

Hybrid procedure: percutaneous atrial septal defect closure and off-pump surgical repair of left-sided partial anomalous pulmonary venous connection

Procedura hybrydowa: przezskórne zamknięcie ubytku przegrody międzyprzedsionkowej oraz korekcja częściowo nieprawidłowego lewostronnego spływu żył płucnych bez użycia płucoserca

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Abstract

Partial anomalous pulmonary venous connection of the left upper pulmonary vein with atrial septal defect is quite a rare congenital defect. Standard treatment of this combined defect is cardiopulmonary bypass surgery through a median sternotomy. We describe a hybrid procedure for a 12-year-old female patient where the atrial septal defect was closed percutaneously and anomalous pulmonary venous connection of the left upper pulmonary vein was corrected through a minimal off-pump surgical approach during the same anesthesia.

Key words: congenital heart diseases, hybrid procedure.

Streszczenie

Częściowo nieprawidłowy spływ lewej górnej żyły płucnej wraz z ubytkiem przegrody międzyprzedsionkowej jest rzadką wrodzoną wadą serca. Standardem w leczeniu tej kombinowanej wady serca jest operacja wykonywana poprzez sternotomię z użyciem płucoserca. W pracy przedstawiono procedurę hybrydową wykonaną u 12-letniej pacjentki podczas zastosowania jednego znieczulenia. Zabieg obejmuje przezskórne zamknięcie ubytku przegrody międzyprzedsionkowej i korekcję górnej żyły płucnej z zastosowaniem minimalnego chirurgicznego podejścia bez użycia płucoserca.

Słowa kluczowe: wrodzone wady serca, procedura hybrydowa.

Introduction

Partial anomalous pulmonary venous connection (PAPVC) is a rare congenital condition present in 0.4% to 0.7% of postmortem examinations where one or more (but not all) of the pulmonary veins are connected to the venous circulation. This anomaly usually occurs in the right lung, with only 7-10% of anomalous pulmonary veins originating from the left lung [1, 2]. The most common left-sided PAPVC is for one or more left pulmonary veins to enter a vertical vein that drains superiorly into the innominate vein [2]. This congenital defect may go undiagnosed or undetected as it often presents with few clinical symptoms [3]. Often PAPVC

is associated with atrial septal defects (ASD) [1, 2]. If left untreated patients may develop pulmonary hypertension, pulmonary obstructive disease, or right ventricular failure [2]. Standard treatment is surgical redirection of the anomalous pulmonary vein to the left atrium and ASD closure via a median sternotomy with cardiopulmonary bypass [1]. We describe a hybrid treatment by closing the ASD percutaneously and repairing left-sided PAPVC surgically through left posterolateral thoracotomy without cardiopulmonary bypass during the same anesthesia. To our knowledge this is the first report for this hybrid approach to such a combined congenital defect in the literature.

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Case

An otherwise healthy 12-year-old girl was suspected of congenital defect incidentally when a heart murmur was observed during the ordinary screening a month before admission to our hospital. Transthoracic echocardiography showed a hemodynamically significant 20 mm ostium secundum ASD, moderate right-sided enlargement, mild mitral regurgitation, and preserved heart function. The pulmonary flow to systemic flow ratio was 3.68. Transesophageal echocardiography confirmed the diagnosis. No evidence of PAPVC was detected. The patient was short-listed for a device closure of the ASD. An anomalous left upper pulmonary vein connection to the left innominate vein via the vertical vein was detected during cardiac catheterization (Fig. 1A). The procedure was postponed. It was decided to perform a hybrid procedure by closing the ASD percutaneously and correcting PAPVC surgically. Intravenous analgesia and endotracheal intubation was applied. ASD was closed percutaneously using 24 mm Occlutech Figulla ASD Flex Occluder (Occlutech GmbH, Jena, Germany) in the standard manner [4] in the catheterization laboratory (Fig. 1B). Then intubated patient was transferred to the operating room. A less invasive off-pump surgical technique was applied as described by Andrew W. ElBardissi and colleagues [2] with some adaptation. The chest was opened through left posterolateral mini-thoracotomy. The pericardium was opened posterior to the left phrenic nerve (Fig. 2A). The left upper pulmonary vein and vertical vein were mobilized and dissected from the inflow to the innominate vein. After heparinization the auricle of the left atrium and anomalous vein were clamped and opened. The mobilized left upper pulmonary vein was anastomosed with the left auricle

using continuous 6-0 polydioxanone suture. Air was expelled from the anastomosis and it was unclamped (Fig. 2B). A chest drain was placed in the left pleural cavity. The postoperative period was uneventful. The patient was extubated after a few hours and discharged home on the fifth day. 200 mg of aspirin was prescribed for the patient for 6 months. Follow-up transthoracic echocardiography showed mild right-sided enlargement, leak proof atrial septum, and no evidence of left pulmonary vein obstruction or stenosis.

Discussion

Combined significant ASD and PAPVC of the left upper pulmonary vein are rare [1, 2]. Individuals with a pulmonary flow to systemic flow shunt ratio higher than 1.5:1 should undergo surgical correction of these congenital defects [1]. The standard approach through median sternotomy produces excellent results [1, 2]. For cosmetically anxious patients a hybrid approach is possible, which was originally suggested by Andrew W. ElBardissi and colleagues [2]. But we did not find a report of an accomplished procedure of the hybrid approach for such type of combined congenital defect in the literature. Percutaneous ASD closure is the routine procedure nowadays [4], and surgical repair of isolated left-sided PAPVC through the left thoracotomy is straightforward [1]. The hybrid procedure of percutaneous closure and off-pump posterolateral thoracotomy could be advantageous for a number of reasons, including cosmesis [5, 6] and avoidance of cardiopulmonary bypass risks [7]. Posterolateral thoracotomy compared with anterolateral may be more advantageous in children as there is less possibility for breast deformation [5]. However, the surgical approach through lateral thoracotomy has its own risks,

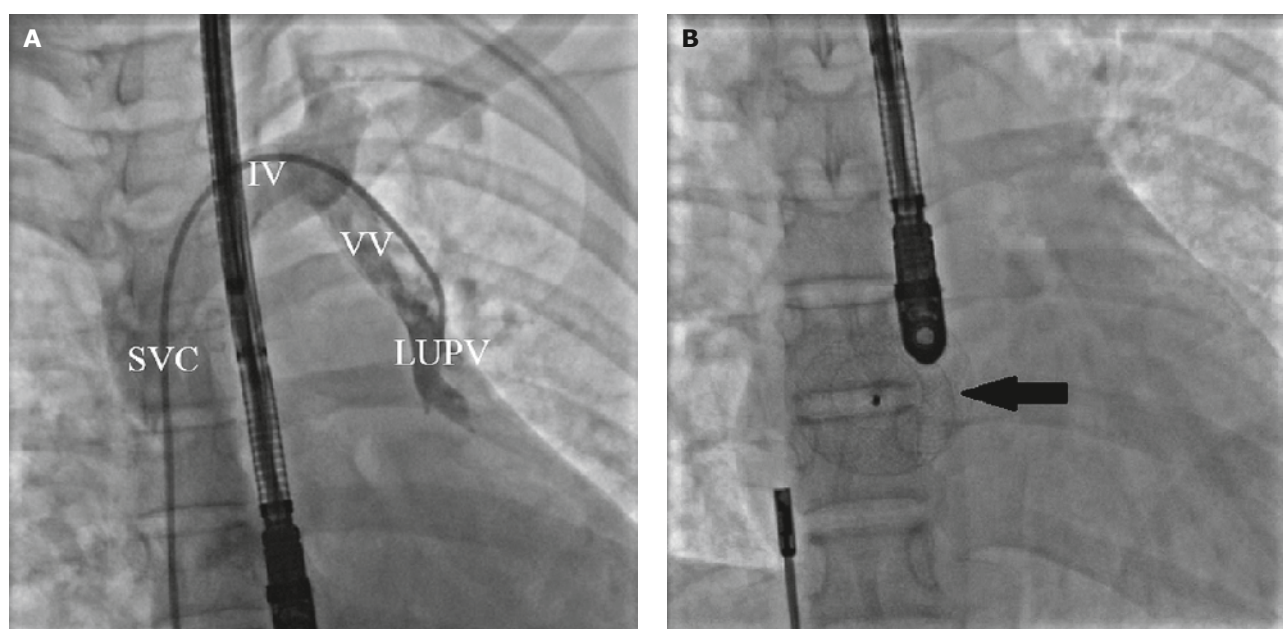


Fig. 1A–B. Anomalous left upper pulmonary vein connects to innominate vein through vertical vein (A). Atrial septal defect is closed with a 24 mm Occlutech Figulla ASD Flex Occluder (black arrow) (B). LUPV – left upper pulmonary vein; VV – vertical vein; IV – innominate vein; SVC – superior vena cava

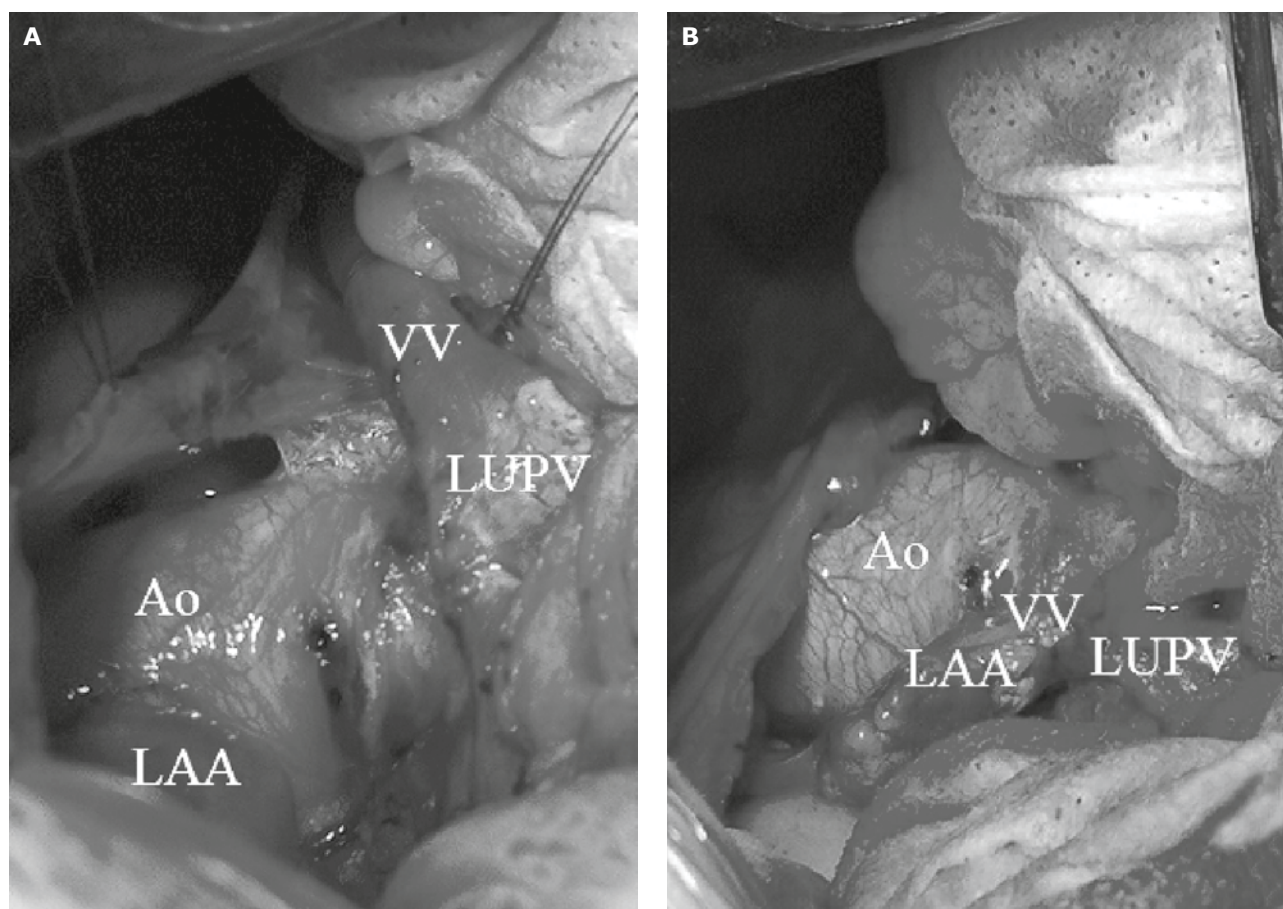


Fig. 2A–B. The left upper pulmonary vein is seen connected to a vertical vein (A). After correction the pulmonary vein is anastomosed to the left atrial appendage (B). LUPV – left upper pulmonary vein; VV – vertical vein; Ao – aorta; LAA – left atrial appendage

such as lung damage, phrenic nerve damage or winged scapula. Also it is not quite clear whether the surgical manipulation of the left auricle could destabilize or dislocate a just implanted occluder and which procedure – surgical or interventional – should be done first. Moreover, this novel procedure is more challenging as it requires a multidisciplinary approach. Both a cardiac surgeon and an interventional cardiologist must arrange the plan of the procedure.

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