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Title
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Author(s)
NAKATA Tomohiro / TACHI Maiko / YASUDA Kenji / TANABE Junya / Kawanami Yuka / YAMAZAKI Kazuhiro

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We encountered an adult patient with right hemitruncus with pulmonary hypertension (PH) and right PAVF. We successfully performed a definitive repair and follow-up selective contrast echocardiography showed significant regression of PAVF.

Case Report

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Cardiac catheterization demonstrated that the pressure of the left pulmonary artery (LPA), the RPA, and the aorta was 77/13/45, 151/70/103, and 146/70/102 mmHg, respectively. The pulmonary vascular resistance estimated by cardiac catheterization and magnetic resonance imaging was 8.5 WU/m^2. Selective contrast echocardiogram (agitated saline injection into the RPA) during cardiac catheterization demonstrated a moderate amount and prompt appearance of echo contrast in the left atrium. Echocardiography demonstrated moderate mitral regurgitation (MR) (central regurgitation) with a left ventricle end-diastolic diameter (LVDd) of 66 mm.

The patient was placed on cardiopulmonary bypass in the standard fashion, using ascending aortic and bicaval cannulation. Ligation and division of the PDA were performed, and then cardiac arrest was established and an atrioseptostomy of 4 mm was created through a right atriotomy. Next, a woven Dacron graft (12 mm in diameter) was employed to connect the RPA and main pulmonary artery via an end-to-side anastomosis (5-0 polypropylene, double-stitch; Fig. 2).

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Epoprostenol infusion and nitric oxide inhalation were discontinued on postoperative day 2 and 3, respectively. Tadalafil, macitentan, selexipug, aspirin and warfarin were initiated during hospitalization. Right heart catheterization on postoperative day 27 demonstrated that the pressure of the LPA and RPA were 46/11/26 and 42/11/25 mmHg, respectively. The pulmonary vascular resistance estimated by cardiac catheterization was 4.1 WU/m^2. The patient was discharged without major complications on postoperative day 32.

A thorough postoperative examination was performed 6 months after surgery. Enhanced CT (Fig. 1B) and cardiac catheterization demonstrated unobstructed flow within the graft. The pressure of the LPA and RPA was 42/9/21 and 30/9/17, respectively. The pulmonary vascular resistance estimated by cardiac catheterization was 3.8 WU/m^2. Selective contrast echocardiogram showed no residual PAVF. Echocardiography demonstrated mild MR with LVDd of 51 mm. A lung perfusion scan showed the ratio of right and left pulmonary blood flow had improved from 10.4:89.6 (predischarge) to 16.3:83.7 (6 months post-surgery). A lung perfusion scan revealed that the estimated shunt ratio ([wholebody – lung] / wholebody) had also improved from 11.2% (predischarge) to 5.0% (6 months post-surgery). The triple therapy (tadalafil, macitentan and selexipug) is being continued at the time of writing and we plan to

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discontinue these drugs one by one after the next follow-up cardiac catheterization.

Discussion

Although the cause of diffuse PAVF is unknown, the absence of HF in pulmonary blood flow is believed to be a predisposing factor [4–6]. Usually, the initial symptom of PAVF is hypoxia. However, in patients with right hemitruncus, the right lung is perfused with fully oxygenated blood and the progression of PAVF never causes hypoxia. This feature might result in delayed diagnosis of PAVF. If the selective contrast echocardiography is performed routinely in patients with hemitruncus, it is possible that diffuse PAVF will be found more frequently.

We speculated that HF was consumed mostly by the left lung and was excluded from the right lung in patients with right hemitruncus [6]. PAVF in this patient was not localized [7] but diffuse (undetectable by enhanced computed tomography); therefore, we predicted that this diffuse PAVF could be acquired (not congenital PAVF) due to the lack of HF; therefore, this PAVF could be improved by inclusion of HV blood flow to the RPA by the definitive repair.

It may be argued that a right pulmonary artery banding (RPAB) be less invasive than the definitive repair [8]. The definitive repair of hemitruncus in adulthood is a

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We assumed that after the RPA reconstruction in the presence of PAVF, right-to-left shunt could result in desaturation. However, the extent of desaturation

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Abbreviations

CT	computed tomography
HF	hepatic factor
LPA	left pulmonary artery
LVDd	left ventricle end-diastolic diameter
MR	mitral regurgitation
PAVF	pulmonary arteriovenous fistula
PDA	patent ductus arteriosus
PH	pulmonary hypertension
RPA	right pulmonary artery
RPAB	right pulmonary artery banding

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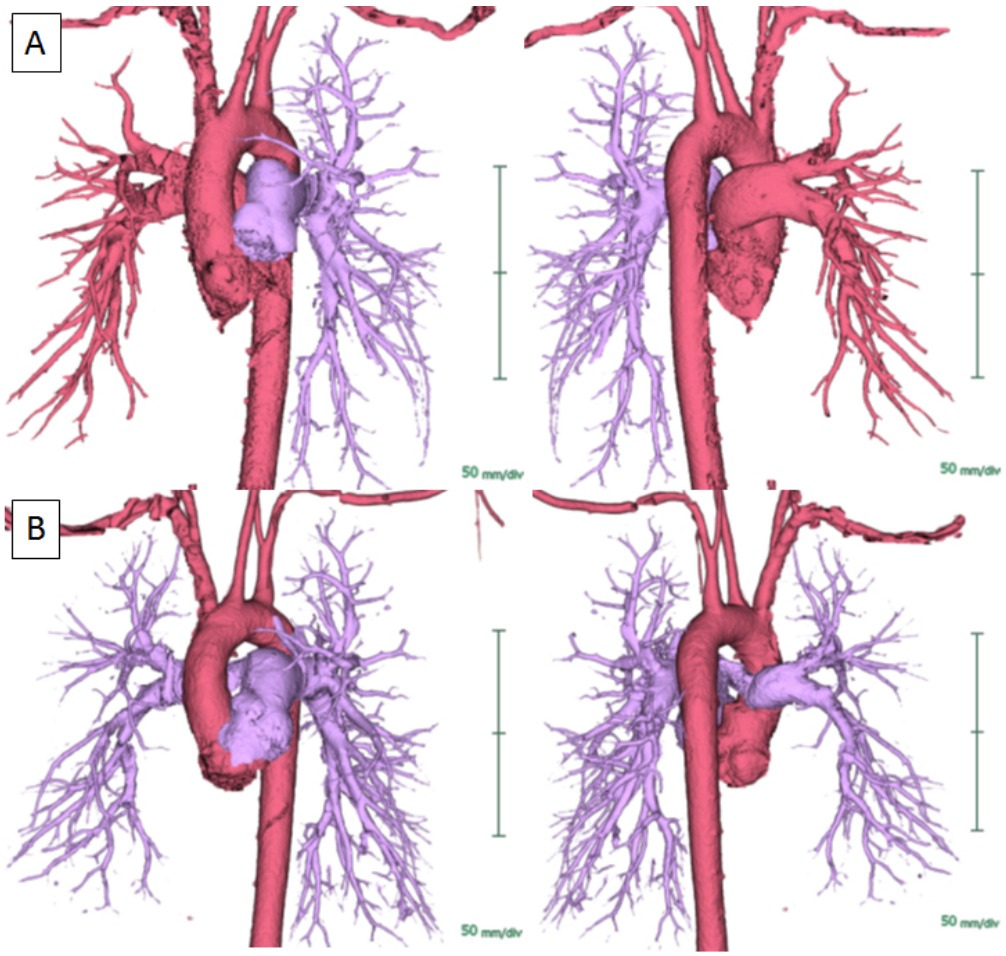


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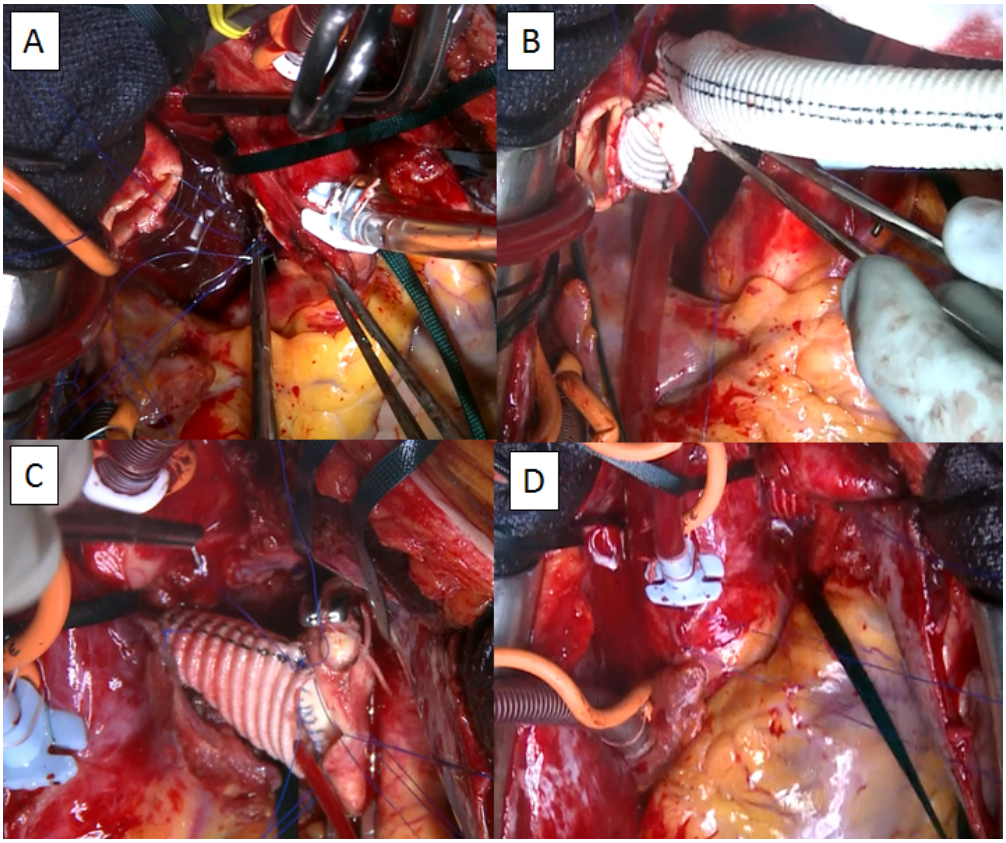


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91x75mm (200 x 200 DPI)