# Effectiveness of posterior aortopexy for the left pulmonary vein obstruction between the left atrium and the descending aorta

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## Abstract

Background: Left pulmonary vein (PV) obstruction can occur due to compression between the left atrium (LA) and the descending aorta (DA). One of the effective solutions for this problem is posterior aortopexy. In this study, we have reported five cases of posterior aortopexy to relieve left PV obstruction between the LA and the DA. Methods: Since August 2012, five patients have undergone posterior aortopexy for compression of the left PV between the LA and the DA. The median age and weight of the patients at the time of operation were 5.5 months (range, 1-131 months) and 5.2 kg (range, 4.2-29.5 kg), respectively. The left PV obstruction was initially diagnosed on echocardiography in four patients and computed tomography angiography in one patient. The median peak pressure gradient across the obstructed left PV was 7.3 mmHg (range, 4-20 mmHg). Concomitant procedures were ventricular septal defect closure in one patient and patent ductus arteriosus ligation in one patient. Results: There was no PV obstruction on echocardiography in any of the patients after the operation except in the case of one patient who had diffuse pulmonary vein stenosis. The median follow-up duration was 34 months (range, 14-89 months), and during follow-up no incidence of the left PV obstruction was observed in any of the surviving patients. Conclusions: The posterior aortopexy technique could be a good surgical option for the left PV obstruction caused by compression between the LA and the anteriorly positioned DA.

#### Introduction

Pulmonary vein (PV) obstruction is a known prognostic risk factor in the congenital heart surgery <sup>1</sup>. Left PV obstruction can occur due to compression between the left atrium (LA) and the descending aorta (DA), which can be a result of a combination of cardiomegaly and the anteriorly positioned DA or one of them  $^{2-5}$ . One of the effective solutions for this problem can be a posterior aortopexy, which is generally used for the management of airway obstruction  $^{6-9}$ . In this study, we have investigated the effectiveness of the posterior aortopexy technique to relieve left PV obstruction caused by compression between the LA and the DA.

#### Materials and methods

The institutional review board at Pusan National University Yangsan Hospital approved the present study, and the patient consent was waived for the retrospective data analysis (IRB No. 05-2018-178). We have adopted the posterior aortopexy technique to treat left PV obstruction caused by compression between the LA and the DA in five patients (three female) since August 2012. The median age and weight of the patients at the time of operation were 5.5 months (range, 1-131 months) and 5.2 kg (range, 4.2-29.5 kg), respectively. When the patient revealed a turbulence flow or peak pressure gradient (PG) of 10 mmHg or more, or showed a continuous, non-phasic flow pattern on the echocardiography, the condition was deem to be an obstruction of the corresponding  $PV^{1,10}$ , and then the position and shape of the pulmonary vein obstruction was confirmed by using computed tomography (CT) angiography <sup>11</sup>. All five patients but one were initially diagnosed with the left PV obstruction on the echocardiography. In one patient, the left PV obstruction

was diagnosed with CT angiography and cardiac catheterization. This patient showed a distended inferior vena cava in follow-up echocardiography after the Fontan procedure. The CT angiography performed for further evaluation showed left PV obstruction between the LA and the DA (Fig. 1A). A sharp demarcation of the left PV flow at the DA border was also noticed in the levophase of pulmonary angiogram (Fig. 1B). The compressed PV pattern was identified on preoperative CT angiography: focal stenosis in three patients, focal and diffuse stenosis in one patient, and diffuse stenosis in one patient. The median preoperative peak PG across the obstructed left PV was 7.3 mmHg (range, 4-20 mmHg) in all five patients, and concomitant procedures were performed in two patients. Table 1 summarizes the baseline characteristics of the patients.

Posterior aortopexy was carried out through left thoracotomy (the fourth intercostal space (ICS) in one patient, the fifth ICS in three patients, and the sixth ICS in one patient). The DA was fixed to the posterior chest wall using Teflon pledget-buttressed 3-0 or 4-0 polypropylene sutures (Fig. 2A and B) <sup>6,12</sup>. To evaluate the appropriateness of the posterior aortopexy technique, we measured the change in the angle between the DA and the vertebra on CT angiography before and after the operation. A horizontal line was made from the center of the vertebra, and the other line was drawn from the center of the vertebra to the center of the DA in the axial slice at the level of left PV obstruction (Fig. 3A, inlet). Subsequently, we checked the angle between the two lines before and after the operation on CT angiography, and the change in the angle (Za) was calculated by using the formula mentioned below.

Za = a - a'

a = preoperative angle between the DA and the vertebra (Fig. 3A)

a' = postoperative angle between the DA and the vertebra (Fig. 3B)

If the value of Za is positive, the posterior aortopexy is considered to be performed effectively.

The data were collected and managed with Microsoft Excel 2016 (Microsoft, Redmond, Wash) and the Statistical Package for Social Sciences, version 25 (SPSS, Inc., Chicago, Ill). The continuous variables are expressed as a median and range.

## Results

There was no early death during the study period. The median postoperative ventilator support time was 10 hours (range, 0-174 hours). The median intensive care unit stay was 2 days (range, 1-227 days), and the median hospital stay was 7 days (range, 6-257 days). None of the patients showed a phrenic nerve palsy following the operation (Table 2). The change in the angle between the DA and the vertebra (Za) was measured, and effective aortopexy (Za > 0) was observed in four patients (80%). A possible reason for the inadequate posterior aortopexy in patient 5 was related to the surgical approach. We adopted a posterior aortopexy through the fourth ICS thoracotomy for concomitant patent ductus arteriosus (PDA) ligation and relief of airway obstruction on the patient 5. This might have led to an inadequate posterior aortopexy (Fig. 4A and B, Za = -1). The median postoperative peak PG across the left PV was 3.1 mmHg (range, 0-20 mmHg), and all but one patient showed a decreased PG after posterior aortopexy (Table 3).

The median follow-up duration was 34 months (range, 14-89 months). There was one case of late mortality. Patient 2 had the Rastelli operation for truncus arteriosus, atrial septal defect, and partial anomalous pulmonary venous return (PAPVR) at the age of 51 days. The patient showed persistent pulmonary edema and severe pulmonary hypertension, and the left PV obstruction was identified between the LA and the DA on postoperative follow-up echocardiography and CT angiography (Fig. 5A). In this patient, posterior aortopexy for the left PV obstruction was performed at the age of 5 months. The PV obstruction pattern was diffuse, and the PG across the obstructed left PV was still high after the procedure (20 mmHg, Fig. 5B). The patient also showed a moderate truncal valve regurgitation, and truncal valve and PAPVR repair were performed subsequently. However, pulmonary hypertension and PV obstruction persisted, and the patient eventually died at the age of 14 months. The median cardiothoracic ratio (CTR) at the last follow-up chest radiography was 0.54 (range, 0.47-0.63) in all five patients. Patient 4 and 5 demonstrated a decrease in the last follow-up CTR as compared to the preoperative results (0.57 vs. 0.7 and 0.52 vs. 0.62). In the

last follow-up echocardiography, there was no PV obstruction in any of the patients except for one case of mortality (Patient 2) (Table 4).

### Discussion

The left PV obstruction caused by compression between the LA and the DA has been rarely reported <sup>2-5,11</sup>. Previous reports demonstrate that the left lower PV is usually involved, and four out of five patients exhibited left lower PV obstruction in the present study as well. Some of the possible factors that affect the left PV obstruction caused by compression between the LA and the DA should be considered. First, the abnormally anterior location of the DA can be a cause. The left PV can be narrowed due to compression from the back by the DA located forward <sup>2</sup>. The PV has clearly low pressure as compared to the DA. Therefore, if the anteriorly located DA passes just behind the PV, it can cause narrowing of the involved PV. Second, an enlarged LA can compress the left PV from the front in the space between the LA and the DA. This phenomenon can be observed in the heart with left-sided volume loading, such as atrioventricular valvular (AVV) regurgitation or large left-to-right shunt lesions<sup>4,5</sup>. The third possible factor can be a cardiac compression that is seen posteriorly due to chest wall deformity such as the pectus excavatum

Direct surgical repair <sup>13-15</sup>, balloon dilatation<sup>16,17</sup>, and stent implantation <sup>18,19</sup> can be the treatment options for the PV obstruction caused by compression between the LA and the DA. Kotani et al. reported 15 patients with left PV obstruction among 494 consecutive single-ventricular patients. The authors performed the sutureless repair in seven patients and failed to achieve the Fontan operation. Posterior aortopexy was not attempted in any of the patients <sup>4</sup>. O'Donnell et al. reported 29 cases (1.5%) of 1995 patients who underwent hemodynamic catheterization. Of these, 26 had a left aortic arch and a left lower PV compression, and three had a right aortic arch, right-sided descending aorta, and a right lower PV compression. Nineteen of 29 patients had a single-ventricular physiology. The authors performed PV stenting in two patients who had single ventricular physiology with poor results. Other surgical interventions for relief of PV obstruction including posterior aortopexy were not performed in any of the patients <sup>5</sup>. We had a case of successful relief from PV obstruction with a posterior aortopexy in a post-Fontan patient (patient 1). Kotani et al. reported a case of successful relief from the left PV obstruction by posterior aortopexy and plication of a redundant LA wall in a patient with the complete atrioventricular septal defect and persistent left superior vena cava. The posterior aortopexy technique for patients with PV obstruction caused by compression between the LA and the anteriorly positioned DA has several advantages as mentioned earlier by Kotani et al.  $^{3}$ . First, external compression of the PV by the anteriorly positioned DA could be effectively treated. Second, the posterior aortopexy technique is a less invasive procedure as compared to the intra-cardiac repair of the PV under cardiopulmonary bypass. Third, avoiding direct surgical manipulation of the PVs could prevent post-repair PV stenosis.

Using the posterior aortopexy technique we have treated five patients with left PV obstruction between the LA and the DA. In two patients (Patient 1 and 3) with focal PV obstruction that was mainly caused by the anterior location of the DA, posterior aortopexy was effectively performed. The changes in the angle between the DA and the vertebra (Za) were 37 and 38, respectively. In the patients with LA enlargement caused by left heart volume loading (Patient 4 and 5), the effectiveness of the posterior aortopexy was somewhat unclear. In patient 4, the improvement in the left PV obstruction occurred immediately after the operation. This improvement can be attributed to either posterior aortopexy was not effectively performed (Za = -1). The improvement in the left PV obstruction observed in patient 5 was thought to be due to the reduction in the volume loading of the heart by the PDA ligation. Finally, in patient 2, a preoperative CTR was 0.66. The left PV obstruction did not improve after the posterior aortopexy. In this patient, diffuse left PV obstruction was observed. The immediate postoperative CTR was still 0.64, showing the cardiomegaly, and the postoperative PG across the left lower PV was 20 mmHg. The cause of the left PV stenosis could be an intrinsic diffuse PV stenosis.

Based on our experiences with the five patients, it is hypothesized that posterior aortopexy is an effective option for the management of the PV obstruction caused mainly by the anterior location of the DA in the patients without left heart volume loading. However, the effectiveness of posterior aortopexy is somewhat unclear in patients with severe left heart volume loading. A sudden decrease in left lung blood flow can mask the effectiveness of the posterior aortopexy.

Our current study has some clear limitations. It is a retrospective study investigating a small number of patients with heterogeneous heart diseases, and there were no definite operative indications for the procedure. Also, we measured the CTR to evaluate the volume loading and enlarged LA of the patients, but it is not enough to show them sometimes. Moreover, the definition of focal and diffuse stenosis is not clear. To overcome these limitations, we performed preoperative and postoperative CT angiography for all 5 patients.

In conclusion, posterior aortopexy could be considered as an effective treatment option for patients with left PV obstruction that is mainly caused by the anterior location of the DA without the left heart volume loading. In patients with large left heart volume loading, the posterior aortopexy could be recommendable in cases where PV obstruction persists even after removal of the volume loading.

#### Figures

Figure 1. Patient 1 showed left pulmonary vein (PV) obstruction in follow-up computed tomography (CT) angiography and cardiac catheterization. (A) The follow-up CT angiography showed left PV obstruction (arrow). (B) Cardiac catheterization showed a sharp demarcation of the left PV flow at the descending aorta border.

Figure 2. Posterior aortopexy technique. (A) Descending aorta (DA) was fixed to the posterior chest wall using Teflon pledget-buttressed polypropylene sutures. (B) The DA was relocated posteriorly after snares were tightened.

Caud = caudad; Ceph = cephalad.

Figure 3. Computed tomography (CT) angiography findings (A) before (inset, schematic drawing of the axial CT image at the level of the left pulmonary vein obstruction) and (B) after posterior aortopexy in patient 3 (preoperative (a) and postoperative (a') angle between the descending aorta and the vertebra)

DA = descending aorta; LA = left atrium; PV = pulmonary vein; V = vertebra.

Figure 4. Computed tomography angiography findings (A) before and (B) after posterior aortopexy in patient 5.

Figure 5. Computed tomography angiography findings (A) before and (B) after posterior aortopexy in patient 2 show diffuse left lower pulmonary vein stenosis (arrows)

## Tables

Table 1. Baseline characteristics of the patients

Patient no.	Diagnosis	Age (months)	Weight (kg)	Sex	Initial PVO diagnosis tool	Affected PV pattern	Peak PG across left PV (mmHg)	Concom procedu
1	PA/IVS s/p Fontan operation	131	29.5	F	CT angiog- raphy and cardiac catheterizatio	LPV, focal	4	None

Patient no.	Diagnosis	Age (months)	Weight (kg)	Sex	Initial PVO diagnosis tool	Affected PV pattern	Peak PG across left PV (mmHg)	Concor proced
2	Truncus arterio- sus s/p Rastelli operation	5.5	4.7	М	Echocardiogr	арЛуРV, diffuse	20	None
3	CoA with VSD s/p total correction	80.5	15.9	F	Echocardiogr	а <b>µЦу</b> РV, focal	9	None
4	VSD, ASD, and PHT	1	4.2	М	Echocardiogr	a <b>þlíy</b> PV, focal + diffuse	7.3	VSD closure
5	PDA, left main bronchus stenosis	2.4	5.2	F	Echocardiogr	афИуРV, focal	6.6	PDA ligation

ASD = atrial septal defect; CoA = coarctation of the aorta; CT = computed tomography; IVS = intact ventricular septum; LLPV = left lower pulmonary vein; LPA = left pulmonary artery; LPV = left pulmonary vein; PA = pulmonary arteria; PDA = patent ductus arteriosus; PG = pressure gradient; PV = pulmonary vein; s/p = status post; VSD = ventricular septal defect.

Table 2.	Postoperative	findings
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Patient no.	Postop. Ventilator care (hours)	ICU stay (days)	Hospital stay (days)	Postop. Phrenic n. palsy
1	0 (extubation in OR)	1	6	-
2	174	227	257	-
3	4	1	7	-
4	48	5	8	-
5	10	2	6	-

ICU = intensive care unit; OR = operating room.

Table 3.	Clinical	data	before	and	after	posterior	aortopexy
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	Angle between DA a vertebra Preop. (a)	and	Postoperative peak PG
Patient no.	Postop. (a')	Za	across PV (mmHg)
1	50° 13°	37	0
2	$18^{\circ} \ 0^{\circ}$	18	20

	Angle between DA and vertebra Preop. (a)		Postoperative peak PG
Patient no.	Postop. (a')	Za	across $PV (mmHg)$
3	60° 22°	38	2.5
4	31° 11°	20	5.8
5	25° 26°	-1	3.1

DA = descending a orta; LLPV = left lower pulmonary vein; LPV = left pulmonary vein; PG = pressure gradient; PV = pulmonary vein.

Patient no.	Preop. CTR	Immediate Postop. CTR	Immediate postop. PV obstruction	F/U duration (months)	Last $F/U$ CTR	PV obstruction in last F/ echocardie	
1	0.44	0.44	-	89	0.47	-	-
2	0.66	0.64	+	14	0.63	+	+
3	0.46	0.51	-	45	0.54	-	-
4	0.7	0.71	-	22	0.57	-	-
5	0.62	0.58	-	34	0.52	-	-

Table 4. Follow up data of the patients undergoing posterior aortopexy

CTR = cardiothoracic ratio; F/U = follow-up; PV = pulmonary vein.

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