ORIGINAL ARTICLE

Hybrid Strategy in the Management of Congenital Heart Defects. Initial Results in a Multicenter Experience

Estrategia híbrida en el manejo de cardiopatías congénitas. Resultados iniciales en una experiencia multicéntrica

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ABSTRACT

Background: The hybrid approach implies the close collaboration between surgical and interventional cardiology teams for the treatment of complex congenital heart diseases, to reduce the number of interventions by using less invasive techniques, avoid the limitations due to the size of vascular accesses and the use of cardiopulmonary bypass.
Objectives: The aim of this study is to evaluate the indications, techniques, initial results and short-term follow-up of the hybrid approach.
Methods: We conducted a retrospective analysis of patients treated with hybrid approach between March 2014 and September 2020. Fourteen procedures were performed in 13 patients. Mean age was 16.8 months (1 day-13 years); 50% were neonates, 57% were male and mean weight was 9 kg (1.9-70 kg). The hybrid procedures performed included stenting in coarctation of the aorta (4 patients), ductal stenting (3 patients), stenting of pulmonary artery branches (1 patient), neonatal aortic valvuloplasty (4 patients) and perventricular closure of muscular ventricular septal defect (2 patients). The access used was dissection of the carotid artery in 8 procedures and sternotomy in the remaining 6.
Results: All the procedures were successfully completed, except for two patients: one died due to ventricular fibrillation after the procedure, and another in the immediate postoperative period due to severe coagulation abnormality. Mean follow-up of the remaining 12 patients was 19 months with favorable outcome.
Conclusions: The initial results with the use of a hybrid strategy were satisfactory in terms of survival and improvement of the hemodynamic parameters. The selection of cases, teamwork and appropriate follow-up are key factors to achieve satisfactory results.

Key words: Cardiac Surgical Procedures / methods - Heart Defects, Congenital / surgery

RESUMEN

Introducción: El abordaje híbrido implica la colaboración estrecha de los equipos quirúrgico y de hemodinamia para el tratamiento de cardiopatías congénitas complejas, con el objetivo de disminuir el número de intervenciones a través de técnicas menos invasivas y de evitar las limitaciones dadas por el tamaño de los accesos vasculares y la utilización de circulación extracorpórea.
Objetivos: Evaluar indicaciones, técnicas, resultados iniciales y seguimiento a corto plazo del abordaje híbrido.
Material y métodos: Revisión retrospectiva de pacientes abordados de manera híbrida desde marzo de 2014 hasta septiembre de 2020. Se realizaron 14 procedimientos en 13 pacientes. Edad media de 16,8 meses (1 día-13 años), el 50% fueron neonatos y el 57% de sexo masculino; el peso medio fue 9 kg (1,9-70 kg). Los procedimientos híbridos realizados incluyeron implante de stent en coartación de aorta (4 pacientes), implante de stent ductal (3 pacientes), implante de stent en ramas pulmonares (1 paciente), valvuloplastia aórtica neonatal (4 pacientes) y cierre de comunicación interventricular muscular (2 pacientes). El acceso se realizó mediante disección carotídea en 8 procedimientos y esternotomía en los 6 restantes.
Resultados: Todos los procedimientos pudieron completarse exitosamente, aunque un paciente presentó fibrilación ventricular posprocedimiento y otro falleció durante el posoperatorio inmediato debido a trastorno grave de la coagulación. El seguimiento de los 12 pacientes restantes fue en promedio de 19 meses y la evolución fue favorable.
Conclusiones: Utilizando una estrategia híbrida, los resultados iniciales en términos de sobrevida y mejoramiento de parámetros hemodinámicos fueron adecuados. La selección de casos, el trabajo en equipo y el seguimiento apropiado son cruciales para lograr resultados satisfactorios.

Palabras clave: Procedimientos quirúrgicos cardíacos/métodos - Cardiopatías congénitas/cirugía

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Abbreviations

CCHD	Complex congenital heart disease	**HLHS**	Hypoplastic left heart syndrome
CPB	Cardiopulmonary bypass	**VSD**	Ventricular septal defect
CoA	Coarctation of the aorta	**PDA**	Patent ductus arteriosus
RCoA	Recoarctation of the aorta		

INTRODUCTION

During the last decade, the surgical results of complex congenital heart diseases (CCHDs) have improved significantly due to the increased use of interventional procedures to correct these conditions. The hybrid approach demonstrates the close collaboration between surgical and interventional cardiology teams, where each team uses the best techniques in their respective fields in order to reduce the number of interventions through less invasive procedures, avoid the limitations due to the small size of vascular accesses in neonates and infants, and reduce cardiopulmonary bypass (CPB) and circulatory arrest times. The hybrid approach also allows timely planning of the most challenging cases that would benefit from a combined procedure and provides treatment options that are tailored to individual needs. (1-4)

The aim of our study was to evaluate the indications, techniques, initial results, and short-term follow-up of the hybrid approach in different centers dedicated to the treatment of patients with CCHDs.

METHODS

We conducted a retrospective, multicenter, descriptive, observational study of patients undergoing a hybrid approach from March 2014 to May 2020 in different medical centers in the city of Córdoba. All the procedures were performed by the same team. The indication of hybrid approach in each patient was decided by agreement between cardiologists, cardiovascular surgeons and interventional cardiologists of each participant center. Demographic characteristics, type of congenital heart disease, type of access, hybrid procedure performed, its complications, initial results and short-term follow-up were included in the analysis.

Statistical analysis

Categorical variables are expressed as percentage and continuous variables as mean and interval. All the statistical calculations were performed using InfoStat/P software package (*Universidad Nacional de Córdoba*, 2018).

Ethical considerations

The study was conducted following the recommendations of the Declaration of Helsinki for observational studies and was approved by the institutional review board of the participant centers, but it did not include an informed consent form signed by the patients.

RESULTS

From March 2014 to September 2020, 14 hybrid procedures were performed in 13 patients (Table 1). Mean age was 16.8 months (interval: 1 day-13 years) and 50% were neonates. Mean weight was 9 kg (range: 1.9-70 kg) and 57% corresponded to the male sex. The procedures performed included hybrid stenting of coarctation of the aorta (CoA) (28%), ductal stenting (20%), stenting of pulmonary artery branches (7%), neonatal aortic valvuloplasty (28%) and perventricular closure of muscular ventricular septal defect (VSD) (14%) (Table 3).

Four stents were implanted for CoA, 2 in patients with diagnosis of native long-segment CoA and the other 2 in patients with recoarctation of the aorta (RCoA): one after surgery with severe hemodynamic impairment and the other in a patient with a variant of hypoplastic left heart syndrome (HLHS) with severe distal RCoA during the Norwood procedure, developed after the stage of bidirectional Glenn procedure.

A patient with tetralogy of Fallot after complete repair developed severe stenosis of the conduit placed between the right ventricle and the pulmonary artery branches, associated with severe diffuse stenosis of the left pulmonary branch, requiring stent implant in that branch during surgery.

Three stents were implanted in patients with patent ductus arteriosus (PDA)-dependent systemic flow. One stent was implanted to a neonate with diagnosis of fetal critical aortic valve stenosis and severe left ventricular dysfunction/dilation who underwent hybrid neonatal aortic valvuloplasty. Another patient underwent ductal stenting and bilateral pulmonary artery branch banding during the same procedure due to significant aortic valve and aortic arch hypoplasia associated with large malalignment-type VSD. The third case was a patient with diagnosis of HLHS with a history of bilateral pulmonary artery branch banding and atrial septostomy.

Two patients underwent perventricular closure of muscular VSD; the procedure was associated with removal of the pulmonary artery band in one patient and with PDA ligation in the other.

Finally, four neonates with severe aortic valve stenosis underwent hybrid aortic valvuloplasty.

The hybrid approach was performed with dissection of the right carotid artery in 8 patients: as vascular access for aortic valvuloplasty in 4 patients and for stenting of coarctation of the aorta in the other 4. Median sternotomy was used in 6 patients (for perventricular closure of VSD in 2 patients, ductal stenting in 2 patients with right ventricular outflow tract puncture and ductal stenting with puncture of the main pulmonary artery in another patient, and 1 stenting of the left pulmonary artery branch through surgical opening of the main pulmonary artery). Two of the interventions were performed in patients who

Table 1. Clinical data and detail of the hybrid procedures

P	Age	weight (kg)	Diagnosis	Procedure	Approach	Associated cardiac anomalies	Additional simultaneous procedures	Complications
1	15 d	2.5	Critical ReCoA of distal transverse arch	Aortic Stenting	RCA dissection			
2	7 m	5.2	Critical juxtaductal CoA + hypoplasia of the isthmus	Aortic Stenting	RCA dissection			VF, torsade de pointes
3	12 m	11	Critical juxtaductal CoA + hypoplasia of the isthmus	Aortic Stenting	RCA dissection			
4	22 d	8	Critical RCoA	Aortic Stenting	RCA dissection	HLHS variant - Norwood procedure, Glenn procedure		
5	13 y	70	Critical LPAB stenosis	LPAB stenting	Re-sternotomy (CPB)	Corrected TOF		Massive bleeding Died
6	27 d	2.5	Severe hypoplasia of the AV, AoA, VSD, PDA	Ductal stenting	Sternotomy	OS ASD	Bilateral PA banding	
7	17 d	2.3	Critical AV stenosis after hybrid valvuloplasty	Ductal stenting	Re-sternotomy	PA branch banding, restrictive PDA		
8	2 m	1.9	HLHS	Ductal stenting	Re-sternotomy	PA branches banding, atrial septostomy		
9	1 d	2.5	Critical AV stenosis	Aortic valvuloplasty	RCA dissection			
10	1 m	2.7	Critical AV stenosis	Aortic valvuloplasty	RCA dissection			
11	2 m	4	Critical AV stenosis	Aortic valvuloplasty	RCA dissection			Mild to moderate AVR
12	22 d	3	Critical AV stenosis	Aortic valvuloplasty	RCA dissection	Shone's syndrome		
13	7 m	4.5	Apical muscular VSD	Perventricular VSD closure	Sternotomy	CoA repaired, main PA banding	Removal PA banding	
14	22 m	8	Midmuscular VSD	Perventricular VSD closure	Sternotomy	PDA	PDA ligation	

AoA: aortic arch; OS ASD: ostium secundum atrial septal defect; AV: aortic valve; AVR: aortic valve regurgitation; CoA: coarctation of the aorta; CPB: cardiopulmonary bypass; HLHS: hypoplastic left heart syndrome; LPAB: left pulmonary artery branch; VSD: ventricular septal defect; PA: pulmonary artery; PDA: patent ductus arteriosus; RCA: right carotid artery; RCoA: recoarctation of the aorta; TOF: tetralogy of Fallot; VF: ventricular fibrillation.

Table 2. Demographic characteristics and approaches

Age	16.8 months (1 day-13 years)
Weight	9 kg (1.9-70 kg)
Sex	57% male/ 43% female
Access	Dissection of the right carotid artery: 57%
	Sternotomy: 43%

Table 3. Hybrid procedures performed

Aortic stenting	4 (28.5%)
Ductal stenting	3 (21.4%)
Pulmonary artery branch stenting	1 (7.1%)
Aortic valvuloplasty	4 (28.5%)
Closure of ventricular septal defect	2 (14.2%)

required reopening of a previous sternotomy and one patient underwent reopening of the sternotomy for the fourth time, a procedure performed with CPB and circulatory arrest. There were no complications associated with the approach.

All the scheduled procedures were successfully completed. One patient with stenting of CoA developed ventricular fibrillation 3 hours after the procedure and another patient presented bleeding in the right internal iliac artery requiring surgical repair 7 days after the initial procedure. In another case, a 13-year-old patient died due to uncontrollable bleeding after the fourth resternotomy for replacement of the conduit between the right ventricle and the main pulmonary artery and hybrid stenting of the left pulmonary branch, a complex procedure performed with CPB and circulatory arrest.

After hospital discharge, all the patients who survived were monitored clinically and by color Doppler echocardiography during an average follow-up of 19 months (1-75 months) and had a favorable outcome. The 4 patients who required stent implant due to aortic arch obstruction did not present significant gradients. The two patients requiring devices for VSD closure evolved without residual shunts. Of the 4 patients with neonatal aortic stenosis, 3 are still free from reintervention. One of these patients presented a significant increase in the pressure gradient that required a second percutaneous aortic valvuloplasty, with satisfactory results. Two of the 3 patients with ductal stenting are waiting for definitive surgery of their CCHD. The remaining patient underwent bilateral pulmonary artery branch banding removal with balloon dilation and percutaneous occlusion of the ductal stent with a vascular plug, with favorable outcome.

DISCUSSION

Historically, the relationship between cardiovascular surgeons and interventional cardiologists has been competitive. With the progress of interventional techniques, together with better surgical results in the management of CCHDs, it is reasonable to think that the new challenges for both disciplines can be addressed and solved more effectively through collaborative efforts. It is clear that there must be absence of professional or economic competition and mutual respect for the individual talents and limitations to achieve this target. In addition, the members of the intervening cardiovascular multidisciplinary team should engage in frank discussions.

In our Children and Adult Congenital Heart Disease Program, we started with hybrid procedures with defined objectives, which included reducing morbidity and mortality of complex patients, improving quality of life, providing more efficient and cost-effective individual care and, above all, reducing the cumulative impact of multiple interventions that these complex patients undergo throughout their lives. This collaborative approach offers several advantages. Percutaneous procedures are quicker because they use more direct and safer accesses, allowing appropriate anatomical inspection so that surgeons can take early action if necessary. Another advantage is the possibility of performing procedures with few limitations in terms of age or weight and that, in many cases, CPB is not required. (5)

In cases requiring intraoperative stenting of pulmonary artery branches, rigid or flexible bronchoscopy can be used to obtain direct "live" visualization of the lesion and for postoperative control. (6) In addition, the hybrid approach avoids proximal lobar and segmental branch involvement, allows for significant decrease in CPB times and avoids extensive retro and para-aortic dissections which are sometimes necessary in cases of surgical repair alone. (7)

In interventions for perventricular closure of muscular VSDs, we eliminated the problem of vascular access in small patients and were able to perform the interventions at earlier ages (patients weighing <5 kg). We also avoided the use of large sheaths to place the device, reduced or eliminated the use of CPB and reduced the persistence of residual shunts in cases of complex interventricular septal defects extending to the ventricular apex. Alternatively, it can be used in patients with contraindications for CPB or in those with failed attempts of percutaneous or surgical closure. (8, 9)

For the treatment of neonatal aortic stenosis, we used the carotid access to perform aortic valvuloplasty, since the diameter of the carotid artery in neonates is greater than that of the femoral artery. This access prevents the complications associated with the femoral access, such as thrombosis in high-risk patients, especially neonates weighing less than 4 kg. It also facilitates catheter advancement in tortuous anatomies or in the presence of acute angles during certain procedures and provides a safer and more effective route towards the lesion to be treated. (10, 11) It also avoids the possible mitral valve injury associated with aortic valvuloplasty via antegrade access. (12-14) The

same hybrid approach via the carotid access for stent placement in CoA and RCoA allows the use of larger sheaths to advance stents that can be easily dilated up to adult vessel diameters. (15, 16)

The hybrid approach is an established treatment option as an initial or stage I palliative treatment of HLHS and its variants, and includes bilateral pulmonary artery banding, PDA stenting and balloon atrial septostomy. It is a useful approach in high-risk patients, as it avoids CPB and hypothermic circulatory arrest in neonates. High risk patients include those with birth weight <2.5 kg, prematurity, ventricular dysfunction, severe hypoplasia of the ascending aorta, genetic abnormalities and other severe comorbidities. (17, 18) The results of the hybrid approach in HLHS are encouraging, confirming the importance of collaboration between the treating teams in the management of these patients with severe CCHDs.

In our program, the hybrid strategy for patients presenting with HLHS is carried out in the catheterization laboratory, where the cardiovascular team performs the sternotomy and initially places banding on both pulmonary artery branches. Next, the right ventricular outflow tract (or the main pulmonary artery) is punctured, and a short sheath is placed for the initial angiography to visualize the indentation of the banding on both pulmonary artery branches and the PDA anatomy/diameter. Finally, the selected stent is advanced through this sheath and implanted in the ductus. If necessary, percutaneous atrial septostomy is performed in a second intervention.

This hybrid approach for HLHS is based on the rationale that avoiding CPB and circulatory arrest during the neonatal period will improve myocardial function in the long term. In addition, delaying aortic arch reconstruction (which requires CPB/circulatory arrest) until the age of 4-6 months will improve long-term neurological outcomes. Although it is still a hypothesis, we believe that an infant younger than 4-6 months leaving the operating room with serial circulation (Glenn anastomosis) is more stable than a neonate leaving the operating room with balanced circulation (Blalock-Taussig or Sano shunt) after a similar procedure. The debate is ongoing, and multiple research studies are currently being conducted to answer these questions.

CONCLUSION

In our pioneering multicenter experience, the initial results of a hybrid strategy for the treatment of CCHDs showed satisfactory results in terms of survival and improvement of hemodynamic parameters. The procedures are technically safe and efficient. Detailed planning and teamwork are essential for the success of the hybrid technique. A larger number of cases with longer follow-up will be required to confirm the initial observations.

Conflicts of interest

None declared.

(See authors' conflicts of interest forms on the website/ Supplementary material)

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