# Systemic-to-pulmonary shunt operation in neonates with ductus-dependent pulmonary blood flow

*Duktusa bağımlı pulmoner akımı olan yenidoğanlarda sistemik-pulmoner şant ameliyatı*

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***Background:*** Systemic-to-pulmonary arterial (S-P) shunts are among the major strategies for palliation of cyanotic neonates with critical pulmonary stenosis or atresia. We evaluated early and mid-term results of S-P shunt operations performed in neonates.

***Methods:*** Systemic-to-pulmonary shunt operations were performed in 25 neonates (mean age 13.5±6.4 days; range 1 to 25 days) with cyanotic congenital heart disease and ductus-dependent pulmonary blood flow. The smallest neonate weighed 1500 grams. Median sternotomy was performed in all but two patients who underwent a left thoracotomy. Shunt anastomoses included the ascending aorta to the main pulmonary (n=8) or right pulmonary (n=8) arteries, or the brachycephalic artery to the right pulmonary (n=7) artery. Graft size was 3.5 mm for the ascending aorta and 4 mm for the brachycephalic artery. The mean follow-up period was 24 months (range 3 to 54 months).

***Results:*** In-hospital mortality occurred in six patients (24%) due to sudden cardiac arrest (n=2) and sepsis (n=4). Shunt-related complications developed in five patients (20%). Three sudden deaths occurred after 9, 12, 17 months, respectively. Four corrective and three palliative operations were performed. The remaining patients were in good clinical condition and were followed-up for further treatment. Emergent operation was found to be a significant predictor for in-hospital mortality ($p<0.05$). None of the risk factors were significantly associated with shunt failure. Actuarial one- and three-year survival rates were 93% and 74%, respectively.

***Conclusion:*** Systemic-pulmonary shunt operation in the neonatal period is a complex procedure, with considerable mortality and morbidity. Satisfactory mid-term palliation can be obtained with median sternotomy, but close follow-up of patients including early elective palliation or correction is mandatory due to high incidence of sudden death.

*Key words:* Anastomosis, surgical; heart defects, congenital; infant, newborn; palliative care; pulmonary artery/surgery.

***Amaç:*** Sistemik-pulmoner (S-P) şant ameliyatları kritik pulmoner stenoz veya atrezi olan siyanotik yenidoğanlarda palyasyon için en önemli stratejilerden biridir. Bu çalışmada, yenidoğanlarda uyguladığımız S-P şant ameliyatlarının erken ve orta dönem sonuçları değerlendirildi.

***Çalışma planı:*** Siyanotik doğuştan kalp hastalığı ve duktusa bağımlı pulmoner kan akımı bulunan 25 yenidoğana (ort. yaş 13.5±6.4 gün; dağılım 1-25 gün) S-P şant ameliyatı uygulandı. Olgular içinde en düşük ağırlık 1500 gr idi. İki hastaya sol torakotomi uygulanırken, diğer hastalarda median sternotomi yaklaşımına başvuruldu. Şant anastomozları çıkan aort ile ana pulmoner arter (n=8) ya da sağ pulmoner (n=8) artere ve brakiosefalik arter ile sağ pulmoner artere (n=7) yerleştirildi. Çıkan aort için 3.5 mm, brakiosefalik arter için 4 mm çapında greft kullanıldı. Ortalama takip süresi 24 ay (dağılım 3-54 ay) idi.

***Bulgular:*** Hastane mortalitesi altı olguda (%24) görüldü; ölüm nedeni iki hastada ani kardiyak arrest, dört hastada sepsis idi. Beş hastada (%20) şanta bağlı komplikasyonlar ortaya çıktı. Üç hasta, ameliyat sonrası 9, 12 ve 17. aylarda ani ölüm nedeniyle kaybedildi. Hastalara dört düzeltici, üç palyatif ameliyat uygulandı. Klinik durumları iyi olan diğer hastalar sonraki tedavileri açısından izlendi. Acil ameliyatın hastane içi mortalite için anlamlı öngördürücü olduğu görüldü ($p<0.05$). Risk faktörlerinin hiçbiri şant başarısızlığı ile ilişkili bulunmadı. Kaplan-Meier yöntemiyle hesaplanan bir ve üç yıllık sağkalım oranları sırasıyla 93% ve %74 bulundu.

***Sonuç:*** Yenidoğan döneminde yapılan S-P şant ameliyatları, önemli mortalite ve morbiditesi olan kompleks ameliyatlardır. Median sternotomi yaklaşımıyla orta dönemde başarılı palyasyon sağlanabilir; ancak, takip sırasında yüksek ani ölüm riski nedeniyle sonraki palyatif veya düzeltici ameliyatlar için hastaların yakından takibi şarttır.

*Anahtar sözcükler:* Cerrahi anastomoz; doğuştan kalp defekti; bebek, yenidoğan; palyatif bakım; pulmoner arter/cerrahi.

*Received:* March 16, 2006 *Accepted:* November 8, 2006

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Neonates with inadequate pulmonary blood flow due to congenital heart anomalies such as critical pulmonary stenosis or atresia have a poor prognosis if intervention is not performed. In these patients, pulmonary circulation is usually ductus-dependent. Mortality may occur due to hypoxia and acidosis when ductus arteriosus closes within hours or days after birth.[1] Most patients have associated cardiac anomalies for which total correction is not convenient during the neonatal period. Systemic-to-pulmonary (S-P) shunt operations between the systemic and pulmonary arteries have been shown to provide an excellent form of palliation in these patients until further interventions or corrective operations are to be performed.[1-3] In neonates, the S-P shunt operation is quite different from that performed in infants and older children due to the small diameters of the subclavian and pulmonary arteries, the presence of high pulmonary vascular resistance, and a high preoperative incidence of severe hypoxia and acidosis. The neonatal age itself (<30 days) has been recognized as an increased risk factor for postoperative mortality and shunt failure.[4,5] Preoperative management, surgical strategy, postoperative management, and graft choice are all important parameters for outcome.

In this study, we evaluated the surgical strategy and early and mid-term results of S-P shunt operations performed in 25 neonates.

## PATIENTS AND METHODS

Between February 2000 and July 2005, 107 patients with various forms of cyanotic congenital heart defects and reduced pulmonary blood flow underwent S-P shunts. Among these, 25 neonates (23.4%) aged less than 30 days (mean age 13.5±6.4 days; range 1 to 25 days) formed the study group. The smallest neonate weighed 1500 grams. Fourteen patients were males. Preoperative diagnosis are presented in Table 1. Three patients had dextrocardia. Two patients had extracardiac anomalies (imperforate anus, single polycystic kidney, and multiple anomalies) and one patient had Di George syndrome. Those who underwent a S-P shunt operation with pulmonary banding as part of a two-stage arterial switch operation for complete transposition and those who underwent the Norwood's operation for hypoplastic left heart syndrome were excluded.

**Table 1. Preoperative diagnoses of the neonates**

| Diagnoses | No |
|---|---|
| Pulmonary atresia with | |
| Tetralogy of Fallot | 9 |
| Intact ventricular septum | 5 |
| Single ventricle | 5 |
| Corrected transposition | 2 |
| Ventricular septal defect | 1 |
| Double outlet right ventricle | 1 |
| Critical pulmonary stenosis with | |
| Single ventricle | 1 |
| Transposition and ventricular septal defect | 1 |
| *Total* | 25 |

All the patients were admitted to intensive care unit with severe cyanosis and significant systemic desaturation. Efforts to provide preoperative stabilization included maintenance of adequate hydration, prevention of hypothermia, assessment of metabolic parameters, and, if any, correction of anomalies. Prostaglandin $E_1$ ($PGE_1$) infusion was administered to maintain ductal patency required for stabilization of saturation and metabolic profile, starting with initial doses of 0.05 µg/kg/min and later tapering off to the lowest possible dose. A detailed preoperative echocardiographic evaluation was carried out to assess intracardiac anatomy and the entire pulmonary artery anatomy. Other anatomic variables of interest included the side of the aortic arch, its branching pattern, the presence of a patent duct and the site of its insertion into the pulmonary arteries, and the presence of stenosis at the site of duct insertion. Preoperative cardiac catheterization was not performed to confirm the diagnosis.

In critically ill patients, mechanical ventilation and invasive monitoring (arterial, central venous lines, Foley catheter) were instituted immediately after admission. Metabolic acidosis was corrected with intravenous bicarbonate, and inotropic infusion was started when necessary. Most patients were referred to our clinic from other hospitals. Three patients arrived intubated and two patients had severe hypoxia and acidosis despite mechanical ventilation and $PGE_1$ perfusion.

***Surgical technique.*** Median sternotomy was performed in all but two patients who underwent a left thoracotomy. The thymus was resected. The pericardium was opened vertically and was suspended. The ascending aorta, brachycephalic artery, innominate vein, main pulmonary artery, branch pulmonary arteries, and ductus arteriosus were prepared and encircled. In cases where cardiopulmonary bypass (CPB) was not used, heparin (100 U/kg) was given before vascular clamping for anastomosis. Before starting the shunt anastomosis, the target pulmonary artery was clamped and $SpO_2$ was observed for three to five minutes. In case progressive desaturation and hypotension occurred, the clamp was removed and the operation was performed under normothermic CPB support. Nine patients needed CPB support during the shunt procedure, including five

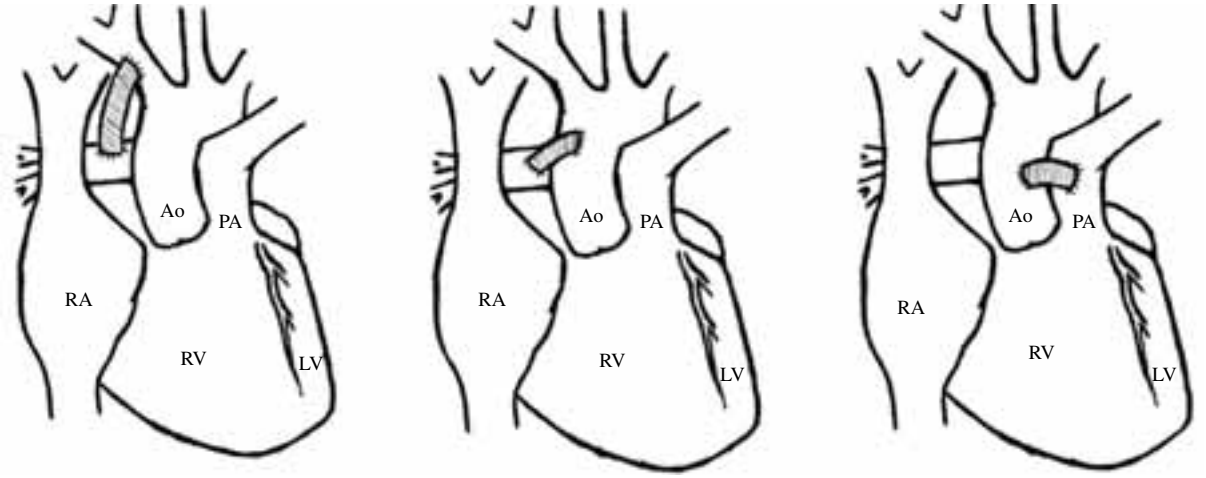


**Fig. 1.** Types of S-P shunts via median sternotomy. (Ao: Aorta; RA: Right atrium; RV: Right ventricle; LV: Left ventricle; PA: Pulmonary artery).

patients who additionally underwent atrial septectomy (n=3), and atrial septectomy and pulmonary valvotomy (n=2). Shunt anastomoses were constructed using continuous 7/0 polypropylene sutures. After removal of the vascular clamps, the ductus arteriosus was closed by a Ligaclip over nylon tape encircling the duct, so that the ductus could be easily reopened if desaturation occurred. Inspired oxygen fraction ($FiO_2$) was decreased to 30% in order to prevent excess blood flow to the lungs and to determine the saturation in room air. The S-P shunt was performed with a thin Gore-Tex PTFE tube graft (WL Gore, Elkton, MD, USA). Shunt anastomoses were performed from the ascending aorta to the main pulmonary (n=8) or right pulmonary (n=8) arteries, or from the brachycephalic artery to the right pulmonary (n=7) artery (or left in patients with dextrocardia) (Fig. 1). Whenever possible, a central shunt was preferred from the ascending aorta to the main pulmonary artery. The grafts were 3.5 mm and 4 mm in size depending of the site of the inflow, i.e. the ascending aorta and the brachycephalic artery, respectively. Where the shunt operation involved the left subclavian to the left pulmonary artery via a left thoracotomy, a 4-mm graft was used.

Postoperatively, the patients received dopamine perfusion at a dose of 5 µg/kg/min to maintain adequate systolic pressure and, after achieving hemostasis, low-dose heparin infusion (5-10 U/kg/hr) to maintain activated clotting time (ACT) at 150-160 sec for 48 hours. Whenever possible, heparin was not reversed intraoperatively in cases in which CPB support was not used. Aspirin (5 mg/kg) was started on the postoperative first day.

***Data collection and statistical analysis.*** All data were collected from hospital records. Patient families were contacted by telephone to assess the clinical status. All variables were expressed as mean ± standard deviation with 95% confidence limits (CL). Risk factors affecting hospital mortality and shunt failure were analyzed with the chi-square test and Fisher's exact test. The Kaplan-Meier method was used to estimate cumulative survival. Statistical analysis were performed with SPSS version 11.5 for Windows. A *p* value of less than 0.05 was considered significant.

## RESULTS

In-hospital mortality occurred in six patients (24%; 95% CL: 9.4 - 45.1%). Two patients died on the postoperative Day 1 and 2 as a result of sudden cardiac arrest and unsuccessful resuscitation. Exploration revealed shunt thrombosis in one patient thereof. Four patients died of sepsis and multiorgan failure, two of whom were operated in emergent conditions due to severe preoperative hypoxia and acidosis.

Shunt-related complications such as acute shunt thrombosis or insufficient pulmonary blood flow occurred in five patients (20%; 95% CL: 6.8-40.7%), resulting in death in one. Two patients underwent a subsequent shunt operation, at which the thrombosed shunt was resected and changed in one patient and, in the other patient with thoracotomy, an additional shunt was placed to the opposite site via a sternotomy in order to increase pulmonary blood flow. Two patients with acute shunt thrombosis underwent right ventricle outflow tract (RVOT) reconstruction under CPB. Severe generalized edema developed in one patient with tricuspid atresia and flap-like foramen ovale after shunt operation. Atrial septectomy was performed on the fifth postoperative day, resulting in resolution of the edema. Postoperative complications are presented in Table 2.

Emergent operation was found as a significant predictor for in-hospital mortality ($p<0.05$). None of the

**Table 2. Postoperative complications**

| | No | % |
|---|---|---|
| Prolonged mechanical ventilation (>48 hr) | 7 | 28 |
| Acute shunt thrombosis/insufficiency | 5 | 20 |
| Peritoneal dialysis | 4 | 16 |
| Pneumonia | 2 | 8 |
| Sternum being left open | 2 | 8 |
| Mediastinitis | 1 | 4 |
| Bleeding requiring revision | 1 | 4 |

risk factors analyzed were found to have a significant effect on the development of shunt failure (Table 3). Eleven patients (44%) had an uneventful postoperative course with short intensive care unit (<3 days) and hospital (<10 days) stay. The mean lengths of intensive care unit stay and hospital stay were 8.1±13.0 days (range 2 to 60 days) and 15.9±16.1 days (6 to 74 days), respectively.

***Follow-up.*** Nineteen survivors were followed-up for a mean period of 24±16.7 months (range 3 to 54 months). Periodic echocardiographic assessments were performed during follow-up to evaluate the condition of the shunt and primary cardiac lesion. When the next stage of treatment was planned, all patients were subjected to cardiac catheterization and angiography to evaluate pulmonary artery anatomy and shunt flow (Fig. 2a, b). Suitability for single- or two-ventricle repair was assessed.

Three patients died suddenly (15.8%) nine, 12, and 17 months after the operation, respectively. Their pathologies were pulmonary atresia (PA) accompanied by intact ventricular septum with right ventricle sinusoids, tetralogy of Fallot with PA, and single ventricle with PA, respectively. In one patient, the shunt was patent three months before the sudden death. No follow-up record was available for the other two patients.

Four patients (21.1%) underwent corrective operations. Two patients with pulmonary atresia and intact ventricular septum underwent biventricular repair with RVOT reconstruction and atrial septal defect closure five months and three years after the initial operation, respectively. Fenestrated extracardiac Fontan procedure was performed in two patients with single ventricle pathology in the fourth postoperative year. One patient died in another center early after Fontan operation due to low cardiac output.

Three patients (15.7%) were not found eligible for a corrective operation due to small age or poor growth of pulmonary vasculature. One patient with increasing cyanosis underwent a second S-P shunt operation for the opposite side. Two patients underwent an elective Glenn anastomosis with the aim of a staged Fontan procedure 17 and 21 months after the initial operation, respectively.

The remaining patients were in good clinical condition, waiting for another palliation or total correction.

**Table 3. Analysis of factors affecting hospital mortality and early shunt failure**

| Factors | | No | Mortality | *p* | Shunt failure | *p* |
|---|---|---|---|---|---|---|
| Diagnosis | TOF-PA* | 11 | 4 | NS | 3 | NS |
| | SV-PA | 5 | – | | – | |
| | PA-IVS | 5 | 1 | | 2 | |
| | c-TGA-PA | 2 | – | | – | |
| | TGA-VSD-PS | 1 | 1 | | – | |
| | SV-PS | 1 | – | | – | |
| CPB use | Yes | 9 | 4 | NS | 2 | NS |
| | No | 16 | 2 | | 3 | |
| Shunt size | 3.5 mm | 16 | 3 | NS | 3 | NS |
| | 4.0 mm | 9 | 3 | | 2 | |
| Shunt site | A-A | 8 | – | NS | 1 | NS |
| | A-R | 8 | 4 | 0.07 | 2 | |
| | B-R | 7 | 2 | | – | |
| | T | 2 | – | | 2 | |
| Emergent operation | Yes | 2 | 2 | <0.05 | 0 | NS |
| | No | 23 | 4 | | 5 | |

*One patient with VSD-PA and one patient with DORV-PA were included into the TOF-PA group; A-A: Ascending aorta-main pulmonary artery; A-R: Ascending aorta-right pulmonary artery; B-R: Brachycephalic artery-right pulmonary artery; c-TGA: Corrected transposition of great arteries; DORV: Double outlet right ventricle; IVS: Intact ventricular septum; PA: Pulmonary atresia; PS: Pulmonary stenosis; SV: Single ventricle; TOF: Tetralogy of Fallot; TGA: Transposition of the great arteries; T: Thoracotomy (left subclavian-left pulmonary artery); VSD: Ventricular septal defect; CPM: Cardiopulmonary bypass; NS: Not significant.

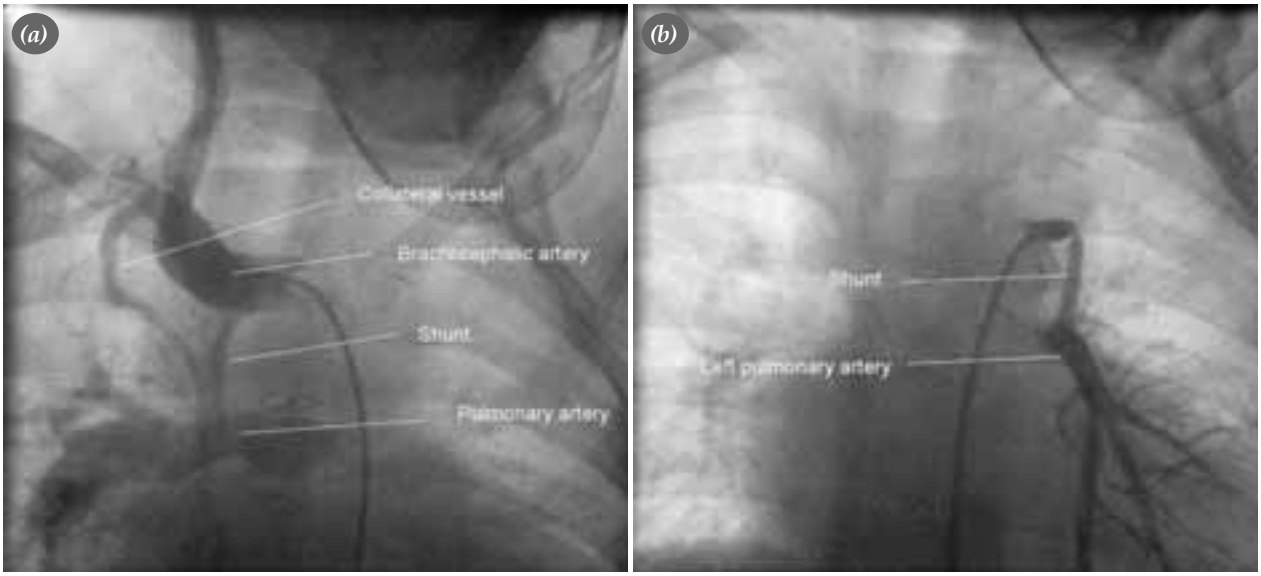


**Fig. 2.** Cardiac catheterization of the patient before fenestrated Fontan operation, who had two 4-mm shunts placed in the neonatal period (a left S-P shunt via a thoracotomy and a right S-P shunt via a sternotomy). **(a)** Right shunt injection. **(b)** Left shunt injection. Proximal stenosis of the shunt is prominent.

Actuarial survival rates at one and three years were 93% and 74%, respectively (Fig 3).

## DISCUSSION

There is a significant number of neonates presenting with cyanotic congenital heart defects that require urgent treatment even though definitive repair may not be possible. Many die when ductus arteriosus closes which may be the only source for pulmonary blood flow.[1] The use of $PGE_1$ has dramatically changed the natural history of these patients, making it possible to transfer critically ill babies to a tertiary center in a more stable clinical condition.[2,3] However, in developing countries such as Turkey, this is still a problem and is directly related with poor outcome. Despite some encouraging results showing a mortality rate of 3.3% following S-P shunts in neonates,[6] mortality after S-P shunt operations is still high in many centers. Tamisier et al.[7] reported early deaths and shunt failure in 21% of patients who were less than three months of age. According to the EACTS (European Association for Cardio-Thoracic Surgery) congenital database report, cumulative hospital mortality of neonatal S-P shunt operations is 8.8%.[8] Mortality of central shunt operations is 14.4% and mortality of other types of neonatal shunt operations is 24.1%. These mortality rates are higher than those of more complex procedures including arterial switch operation (7.7%), repair of total pulmonary venous return (11.3%), and ventricular septal defect closure (1.6%).[8]

The results of shunt operations in infants and children are very different from those performed in neonates. Compared to 24% in this series of patients, hospital mortality was found as 1.2% (1/82) in patients older than one month of age, who were operated on in our center during the same period. Surgical approach was usually thoracotomy and the need for CPB support was exceptional.

Surgical approach and graft choice in neonates differ greatly among centers. Conventional approach has been thoracotomy incision, but median sternotomy offers additional advantages, enabling greater control of the vessels without the risk for lung compression and associated respiratory compromise.[2,9] Cardiopulmonary bypass support can be readily instituted whenever necessary. There are more choices for inflow and outflow routes, namely, the subclavian and brachycephalic arteries or the ascending aorta for inflow, and the right, left, or main pulmonary arteries for outflow shunt anasto-

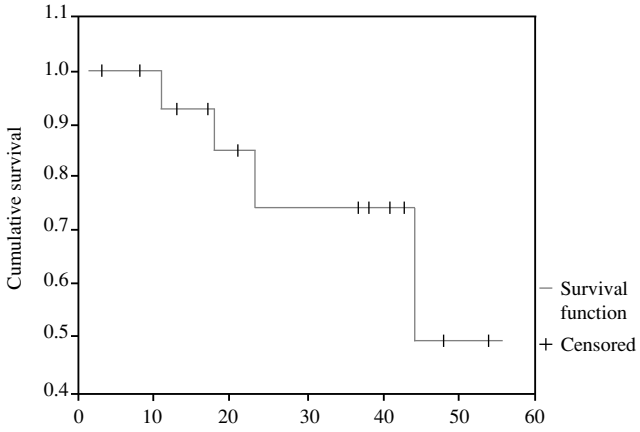


**Fig. 3.** Actuarial survival of the patients.

moses. Surgical strategy may be changed intraoperatively in case of small target vessels or an arcus anomaly. Other advantages of median sternotomy are as follows: Placement of distal anastomosis to the main pulmonary artery or to branch pulmonary artery closer to bifurcation allows an even distribution of blood to both lungs, which is highly desirable.[10] Repair of pulmonary artery during bidirectional Glenn operation is easy, because shunt anastomosis is in the same location for superior vena cava to right pulmonary artery anastomosis. Preferential flow to one or the other lung and unequal growth of pulmonary arteries have been observed in shunts constructed through the thoracotomy approach. Repair of pulmonary artery distortion when needed may be very troublesome in these cases.

In our series, there were two cases in which thoracotomy was performed. Both patients required emergent reoperation due to shunt-related problems in the early postoperative period. Our routine approach has been median sternotomy in neonates for the last four years.

Similar to other reports,[2,3,7] acute shunt thrombosis was the major cause of mortality and morbidity in our patients. High pulmonary vascular resistance and small vessels were responsible for this complication. Another important concern was the development of pulmonary edema and heart failure due to high pulmonary blood flow, but these were not encountered. Inflow vessel diameter and shunt size are important determinants of shunt flow. Gold et al.[11] reviewed their results in 112 shunts with 4-mm or 5-mm grafts in 92 neonates and reported a 21% incidence of congestive heart failure secondary to increased blood flow. We use a 4-mm graft when the brachycephalic artery is the inflow, and a 3.5-mm graft when the ascending aorta is the inflow. Based on our experience, this strategy is suitable for maintaining satisfactory arterial saturation even in the mid-term follow-up, without any risk for pulmonary overflow. Besides graft and inflow vessel choices, early postoperative respiratory management is also very important for shunt flow. Lowering $FiO_2$ and increasing arterial $PCO_2$ levels during mechanical ventilation contribute to increased pulmonary vascular resistance and decreased shunt flow, or vice versa. Therefore, shunt flow should be balanced based on arterial $SpO_2$, pulmonary oscultation, and chest X-ray findings. Some medications may also be used for this purpose. Nitric oxide and $PGE_1$ have been used with success by some authors.[12]

Right ventricle outflow tract reconstruction may be an alternative to shunt operation in selected cases. It should be kept in mind in case of shunt failure and may be a life-saving procedure as in two patients in this series.

In this retrospective analysis, intermediate-term palliation seems satisfactory, but high incidence of sudden death is disappointing. Shunt flow is the only source for pulmonary blood flow in most of the patients. High hemoglobin levels, fever, dehydratation, or cessation of aspirin may cause acute shunt thrombosis and sudden death in these patients.

In conclusion, S-P shunt operation in the neonatal period is a complex procedure, with considerable mortality and morbidity. Preoperative hemodynamic and metabolic stabilization, $PGE_1$ infusion, surgical approach, and postoperative management are all critical factors for successful results. Median sternotomy approach might be the procedure of choice. Satisfactory intermediate-term palliation can be obtained with this technique, without the risk for pulmonary overflow. All patients should be followed-up closely and, due to high incidence of sudden death during follow-up, early elective palliation or correction is recommended.

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