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Case Report

Hybrid Stenting of Aortic Coarctation in Very Low Birth Weight Premature Infant

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A very low birth weight infant with severe aortic coarctation developed progressive left ventricular dysfunction and pulmonary overflow with hemorrhage, while receiving prostaglandins. To avoid morbidity from conventional surgery or percutaneous intervention, a two-step strategy was performed at a weight of 970 g. First vascular access was obtained through sternotomy: a 3/8 mm coronary stent was deployed through a 4 French sheath in the ascending aorta; the arterial duct was clipped. At the age of 5 months, the stent was removed and the aortic arch reconstructed with an end-to-end anastomosis through lateral thoracotomy. This strategy was not associated with morbidity typical for premature infants with congenital heart disease.

Key words: hybrid procedure; stent; aortic coarctation; very low birth weight; premature infant; vascular access

INTRODUCTION

Severe aortic coarctation in very low birth weight infants often includes medical palliation with prolonged prostaglandin (PGE-1) infusion. After several days they frequently develop high output failure with ventricular dysfunction, pulmonary overflow, and marginal systemic perfusion, which predisposes for necrotizing enterocolitis, infections, and excessive delay of surgical repair [1–3].

Primary surgical correction is possible in infants less than 1.5 kg but has a substantial mortality and morbidity [4–6]. Primary stenting of the aortic coarctation with surgical stent removal and end-to-end anastomosis on a more adequate weight has been proven to be a safe strategy in these very small infants [1,2,7,8]. Vascular complications however remain a challenge.

We performed a hybrid procedure in a 970 g infant with severe aortic coarctation through sternotomy, allowing successful stenting of the aortic coarctation and clipping of the arterial duct without significant morbidity.

CASE AND PROCEDURE DESCRIPTION

A male infant (twin, birth weight 670 g, 28 weeks of gestational age, ventilator dependent) presented with severe aortic coarctation with hypoplasia of the isthmus (2 mm). PGE-1 was started with restoration of the perfusion and the dose was maintained at a minimal level of 10 nanogram/kg/hr. The clinical condition deteriorated 1 week later with decrease of systemic

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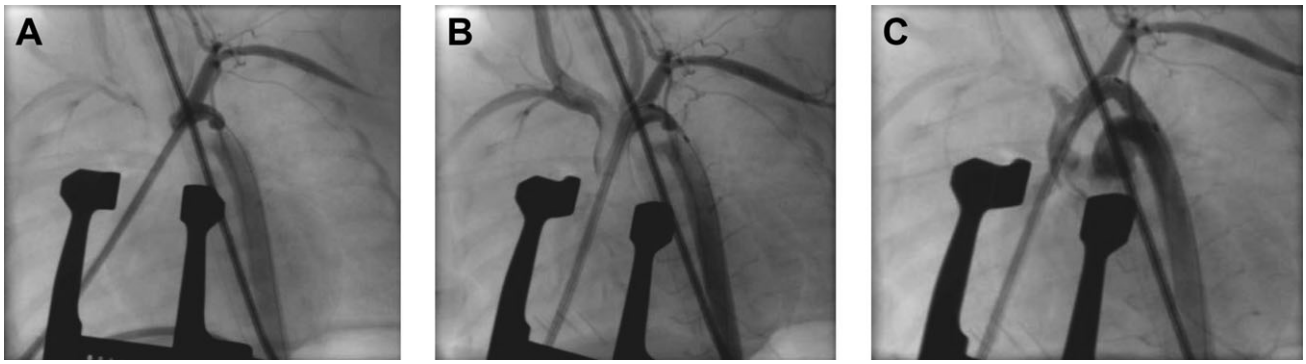

Fig. 1. A: Aortogram through 4 Fr sheath at aortic cross demonstrating critical aortic coarctation. B: Positioning stent asymmetric across the narrowing from left subclavian artery until just beyond the coarctation. C: Aortogram with final result; the patent duct was subsequently closed with a vascular clip.

perfusion, pulmonary overflow, pulmonary hemorrhage, and progressive left ventricular dysfunction. The PGE-1 was stopped without success as the arterial duct remained widely patent. At the age of 29 days and a weight of 970 g an intervention became necessary, but due to the low weight and the brittle condition, the patient was declined for conventional surgical or percutaneous intervention. A two-step strategy was applied to treat the coarctation: first bail-out stenting of the isthmus and coarctation followed later by surgical stent removal and arch reconstruction [2]. Stent size was chosen based on echocardiographic measurements: diameter same as the proximal aortic cross (3 mm), stent length was the distance between distal take-off of left subclavian artery until just beyond the coarctation (8 mm). Access through sternotomy was chosen in order to deal both with the coarctation and the arterial duct at the same time. The procedure was done in the hybrid suite (surgical theatre with monoplane radiology). The surgeon provided access through medial sternotomy and sutured a purse string with Prolene 6/0 on the 5 mm ascending aorta. Puncture of the aorta was done with a 21-gauge needle. A 0.014″ coronary wire (tip mildly angulated at 7 mm of tip, wire cut off at 70 cm) was advanced. The needle was removed and a 4 Fr introducer sheath was advanced into the ascending aorta. A dose of 50 IU/kg of heparin was administered intravenously; all sheath flushes were done with non-heparinized 0.9% saline. Monoplane fluoroscopy was used to position the wire in the descending aorta. The tip of the sheath was advanced until the aortic cross. An angiogram was obtained with a small 1 cc hand injection through the introducer sheath (Fig. 1). A premounted coronary stent 3/8 mm (Liberté Monorail®, Boston Scientific, Nanterre Cedex, France) was positioned over the full length of the isthmus, from just distal of the left subclavian artery until just beyond the coarctation. An angiogram confirmed the correct position. The stent was deployed using an indeflator at 15 atmosphere bringing the stent to a diameter of 3.4 mm. After angiographic check, the sheath was removed and the purse string was closed. The arterial duct was closed surgically with a vascular clip without transsection; pulse oxymetry on the lower limb confirmed persistence of adequate perfusion. The infant remained very stable throughout the procedure. The total procedural time was 16 min. The total fluoro-time was 3 min with a total radiation dose of 250 cGy/cm^2; total contrast volume 3 cc. The infant recovered well from the procedure and ventilator support could be stopped after 6 days.

After a few weeks the femoral pulses became progressively weaker, with increasing gradient across the stent (Doppler peak gradient of 40 mm Hg) and left ventricular hypertrophy. At the age of 3 months and a weight of 2.4 kg a percutaneous balloon dilatation of the stent was performed. We felt confident to safely put a 4 Fr sheath in the femoral artery and dilate the stent to the maximum of 5 mm with a 6 mm balloon (Maverick® Boston Scientific, Nanterre Cedex, France); this maneuver also shortened the stent from 8 to 6 mm length. At the age of 5 months, the stent was removed through a left lateral thoracotomy, and the arch was reconstructed with an oblique end-to-end anastomosis. The further clinical evolution was excellent without neurological sequelae or vascular problems.

DISCUSSION

Primary surgical correction of an aortic coarctation in low birth weight infants(<1.5 kg) is possible but with increased mortality and morbidity, especially if

the infant is in a critical state. The treatment typically consists of a single step extended end-to-end anastomosis, which in this subgroup is associated with a higher risk for recoarctation or residual hypoplasia [1,2,4–6]. Performing a thoracotomy in a low weight infant with unstable hemodynamics and limited respiratory reserve is not well tolerated: poor perfusion and oxygenation may result in cerebral accidents. Local complications such as bleeding, chylothorax, phrenic, or recurrent nerve paralysis are more likely to occur.

Percutaneous and hybrid interventions with stenting of the aortic coarctation have been proven to be safe alternatives [1,2,7,9]. A balloon dilation of the coarctation can be performed through a 3 Fr sheath but has a high incidence of early re- or residual coarctation [3,10]. A stent gives a very predictable result for some weeks to months, but requires a 4 Fr sheath. Accessing the "classical" vessels such as the femoral, carotid, or subclavian/axillary artery in these small infants has a high risk of vascular complications: bleeding, thrombosis, aneurysm, fistula, and even avulsion. The incidence of these problems rises exponentially in infants below 1.2 kg body weight [1–3,7]. Currently such access does not allow to deal with the arterial duct.

Due to progressive left ventricular dysfunction and excessive pulmonary flow, an intervention was necessary at a weight of 970 g. We preferred a two-step approach with "safe" access through sternotomy which allowed us to deploy a 3/8 mm stent in the aortic coarctation though a 4 Fr sheath in the ascending aorta; vascular access problems were thereby avoided. At the same time the surgeon could close the arterial duct using a vascular clip.

As our experience has grown over the past years in stenting the aortic coarctation in small infants, we now choose the length of the stent as short as possible which makes removal easier for the surgeon. Because of the associated isthmal hypoplasia, the stent must cover the whole length from the origin of the left subclavian artery until beyond the coarctation site, implanting the stent asymmetrically across the coarctation. The ideal stent diameter needs to be determined: we now aim for the size of the aortic cross; this implies a significant increase in size at the coarctation site, but the risk of tear of "fetal tissue" appears to be very limited. However we always have a covered stent in back-up on the shelf (Jostent Graftmaster, Abbott Vascular Int, Diegem, Belgium).

Most coronary stents can be dilated up to 5 mm; this allows postponing the corrective surgery to a higher body weight. Dilation of the stent will also reduce the length of the stent. The 2- (or 3-step) strategy increases the anterograde aortic flow which allows catch-up growth of the aortic cross. The final surgical procedure becomes easier with a "simple" end-to-end anastomosis at a higher weight, compared to a more difficult surgery in a very low weight infant requiring an extended end-to-end reconstruction at the proximal cross. In time one might chose for a stent that dissolves or can be dilated up to adult size; such stents have currently still important shortcomings as unpredictable dissolution, or need for multiple dilations with significant shortening and fractures requiring restenting.

All involved clinicians were impressed by the stability and fast recovery during and after this procedure, especially when compared to the anticipated destabilization of a procedure through a thoracotomy (including lung deflation and vascular clamp). The ascending aorta has previously been reported for vascular access [11,12]. We have been using a "dedicated" sternotomy in other situations in premature infants where vascular or cardiac access was required. We now no longer consider sternotomy with direct access as "last resort," but have "upgraded" this access to "first choice entry point" in selected critical ill premature or very small infants. The "price" of a sternotomy is limited, especially when comparing it with all possible complications of a full surgical procedure through thoracotomy or peripheral vascular access in a minute critical infant.

CONCLUSION

A two-step strategy involving a hybrid procedure with vascular access through sternotomy is a safe alternative in very low birth weight infants with important aortic coarctation avoiding many vascular access difficulties and other complications.

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