



Case Report / Olgu Sunumu

Surgical management of late-term pulmonary venous baffle stenosis after Senning operation

Senning ameliyatı sonrası geç dönem pulmoner venöz odacık darlığının cerrahi tedavisi

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ABSTRACT

Although pulmonary venous baffle stenosis is not a common late complication after the Senning operation, surgical treatment is vital, when interventional methods fail to correct the pathology. Our patient was admitted to hospital with recurrent epistaxis and exertional dyspnea 6.5 years after the Senning procedure. The gradient in the pulmonary venous baffle was measured as 34 to 35 mmHg via transthoracic echocardiography. During surgical treatment, the calcified and retracted polytetrafluoroethylene flap was completely excised and porcine pericardial xenograft was used to cover the pulmonary venous baffle. At the end of the procedure, no significant gradient was detected via transesophageal echocardiography.

Keywords: Porcine xenograft; pulmonary venous baffle stenosis; Senning operation.

ÖZ

Senning ameliyatı sonrası pulmoner venöz odacık darlığı sık görülen bir komplikasyon olmamasına rağmen, patolojinin düzeltilmesinde girişimsel yöntemlerin başarısız olması durumunda cerrahi tedavi hayati önem taşır. Olgumuz, Senning ameliyatından 6.5 yıl sonra tekrarlayan burun kanaması ve efor dispnesi ile hastanemize başvurdu. Transtorasik ekokardiyografide pulmoner venöz odacıkta gradyan 34-35 mmHg olarak ölçüldü. Cerrahi tedavi sırasında kalsifik ve geri çekilmiş politetrafloroetilen flep tamamen eksize edildi ve domuz perikard ksenogreft ile pulmoner venöz odacık kapatıldı. İşlemin sonunda transözofageal ekokardiyografide anlamlı bir gradyan izlenmedi.

Anahtar sözcükler: Domuz ksenogreft; pulmoner venöz odacık darlığı; Senning ameliyatı.

The Senning and Mustard procedures are not as widely used as in previous decades since the arterial switch operation (ASO) has become common worldwide. Arterial switch operation is associated with lower complication rates and provides more adequate physiological correction rather than the Senning or Mustard procedure.^[1] However, these procedures are still favored in cases of corrected transposition of the great arteries (c-TGA) or transposition with complex coronary anomalies.^[2] Furthermore, the Senning procedure can be also performed to provide anatomical correction in patients with isolated atrioventricular discordance.^[2-4]

Following the Senning operation, several complications, particularly in late-term, may develop such as arrhythmias, right ventricular systolic dysfunction, tricuspid valve insufficiency, and baffle leak can be seen.^[5] In addition, the complication rate of pulmonary venous baffle stenosis (PVBS) varies from 1.9 to 7.6%,^[5,6] and among patients undergoing double switch operation with the diagnosis of c-TGA, the PVBS rate has been shown to be similar (%5).^[6] However, the majority of patients with PBVS remain asymptomatic and only 2.5 to 3% of patients require surgical treatment.^[7] Herein, we present a patient who underwent reoperation for PVBS caused by a calcified

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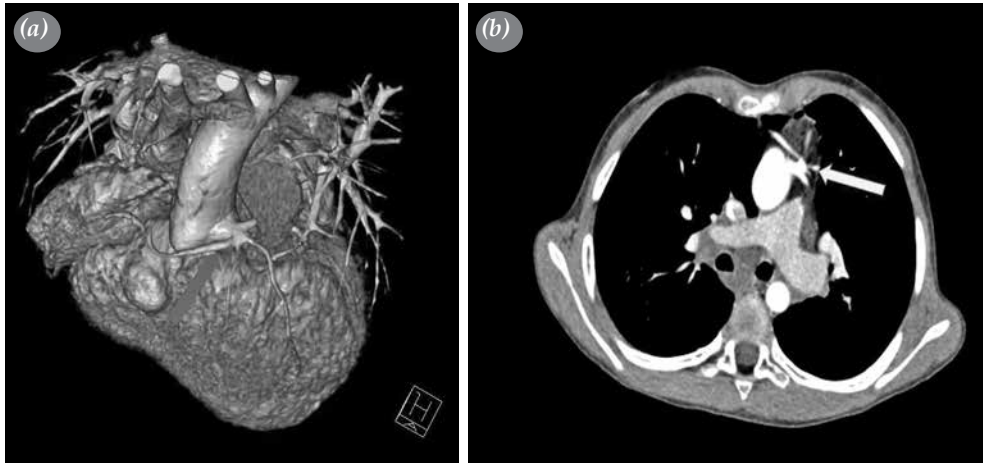


Figure 1. (a) A computed tomography and three-dimensional image showing that all coronary arteries originated from a single left coronary orifice. (b) Left coronary orifice and coronary arteries are marked with arrow.

polytetrafluoroethylene (PTFE) patch 6.5 years after the Senning procedure.

CASE REPORT

Our male case was diagnosed with Taussig-Bing anomaly presenting with transposition of the aorta to the right ventricle and malposition of the pulmonary artery in the antenatal period. Furthermore, there was a coexisting atrial septal defect (ASD). He underwent pulmonary banding operation in the neonatal period via median sternotomy in an external center. The Senning operation was performed, when the patient reached age of 18 months in our clinic.

We were unable to perform ASO due to coronary anomaly and adjacent localization of the great arteries. All coronary arteries were originating from a single left coronary orifice (Figure 1). Thus, the Senning operation was decided rather than ASO. A written informed consent was obtained from each parent.

Due to the adhesions from the initial operation and lack of bioprosthesis pericardial patches, a PTFE patch was used for both the closure of interventricular communication and covering the outer wall of the pulmonary venous baffle rather than autologous pericardium. The surgical procedure was smooth and



Figure 2. (a) A computed tomography and three-dimensional image showing calcification in the polytetrafluoroethylene flap covering the pulmonary venous baffle. (b) The calcified and retracted segment is indicated by the arrow.

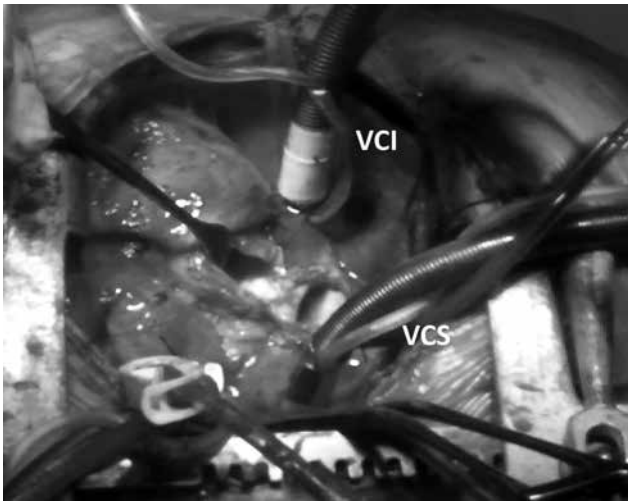


Figure 3. An intraoperative view of calcific and retracted polytetrafluoroethylene flap (The photo was taken by the head-side of the patient).

VCI: Vena cava inferior; VCS: Vena cava superior.

there were no complications until childhood during clinic follow-up visits.

The patient was planned to be examined by transthoracic echocardiography (TTE) annually. Unfortunately, the patient was lost to follow-up and the final imaging was able to be performed two years ago. At the final visit, no significant gradient was measured.

At the age of eight, weighing 21 kg, the patient began to suffer from recurrent epistaxis which occurred particularly in the mornings and exertional dyspnea. The patient was referred for consultation, and no pathological sign was found on nasopharyngeal examination. However, TTE showed PVBS with a mean gradient of 34 to 35 mmHg gradient. In addition, mild insufficiency was observed in systemic atrioventricular valve with a right ventricle ejection fraction (RVEF) of 50%. There is also mild pulmonary arterial hypertension on TTE. Computed tomography angiography showed an enlargement in the pulmonary venous chamber (Figure 2). Based on these evidences, epistaxis was thought to be related to hemoptysis, and surgical treatment was decided. Due to the potential rupture risk of the calcified flap, balloon angioplasty was not considered as a treatment option by cardiologists. A written informed consent was obtained from each parent.

The perioperative transesophageal echocardiography (TEE) confirmed the diagnosis. The heart was accessed through re-sternotomy. The previously inserted PTFE patch was calcified

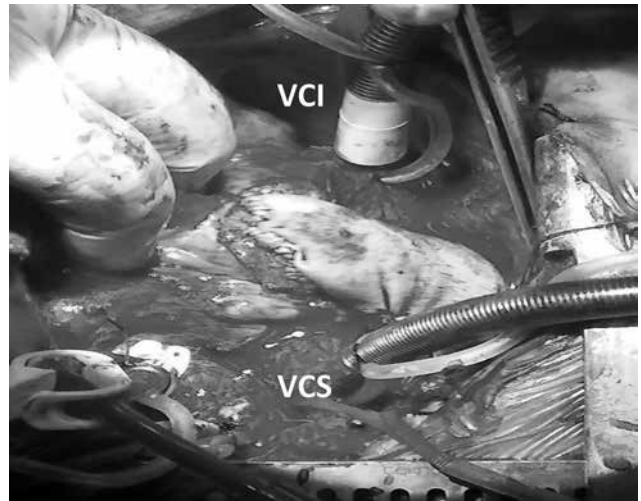


Figure 4. An intraoperative view showing pulmonary venous baffle augmentation with porcine graft (The photo was taken by the head-side of the patient).

VCI: Vena cava inferior; VCS: Vena cava superior.

and retracted with a limited motion. Based on intraoperative findings, asymmetrical and skew tissue development were considered responsible for kinking PTFE graft which rapidly calcified and stenosed. Following bicaval cannulation and antegrade cardioplegia administration, retracted pulmonary venous baffle patch was completely resected (Figure 3), and the pulmonary veins were exposed. No obstruction was observed. The gap was covered by a porcine pericardial biograft using 5/0 polypropylene thread (Figure 4). Total cardiopulmonary bypass time was 75 min and cross-clamp time was 48 min. At the end of the surgical procedure, repeated TEE revealed no gradient at the baffle. Postoperatively, the patient was admitted to the intensive care unit and, then, extubated in the first day without any need of inotropic support during this period. He was, then, referred to the ward on postoperative Day 2. He was asymptomatic and in sinus rhythm. The patient was discharged on postoperative Day 7. The final TTE before discharge was non-specific, and there was no gradient in either systemic or pulmonic pathways. The RVEF was measured as 50%.

At one and three months during follow-up, TTE examinations were performed. The patient was in the New York Heart Association (NYHA) Class I and cardiac rhythm was sinus. There was no measured gradient in the baffles and the RVEF was 50%. All control TTE examinations were non-specific, except for mild insufficiency in the tricuspid and mitral valves.

DISCUSSION

Currently, the Senning operation has been mostly abandoned in favor of ASO. Raissadati et al.^[8] published a nationwide, four decades of results comparing Mustard, Senning, and ASO outcomes. Late mortality was seen in the Senning group with a 20% rate which was significantly higher than the ASO group. In addition, the authors reported a higher 30-year reoperation-free survival with a rate of 84%, and the major causes of mortality were arrhythmias and heart failure. In another study, Kammeraad et al.^[9] showed that atrial fibrillation or atrial flutter were the only associated factors which increased the risk of sudden death, while electrocardiography (ECG), chest X-ray, and Holter ECG findings were not predictive.

Until now, there are several publications reporting complications following the Senning operation. Right ventricle failure may develop in later years with a rate of over 10%.^[5,6,8,10,11] Tricuspid valve insufficiency also is common among patients undergoing the Senning operation. The underlying mechanism which has been proposed is that the tricuspid valve is more pressure-sensitive and loses its usual structure over time.^[10] The reported incidence varies between 8.8 and 17.2%.^[8,11] Baffle leakage has been also reported as an ensuing complication with a rate of about 8%.^[12] Left ventricular outlet tract obstruction (LVOTO) is another reason for reoperation; LVOTO accounted for five of a total of 12 reoperations in the study of Roubertie et al.^[13]

Although PBVS is not a common late complication, it is of utmost importance, as it is associated with low cardiac output and sudden death in the presence of ventricular tachycardia.^[14] Although PBVS may occur in short-term, the surgical technique used plays a critical role. In later years, the development of stenosis is more related to calcification or retraction caused by non-growing materials. For the management of PBVS, interventional, hybrid, or conventional methods may be chosen according to patient characteristics.

Although PBVS mostly occurs several years after surgery, some authors have suggested recognizing stenosis in the short-term, as well. In a case series of 16 patients with a mean follow-up of six months, Satomi et al.^[15] examined the predictive value of TTE in rapid PBVS development. Three patients with pulmonary venous channel diameter and body surface area rate below 20 mm/m² needed surgical repair. Moreover, Kurokawa et al.^[16] reported a case who was diagnosed with TEE intraoperatively and in whom a

repair was needed. Consequently, they defined this rate as a sign of emerging or existing stenosis. In addition, we used TEE during surgery in our case and the score was measured as 18 mm/m². Therefore, we recommend close follow-up after surgery with regular TTE examinations.

Sareyyupoglu et al.^[17] reported a 28-year-old patient who underwent hybrid PBVS correction via right anterior minithoracotomy through the fourth intercostal space. However, this procedure cannot be frequently performed due to technical difficulties.

Similar to our case, Juaneda et al.^[18] presented a patient with PBVS caused by calcification and retraction of the pulmonary venous baffle flap. After an unsuccessful balloon angioplasty procedure (15×40 mm), they used a PTFE flap for enlargement. Elder and Hellenbrand^[19] reported a similar case who had a new-onset of tachypnea, cough, and exertional dyspnea, classically suggesting pulmonary edema and congestive heart failure secondary to pulmonary venous obstruction after an uneventful four years following the initial surgery. Unfortunately, their surgical method has not been described in detail in their report. However, Dhawan et al.^[20] reported a nine-year-old case who was uneventful for seven years following the Senning procedure, and lately began to suffer from recurrent episodes of cough with expectoration and hemoptysis. The authors performed the correction procedure via homologous pericardium.

In conclusion, surgical treatment is essential, when interventional methods are not able to correct pulmonary venous baffle stenosis. Therefore, as in our case, a porcine graft as a flap can be used to avoid reimplantation of a polytetrafluoroethylene graft in the absence of autologous pericardial tissue.

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