Stenting of the Arterial Duct and Banding of the Pulmonary Arteries
Basis for Combined Norwood Stage I and II Repair in Hypoplastic Left Heart

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Background—Outcome of patients with hypoplastic left heart (HLH) is mainly influenced by the successful first-step palliation according to the Norwood procedure. An alternative approach is heart transplantation (HTX). The feasibility of ductal stenting in newborns with duct-dependent systemic blood flow and bilateral pulmonary artery banding has been reported. But it remains to be elucidated whether this approach allows a new strategy for patients with HLH.

Methods and Results—In patients with various forms of HLH (n=11) and prostaglandin E-1 administration, ductal stenting was performed with balloon expandable Jo stents or Saxx stents. Bilateral pulmonary artery banding was surgically accomplished 1 to 3 days after the transcatheter procedure. Unrestricted blood flow through the interatrial septum was secured by balloon dilatation atrial septotomy, as required. Interventional procedures were performed with no mortality. Stent and ductal patency were achieved for up to 331 days. Two patients underwent HTX, and 8 patients had a palliative 1-stage procedure with reconstruction of the aortic arch and bidirectional cavopulmonary connection at the age of 3.5 to 6 months. There were 2 deaths. One patient with preoperative right heart failure died after the reconstructive surgery, and 1 patient died 4 months after ductal stenting and bilateral banding awaiting HTX.

Conclusions—The present study is the first clinical trial showing that stenting the duct followed by bilateral pulmonary artery banding in newborns with HLH allows the combination of neoaortic reconstruction, which is part of first-stage palliation of HLH, with the establishment of a bidirectional cavopulmonary connection. Additionally, it allows the chance for HTX after extended waiting periods. (Circulation. 2002;105:1099-1103.)

Key Words: ductus arteriosus, patent ■ stents ■ pediatrics ■ heart defects, congenital

Newborns with congenital heart disease and duct-dependent systemic blood flow are set on prostaglandin E-1 (PGE-1) to reopen the constricting ductus arteriosus (DA). In some patients, particularly those with cardiogenic shock, pharmacological treatment may be ineffective. In these patients, patency of the DA can be achieved by stent implantation.1,2 The limited prognosis of patients with hypoplastic left heart (HLH) is caused by a still high mortality during stage I of the Norwood procedures.3 Additionally, a significant number of patients die in the period between the first and second step of the staged procedure.4,5 Patients with HLH selected for heart transplantation (HTX) have to be withdrawn from the waiting list because of high pulmonary artery resistance mostly fixed after 6 months of life.

Attempts of maintaining ductal patency by placement of balloon-expandable stents started in animal studies in 1991,6 and the experience was transferred to human congenital heart disease with duct-dependent pulmonary circulation.7 Recently, reports of stenting the DA in HLH1,2 before heart transplantation proved applicability in patients with right to left shunting for systemic circulation.

Stenting of the DA combined with banding of the pulmonary arteries and atrial septectomy or septostomy as a new approach to palliation for HLH was reported for the first time by Gibbs et al.8 This technically feasible approach offered the hope for an effective palliation for the HLH. However, the question was raised whether this concept is a reasonable alternative for those patients. The present study demonstrates in a series of patients that ductal stenting and bilateral pulmonary banding allow a new surgical strategy in newborns with HLH.

Methods

Patients
Between May 1998 and December 2000, 11 newborns with weight ranging from 2.6 to 3.8 kg were included. Ten had typical HLH, 1

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accompanied by extreme coronary fistula. One patient had aortic valve atresia with hypoplasia of the aortic arch in corrected transposition and double-inlet left ventricle. Prenatally unknown diagnosis was made by standard ECG in all. At the time of admission, all were on conventional PGE-1 therapy. Compassionate therapy, palliative reconstructive surgery, or HTX were offered as principal therapeutic options, as is standard in our institution. F If parents opt for surgical treatment, we accept the family’s preference for transplant versus reconstructive surgery, because from the medical point of view, either approach is applicable. Continued impairment of right ventricular function, severe tricuspid regurgitation, or coronary fistulae leaves HTX as the treatment of choice. The postnatally performed Norwood 1 procedure is offered at our institution on an individual basis, with preference to patients with single dominant left ventricular anatomy. Additionally, palliation with the presented combination of interventional and surgical procedures was supplied after principal approval of the institutional review board of the university hospital. All parents opted for an attempt at palliation, and fully informed consent was obtained.

**Cardiac Catheterization and Stent Placement**

Heart catheterization was performed in 2 patients under general anesthesia and in 9 patients with local anesthesia and standard analgesia and sedation. The left femoral artery and the right femoral vein were catheterized with 4-F and 5-F open-tip balloon or cobra catheters, respectively. Pulmonary angiography in lateral projection was used to delineate the ductal anatomy. Ductal length ranged from 12 to 22 mm (median, 17 mm), and the diameters were 5 to 8 mm (median, 7 mm) at the aortic end and 3 to 7 mm (median, 6 mm) at the pulmonary artery end. The aortic catheter was advanced to the level of the duct for use as a landmark. We used balloon-expandable peripheral Jo stents (JoMed) or Saxx stents (Devon) with a length of 12, 17, or 20 mm, respectively. The stents were mounted on 4-F or 5-F 20-mm-long compliant balloon catheters (Osypka). The final balloon diameter ranged from 7 to 9 mm. The hand-crimped stents were advanced transvenously over a stiff guidewire (0.018 inch) into the DA, guided by landmarks or hand injections of contrast through an aortic catheter via pulmonary artery route without guidance of a long sheath. Satisfactory stent position was assessed by angiography (Figure 1), and PGE-1 infusion was discontinued. In 3 patients, ductal obstruction.

Figure 1. Ductal stenting in 1 patient (lateral view fluoroscopy). A, Feature of the DA for measurement of ductal dimensions. B, Location of the balloon-stent unit within the DA before expansion of the transvenously advanced stent. C, Inflated balloon with a fully expanded 17-mm Jo stent. D, Final angiography to demonstrate patency of the DA and exclusion of any site of obstruction.

length necessitated the use of 2 stents (12- and 17-mm Jo stents, respectively). The second stent was delivered by telescopic placement, tailoring the overall stent length to the individual ductal anatomy. Balloon dilation atrial septotomy was performed in 5 patients who did not have unrestricted blood flow through a large atrial septal defect. Balloon catheters with final diameters of 10 or 12 mm were used. Heparin 100 U/kg was given as a single dose after stent placement. Anticoagulation was continued with 300 U/kg per day in addition to a prophylactic antibiotic treatment (cefuroxime) both for 24 hours. Before discharge home, 8 to 21 days after the interventional surgical approach in all patients, hemodynamics were reevaluated by additional catheterization. Reevaluation of their clinical course, echocardiography, and ECG were performed every 1 to 2 weeks as outpatients. Echocardiographic findings of restrictive blood flow through the atrial septal defect or an excessively banded pulmonary artery were indications for balloon dilation of the interatrial communication or the banded pulmonary artery branch, respectively. Preoperative cardiac catheterization was done routinely 3 to 5 months after the initial interventional surgical approach (Figure 2) before reconstructive surgery was performed. Patients scheduled for HTX were not routinely evaluated by heart catheterization. Except in 1 patient, the final decision in favor of reconstructive surgery was based on the clinical, echocardiographic, and angiographic reevaluation.

**Surgical Techniques**

One to 3 days after the interventional procedures, bilateral banding of the pulmonary arteries was accomplished to reduce the systolic pulmonary artery pressure on both sides to less than half of the systemic blood pressure, decreasing the oxygen saturation (SaO2) to a range of 80±5% at room air and to obtain a systolic-diastolic flow pattern with a maximum velocity of about 4 m/sec on CW-Doppler. For this purpose, via a median sternotomy, the proximal right and left pulmonary arteries were banded while the distal pulmonary blood pressure was monitored by a 2.5-F catheter placed through the pulmonary artery trunk. Each band of Dacron mesh material was held in place by 2 Prolene 6.0 sutures.

Aortic arch reconstruction and bidirectional cavopulmonary connection (BCPC) combined in 1 cardiopulmonary bypass (CPB) procedure were performed 3.5 to 6 months later through a median sternotomy. CPB was established by cannulation of the main pulmonary artery as well as the superior and inferior vena cava,
respectively. The main right and left pulmonary artery branches were de-banded, but they immediately occluded. Deep hypothermia with circulatory arrest (average time, 62±13 minutes) was used for neo-aortic reconstruction. Arterial cannula was then removed from the heart. The main pulmonary artery was transected just proximal to the bifurcation. Additionally, the stented DA was transected at the pulmonary end. The insertion of the duct and the pulmonary arteries, opened from the superior vena cava to the left upper lobe branch, were patched with autologous pericardium. The descending thoracic aorta was extensively mobilized. All ductal tissue, including the stents within the DA, was excised. The aortic arch was opened inferiorly back down into the ascending aorta to the level of the right common arterial trunk in the first 4 patients, in whom a complete tube of arterial allograft was placed from the proximal divided main pulmonary artery to the arch and proximal descending aorta. In the following patients, the arterial wall allograft was used to supplement the Anastomosis between the proximal divided main pulmonary artery and the ascending aorta, aortic arch, and proximal descending aorta. The procedure was completed by an atrial septectomy. During rewarming, BCPC was performed and the superior vena cava was connected to the superior aspect of the right pulmonary artery. In 1 patient, the left pulmonary artery branch was intraoperatively stented using a 17-mm Jo stent mounted on a 6×20-mm balloon catheter. Alternatively, patients received HTX. The technical details have been described previously.10

Postoperative Management
After 1-stage combined reconstruction, infants were returned to the pediatric intensive care unit with open sternotomy on a fentanyl infusion and neuromuscular blockade. They were kept on volume control ventilation at a tidal volume of about 20 mL/kg and a positive end-expiratory pressure of 4 cm H2O. The breathing rate was adjusted to a well-balanced Paco2. Considering our perioperative management of pulmonary hypertension after cardiac transplantation in children,11 NO was continuously inhaled by all patients, the concentrations ranging from 8 to 25 ppm. Additionally, intravenously infused vasodilators, inotropic support, and volume as well as diuretics were given as needed.

Results
In all 11 patients with various forms of HLH, stent implantation in the DA was successful, and bilateral pulmonary artery banding was effective. Eight patients received palliative surgery with reconstruction of the aortic arch and BCPC in 1 operation at the age of 3.5 to 6 months. Two patients underwent HTX, 66 and 331 days after duct stenting and pulmonary artery banding, respectively. One patient, chosen for HTX because of postnatal multiorgan failure, died because of continued compromised right ventricular function 4 months after ductal stenting and bilateral banding. Patient characteristics at the time of surgery are summarized in the Table. The average preoperative mean pulmonary artery pressures distal to the banding ranged between 10 and 19 mm Hg (median, 14 mm Hg), and right ventricular end-diastolic pressure ranged between 3 and 15 mm Hg (median, 9 mm Hg).

Percutaneous procedures were performed with no mortality. Cardiopulmonary resuscitation was necessary in 1 patient. Two patients needed epinephrine because of transient bradycardia while advancing the stents. With a mean stent to duct ratio of 1.44±0.27, a single procedure was effective. Redilation was necessary in 1 patient, but for covering the DA completely, a second stent was necessary in 3 patients. Stent and ductal patency were achieved for almost 1 year. Pulmonary artery branches surgically banded to a diameter of 2 to 3 mm achieved mean pulmonary artery pressures ranging between 12 to 21 mm Hg, depending on the actual blood pressure in the pulmonary trunk. In 2 patients, the banded right pulmonary artery narrowed to <2 mm was successfully dilated with a 3-mm balloon catheter. Three patients developed significant cyanosis because of interatrial flow restriction at the age of 6 weeks to 3.5 months, which was resolved by balloon dilatation atrial septotomy. One patient aged 3 months developed low cardiac output at home attributable to a stenosis at the aortic part of the DA not covered by the stent. Emergency balloon dilatation was performed, but the patient remained in right heart failure. Parents refused HTX for religious reasons, so reconstructive surgery was performed as a rescue approach, but the infant died because of right heart failure.

Surgical removal of the ductal stent was uneventful in all patients, but 1 developed transient diaphragmatic paralysis. Reconstruction of the aortic arch by placement of an allograft tube was associated with significant postoperative residual, such as significant stenosis of the left pulmonary artery and distal arch obstruction. Pulmonary artery stenosis was observed in 1 patient intraoperatively and in 2 patients during the early postoperative period. Stent implantation was effective in all patients. In 3 of the patients, neo-aortic arch obstruction was observed at midterm follow-up, also effectively managed by balloon dilatation and stent placement, respectively (Table). The BCPC performed at the site of the

<table>
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<tr>
<th>Patient</th>
<th>Diagnosis</th>
<th>Age, d</th>
<th>Weight, kg</th>
<th>SaO2, %</th>
<th>Outcome</th>
<th>Additional Postoperative Procedures</th>
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<td>8</td>
<td>L-TGA/AA</td>
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<td>5.3</td>
<td>84</td>
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MS indicates mitral stenosis; AS, aortic stenosis; MA, mitral atresia; AA, aortic atresia; L-TGA, corrected transposition of the great arteries; LPA, left pulmonary artery; CoA, coarctation of the aorta; Pre, preoperatively; Post-A, 6 h postoperatively; and Post-B, at discharge.
previous right pulmonary artery banding was uneventful, and no patient had pleural effusions or excessive chest tube drainage for >5 days after surgery. Postoperative oxygenation with an mean arterial oxygen saturation of 68% on arrival in the intensive care unit improved over the first 48 hours and to 81% at time of extubation. This level of oxygen saturation remained unchanged through hospital discharge. Total hospital stay was in median 17 days after PA banding and 23 days after the combined Norwood operation.

The overall survival rate is now 82%. In clinical follow-up of 6 months to almost 3 years, all of the patients except 1, who was postnatally in severe cardiogenic shock, showed no major neurological complications. However, no patient has continued to Fontan completion yet.

**Discussion**

The present preliminary experience demonstrates in a series of 11 patients that the approach of ductal stenting and bilateral pulmonary artery banding allows a new surgical strategy of combining neoaoartic reconstruction, which is part of first-stage palliation of HLH, together with BCPC in a single operation with cardiopulmonary bypass beyond the neonatal period as well as HTX after an extended waiting period. The decision concerning the individual strategy in patients with HLH has changed with increasing experience in making prenatal and postnatal diagnosis as well as in surgical palliations and is now based on medical and social considerations. In Giessen, Germany, we started a pediatric HTX program in 1988, and since 1993 we have additionally offered Norwood operations at our institution. In 1998, we introduced the interventional surgical procedure, described here, as we gained more experience in ductal stenting in various forms of complex heart malformations. In our experience with highly flexible stents and an altered technique, ductal stenting can be recommended in patients with duct-dependent systemic circulation, such as those with HLH, particularly in patients with restrictive DA not responding to conventional PGE-1 therapy. Furthermore, this type of stenting is contemplated in centers with a successful neonatal transplant program where potential recipients must await a matching donor heart, as at our institution.

As a matter of principle, patients being considered for univentricular palliation with Norwood’s staged procedures do not need a ductal stent, because the sooner those patients undergo surgical reconstruction, the better their outcomes. However, despite substantial improvements of the Norwood operation and its modifications as well as intensive care management, patients with HLH remain at significant risk of surgical or nonsurgical mortality while awaiting additional steps of the staged Norwood procedure. In addition, neurodevelopmental outcome is extensively influenced by the preoperative course as well as intraoperative complications during the Norwood I procedure.

We believe that delayed surgery beyond the neonatal period might have many advantages. The use of early BCPC on an elective basis with relief of volume load for the single right ventricle by establishing serial instead of parallel pulmonary and systemic circulations seems to be most advantageous after aortic reconstruction with hypothermic cardiac arrest. Although a potential obstacle to a successful BCPC in the young infant might be pulmonary vascular resistance, our strategy for overcoming elevated pulmonary vascular resistance was highly successful. It seems possible to perform the BCPC even closer to the neonatal period. Encouraging early results, particularly in infants with HLH, were described. In addition, the surgical technique of reconstructing the aortic arch in HLH is easier to perform in infants 3 to 6 months of age who weigh at least 5 to 6 kg rather than immediately after birth. However, the technique used in the first 3 patients of our series, with placement of an allograft tube from the proximal divided main pulmonary artery to the arch and proximal descending aorta, was associated with the development of postoperative coarctation of the aorta as well as extrinsic pulmonary artery stenosis of the left main branch. Nevertheless, removal of the stented DA does not seem to have provoked recoarctation until now. Both major complications are strongly associated with the surgical procedure. But these well-known surgical complications can be effectively addressed by balloon angioplasty and stent implantation. Extended augmentation of the aortic arch and patch augmentation of the pulmonary arteries just proximal to the left and right bifurcation solved the problem of coarctation at both sides in the last 4 of our patients, implying a learning curve for the surgeons as well, which should not be judged as an argument against that new approach.

Prenatal diagnosis of HLH is associated with improved survival after first-stage palliation in comparison with patients diagnosed after birth. In this context, an additional advantage of the presented strategy seems to be relevant in patients with HLH suffering postnatal cardiogenic shock, because it offers a chance of complete recovery from compromised heart function or multiorgan failure. The decision for additional surgical or nonsurgical treatment can be made by physicians and parents without the pressure of time. In the same period of the present study, in 4 neonates, 3 of them with preferential single left ventricle anatomy, the modified Norwood I procedure was carried out successfully, and 5 other patients with HLH admitted to our institution died after compassionate care. The parents refused either surgical treatment after our comprehensive information concerning short- and long-term risks of each surgical strategy. However, the long-term implication of the described approach has to be elucidated in additional studies. A significant number of patients who are not suitable for reconstructive surgery and whose parents opt for transplantation die awaiting a donor heart for reasons of heart failure or increasing pulmonary artery resistance. Stent implantation in the DA provides the possibility of long-term ductal patency. At the same time, protection of lung vasculature from high pressure by bilateral pulmonary artery banding with low resistance enables transplantation as long as 11 months later. Patients undergoing this procedure may be followed as outpatients, as was possible in 9 patients who were discharged until the combined Norwood I and II stage or HTX was performed. Close follow-up is necessary to detect restrictive interstitial communication, narrowing of the duct, or compromised pulmonary blood flow early. In addition, the development of a significant aortic coarctation with compromised retrograde blood flow has to
be detected early. Meanwhile, in our cohort of 46 patients with duct-dependent systemic blood flow in whom the DA was stented, this severe problem (not shown here) was observed twice, making stent placement in the narrowed isthmus necessary. Altogether, maintenance of ductal patency by stent implantation is attractive, but in practice, quality of palliation requires much experience in interventional pediatric cardiology. Disappointing results, as reported by Gibbs et al., may be attributable in part to the strategy used, procedural difficulties, and, not least of all, technical differences such as stent design. Particularly, the surgical septostomy of the interatrial septum using cardiopulmonary bypass is likely to depress heart function excessively. Alternatively, balloon septotomy of the interatrial septum is in our opinion very effective to dilate the interatrial septum and can be repeated or extended by stent placement if necessary.

**Conclusion**

In patients with HLH, stent implantation in the DA and subsequent bilateral pulmonary artery banding provide the opportunity of a combined first and second stage of the Norwood procedure beyond the neonatal period or for delayed HTX. In the case of the reconstructive strategy, I palliative procedure with the necessity of cardiopulmonary bypass can be avoided. Additionally, early bidirectional cavopulmonary shunt on an elective basis in young infants may reduce the deleterious sequelae of long-term venous volume overload and chronic hypoxemia. If this strategy reduces overall mortality and morbidity with improved neurological outcome, then it warrants additional investigation.

**References**


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