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“Intraoperative hybrid stenting of recurrent coarctation and arch hypoplasia with large stents in patients with univentricular hearts”

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ABSTRACT

Background: Obstruction of the reconstructed aortic arch, tubular hypoplasia and recurrent coarctation (RC) is an important risk factor in univentricular physiology. For the past two years we have adopted the concept of intraoperative hybrid stenting of RC and arch hypoplasia with large stents in patients with univentricular hearts as standard care procedure.

Method/Result: Retrospective analysis of the anatomy and procedural outcome of 14 patients was scheduled for intraoperative stenting of the aortic arch (12 during surgery for BCPS, 2 during Fontan completion). The median age was 5.3 months, weight 5.5 kg, height 62 cm. Five patients had tubular hypoplasia and 9 patients had distal stenosis of the aortic arch. Nine patients had a previous balloon dilatation. The mean diameter of the distal arch was 11.0 mm, at the coarctation 5.1 mm, at the level of the diaphragm 8.2 mm (CoA-index 0.62). Intraoperative stenting was performed in 13/14 patients. Stents were implanted with a mean balloon diameter of 10.8 mm (SD 3.4 mm). The achieved final mean diameter was 9.8 mm (mean, SD 2.8 mm) with an oversized Coa-index of 1.2. There was no re-coarctation at a mean follow-up of 7.3 months (range 3 to 24), the maximum flow velocity of 2 m/s across the stented lesion assessed by ECHO.

Conclusion: This hybrid approach is an easy and safe concept to manage recurrent aortic arch hypoplasia and stenosis. The use of large stents allows redilatation to adult size diameters later on.

1. Introduction

Obstruction of the reconstructed aortic arch, tubular hypoplasia and recurrent coarctation (RC) is known to be an important risk factor for patients after the Norwood procedure for hypoplastic left heart syndrome (HLHS) or similar lesions [1–3]. In contrast to native coarctation, the diagnosis of RC after the Norwood procedure is significantly more difficult. The reported incidence of RC ranges from 5% to 37% [4–7], depending on the definition of RC and it occurs when the reconstructed arch is narrowed below a certain limit. Measurement of the narrowest dimension of the distal arch, normalized to the distal diameter of the descending thoracic aorta, defined as “coarctation index” is a useful tool to define RC in this patient group [5]. Although balloon angioplasty (BA) has become the standard therapy for recurrent aortic arch obstruction, pressure gradient relief is often not complete, with a high incidence of restenosis after BA and occurs usually within the first year.

Whereas stent implantation for native or recurrent coactation has been proposed for native and recurrent coarctation or arch obstruction, in small infants with arch obstruction after Norwood I it has been evaluated less frequently [8], In general one very important prerequisite of stenting obstructive lesions of the aortic arch is to adapt the size of the stent to body growth and to have the option to redilate the stents to adult-size diameter. The use of stents without the ability to expand to an adult-size diameter should be reserved for bail-out situations and for acute stabilization of a patient with decreased ventricular function until the clinical situation allows further procedures. Those stents that show these kind of conditions demand for the implantation large implantation sheaths often not suitable for small patients [9–11]. In order to overcome the problem of sheath/vessel mismatch, the technique of anterograde stent implantation has been introduced [8]. This method reduces the risk of potential vascular arterial complications and can be performed in this group of patients with a acceptable risk. However the stents used in the literature so far still have the downside of an inadequate maximal size.

In those patients where percutaneous interventions may be associated with an increased risk due to limited vascular access or a very tortuous catheter course the combination of operative and interventional

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approaches (i.e. hybrid procedures) with direct puncture of the heart or the great vessels may facilitate implantation of even large devices [12, 13]. This technique has widely been used for perventricular VSD closure, intraoperative stenting of branch pulmonary artery stenosis or for the hybrid management of HLHS [14–18]. Intraoperative balloon angioplasty via the ascending aorta was first reported in 2000 [19] and was used in combination with the implantation of a coronary stent in a premature infant [20]. Only few case series exist with the combination of intraoperative hybrid stenting of RC in patients with univentricular physiology [4,12]. For the last two years we have adopted the concept of intraoperative hybrid stenting of RC and distal arch hypoplasia with large stents in patients with univentricular hearts as standard care procedure.

2. Methods

2.1. Data analysis

Retrospective analysis was performed of the files of all patients with univentricular physiology who had an intraoperative hybrid stenting of the distal aortic arch during the period of 1st of July 2013 until 30th of June 2015. Informed and written consent for the procedure was obtained by the parents for the procedure, the retrospective analysis was approved by the local ethical committee (Ref. no.: 36/2015) and additional consent was waived due to the retrospective character of the analysis.

2.2. Initial catheterization study

151 patients with univentricular physiology had a diagnostic catherization before the scheduled next surgical procedure i.e. bidirectional superior cavopulmonary anastomosis (BCPS) or Fontan completion by using an extracardiac conduit (Fontan). Patients with classical forms of the HLHS were treated with a Norwood procedure, patients with other anatomy and physiologically antegrade flow to the ascending aorta received a Damus–Kaye–Stansel operation and arch augmentation wherever necessary. During this catheter investigation hemodynamic assessment was performed including initial pressure gradients across the aortic arch, and, when necessary, balloon dilatation was performed thereafter. In 14/151 patients there was an inadequate result of the anatomy or after balloon dilatation. These patients were eligible for a subsequent hybrid approach. In addition the following anatomical measurements were performed: the diameter of the aortic arch at the level of the left subclavian artery (AoSub), the minimal diameter of the coarctation (CoA), the diameter of the aorta distal to the coarctation (CoApost), the diameter of the aorta at the level of the diaphragm (AoDia) and the ratio of CoA/AoDia i.e. the coarctation index [5]. For the hybrid procedure the appropriate balloon diameter and the stent size were calculated depending on the AoSub and CoApost diameters.

2.3. Measurements during the hybrid procedure

After obtaining access to the aorta a hand injection was performed to delineate the exact anatomy and the measurements were repeated as outlined above. AQrterial catheters had been inserted by the anesthesiologists in the right radial artery and in the right femoral artery and no additional pressure measurements were necessary during the procedure. After stent implantation a final angiography by hand injection was performed to assess the result of the procedure and the previous described anatomical measurements were repeated.

2.4. Hybrid procedure

The procedure was performed in the hybrid suite of the hospital [13]. The patients were anesthetized and surgical preparation was performed using standard techniques. In general the great vessels, the aortic arch and the heart were prepared for venous and aortic cannulation for the subsequent procedure (i.e. BCPS or Fontan completion). The procedure was performed without bypass and with the heart beating before or after the surgical procedure. If the stent implantation was performed first, a purse string suture was placed on the ascending aorta and a short sheath of the adequate size to allow placement of the balloon with the mounted stent was inserted into the ascending aorta (i.e. 8–10 F). Thereafter a hand injection was performed to assess the exact anatomy of the aortic arch (see Figs. 1 and 2). A 0.0035 standard guide wire was then advanced to the descending aorta and the stent mounted on the balloon was placed in the area of the coarctation. A control hand angiography was performed to confirm the accurate position and the balloon was inflated thereafter. After deflation and extraction of the balloon and the guide wire a final control angiography was performed n(see Fig. 1A–D). The sheath was exchanged for the aortic cannula and the surgical procedure was completed. If the stent implantation was performed after the surgical procedure, the aortic cannula had been removed and replaced by the sheath followed by stent implantation as described before.

In those cases with a relatively short arch and hypoplastic aortic arch it was necessary to place the balloon very proximal. To enable exact identification of the aortic wall, a forceps was placed at the outside of the aorta at the puncture site. This maneuver allowed careful maximal retraction of the sheath to the insertion site (see picture 2C and 2D).

If the required length of the balloon did not allow an adequate placement of the stent by using the technique described here, a short period of deep hypothermia with selective perfusion of the head was needed and the stent was placed under direct vision in the distal aortic arch. The stent has then been fixed with a single suture to the aortic wall. After closing the aorta and de-airing the full body perfusion has been instituted and the surgical procedure was completed during the rewarming phase.

2.5. Follow-up

All patients were followed up in our outpatient department by standardized clinical and echocardiographic assessment including non-invasive blood pressure measurement at the arm and leg and Doppler flow measurements across the distal aortic arch.

2.6. Statistics

The results are documented in tables in a descriptive manner and the values are expressed as mean, standard deviation of the mean and median wherever suitable.

3. Results

3.1. Patient characteristics

All patients had univentricular physiology and coarctation, some had additional aortic arch hypoplasia. All patients had undergone previous aortic arch repair. There were 10 patients with hypoplastic left heart syndrome (HLHS), one patient with imbalanced AVSD and coarctation, one patient with a borderline left ventricle, interrupted aortic arch, subaortic stenosis and coarctation, one patient with tricuspid atresia, transposition of the great arteries and aortic arch hypoplasia with coarctation, and 1 patient with double inlet left ventricle transposition and coarctation. Twelve patients received superior bidirectional cavopulmonary anastomosis. And the remaining 2 Fontan completion. The median age at surgery was 5.3 months (mean 8.6, SD 10.1), weight 5.5 kg (mean 6.5, SD 3.0), height 62 cm (mean 65, SD 12). The patient characteristics and anatomical diagnoses are depicted in Table 1.