends to recur with poor long-term results.[3,4] Such an approach was used by Matsuyama and colleagues,[7] dissecting the PV peripherally beyond the pericardial cavity and covering the dissected extrapericardial area around the PV with a bovine pericardial patch after PVO release. However, this tactic may not translate to pediatric post-TAPVC repair patients because the Matsuyama report was in a 52-year-old man. Also, we previously reported another PVO release technique in which the left atrial wall, atrial septum, and stenotic PV were all resected, and an expanded polytetrafluoroethylene patch was used to make a left atrial roof.[8] The advantage of this method is versatility as use of artificial materials enables PVO repair even without access to pericardium, although longevity and recurrent PVO are still concerns.

Compared with previous reports, the advantage of this technique is use of autologous tissues. Although enough pericardium and pulmonary pleura must be spared, this technique may lower inflammatory response and thrombogenic reactions vs xenogeneic tissues or artificial materials. Furthermore, this method avoids direct suturing on PV tissue, minimizing risk of recurrent PVO. However, phrenic nerve palsy is the main surgical complication for this technique because the phrenic nerve runs beside the pericardial incision. Thus, protecting the phrenic nerves is of critical importance during repair.

In conclusion, we report on a case of recurrent PVO that involved the upstream PV outside the pericardial sac after TAPVC repair, in which the obstruction was successfully released with a modified, sutureless technique using autologous tissue. Such an approach may be a new useful option for recurrent upstream PVO.

The Supplemental Figure can be viewed in the online version of this article [https://doi.org/10.1016//j.atssr.2024.09.004] on http://www.annalsthoracicsurgery.org.

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DISCLOSURES

The authors have no conflicts of interest to disclose.

PATIENT CONSENT

Consent for publication was obtained from the patient and his parents.

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