

ORIGINAL ARTICLE

Combined Surgical and Interventional Approaches for Treating Patients with Congenital Heart Disease

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ABSTRACT ***Aims:*** **During the last decade the cooperation between surgeons and cardiologists has further expanded by combining surgical and interventional techniques (CCBSI) performed in the operating room, without the use of fluoroscopy. We sought to evaluate the results of our experience with CCBSI.** ***Methods:*** **All children with congenital heart disease (CHD) who underwent a CCBSI in the operating room between June 2007 and January 2014 were enrolled.** ***Results:*** **Sixty-eight patients were included. Median age at CCBSI was five months (range 1–48 months). The three main diagnoses leading to surgery included: (1) tetralogy of Fallot (TOF) (n = 40), (2) muscular ventricular septal defects (VSD) (n = 12), (3) single ventricle with pulmonary artery branch stenosis (n = 4). There were 72 catheter-based procedures associated with surgical maneuvres, including: (1) transatrial balloon dilation (BD) of the pulmonary valve (n = 45), (2) transinfundibular BD of the main pulmonary artery trunk (n = 12), (3) perventricular VSD closure with septal occluder (n = 8), (4) BD of pulmonary artery branches (n = 5), and other less common procedures (n = 2). There were no procedure-related complications and no hospital deaths. Median follow-up time was four years (range 0.95–7.9 years). There was one late death for respiratory distress after transapical balloon dilation of the aortic valve. One patient required BD and stenting of the left pulmonary artery branch 3.6 years after intraoperative BD for residual stenosis.** ***Conclusions:*** **The CCBSI represents a safe and effective treatment for selected patients with complex CHD. It will be helpful in minimizing patients' surgical trauma and in shortening or avoiding the use of cardiopulmonary bypass.** doi: 10.1111/jocs.12595 *(J Card Surg 2015;30:719–723)*

Minimally invasive operations and percutaneous interventions are now standard options for the treatment of congenital heart defects (CHDs).[1,2] In addition, hybrid cardiac surgery, defined as combined catheter-based and surgical interventions in either one setting or in a planned sequential fashion (within 24 hours),[3] has developed during the last decade, widening the spectrum of therapeutic options for patients with CHDs. For such procedures, the use of fluoroscopy is often necessary in the catheterization laboratory or in more sophisticated hybrid surgical operating rooms.[3,4]

During the last decade, following the technological advancements and the development of new surgical therapeutic options,[5–8] the cooperation between surgeons and cardiologists has further expanded, by combining surgical and interventional techniques which are performed in the operating room without the use of fluoroscopy. These procedures represent an emerging field, which combines skills and techniques that are part of the armamentarium of pediatric cardiac surgeons and interventional pediatric cardiologists, with the aim of overcoming the technical limits of each single specialty and therefore widening the spectrum of treatable CHDs.

In this study we sought to evaluate the results of our most recent experience with combined catheter-based and surgical interventions (CCBSI) in the operating room for treating selected children with CHD (Figs. 1–5).

Conflict of interest: The authors acknowledge no conflict of interest in the submission.

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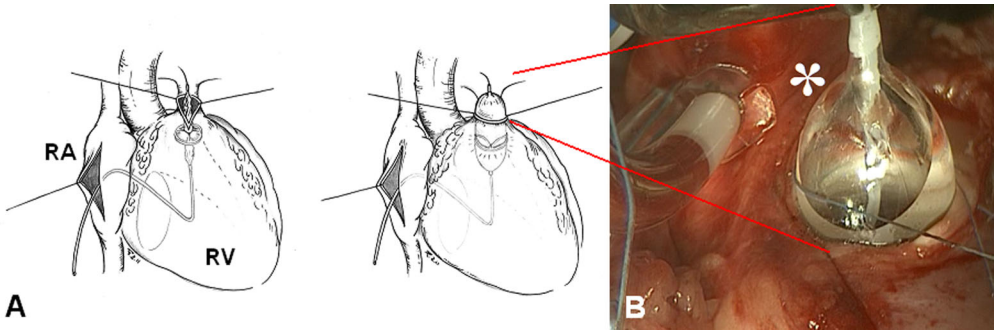

Figure 1. *(A) Intraoperative balloon dilation of the pulmonary valve; (B) intraoperative image showing the high-pressure balloon catheter (*) through the pulmonary valve. RA, right atrium; RV, right ventricle.*

MATERIALS AND METHODS

The clinical investigation committee authorized a review of medical records and the procedures followed were in accordance with the Institutional Guidelines for retrospective record review and protection of patient confidentiality. Patients are not identified, and the chairperson of the Ethics Committee of our institution gave consent for their data to be submitted for publication.

We reviewed data relative to the hospital course and clinical and instrumental follow-up of patients with CHD who underwent a CCBSI in the operating room without the use of fluoroscopy from June 2007 to January 2014. Patients who were followed for less than six months after surgery were excluded from the analysis.

All patients over 5 kg of body weight were monitored in the operating room with transesophageal (TEE) echocardiography. Intraoperative epicardial echocardiogram was routinely performed as an alternative method in patients with a body weight below 5 kg. The use of transthoracic 2D-echocardiography was also adopted in selected patients (i.e., in a premature patient with critical aortic stenosis).

Variables included patient's sex, age at surgery, type of congenital heart disease, type of surgical treatment, type of interventional procedure, the need for cardiopulmonary bypass and ischemic cardiac arrest and the occurrence of postoperative complications. Follow-up variables included patient's clinical data at last physical examination and postoperative instrumental evaluation.

Primary outcomes of this study included the evaluation of early and late surgical results, including patient's clinical status and the need for further hemodynamic and/or surgical procedures following the CCBSI.

Quantitative variables were summarized as median and range. Fisher's exact test was used for categorical variables, and Wilcoxon Rank Sum Test for quantitative variable. Data were analyzed using SAS software (SAS Institute Inc. SAS Stat 9.2®, Cary, NC, USA) and p-values below 0.05 were considered significant.

RESULTS

Sixty-eight patients were included. Median age at CCBSI was five months (range 1–48 months), median body weight 6 kg (range 0.8–12 kg). The most frequent diagnoses leading to CCBSI included: (1) tetralogy of Fallot (TOF) (n = 40), (2) ventricular septal defect +/− pulmonary artery banding (PAB) (n = 12), and (3) single ventricle with pulmonary artery branch stenosis (n = 4). Other diagnoses leading to surgery (n = 10) are listed in Table 1.

Sixty-six patients underwent 136 surgical procedures including pulmonary valve plasty (consisting of pulmonary valve commissurotomy, leaflet's delamination, augmentation, and resuspension) (n = 45), repair of tetralogy of Fallot (n = 40), main pulmonary artery "debanding" (n = 12), ostium secundum atrial septal defect closure (n = 9), ventricular septal defect closure (n = 8), ligation of patent ductus arteriosus (n = 6), bidirectional

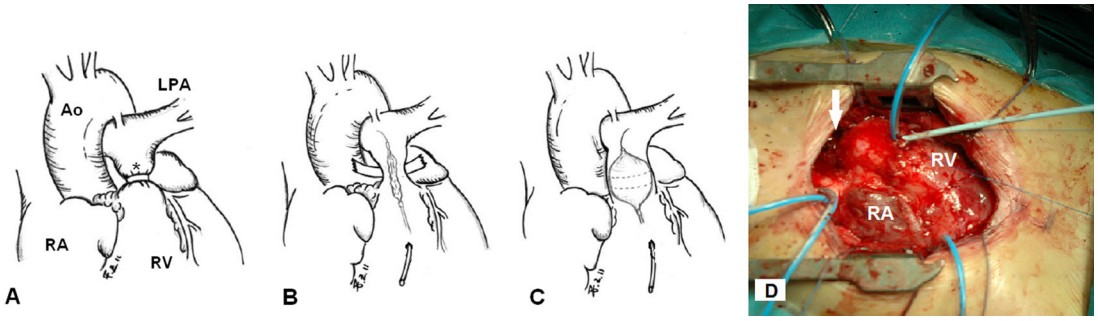

Figure 2. *(A–C) 2D-echo-guided intraoperative balloon dilation of the main pulmonary trunk through the infundibulum of the right venticle after surgical removal of the band. (D) Intraoperative image showing the balloon catheter insertion site (right ventricular infundibulum). RA, right atrium; RV, right ventricle; LPA, left pulmonary artery; Ao, ascending aorta. White arrow indicating the site of main pulmonary artery banding.*

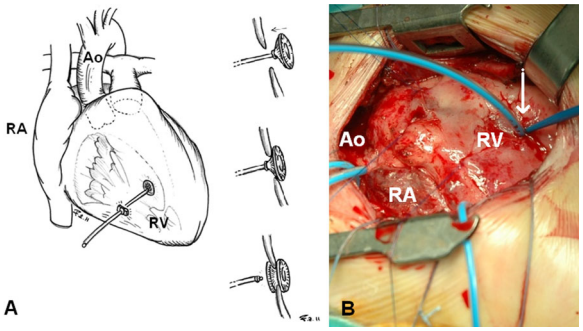

Figure 3. *(A) Intraoperative 2D-echo guided perventricular closure of a muscular venticular septal defect; (B) intraoperative image showing the right ventricular free wall and the catheter insertion site (white arrow). RA, right atrium; RV, right ventricle; Ao, ascending aorta.*

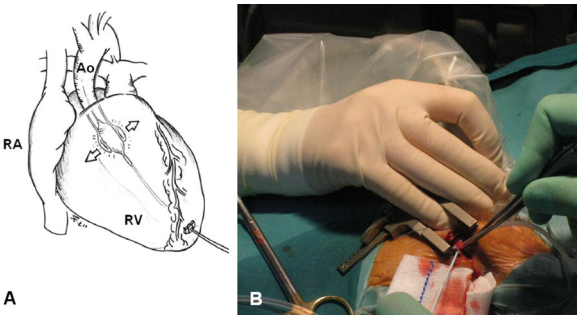

Figure 5. *(A) 2D-echo transapical ballon dilation of the aortic valve; (B) intraoperative image showing the apex of the left ventricle through a left anterior thoracotomy access (site of the insertion for the balloon catheter). LV, left venticle; RV, right venticle; Ao, ascending aorta.*

cavopulmonary anastomosis,[4] arterial switch operation (n = 2), double outlet right ventricle repair (n = 2), complete atrioventricular septal defects repair (n = 2), subaortic fibromuscular resection (n = 2), coronary artery plasty (n = 1), Fontan operation (n = 1), PA-IVS repair (PV commissurotomy + RVOT muscle band resection) (n = 1), and mitral valve plasty (n = 1).

There were 72 catheter-based procedures associated with surgical repair (Table 3) including: (1) open transatrial PV balloon dilation (n = 45), (2) 2D-echo-guided transinfundibular pulmonary trunk balloon dilation (BD) (status post-pulmonary artery banding) (n = 12), (3) 2D-echo guided trans-right ventricle VSD closure with septal occluder (n = 8), (4) open balloon dilation of pulmonary artery branches (n = 5). Other catheter-based procedures associated with surgical repair are listed in Table 2. Two patients (3%) underwent a longitudinal mid-line sternotomy in the operating room followed by transapical right ventricular outflow tract stent implantation and a left anterior thoracotomy followed by a transapical balloon dilation of the aortic valve, respectively.

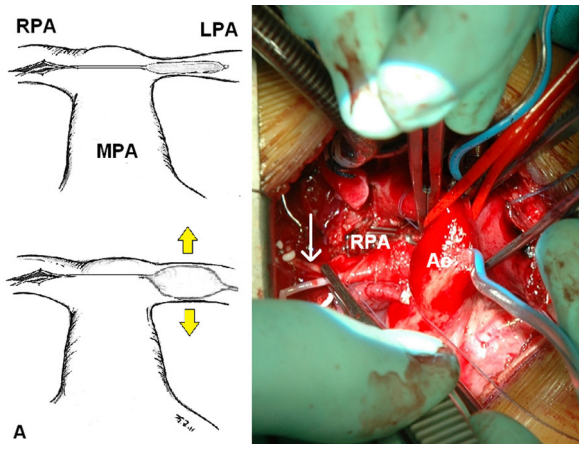

Figure 4. *(A) Intraoperatory balloon dilation of the left pulmonary artery branch through a right pulmonary branch access; (B) intraoperative image showing the balloon catheter insertion site (white arrow). LPA, left pulmonary artery; RPA, right pulmonary artery; MPA, main pulmonary artery; Ao, ascending aorta.*

In 62 patients (91.8%), the median cardiopulmonary bypass (CPB) time was 134 minutes (range 67–237 minutes). In six patients (8.8%) where CPB was not employed, a TEE 2D-echo guided trans-right ventricle VSD closure and the balloon dilation of the pulmonary trunk after banding was performed in four, a transthoracic 2D-echo guided transapical balloon dilation of the aortic valve in one, and a combined TTE and epicardial 2D-echo guided trans-right ventricular apex perforation of the pulmonary valve and position of a stent in the right ventricular outflow tract, in another. Five patients, who underwent intraoperative balloon dilation of either pulmonary artery branches, underwent a concomitant bidirectional cavopulmonary anastomosis (n = 4) and an extracardiac conduit Fontan palliation (n = 1).

There were no procedure-related complications and no early deaths. Median intensive care unit stay was three days (range 1–19 days). Fourteen patients (20%) had 21 postoperative/postprocedural complications including: arrhythmias (n = 9), low-output syndrome requiring inotrope infusion (n = 4), acute renal failure requiring peritoneal dialysis (n = 2), urinary tract infections (n = 2), pleural effusion requiring drainage (n = 2), pneumonia (n = 1), bleeding requiring surgical revision (n = 1). All patients were discharged home in good clinical condition without significant residual intracardiac lesions.

Follow-up

Median follow-up time was four years (range 0.95–7.9 years). There was one late death in a premature 890-g patient who died from severe respiratory distress 40 days after a successful transapical balloon dilation of the aortic valve.[9] The remaining 67 patients were asymptomatic, in good clinical condition and did not require further reoperation following the CCBSI. One patient had lung underperfusion (<30%) at routine scintigraphy 3.6 years after intraoperative balloon dilation, required a repeated transcutaneous left pulmonary artery balloon dilation and stenting. Follow-up outcomes according to the type of combined hemodynamic procedure are summarized in Table 3.

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TABLE 1
Main Diagnosis Leading to Combined Approach (n = 68 Patients)

Main Diagnosis	Number of Patients
Tetralogy of Fallot	40
Ventricular septal defect (VSD) Apical (n = 3)/multiple (n = 7) VSD s/p pulmonary artery banding (n = 9*) VSD with associated congenital heart disease (n = 3**)	12
Single ventricle hearts With pulmonary artery branches stenosis (n = 4***) With pulmonary artery atresia (n = 1) s/p pulmonary artery banding (n = 1)	6*
Transposition of the great arteries with muscular ventricular septal defect	2
Double outlet right ventricle With multiple ventricular septal defect (n = 1) With severe pulmonary valve stenosis (n = 1)	2
Complete atrioventricular septal defects s/p pulmonary artery banding (n = 1) With pulmonary valve and infundibular stenosis (n = 1)	2
Pulmonary stenosis and atrial septal defect	1
Pulmonary atresia-intact ventricular septum	1
Critical aortic stenosis (in a 890 g premature patient)	1
Coronary artery stenosis, subaortic stenosis, pulmonary valve, and subpulmonary stenosis	1

*Three patients underwent also a previous coartectomy.
**Mitral valve dysplasia s/p PAB (n = 1), pulmonary valve stenosis (n = 1), right pulmonary artery stenosis (n = 1).
***Left pulmonary artery stenosis (n = 2), right pulmonary artery stenosis (n = 3).

TABLE 2
Hemodynamic Procedures (n = 72 in 68 Patients)

Hemodynamic Procedure	Number of Patients
Transatrial PV BD	45
Transinfundibular BD of the MPAT	12
Perventricular VSD closure with septal occluder	8
BD of pulmonary artery branches	5
Transapical RVOT stent implantation	1
Transapical BD of the aortic valve	1

BD, balloon dilation; MPAT, main pulmonary artery trunk; PV, pulmonary valve; RVOT, right ventricular outflow tract; VSD, ventricular septal defect.

DISCUSSION

Improved surgical and catheter intervention techniques have stimulated surgeons and interventional cardiologists to further expand the concept of minimally invasive treatment.[1,10,11] Consequently, during the last few years, we employed CCBSI in selected patients with the aim of overcoming the technical limits of each single specialty. The development of these new treatment strategies allowed us to reduce interventional complications and achieve good clinical early and mid-term functional results. Furthermore it allowed us to widen the spectrum of treatable CHDs by enabling new treatment options, particularly in patients with complex CHDs or complex vascular situations.[2,4]

In the absence of sophisticated hybrid suites and fluoroscopic imaging, such procedures have been performed and developed in tandem by pediatric cardiac surgeons and cardiologists, either in the operating room or in the catheterization laboratory, with the aid of 2D-transesophageal, epicardial, or transthoracic echography. Moreover, the involvement of the hemodynamic expertise in the operating room led to the development of new strategies aimed to improve both early and long-term outcomes (i.e., the preservation of the PV integrity and function during early repair of TOF). In such patients, a balloon PV annuls dilation during standard early trans-atrial/transpulmonary repair proved to be effective in preserving both PV integrity and RV function, in the mid-term, in this way improving the results of ''conventional'' TOF repair by means of a transannular valved patch.[5,6] Furthermore, during our more recent experience with this innovative technique we have been able to preserve the PV function even in more severe forms of TOF (with PV Z-score <-3) by adding complex PV surgical reconstruction maneuvres, following effective annular dilation.[6] Moreover, as our experience with this technique developed further, we were able to preserve very stenotic and hypoplastic PVS associated with other congenital heart malformations (i.e., double outlet right ventricle, complete atrio-ventricular septal defects, atrial septal defect).

During the last few years, we have further promoted the applicability of new CCBSI techniques in selected patients, and we have further extended the concept of interaction between cardiologists and cardiac surgeons both in the operating room, and in the catheterization laboratory, that is, with the combined transinfundibular balloon dilation of the pulmonary trunk following pulmonary artery banding and the transventricular VSD closure without the need of CPB,[12–14] or the intraoperative balloon dilation of the pulmonary artery branches in patients with central stenosis, after initial palliative staged approach (i.e., systemic-to-pulmonary shunts, PDA stenting etc.).

With regards to the transinfundibular balloon dilation of the pulmonary trunk following pulmonary artery banding, the main advantage of this technique, over a more conventional pulmonary artery de-banding and patch augmentation of the pulmonary trunk, clearly avoids or reduces the use of CPB, as well as avoiding the use of any patch material. This procedure can be performed safely with good mid-term results; in fact none of our patients in this specific group required a reintervention or a re-operation for residual supravalvar pulmonary stenosis.

We have had very limited experience (n = 5 patients) with pulmonary artery branch dilatation during staged approaches in patients with single ventricle physiology. However, we suspect that this procedure can be carried

TABLE 3
Follow-up Outcomes (n = 67 Patients)

Hemodynamic Procedures (no. of pts)	Median Follow-up Time, Years (Range)	Status
Transatrial PV BD (n = 45)	3.7 (0.75–7.5)	All alive PV regurgitation was none/mild in 40 patients (85%) and moderate in seven patients (15%)
Transventricular BD of the MPAT (n = 12)	6 (1–7.6)	All alive Median residual gradient of 15 mmHg (range 10–25)
Transventricular VSD closure (n = 8)	5.6 (1–7.6)	All alive No residual intracardiac shunts
BD of pulmonary artery branches (n = 5)	5.4 (1.2–6.6)	All alive No residual significant stenosis*
Transapical RVOT stent implantation (n = 1)	3.4	Alive underwent successful BCPS

BCPS, bidirectional cavopulmonary shunt; BD, balloon dilation; MPAT, main pulmonary artery trunk; PV, pulmonary valve; RVOT, right ventricular outflow tract; VSD, ventricular septal defect.
*One patient underwent balloon dilation and stenting of the left pulmonary artery branch for residual stenosis.

out easily under direct vision in the operating room (and without the use of fluoroscopy) allowing an optimization of pulmonary artery diameter, which is a fundamental step toward Fontan repair. Pulmonary artery branches are usually very delicate and fragile in patients after previous surgical interventions. We never isolate the pulmonary artery branches extensively before intraoperative balloon dilation, since the external radial force generated by the scar tissue surrounding the vessel is helpful in balancing the internal force of the balloon during the dilation phase, especially in cases of moderate-severe or long stenosis. Other potential advantages of this technique are the avoidance of an extensive PA tissue dissection, which may be responsible for future re-scarring with consequent external luminal compression, and refraining from the use of prosthetic materials, especially in cases of a small pulmonary vascular tree.[14]

This study has a few limitations: (1) it is a retrospective data examination of the patients who underwent CCSBI for treating their CHD and (2) patient population is small in a context of a wide spectrum of applications. Therefore, we cannot reach definitive conclusions on the efficacy of each different treatment strategies. Nonetheless we were able to safely combine different surgical and catheter-based techniques with the aid of 2D-echo in the operating room for the treatment of patients with simple and complex CHD, with good mid-term results, and has led to improved outcomes compared to standard surgical correction in patients with TOF.[6]

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