Fate of the Stented Arterial Duct

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Background—The technical aspects of ductal stenting have been reported, but little is known of the fate of the duct after stent implantation.

Methods and Results—Nineteen patients underwent stent implantation to maintain ductal patency. Eight had hypoplastic left heart (HLH) syndrome, 10 had pulmonary atresia, and 1 had tricuspid atresia. Median survival with HLH was 57 (12 to 907) days. Stent implantation was successful in all cases of HLH, but there were no long-term survivors. Two well-palliated infants died at transplantation. Median survival with duct-dependant pulmonary flow was 183 (0 to 1687) days, with 3 patients well at latest follow-up (56, 55, and 9 months, respectively). There were 2 operative deaths due to ductal spasm and 4 late deaths, 1 due to duct thrombosis, 1 due to chronic lung disease, and 2 of unknown cause. Stent implantation failed in 4 of the 11 cases. Assessment of endothelialization was possible in 13 cases; the stent was partially covered in 3 and fully endothelialized in all 10 cases assessed 8 weeks after implantation. In patients stented for inadequate pulmonary flow, ductal intimal hyperplasia occurred by 9 months in all 3 survivors but responded to repeated dilation.

Conclusions—Ductal stenting cannot be recommended. In patients with HLH, it provides only short-term palliation even when combined with pulmonary artery banding. With duct-dependent pulmonary blood flow, the procedure carries high risk, and duration of palliation is poor. In patients with bilateral ducts and absent central pulmonary arteries, good palliation may be achieved, but repeated angioplasty is necessary to counteract intimal hyperplasia. (Circulation. 1999;99:2621-2625.)

Key Words: stents ■ arteries ■ pulmonary heart disease ■ pediatrics

Animal studies have suggested that ductal patency may be maintained by stent implantation, but only preliminary human studies with short-term results have been reported. This report describes our experience with ductal stenting over 6 years, with maximum patient follow-up of 53 months.

Methods

Nineteen infants aged 4 to 78 days, with weight ranging from 2 to 4.1 kg, were included. Eight had hypoplastic left heart syndrome (HLH), 10 had pulmonary atresia, and 1 had tricuspid atresia with a restrictive ventricular septal defect. Johnson & Johnson stainless steel stents were used in all cases. The technical details of stent implantation have been described previously. Ethics committee approval was obtained for the studies.

Results

Duct-Dependent Systemic Blood Flow

In all 8 patients with HLH, stent implantation was successful. Seven also had pulmonary artery banding and atrial septostomy. There was no procedure-related death in this group. Survival ranged from 12 days to 30 months (median, 51 days). Two patients survived in good health until transplantation at age 2 and 6 months, respectively, but both died during surgery. The single patient who did not have pulmonary artery banding died during surgical reconstruction of the aortic arch at age 12 days. Four patients aged between 2 and 10 weeks died of right ventricular failure due to excessive pulmonary blood flow despite bilateral pulmonary artery banding. One died at the age of 30 months of abrupt right ventricular failure despite redilation of the stent and effective banding.

Assessment of duct morphology was possible at surgery or autopsy in 7 of the 8 patients. All the stents appeared widely patent, but in 1 case there was a minor degree of constriction at the aortic end of the duct, the end of the stent being 2 mm proximal to the junction of the duct and aorta. As early as 2 weeks, there was partial endothelialization (Figure 1). All stents examined >3 months after implantation were fully endothelialized. In 1 patient, the duct became mildly restrictive (withdrawal gradient of 20 mm Hg) owing to growth and intimal hyperplasia. This responded to repeat dilation at the age of 8 months.

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Duct-Dependent Pulmonary Blood Flow

Stent implantation was successful in 7 of the 11 patients with duct-dependent pulmonary blood flow. In 2 patients, death occurred during catheterization owing to duct spasm, and in 2 others, implantation failed because of duct tortuosity.

Duct patency was maintained after a single procedure in 3 patients, but 2 procedures were required in 1 case, 3 procedures in 2, and 5 procedures in 1. Of the 9 ducts stented in 7 patients, a single stent was required in 5, 2 stents were required in 1, 3 stents in 3 ducts, and 4 (with some overlap) in 1.

That the duct was not initially fully stented only became apparent between 4 and 10 days after prostaglandin E was stopped. Constriction occurred at the aortic end in 1 case and at the pulmonary artery end in the other 4 cases. Accurate stent positioning was difficult because of high flow in the duct and anatomic distortion caused by the guidewire and catheter.

Of the 7 early survivors, 2 died suddenly in their cribs at the age of 6 and 11 weeks, respectively. No clear cause was found, and both had patent ducts at autopsy (1 had situs ambiguous and the other an extensive cleft palate). One child died suddenly at the age of 11 months; autopsy showed acute thrombosis of the duct with some constriction at the pulmonary artery, the distal end of the stent stopping 2 mm from the distal end of the duct. One patient with bronchopulmonary dysplasia became increasingly cyanosed at the age of 4 months; a bridged stent had been used, and there was mild stenosis adjacent to the bridge. We were unable to cross the stent with a balloon, so the patient had a modified Blalock Taussig shunt but died postoperatively owing to chronic lung disease despite a patent duct and patent shunt.

Three patients remain alive and well. Two have absent central pulmonary arteries with bilateral ducts and have needed 3 and 1 elective repeat catheterizations, respectively, to maintain adequate saturations (around 80%) and adequate pulmonary artery growth (Figure 2) at their current respective ages of 5 years and 16 months. One patient with tricuspid atresia underwent 2 repeat dilations (1 during treatment for endocarditis), remaining well at the age of 5, when she had a successful bidirectional Glenn shunt implanted (the duct was left alone at operation).

Increasing cyanosis in the 3 long-term survivors appeared to be due in part to the patients’ growth, but angiography showed endothelial proliferation to be a factor in all (Figure 3). In the 4 patients who died, autopsy showed full endothelialization of the stent, with a thin, complete layer of endothelium formed as early as 6 weeks (Figure 4). Microscopic thickness of the neoendothelium varied from 0.04 to 0.09 mm over the mesh itself and from 0.15 to 0.28 mm between the mesh.

Discussion

Duct-Dependent Systemic Blood Flow

The high mortality rate associated with the Norwood operation after its introduction in the United Kingdom prompted a search for alternative palliation, particularly with a view to later transplantation. Ductal stenting as a bridge to transplantation met with some success in California, but the small UK donor pool suggested that duct patency without control of pulmonary blood flow would not allow sufficient time to find a donor organ. We therefore chose to band both pulmonary arteries and decompress the left atrium as well as to stent the duct. Ductal stenting in HLH proved to be technically straightforward, with no procedural mortality in this group and excellent medium-term maintenance of ductal patency, with little intimal proliferation. Controlling pulmonary blood flow proved very difficult, with 4 of the 8 babies dying with heart failure and excessive pulmonary flow despite the bandings. The 1 long-term survivor (30 months) was well until he abruptly developed fatal right ventricular failure.
Duct-Dependent Pulmonary Blood Flow

Stenting the duct when it is long and tortuous (as is common with pulmonary atresia with ventricular septal defect) is technically demanding, and even gentle passage of a guide-wire may cause fatal ductal spasm. Even after apparent success, duct constriction may occur after prostaglandin withdrawal if a short section of the duct is unstented. Unexplained sudden death occurred in 2 patients who had patent ducts at autopsy. Sudden death may occur after palliation with an aortopulmonary shunt, and it seems likely that this may be related to the underlying disease rather than specifically related to the mode of palliation.

Redilation of the duct was relatively straightforward, only failing in 1 case, and effectively prolongs palliation. However, the need for repeated redilations raises concern about cumulative radiation dosage.

Endothelialization

A thin but complete layer of endothelium forms over the stent as early as 1 month. The thickest endothelium occurred between the mesh of the stent, with a thinner layer covering the mesh itself. Neointimal proliferation played an important role in duct-dependent pulmonary blood flow. Although redilation was successful, there was a gradual recurrent fall in oxygen saturation as the duct again became neonatal stent implantation. Middle and bottom, Selective angiography in left (LPA; middle) and right (RPA; bottom) ducts at age of 4 years after neonatal bilateral ductal stenting for pulmonary atresia with absent pulmonary artery confluence. Pulmonary arteries are well developed, and there are mild stenoses in both ducts (arrowed), related in part to intimal hyperplasia and in part to the stent becoming restrictive as the child has grown. Pulmonary artery pressures were low (means of 10 and 12 mm Hg).
Figure 4. Appearances of endothelialization of stents at different periods after implantation. Top, Autopsy appearance of duct in a patient with pulmonary atresia and a complete AV septal defect who died suddenly 42 days after stenting. Stent is fully endothelialized apart from distal tips, which protrude slightly into pulmonary artery. LPA and RPA indicate left and right pulmonary artery, respectively. Magnification ×400. Middle, Photomicrograph of endothelium over stent mesh 39 days after stent implantation in a case of pulmonary atresia. There is a single layer of neoendothelial cells 0.09 mm in thickness overlying the steel strands, which were removed before sectioning. Bottom, Duct wall in the same case. Media (M) has a normal appearance. Intima (I) between steel strands is several cell layers thick, measuring 0.28 mm in contrast to the 0.09-mm monolayer of cells overlying the steel itself. Gap in intima (●) is where the steel mesh of the stent has been removed. Magnification ×40.
compromised by neoendothelium. Future changes in stent design might reduce endothelial reaction, but at present repeat dilation offers temporary treatment for endothelial proliferation in the stented duct, as it does in other parts of the circulation.\textsuperscript{8,9}

Conclusions
Maintenance of duct patency by stent implantation is theoretically attractive but in practice is disappointing. With HLH, the duct may be kept open, but quality of palliation is poor because of difficulty in controlling pulmonary blood flow even after banding of the pulmonary arteries; the most recent UK results of the Norwood operation and its modifications are clearly superior.

With duct-dependent pulmonary blood flow when the duct is tortuous, there is a risk of fatal ductal spasm. Even when stenting initially appears successful, early duct stenosis may occur unless stent positioning has been exact. However, patients with “disconnected” pulmonary arteries and bilateral ducts, who are difficult to palliate surgically, often have less tortuous ducts, allowing good short- and medium-term palliation to be achieved by stenting; it is also possible to enlarge the stent by repeated balloon dilation as the child grows. The technique may be justified in this rare subgroup of patients with pulmonary atresia, but for the majority of infants with duct-dependent pulmonary or systemic blood flow, ductal stenting cannot be recommended.

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References
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