Journal of Cardiology Cases 8 (2013) e93–e94

Contents lists available at www.sciencedirect.com

Journal of Cardiology Cases

journal homepage: www.elsevier.com/locate/jccase

Editorial

Stenting as a possible new therapeutic strategy to the obstructed TAPVC

Keywords:
Total anomalous of pulmonary vein connection
Stent
Catheter intervention

Total anomalous pulmonary vein connection (TAPVC), one of the diseases with extremely poor prognosis, is defined as the anomaly which the pulmonary veins have no connection with the left atrium but rather connect directly to one of the systemic veins or drain into the right atrium. The frequency of TAPVC is reported from 5.9 to 7.1 per 100,000 live births.

Independent risk factors for death of this anomaly from an international study comprised of earlier age at surgery, hypoplastic/stenotic pulmonary veins, associated complex cardiac lesions, postoperative pulmonary hypertension, and postoperative pulmonary vein obstruction [1]. The existence of obstructed lesion is the most critical for determining prognosis of this anomaly. An infant with obstructive TAPVC usually is acutely plunged into marked respiratory distress, cyanosis, and metabolic acidosis. Therefore, immediate relief of the obstruction is required and corrective surgery should be performed as soon as possible for this complicated cardiac anomaly as a basic idea.

Although intrinsic narrowing in the walls of the anomalous vessels occurs, extrinsic pressure frequently results in narrowing of the venous structure. The vertical vein in TAPVC to the innominate vein may be compressed between the left main pulmonary artery and left main bronchus. Similarly, the anomalous pulmonary vein in TAPVC to the SVC may be obstructed by the right pulmonary artery and trachea. Balloon dilation of extrinsic obstructed anomalous venous channels may be unsuccessful as Lock et al. [2] described. When the anomalous connection is to the portal vein, the hepatic sinusoids are interposed in the pulmonary venous channel and result in increased resistance to pulmonary venous return. Another factor that may contribute to impedance of pulmonary venous return is the length of the ascending or descending vertical venous pathway. Almost all these situation in infracardiac type TAPVC are hardly any candidates of catheter interventions.

On the other hand, it can be said that the many cases of TAPVC have obstruction mainly at the position where catheter intervention can be performed relatively with ease, because pulmonary venous obstruction is known to present in about 50% of supracardiac type [3]. As this report by Matsui et al. [4], the obstructed portions of the supracardiac type are usually approached from the internal jugular vein with ease.

In general, pulmonary venous angioplasty (class IIa) or stenting (class IIb) should be considered after surgical repair for TAPVC as an evidence level of C. In the American Heart Association (AHA) recommendation, pulmonary venous angioplasty and stenting should not be considered in the management of pulmonary vein stenosis associated with other congenital heart diseases that requires surgical intervention (class III) under the evidence level of C [5].

However, the idea of preoperative stabilization by temporarily relieving the obstruction is emerged recently to improve outcomes for neonatal obstructed TAPVR. Moreover, in the low birth weight neonate, percutaneous balloon angioplasty or stent placement maybe the only therapeutic options [6]. Palliative stent placement in obstructed TAPVC was technically feasible and efficacious in relieving pulmonary venous obstruction in low birth weight infant. Review of literature suggests that stent placement in obstructed TAPVC adequately improves obstruction and may be an effective temporizing measure prior to surgical repair in a more favorable clinical status [7,8]. Although AHA statement does not refer the management against native obstructed TAPVC, the guideline edited by Japanese Pediatric Inteventional Cardiaology (JPIC) added another comment as class III that, in the severe cases of obstructive TAPVC with complex cardiac anomaly, the pulmonary venous stenting may be allowed to avoid early surgical intervention which results in a very poor outcome as the evidence level of C [9].

Stent implantations were acutely effective in the focal relief of stenosis, with few procedural complications. However, stent obstruction and reintervention were common. Thus, stent implantation appears to serve a primarily palliative purpose in most patients with pulmonary vein stenosis [10]. Previous reports have suggested that stent diameter was required at least around 6–7 mm for prolonged freedom from restenosis in case of using bare metal stents [10]. It is thought that early restenosis is common after bare metal stent implantation for obstructive TAPVC in small babies who have vertical veins of much smaller than 9 mm in diameter. Therefore, when one uses bare metal coronary stent for obstructed lesion of neonatal pulmonary vein such as a Multi-Link VISION coronary stent Matsui et al. applied, restenosis must be recognized to occur within a short period.

Although the drug-eluting stent (DES) against obstructed TAPVR has been reported effectiveness for prevention of restenosis [11], limitation of the size of DES no larger than around 5 mm could not realize effective dilatation of stenotic lesion of pulmonary veins [9]. However, the issue of restenosis may be ignorable for bare-metal stenting to the obstructed TAPVR because stenting against this lesion must be basically alternative and bridging use in a neonatal period.

The obstructed TAPVR is frequently required urgent surgical intervention to neonates under critical conditions. It is obvious, as

DOI of original article: http://dx.doi.org/10.1016/j.jccase.2013.04.001.

http://dx.doi.org/10.1016/j.jccase.2013.05.001

the authors mentioned, that surgical intervention with cardiopulmonary bypass for critical patients is an inducer of worsening pulmonary edema, intracranial hemorrhage, and organ ischemia. Although the patient in this article was undertaken urgent stenting for releasing obstruction of pulmonary vein tract, the procedures the authors shown was safe as far as under well disciplined team. This provides that the stent implantation for obstructed TAPVR has possibility as the first choice treatment against neonates under extremely critical conditions.

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30 April 2013