BRIEF REPORT

Ductus Arteriosus Patency With Stenting in Critical Pulmonary Stenosis and Pulmonary Atresia With Intact Interventricular Septum

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We report our experience with stenting of the ductus arteriosus in 3 neonates. Two patients had pulmonary atresia with an intact interventricular septum and 1 had critical pulmonary stenosis. Radiofrequency ablation was used to open the atretic pulmonary valve in 2 patients. In all 3 patients implanting a stent avoided the need for surgical creation of a Blalock-Taussig shunt. In two patients the procedure was scheduled as elective surgery, and in 1 it was done as an emergency procedure. Ductus arteriosus stenting is an alternative to palliative cardiac surgery.

Key words: Congenital heart disease. Catheterization. Ductus arteriosus. Stent. Pulmonary stenosis.

Permeabilidad del conducto arterioso con *stent* en la estenosis pulmonar crítica y atresia pulmonar con septo interventricular intacto

Presentamos la implantación de *stent* en el conducto arterioso neonatal en 3 niños, 2 de ellos con atresia pulmonar y septo interventricular intacto, el tercero con estenosis pulmonar crítica. En los niños con atresia pulmonar, la apertura valvular se consiguió mediante radiofrecuencia. En los 3 casos, la implantación del *stent* evitó la fístula de Blalock-Taussig. En 2 de ellos se hizo de manera electiva y en 1 de manera urgente. La implantación de *stent* ductal es una alternativa a la cirugía cardíaca paliativa.

Palabras clave: Cardiopatías congénitas. Cateterismo cardíaco. Conducto arterioso. Stent. Estenosis pulmonar.

INTRODUCTION

Intravascular stent implantation for the treatment of vascular stenoses has advanced considerably in recent years, although use of this technique for congenital heart disease in newborns is extremely limited.

In cases of pulmonary atresia with intact ventricular septum (PA/IVS) and in critical pulmonary stenosis (CPS), pulmonary circulation after birth depends on the ductus arteriosus, which requires intravenous use of prostaglandin E_1 (PGE1) to maintain patency. Pulmonary balloon valvuloplasty is performed for both these conditions, although in PA/IVS the valve must

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Received June 23, 2004. Accepted for publication October 5, 2004. be perforated during cardiac catheterization, either mechanically^{1,2} or with radiofrequency^{3,4} or laser⁵ techniques. Once the obstruction of the right ventricle is released, biventricular physiology is not always achieved immediately, making it necessary to continue PGE1 therapy and in some cases to create a Blalock-Taussig shunt (BTS). This last option has been associated with complications, such as phrenic or vagal nerve paralysis, chylothorax, distorted pulmonary artery growth, pulmonary branch stenosis, and surgical adhesions. Hence, a nonsurgical option for BTS⁶ is necessary. Stent implantation to maintain ductus arteriosus patency^{7,8} has been used in diseases with duct-dependent systemic blood flow or pulmonary blood flow, with much better results in the latter indication. In fact, the best results are obtained in diseases where the ductus arteriosus is short and less tortuous, such as PA/IVS and CPS.^{6,9} We report on our experience with stent implantation to maintain the patency of the ductus arteriosus and thereby avoid pal-

ABBREVIATIONS

PA/IVS: pulmonary atresia with intact ventricular septum.CPS: critical pulmonary stenosis.PGE1: prostaglandin E1.BTS: Blalock-Taussig shunt.

liative surgery.

PATIENTS AND METHODS

Patients

Three patients (Table 1) were studied by echocardiography prior to catheterization and considered to have a tripartite right ventricle. In patients 1 and 2, right ventriculography was performed during catheterization, and the coronary-ventricular fistulas were considered non-significant.

Technique

Informed consent was obtained in all cases before the procedure. Stent implantation was performed by an anterograde approach. A 0.014-inch guidewire was advanced through the femoral vein to the femoral artery, across the right atrium, through the pulmonary artery, ductus arteriosus and descending aorta, and ultimately exteriorized through the femoral artery to establish a venoarterial loop. All stents were premounted on a balloon (Stent BX Sonic®, 4×18 mm, Cordis/Johnson & Johnson) and advanced coaxially over a 0.014-inch guidewire. Stent implantation was done under radioscopic control, and a lateral aortogram was performed to verify the stent position. PGE1 perfusion was discontinued in all cases before stent implantation, in order to visualize on angiography the area of maximum ductal constriction, which was used as a reference to adjust to optimal stent position during expansion. During the catheterization, a control echocardiogram was

Patient Birth Weight, g Sex Diagnosis Initial Treatment Tricuspid Valve Diameter, Z-Value Age, Days PA/IVS RF+PV 2907 Μ 14 mm (-1.32) 1 11 F RF+PV 2 3900 PA/IVS 12 mm (-1.88) 7 3300 Μ CPS ΡV 3 1 mm (-2.56) 8

*RF indicates radiofrequency perforation of pulmonary valve; PV, pulmonary valvuloplasty; M, male; F, female; PA/IVS, pulmonary atresia in intact ventricular septum; CPS, critical pulmonary stenosis

performed to ensure that the stent was correctly positioned in the duct. All children received intravenous cefazolin during the procedure. In addition all received aspirin 5 mg/kg/day and dipyridamole 3 mg/kg/day for 3 months.

RESULTS

Patient 1

A newborn with PA/IVS underwent a procedure to open the pulmonary valve, but continued to depend on PGE1 to maintain a SatO₂ of 85%-90%. Since it was impossible to discontinue therapy, a decision was made to implant a ductal stent 17 days later. PGE1 perfusion was discontinued during implantation of the ductal stent; SatO₂ at the end of the procedure was 92%.

Patient 2

A newborn with PA/IVS underwent radiofrequency ablation and balloon valvuloplasty. Echocardiography performed in the interventional cardiology suite showed slow anterograde flow through the pulmonary valve and ductal constriction despite PGE1. A decision was made for immediate placement of a ductal stent and discontinuation of PGE1 perfusion. SatO₂ at the time of discharge was 88%.

Patient 3

An infant with CPS required PGE1 perfusion after valvuloplasty to maintain $SatO_2>90\%$; 2 days later, he presented clinical symptoms of ductal closure (Figure, A) and, despite high PGE1 doses, $SatO_2$ persisted at 68%-70%. A ductal stent was implanted urgently and SatO₂ at the time of discharge was 90%-93%.

Follow-up

None of the patients required resumption of PGE1 perfusion or emergency BTS during the acute period. The follow-up data are shown in Table 2; in all patients, the ductus arteriosus remained open and there were no clinical symptoms of pulmonary overflow or any need for diuretics. Pulmonary pressure was nor-

Mortera C, et al. Stenting the Ductus Arteriosus in Critical Pulmonary Stenosis and Atresia



Figure. Patient with critical pulmonary stenosis. A) aortography, see other catheter (arrow) in the pulmonary artery showing ductal constriction. B) and C) balloon-mounted stent (arrow) prior to inflation; side and anteroposterior view. D) aortography after stent placement. E) chest x-ray in which the stent can be observed (arrow).

mal in all patients, and right ventricular pressure was also normal except in the patient with CPS, who had residual pulmonary stenosis on echocardiography.

DISCUSSION

Two representative series^{10,11} of patients with PA/IVS who underwent valve perforation by radiofrequency reported that 52% (14/27 and 17/33) of the patients required a BTS between 2 and 24 days after therapeutic catheterization. In CPS it has been observed that 7% of the patients needed a BTS after pulmonary valvuloplasty.¹²

At the present time, the factors predicting which patients will need a BTS after releasing the obstruction of the right ventricular outflow tract are not accurately known.¹³ Identifying these factors would allow us to stratify the patients and create a BTS without waiting too long or doing it as elective surgery. The effectiveness of stenting in certain lesions with duct-dependent pulmonary circulation was questioned by Gibbs et al.⁸ although all patients in this series had a diagnosis of pulmonary atresia with ventricular septal defect (VSD), a disease in which the ductus arteriosus is generally long and tortuous.⁶ In contrast, Schneider et al⁹ reported that stent implantation was successfully performed in their series of eight patients with a history of PA/IVS or CPS, with no mortality or need to resume prostaglandin therapy. This results from the fact that the ductus arteriosus in CPS and PA/IVS is short and straight.

The ductal stent has been observed to be completely endothelialized at 30 days.⁸ Stent patency decreases over time; in 4 of 6 patients it was completely occluded between 4.5 and 17 months (mean, 10 months), a desirable outcome in these patients since it indicates that the right ventricle has already adapted to biventricular physiology.⁹

Our brief experience reflects three different situations. In patient 1, it was necessary to implant the stent electively 17 days later because PGE1 perfusion could not be discontinued. Patient 2 required immediate stent implantation because the echocardiogram in the catheterization room showed that, although the pulmonary valve had opened completely, there was al-

TABLE 2. Clinical Fo	llow-up	,
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Patient	Follow-up, Days	% SatO ₂ of Hb (Last Visit)	Patent Ductus	RV Systolic Pressure†, mm Hg
1	510	96	Yes	20-25
2	88	90	Yes	28
3	78	95	Yes	40

*RV indicates right ventricle.

†Estimate on echocardiographic study.

most no pulmonary anterograde flow and ductal restriction existed despite PGE1 infusion. In the third patient, sudden closure of the ductus required urgent stent implantation 2 days later.

COMMENTS

Although our initial experience is limited, we can suggest the advantages offered by stent placement in this group of patients: a) avoidance of surgical creation of BTS; b) relatively simple implantation, further facilitated when the femoral vessels are still canalized after the outflow tract is completely opened; c) temporary implantation: both ductus arteriosus stenting and the BTS are temporary solutions, since we hope to achieve biventricular viability in these patients (in patients in whom this evolution is not possible, the next step after several months would be a Glenn shunt as an initial step to single-ventricle repair or one and a half ventricular repair; d) possibility to redilate the ductus arteriosus stent in cases where it becomes restrictive; e) easier closure of the ductus arteriosus when it is no longer necessary, using percutaneous techniques; f) avoidance of known distortion of the pulmonary arteries after BTS; and g) avoidance of prolonged PGE1 therapy.

One problem we found in this article that should be studied in the future is how to identify the group of patients (about 50% and 7% in PA/IVS and CPS, respectively) who will need prolonged support with PGE1, BTS, or a ductal stent after the right ventricle is open.

REFERENCES

- 1. Latson LA. Nonsurgical treatment of a neonate with pulmonary atresia and intact ventricular septum by transcatheter puncture and balloon dilation of the atretic valve membrane. Am J Cardiol. 1991;68:277-9.
- Alcíbar J, Cabrera A, Pena N, Baraldi C, Arriola J, Aramendi J. Valvulotomía mecánica percutánea dirigida en la atresia pulmonar con septo íntegro. Rev Esp Cardiol. 2003;56:822-5.
- Rosenthal E, Qureshi SA, Chan KC, Martin RP, Skehan DJ, Jordan SC, et al. Radiofrequency-assisted balloon dilatation in patients with pulmonary valve atresia and an intact ventricular septum. Br Heart J. 1993;69:347-51.
- Camino M, Brugada J, Mortera C, Thio M, Rovirosa M, Bartrons J. Valvulotomía pulmonar percutánea mediante radiofrecuencia en la atresia pulmonar con septo interventricular íntegro. Rev Esp Cardiol. 2001;54:243-6.
- Rosenthal E, Qureshi SA, Kakadekar AP, Anjos R, Baker EJ, Tynan M. Technique of percutaneous laser-assisted valve dilatation for valvar atresia in congenital heart disease. Br Heart J. 1993;69:556-62.
- Gewillig M, Boshoff DE, Dens J. Mertens L, Benson LN. Stenting the neonatal arterial duct in duct-dependent pulmonary circulation: New Techniques, Better Results. J Am Coll Cardiol. 2004;43:107-12.
- Ruiz CE, Bailey LL. Stenting the ductus arteriosus. A "wannabe" Blalock-Taussig. Circulation. 1999;99:2608-9.
- Gibbs JL, Uzum O, Blackburn MEC. Fate of the stented arterial duct. Circulation. 1999;99:2621-6.
- Schneider M, Zartner P, Sidiropoulos A, Konertz W, Hausdorf G. Stent implantation of the arterial duct in newborns with duct-dependent circulation. Eur Heart J. 1998;19:1401-9.
- Humpl T, Soderberg B, McCrindle BW, Nykanen DG, Freedom RM, Williams WG, et al. Percutaneous balloon valvotomy in pulmonary atresia with intact ventricular septum: impact on patient care. Circulation. 2003;108:826-32.
- Agnoletti G, Piechaud JF, Bonhoeffer P, Aggoun Y, Abdel-Massih T, Boudjemline Y, et al. Perforation of the atretic pulmonary valve. Long-term follow-up. J Am Coll Cardiol. 2003;41:1399-403.
- Benito Bartolomé F, Sánchez Fernández-Bernal C, Torres Feced V. Valvulotomía percutánea de la estenosis pulmonar crítica neonatal. Resultados y seguimiento a medio plazo. Rev Esp Cardiol. 1999;52: 666-70.
- 13. Weber HS. Initial and late results after catheter intervention for neonatal critical pulmonary valve stenosis and atresia with intact ventricular septum: a technique in continual evolution. Catheter Cardiovasc Interv. 2002;56:394-9.