CONGENITAL HEART DISEASE

Stenting of the ductus arteriosus and banding of the pulmonary arteries: basis for various surgical strategies in newborns with multiple left heart obstructive lesions

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Objective: To present an institutional experience with stent placement in the arterial duct combined with bilateral banding of the pulmonary artery branches as a basis for various surgical strategies in newborns with hypoplastic left heart obstructive lesions.

Design: Observational study.

Setting: Paediatric heart centre in a university hospital.

Patients: 20 newborns with various forms of left heart obstructive lesions and duct dependent systemic blood flow.

Interventions: Patients underwent percutaneous ductal stenting and surgical bilateral pulmonary artery banding. Atrial septotomy by balloon dilatation was performed as required, in one premature baby by the transhepatic approach.

Main outcome measures: Survival; numbers of and reasons for palliative and corrective cardiac surgery.

Results: One patient died immediately after percutaneous ductal stenting. One patient died in connection with the surgical approach of bilateral pulmonary banding. Stent and ductal patency were achieved for up to 331 days. Two patients underwent heart transplantation and two patients died on the waiting list. Ten patients had a palliative one stage procedure with reconstruction of the aortic arch and bidirectional cavopulmonary connection at the age of 3.5–6 months. There was one death. One patient is still awaiting this approach. Two patients received biventricular repair. In one, biventricular repair will soon be provided.

Conclusions: Stenting the arterial duct combined with bilateral pulmonary artery banding in newborns with hypoplastic left heart or multiple left heart obstructive lesions allows a broad variation of surgical strategies depending on morphological findings, postnatal clinical conditions, and potential ventricular growth.

METHODS

Patients

Between May 1998 and March 2002, 20 newborns with a birth weight ranging from 1.6–3.7 kg were studied. Sixteen had typical hypoplastic left heart. One of these had a residual antegrade blood flow through a moderate hypoplastic aortic valve annulus with a diameter of 4 mm, associated with mild mitral valve hypoplasia. Two patients had aortic valve atresia with severe hypoplasia of the aortic arch, one of them with corrected transposition, and double inlet left ventricle. Two patients had interrupted aortic arch with moderate to large ventricular septal defects and hypoplastic aortic valve annulus (< 4.5 mm), and in one with bicommissural aortic valve (table 1). Non-cardiac diagnoses were renal tumour in one and CATCH 22 syndrome (cardiac defects, abnormal facial features, thymic hypoplasia, cleft palate, and hypocalcaemia) in another premature infant. Diagnoses were unknown.
Table 1: Patient data at the time of ductal stenting and postnatal variables

<table>
<thead>
<tr>
<th>Patient</th>
<th>Diagnosis</th>
<th>Age (days)</th>
<th>Weight (kg)</th>
<th>Stents used</th>
<th>Outcome</th>
<th>Additional postoperative procedures</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>MS/AS</td>
<td>10</td>
<td>3.8</td>
<td>7 × 17 mm Jo-Med + 6 × 17 mm Jo-Med</td>
<td>Combined Norwood 1 and 2 repair</td>
<td>168 days after ductal stenting Stent placement, coarctation of the aorta</td>
</tr>
<tr>
<td>2</td>
<td>MS/AS</td>
<td>7</td>
<td>3.2</td>
<td>7 × 17 mm Jo-Med</td>
<td>Combined Norwood 1 and 2 repair</td>
<td>135 days after ductal stenting Stent placement left pulmonary artery</td>
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<td>3</td>
<td>MA/AA</td>
<td>9</td>
<td>3.3</td>
<td>8 × 17 mm Jo-Med</td>
<td>Died &lt;24 hour after combined Norwood 1 and 2 repair</td>
<td>108 days after ductal stenting None</td>
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<tr>
<td>4</td>
<td>MS/AS</td>
<td>22</td>
<td>3.4</td>
<td>7 × 17 mm Jo-Med</td>
<td>Combined Norwood 1 and 2 repair</td>
<td>87 days after ductal stenting Stent placement left pulmonary artery, coarctation of the aorta</td>
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<tr>
<td>5</td>
<td>MS/AA</td>
<td>6</td>
<td>2.9</td>
<td>7 × 17 mm Jo-Med</td>
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<td>111 days after ductal stenting None</td>
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<td>6</td>
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<td>4</td>
<td>3.5</td>
<td>7 × 20 mm Saxx</td>
<td>Combined Norwood 1 and 2 repair</td>
<td>174 days after ductal stenting None</td>
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<td>3.6</td>
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<td>146 days after ductal stenting None</td>
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<td>54</td>
<td>3.0</td>
<td>7 × 18 mm Omnilink</td>
<td>Combined Norwood 1 and 2 repair</td>
<td>89 days after ductal stenting Stent placement superior vena cava pulmonary artery connection, coil</td>
</tr>
<tr>
<td>9</td>
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<td>27</td>
<td>2.9</td>
<td>7 × 13 mm Saxx + 7 × 13 mm Saxx</td>
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<td>168 days after ductal stenting None</td>
</tr>
<tr>
<td>10</td>
<td>AA/MA</td>
<td>91</td>
<td>2.3</td>
<td>7 × 18 mm Omnilink</td>
<td>Died after catheterisation</td>
<td>NA</td>
</tr>
<tr>
<td>11</td>
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<td>7 × 17 mm Jo-Med</td>
<td>Heart transplantation</td>
<td>57 days after ductal stenting None</td>
</tr>
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<td>3.2</td>
<td>7 × 17 mm Jo-Med</td>
<td>Heart transplantation</td>
<td>57 days after ductal stenting None</td>
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<tr>
<td>13</td>
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<td>Heart transplantation</td>
<td>62 days after ductal stenting None</td>
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<td>7 × 17 mm Jo-Med</td>
<td>Heart transplantation</td>
<td>193 days after ductal stenting None</td>
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<td>15</td>
<td>IAA/VSD</td>
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<td>3.7</td>
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<td>Waiting for combined Norwood 1 and 2 repair</td>
<td>NA</td>
</tr>
<tr>
<td>16</td>
<td>HLH/MS</td>
<td>13</td>
<td>3.4</td>
<td>7 × 17 mm Jo-Med</td>
<td>Waiting for biventricular repair</td>
<td>NA</td>
</tr>
</tbody>
</table>

Figure 1: Ductal stenting in patient 9 (lateral view fluoroscopy).
(A) Feature of the patent ductus arteriosus (DA) for measurement of ductal dimensions. Angiography is performed through the long sheath. (B) Location of the balloon stent unit not yet exactly positioned within the DA. (C) Angiography by the aortic 4 French cobra catheter. Inflated balloon with a fully expanded 18 mm Omnilink stent. The aortic catheter positioned in the descending part of the aortic arch was used as a landmark. (D) Final angiography showing patency of the DA and excluding any site of obstruction.
In addition to the previously described surgical techniques, surgical techniques with continuous administration of PGE1 (two patients), to 1–3 days afterwards (15 patients), to before varied from the same day of the catheterisation procedure as described by Shim and colleagues. Balloon catheters with final diameters of 10 or 12 mm were used. In one, the transhepatic liver parenchyma peripheral to the hepatic vein. Dilatation. Bottom right, steel coil inserted through the sheath into the low right atrium guided by a previously placed wire. Bottom left (lateral view fluoroscopy), inflated balloon catheter placed in the restricted patent foramen ovale necessitates further gradual balloon dilatation. Bottom right, steel coil inserted through the sheath into the liver parenchyma peripheral to the hepatic vein.

Surgical techniques
In addition to the previously described surgical techniques, the timing of the bilateral banding of the pulmonary arteries varied from the same day of the catheterisation procedure (two patients), to 1–3 days afterwards (15 patients), to before the intervention with continuous administration of PGE1 (three patients). The Dacron mesh material of the banding was held in place by two prolene 6.0 sutures to avoid migration of the banding. Furthermore, in infants with classic hypoplastic left heart syndrome or with aortic atresia, in whom arch reconstruction and BCPC combined in one cardiopulmonary bypass procedure were performed 3.5–6 months later, complete circulatory arrest was avoided by means of cerebral perfusion through innominate artery cannulation. Pump flow was maintained at 90–110 ml/min, despite deep hypothermia. Additionally, the management during weaning and the early post-bypass period was changed by introducing the vasodilators phentolamine and nitroprusside in our postoperative strategy. In patients with biventricular repair, reconstruction of the interrupted aortic arch was combined with ventricular septal defect repair following bilateral debanding of the pulmonary arteries and removal of the stented ductus arteriosus.

Technical details of transplantation procedure in patients chosen for heart transplantation have been described previously.

RESULTS
Stenting the duct combined with bilateral pulmonary artery banding in newborns with hypoplastic left heart or multiple left heart obstructive lesions allowed various surgical strategies in 20 patients (fig 3).

Table 1 summarises the patients’ characteristics at the time of ductal stenting.

Percutaneous procedures were performed with a mean procedural time of 95 minutes and a mean fluoroscopy time of 21 minutes. There was one death: patient 12 (table 1) died minutes after the ductal stent was placed and the interventional
procedure was already finished. Acute ischaemia led to cardiac arrest not responding to medical or mechanical resuscitation, although the stented duct allowed reasonable systolic arterial pressure generation by chest compression. In this premature infant the ascending aorta and root were extremely diminutive, with a diameter < 1 mm at the site of the innominate artery with a diameter of the inner lumen of < 1 mm.

Figure 4 The necropsied specimen. The opened aortic arch shows a diminutive orifice of the ascending aorta at the level of the innominate artery and a diameter of < 1 mm. The opened aortic arch shows a diminutive orifice of the ascending aorta at the level of the innominate artery and a diameter of < 1 mm.

Cardiopulmonary resuscitation was necessary in one further patient. Two patients needed adrenaline (epinephrine) because of transient bradycardia while the stents were advanced through the right heart. Single stent placement was effective in 15 patients. Stent redilatation was necessary in two patients and a second stent was necessary in five patients to cover the ductus arteriosus completely. Stent and ductal patency were achieved for almost one year. The use of premounted stents advanced through a 5 French Terumo sheath increased safety and feasibility of the interventional procedure. The 5 French long sheath induced neither relevant tricuspid valve nor pulmonary valve regurgitation.

Bilateral pulmonary artery banding was accomplished surgically after the transcatheter procedure in 17 patients and before ductal stenting in three patients. The technical details have been described previously. Two patients needed bilateral banding of the pulmonary arteries immediately because of excessive pulmonary run off, on the same day following the interventional procedures—that is, after atrial septotomy. One patient died a few hours after the surgical bilateral pulmonary banding as a result of stent-duct obstruction that was not immediately recognised. Pulmonary artery branches surgically banded to a diameter of about 2–3 mm achieved mean pulmonary artery pressures ranging between 12–27 mm Hg depending on the actual blood pressure in the pulmonary trunk. In two patients, the banded right pulmonary artery narrowed to less than 2 mm and was successfully dilated with a 3 mm balloon catheter. Five patients developed significant cyanosis caused by interstitial fluid retention at the age of 5 weeks to 4 months, which was resolved by repeated balloon dilatation atrial septotomy. In one, the transseptal venous access was adversely affected after an immediate postnatal Rashkind procedure at 31 weeks’ gestation, with a body weight of 1660 g. In this patient aged 2 months with a body weight of 2.1 kg, transhepatic venous cardiac catheterisation was performed twice within eight days to achieve a widely patent atrial septum defect (fig 2).

Two patients aged 3 and 5 months developed low cardiac output at home because of a stenosis at the aortic part of the ductus arteriosus, which was not fully covered by the implanted stent. Emergency balloon dilatation was performed but both patients continued to have right heart failure. One of them finally died waiting for a donor heart. The parents of the other one refused heart transplantation for religious reasons, so reconstructive surgery was performed as a rescue approach. This infant died of postoperative right heart failure.

Surgical removal of the ductal stent was uneventful in all patients, but one developed transient diaphragmatic paralysis. As described before, reconstruction of the aortic arch by placement of an allograft tube was associated with significant postoperative sequelae such as significant stenosis of the left pulmonary artery and distal aortic arch obstruction. Stenosis at both sides was effectively managed by balloon dilatation and stent placement, respectively. Extended augmentation of the aortic arch avoided neoaortic arch obstruction in the last six of our patients. The BCPC performed at the site of the previous right pulmonary artery banding was uneventful in all but one. In one patient the narrowed cavopulmonary connection was stented and venovenous collaterals were embolised by metallic coils (Cook, Bloomington, Indiana, USA) for treatment of significant postoperative oxygen desaturation. No patient had pleural effusions or excessive chest tube drainage for more than five days after surgery. Postoperative oxygenation with a mean arterial oxygen saturation of 65% on arrival at the intensive care unit improved over the first 48 hours to 75–85% at time of hospital discharge. In the postoperative period haemodynamic stability was secured mainly by vasodilatation of the pulmonary vascular bed and systemic arteries.

The overall survival rate of all patients with ductal stenting combined with bilateral pulmonary banding is now 75%. The presented surgical modification in hypoplastic left heart lesions has been performed with a success rate of 90%. In clinical follow up of one month to three years only one patient, who was postnatally in severe cardiogenic shock, developed major neurological complications. One patient is awaiting total cavopulmonary connection. In the same period at our institution 10 patients with left heart obstructive lesions underwent the classic Norwood 1 operation, four died in the early postoperative period, and another received a donor heart four weeks after the Norwood 1 operation. During the review process of our article, one patient (patient 20 in table 1) underwent biventricular repair and the patient awaiting total cavopulmonary connection has meanwhile undergone a successful operation.

DISCUSSION

Stenting of the arterial duct combined with bilateral pulmonary artery banding in newborns with hypoplastic left heart or multiple left heart obstructive lesions allows a variety of surgical strategies. Recently, we described this interventional surgical procedure as a basis for a modified surgical technique—that is, neoaortic reconstruction, part of first stage of palliation of hypoplastic left heart, together with BCPC in a single operation with cardiopulmonary bypass beyond the neonatal period, as well as heart transplantation after an extended waiting period. Additionally, this still preliminary experience in a series of 20 patients shows that the approach of ductal stenting and bilateral pulmonary banding provides the opportunity not only for a combined first and second stage procedure or for delayed heart transplantation but also for biventricular repair in newborns with multiple left heart obstructive lesions. In this context, an additional advantage of the presented strategy seems to be relevant in patients with various left heart hypoplastic or obstructive lesions suffering postnatal cardiogenic shock or in whom additional reasonable non-cardiac problems, as described under Methods, are
present. The potential of complete recovery of heart function, multiorgan failure, and neurological complications is the clue for better long-term outcome regardless of the cardiac surgery. The established treatment strategies do not give the opportunity of waiting until a prospective decision can be made. Even patients with multiple left heart obstructive lesions, in whom the immediate postnatal decision regarding single ventricle palliation or biventricular approach is unclear, may benefit from the presented strategy. The controversy regarding whether the presence of antegrade flow in the ascending aorta is predictive of a successful biventricular approach, as shown in one series but not supported by another, highlights additionally the importance of selecting the appropriate strategy in early infancy. The interventional surgical approach presented here may be considered in postnatal decision making of borderline left heart obstructive lesions.

On the basis of a critical analysis of the 20 patients and our recently published data, we offer the following recommendations regarding interventional and surgical techniques and strategy.

Considerations regarding cardiac catheterisation and stent placement

Firstly, a newborn with a hypoplastic left heart lesion admitted with PGE1 infusion with a wide open duct with no detectable ductal narrowing and unrestricted interatrial communication would be considered for bilateral banding within the first 3–5 postnatal days or earlier, if a pulmonary run off in consequence of a postnatal decrease in pulmonary vascular resistance is observed. Ductal stenting, as we described recently, as well as re-evaluation of the effectiveness of the bilateral banding, would be performed 2–3 days before the patient should be discharged home. In such a case, we would discontinue the PGE1 infusion 2–3 hours before catheterisation to achieve a smaller and narrowed duct, facilitating stent placement and preventing stent dislocation.

A newborn with a restrictive patent foramen ovale (mean gradient > 5 mm Hg) would be catheterised before bilateral banding is provided. In case of an already recognisable narrowing at any part within the duct, even if no pressure gradient is detectable, a stent would be placed immediately before the patent foramen ovale is dilated. Effective patent foramen ovale dilation can be achieved with a balloon diameter of 10–12 mm. If the femoral vein is not accessible for cardiac catheterisation, the percutaneous transhepatic venous approach is feasible especially in premature infants.

Any haemodynamically relevant ductal stenosis, particularly in patients with cardiogenic shock, and ineffective pharmacological treatment are, in our view, indications for balloon dilatation or stent implantation in the arterial duct, and also as a rescue procedure.

Secondly, the technical aspects of ductal stenting have changed over time, not least with respect to improved stent design. We started with the technique as described by Gibbs and colleagues, followed by the technique described previously. We also used for the first time premounted stents with balloon diameters useful for ductal stenting in patients with duct dependent systemic blood flow. These stents can be advanced through a 25 mm long French Terumo sheath, which can easily be placed in the pulmonary trunk without any haemodynamic compromise, as we have seen using French sheaths as described by Gibbs and colleagues. Additionally, the premounted stents have a low profile, which makes them very flexible and allows multiple repositioning manoeuvres contributing to a safer procedure. Therefore, we would recommend the stent design and implantation procedure described here, particularly when starting with ductal stenting.

Considerations regarding the surgical techniques

Firstly, bilateral pulmonary artery banding is principally easy with the technique we described previously. We experienced narrowing of the duct and displacement of the stent during the banding operation in one patient, who died a few hours after the easily performed bilateral banding led to the following change of strategy. Pulmonary artery banding was performed in all patients with a wide open duct before the ductal stent was placed. In cases where the catheterisation has to be performed first, the surgeon has to be very careful to avoid stent obstruction or displacement. Too much traction while the band is being placed and secured to the adventitia of the pulmonary artery branch or uncontrolled impingement of the stented duct may result in deleterious restriction of ductal flow to the coronary arteries or the descending aorta.

Secondly, extended augmentation of the aortic arch and patch augmentation of the pulmonary arteries just proximal to the left bifurcation may solve the problem of coarctation on both sides. Additionally, avoiding complete circulatory arrest by means of cerebral perfusion through the innominate artery and introducing the arterial and venous vasodilators phenolamine and nitroprusside, respectively, during weaning, as well as intensive care management, may be why in individual cases no further inotropic support is necessary other than the already routinely used phosphodiesterase inhibitor milrinone.

However, the long-term implication of the described approach has to be elucidated in further studies. A significant number of patients who are not yet suitable for reconstructive surgery and whose parents opt for transplantation die awaiting a donor heart because of heart failure or increasing pulmonary artery resistance. In principle, stent implantation in the ductus arteriosus provides the possibility of long-term ductal patency. At the same time, protection of the lung vasculature from high pressure by bilateral pulmonary artery banding with low resistance enables transplantation as long as 11 months later. But close follow up is necessary to detect restrictive interatrial communication, narrowing of the duct, or compromised pulmonary blood flow before heart function worsens. In addition, the development of significant aortic coarctation with compromised retrograde blood flow and coronary perfusion at risk must be diagnosed very early. Altogether, maintenance of ductal patency by stent implantation is attractive but in practice the quality of palliation requires much experience in interventional paediatric cardiology, as well as experience in the echocardiographic and clinical assessment of the haemodynamics in follow up before further surgery is undertaken.

Conclusion

Stenting of the arterial duct and banding of the pulmonary arteries expand the possible surgical strategies in newborns with multiple left heart obstructive lesions. In particular, moderate hypoplasia of the left ventricle or left sided valves may mature with time and allow biventricular repair after some months. For these patients the presented approach is a “bridging procedure”. The same is true for newborns with associated non-cardiac disorders with significant relevance concerning total outcome, where the decision regarding definitive cardiac repair can be postponed.

However, with respect to the deaths on our heart transplantation waiting list, one of our goals is to evaluate the combined interventional and surgical approach as a bridging procedure for transplantation as well. Regarding the experience with the Norwood procedure without circulatory arrest, as described by Imoto and colleagues, we hypothesise that cardiopulmonary bypass, which also allows complete avoidance of circulatory arrest and deep hypothermia in the combined Norwood 1 and 2 repair, may also enable this surgical technique to be used as a bridging method. In this context, the early use of BCPC with relief of volume load for a weak single right ventricle by establishing serial instead of parallel pulmonary and systemic circulations may support this strategy. Changes in surgical techniques, as well as in perioperative management, imply a
learning curve for the whole team. Improvement has been achieved and can be expected in the future.

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REFERENCES

IMAGES IN CARDIOLOGY

Thirty years after percutaneous closure of patent ductus arteriosus*

A 68 year old woman presented with new onset of dyspnoea and fatigue. Thirty years previously she was diagnosed as having a patent ductus arteriosus. At that time the left-to-right shunt across the patent ductus was found to be 33%, and no evidence of pulmonary vascular disease was present. A transcatheter closure of the patent ductus arteriosus was performed percutaneously using an Ivalon plug with a metallic skeleton as occluder device (Porstmann and Wierny: Z Gesamte Inn Med, 1968).

Thirty years later the patients lung and renal function were well preserved; the ECG and laboratory values did not show any abnormalities and could not help elucidate the patient’s symptoms. Trans-thoracic echocardiography showed normal systolic function but reduced early transmitral velocity compared with increased atrial velocity and shortened deceleration time.

However, multiplane transoesophageal echocardiography (panel A) demonstrated the correct position of the Ivalon occluder (size 11 × 13 × 8 mm) (arrow). Between the pulmonary trunk (P) and the proximal descending aorta (Ao) just distal to the left subclavian artery, the smooth wrapping (envelope) of the Ivalon plug is seen. A left-to-right shunt was excluded. The diastolic dysfunction was confirmed with left and right heart catheterisation. The angiogram of the aorta (panel B) demonstrates the metal nucleus (skeleton) of the occluder device (arrow). There was no evidence of coronary artery disease, valvar heart disease, left-to-right or right-to-left shunt with correct position of the ductus occluder more than three decades after percutaneous placement.

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*In memorial of Professor Werner Porstmann (1921–1982), who introduced this method and was the first to perform this procedure at the Charité University Hospital Berlin in 1966
Thirty years after percutaneous closure of patent ductus arteriosus

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