External shunt versus internal shunt for off-pump Glenn

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Background
Off-pump bidirectional Glenn (BDG) operation can be associated with elevation of superior vena cava (SVC) pressure that may lead to neurological damage.
Aim
Off-pump BDG operation was done using either a veno-atrial shunt or external shunt to decompress SVC during clamping.
Patients and methods
A prospective, randomized comparative study in a single tertiary care cardiac center where 30 patients with functional single ventricle underwent off-pump BDG. Group I with a veno-atrial shunt (internal) and group E with an external shunt.
Measurements and main results
There was no early hospital mortality. The mean SVC pressure during clamping was 40.4±3.4 mmHg before and 28.5±3.8 mmHg after shunt opening in group I and 37.6±4.5 mmHg before and 26.4±2.1 mmHg after shunt opening in group E. The mean clamp time was 19.8±3.5 min in group I and 16.9±4.4 min in group E. The transcranial pressure gradient was 58.1±6.89 mmHg in group I, while 54.86±9.1 mmHg in group E. There were no major neurological complications apart from treatable convulsions in one (3%) case in group I and delayed recovery in one (3%) case in group E.
Conclusions
Off-pump BDG can be safely performed with either external or internal shunt avoiding cardiopulmonary bypass complications.

Keywords:
bidirectional Glenn shunt, congenital cyanotic heart disease, internal shunt, off-pump surgery, univentricular heart

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Introduction

The bidirectional Glenn (BDG) shunt is performed for cyanotic congenital heart defects, with single-ventricle pathology [1,2]. The end-to-side anastomosis of the superior vena cava (SVC) to the right or left pulmonary artery may be converted to a total cavopulmonary connection later and effectively increases arterial blood oxygen saturation and decreases the ventricle volume overload.

The BDG is usually performed with cardiopulmonary bypass (CPB) with its associated complications. The conduct of this operation without CPB can be associated with elevation of SVC pressure that may lead to neurological damage.

However, the safety of performing BDG without CPB was reported by many authors, but with some decompression techniques of the SVC at the time of clamping [3–11].

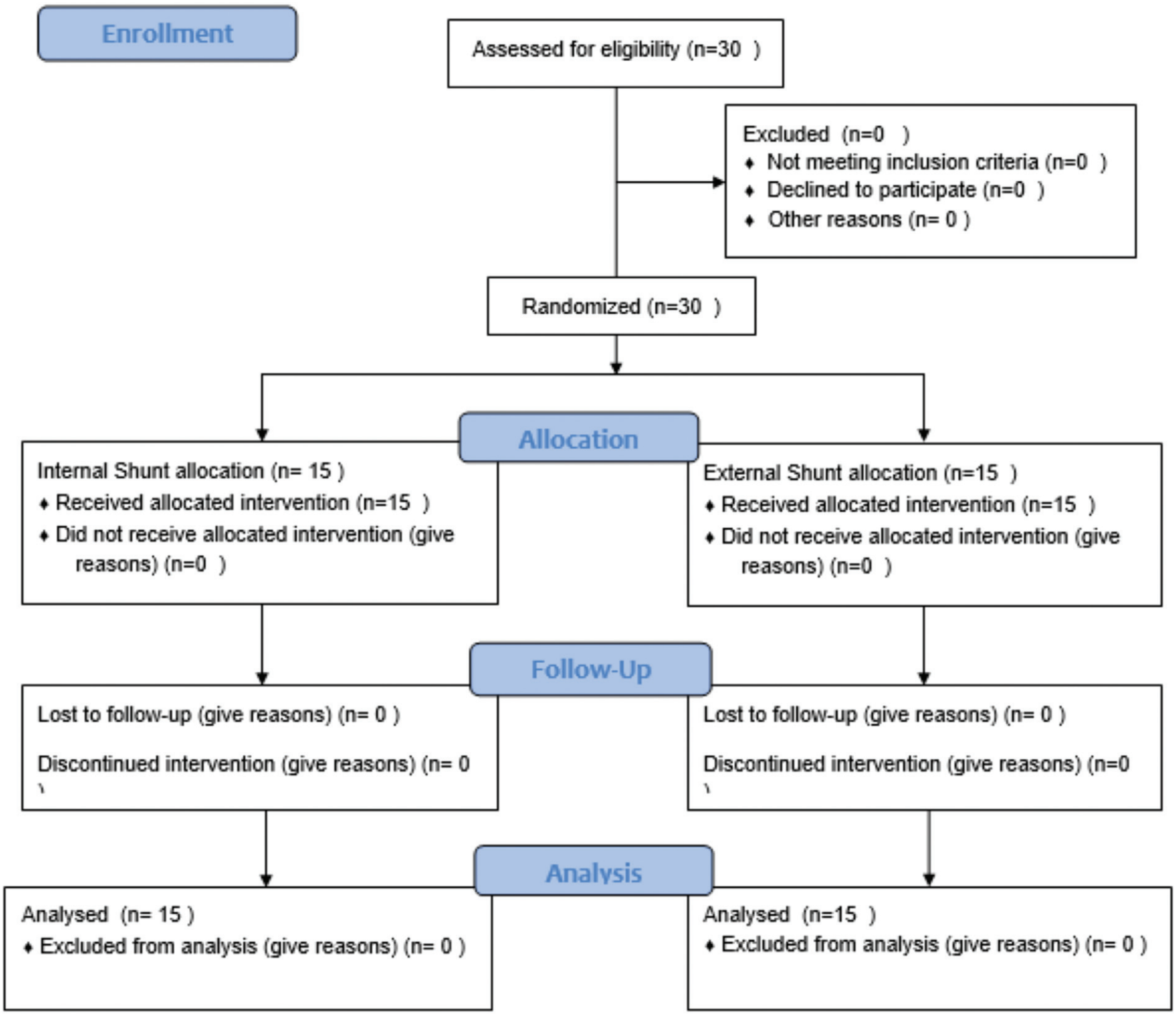
We reported our results with off-pump BDG with two various techniques used to drain the SVC blood during clamping.

Patients and methods

The trial has been registered on the ClinicalTrials.gov platform with identifier NCT05294718 after approval by the local ethical committee on clinical investigation and guardian written informed consent. The protocol structure was written in compliance with the Consolidated Standards of Reporting Trials (CONSORT) 2010 Statement guidelines.

A prospective randomized study of 30 patients suffering from functionally single ventricle who were palliated using a BDG shunt was conducted. The decision to do the procedure without CPB was made after a complete evaluation with echocardiography. They were randomly assigned into two groups according to the shunt technique, 15 of them with a veno-atrial shunt and 15 with an external shunt. Randomization was established by allocation concealment (Fig. 1).

Figure 1



Patient randomization, follow-up, and analysis of populations.

Inclusion criteria were single-ventricle lesions, good-sized branch pulmonary arteries (Magoon's index >1.5), PA pressures less than 20 mmHg, SO_2 less than 80%, age range from 6 months to 5 years, and weight range from 6 to 21 kg. All these patients had adequate atrial septal defects and none of these patients required any intracardiac repair. Exclusion criteria were previous cardiac operation, known intracranial pathology, neurologic disease, or craniofacial anomalies.

Anesthetic management

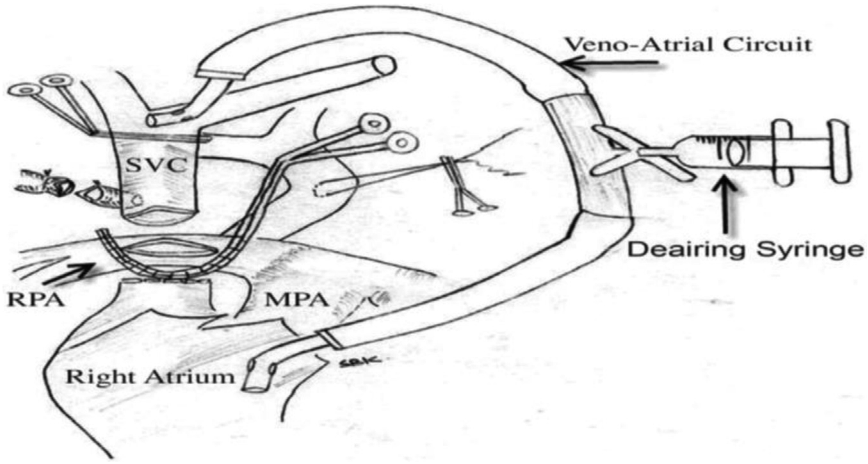
Preoperative evaluation through history, clinical examination, chest radiograph, echo Doppler, and laboratory investigation was done.

The patient is kept on minimal fasting as per standard guidelines after premedication with atropine (0.02 mg/kg), oral midazolam (0.2 mg/kg), and ketamine (2 mg/kg).

Intravenous induction with ketamine and/or opioid generally was performed after intravenous line placement, it was normal practice to administer ringer 10–15 ml/kg, to counteract patient dehydration. The FIO_2 was decreased to 0.5 after endotracheal intubation and patients ventilated to normocapnia. The operating room temperature should not be unduly warm for better brain protection.

Opioid-based anesthesia (fentanyl 10 μg/kg) was supplemented with isoflurane as tolerated, and the neuromuscular blockade was achieved with atracurium infusion (0.5 mg/kg/h). Systemic heparin (2 mg/kg) was administered to achieve an activated clotting time of 180 s or more. The head was turned just off the midline to prevent pressure or movement on the endotracheal tube by the surgical team while avoiding the possible effects of extremes of lateral head position on cerebral blood flow and venous drainage. The head of the operating table was

Figure 2



The veno-atrial shunt.

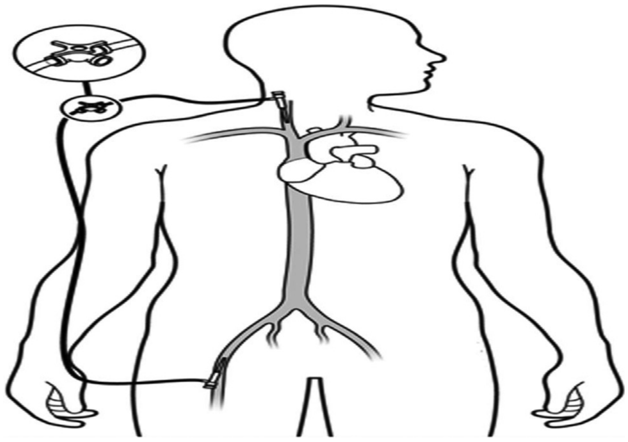
elevated 30° upward to allow better venous drainage by gravity through the collateral pathways.

In addition to routine monitors (oximetry, ECG, temperature, NIBP, invasive pressure monitoring, and urine output), a central venous catheter was placed via the right internal jugular veins for perioperative monitoring of SVC pressure and O_2 saturation in the internal jugular vein ($ScvO_2$), the femoral vein was cannulated with triple-lumen catheter, it is used for supplying the patient with fluids and drugs all through the operation, especially during clamping of SVC, monitoring of central venous pressure (CVP), and for external shunt during Glenn anastomosis to drain the high-pressure SVC during clamping.

A BDG procedure was performed with standby CPB, elective dobutamine (10 μg/kg/min) started, and volume load (5 ml/kg) of colloid (fresh frozen plasma or blood according to the hemoglobin level) was given, with intermittent boluses of noradrenaline 1–2 μg to elevate the mean arterial pressure to maintain an adequate transcranial pressure gradient (30–40 mmHg). All patients received 30 mg/kg methylprednisolone before clamping the SVC. The pulmonary artery was clamped electively for 2 min to test for any hemodynamic disturbances or changes in the oxygen saturation. The FIO_2 was increased to one during clamping and decreased again after declamping.

In total, 15 patients had the Glenn with the internal shunt (I) (veno-atrial shunt) where the surgeon established a shunt between distal SVC and the right atrium (Fig. 2), establishing a veno-atrial shunt. We ensured adequate matching of the

Figure 3



Percutaneous jugulo-femoral SVC bypass technique. SVC, superior vena cava.

cannulae to the patient's age and weight. We attempted to cannulate the SVC vein and the right atrium in a proper position for proper drainage.

The other 15 had the Glenn with the external shunt (E) where the anesthesiologist connected the internal jugular venous cannula that represents the SVC with the main lumen of the femoral cannula, which represents the IVC through long venous extension in Fig. 3.

In both shunts, the upper and lower catheters were connected to a three-way connection and after clamping of SVC, the SVC was decompressed by opening the three-way allowing blood to flow passively by the pressure gradient. But if SVC pressure increased above 30 mmHg, a 20-ml Luer lock was connected to the three-way with sterile precautions and care to avoid air, blood was actively aspirated from the upper catheter and injected to the lower one around 20 times per minute, these maneuvers lowered SVC pressure.

After transection of SVC, the closure of the right atrial stump is done after the anastomosis to minimize the duration of the SVC clamp, leaving the azygous vein open during the construction of the anastomosis acting as an internal shunt. Otherwise, no changes in the anesthetic technique or the surgical technique over the time of the study, and the anesthetic implications of off-pump BDG are summarized in Table 1.

Postoperatively, the patients were stabilized in the ICU and after monitoring the SVC pressure for 12 h, the internal jugular vein cannula was removed to prevent any jugular vein thrombosis. The patients were started on aspirin (5 mg/kg/day), which was continued indefinitely.

Data collection

Data were independently collected by a researcher who was blinded to treatment assignment and also not involved in clinical care decision-making. Baseline characteristics included age, sex, weight, diagnosis, and baseline hemoglobin. Intraoperative data included the duration of the SVC clamp. Postoperative data included the intubation time, inotropic support time, and the length of ICU stay.

Study outcomes

Primary outcome: SVC pressure during clamping. Secondary outcomes: intraoperative data were collected and analyzed at the following time points: preclamping (postinduction), during clamping, and after declamping of SVC. Variables such as arterial oxygen saturation (SaO_2), systolic blood pressure (SBP), SVC pressure, cerebral perfusion pressure (CPP=MAP-CVP), and $ScvO_2$ were analyzed.

Table 1 Anesthetic implications during off-pump Glenn

Perfusionist standby
Cerebral protection
Avoid a warm operating room environment
Transcranial pressure gradients (systolic BP–SVC pressure) >30 mmHg
Decompress SVC (shunts)
Avoid arrhythmias
Avoid desaturation
Head-end elevation
Parallel cannulae without raising them above the body level
Test-clamp the PA
Test-clamp the SVC
Place the clamp below the azygous
Maintain a source of pulmonary blood flow till Glenn is complete

BP, blood pressure; PA, pulmonary artery; SVC, superior vena cava.

Sample size

The sample size was calculated using MedCalc software version 14 (MedCalc Software bvba, Ostend, Belgium). Our primary outcome was SVC pressure during clamping. A previous study reported that SVC pressure was 24±4.4 mmHg during off-pump Glenn with Veno_venous shunt [12]. At an alpha error of 0.05, we calculated that 28 children would give 80% power to detect a 20% difference in the SVC pressure in off-pump Glenn with shunt between the two groups. And the required sample size increased to 30 patients (15 patients per group) to compensate for possible dropouts.

Statistical analysis

Statistical analysis was conducted using IBM SPSS Statistics (version 21.0; IBM, Armonk, New York, USA). All data will be expressed as mean±SD, number, or percentage as appropriate. The two groups will be compared using the Student *t* test. Multiple measurements will be compared using analysis of variance, and the results will be considered statistically significant if the *P* value less than or equal to 0.05.

Results

Study population

This study was carried out on 30 patients with physiologic univentricular hearts who underwent a bidirectional cavopulmonary Glenn shunt. There were 15 cases (group I) in which the operations were done using a temporary veno-atrial shunt done by the surgeon (internal shunt). In the other 15 cases (group E), the operations were done using an external shunt by the anesthesiologist.

In our study, the sex, age, weight, type of pathology, preoperative variables, and intraoperative variables' distribution between the two groups was not significant (Table 2).

There was no hemodynamic instability during any of the procedures and oxygen saturation was maintained at more than 65–70% throughout the procedure (Table 3).

There was no significant difference between the two groups in terms of mean SVC pressure before, during, and after SVC clamping ($P>0.05$), as shown in Table 3. The transcranial pressure gradient was 58.1

±6.89 mmHg in group I, while 54.86±9.1 in group E was comparable between the groups (*P*>0.05) (Table 3).

There was no significant difference between the two groups in terms of $ScvO_2$ before, during, and after SVC clamping (*P*>0.05), as shown in Table 3.

There was no significant difference between the two groups in postoperative variables (the mean mechanical ventilation time, the mean inotropic support time, and the mean ICU stay) (Table 2).

Table 2 Demographic, pathological, and surgical data

	Group I Internal shunt (*N*=15)	Group E External shunt (*N*=15)
Age (months)	26±9.6	20.4±7.1
Weight (kg)	13.2±3.3	11.3±2.5
Sex [*n* (%)]		
Male	8 (54)	9 (60)
Female	7 (46)	6 (40)
Diagnosis [*n* (%)]		
TA	5 (33)	8 (54)
SV&PS	8 (54)	6 (40)
DILV	2 (13)	1 (6)
Preoperative Hb (g/dl)	17.2±1.6	16.32±1.92
SVC clamp time (min)	19.85±3.52	15.3±4.4
PO ventilation (h)	10.8±18	8.2±15.5
Inotropic support time (h)	10.8±18	8.5±3.49
ICU time (days)	5.2±2.1	4.2±1.49

DILV, double-inlet left ventricle; SV&PA, single ventricle and pulmonary stenosis; SVC, superior vena cava; TA, tricuspid atresia.

There were no postoperative dysrhythmias or phrenic paralysis. There was postoperative bleeding in one case in group E that required exploration. There were neurological complications in one case in each group: in group I, one case developed neurological convulsions on the first postoperative day, which were controlled on phenytoin treatment; in group E, one case showed delayed recovery for 8 h. After reaching the ICU, there was one case in group I that showed wound infection that responded to antibiotic therapy according to culture and sensitivity, wound drainage, and frequent dressing. All patients survived to discharge.

Discussion

The BDG shunt is a method of palliation for single-ventricle physiology and an intermediate step before the Fontan-type operations. It has the advantage over the BT shunt of improving oxygenation without overloading the ventricle, increasing pulmonary vascular resistance, or pulmonary artery distortion. Attempts were tried to perform this operation off-pump [3–18] to eliminate CPB's detrimental effects. It is proposed that performing the surgery off-pump would lead to better pulmonary function, better hemodynamics, shorter hospital stays, and lower cost.

Many criteria of the patients have been described for performing BDG shunt. The current accepted optimal age for the BDG is 3–9 months [19]. The mean pulmonary artery pressure should be less than 16

Table 3 Outcomes

	Group I Internal shunt (*N*=15)	Group E External shunt (*N*=15)	*P* value
SO_2 (%)			
Preclamping	75±4.5	72.1±4.5	0.07
During clamping	71.3±1.5	69.6±3.1	0.066
After declamping	90.9±1.8	89.2±2.8	0.06
SBP (mmHg) (the hemodynamic status)			
Preclamping	95.3±4.1	92±4.8	0.052
During clamping	89.3±2.5	87.2±3.5	0.07
After declamping	99.5±4.2	96.5±6.1	0.1
Internal jugular venous saturation (%)			
Preclamping	72.3±1.2	70.6±3.1	0.057
During clamping	68±2.1	67±3.2	0.3
After declamping	87±1.6	85±3.5	0.053
Internal jugular venous pressure (mmHg)			
Preclamping	9±2.2	7.6±1.6	0.056
During clamping			
Before shunt opening	40.4±3.4	37.6±4.5	0.064
After shunt opening	28.5±3.8	26.4±2.1	0.07
After declamping	17.5±2.5	16.2±2.1	0.1
Transcranial pressure gradient (mmHg)			
During clamping	58.1±6.9	54.86±9.1	0.25

mmHg. Most of our patients fulfilled these criteria. Caution is indicated when considering off-pump BDG if patients are too young or have hypoplastic pulmonary arterial branches with a McGoon index of less than 1.4.

During the procedure, the SVC is clamped and the proximal pressures can rise as high as 40–55 mmHg. This impairs blood flow to the brain. The procedure is usually performed at the age of 3–6 months (weighing between 3 and 6 kg) when their cardiac output will be ranging from 0.3 to 0.6 l/min. About 50–60% of the cardiac output returns via the SVC during infancy. This exaggerates the rise in the pressure after the application of the clamp. The drainage per minute by whatever means, should be equal to this flow to minimize the rise in SVC pressures and maintain adequate cerebral perfusion pressure. Flow depends on the viscosity and density of the fluid, the width and the length of the tubing, and the height difference.

We agreed with the opinion that clamping the SVC without a temporary shunt can lead to decreased cerebral blood flow and put the brain at risk [20].

Clamping the SVC can result in cerebral hypoperfusion and cerebral edema. To maintain adequate cerebral perfusion in the face of high cerebral venous pressures, the concept of transcranial pressure has been described [5]. It is the difference between the mean arterial pressure and the mean pressure in the SVC. Since the SVC pressure increases to 40–55 mmHg during clamping, judicious use of inotropes to increase the mean arterial pressure was indicated. Maintaining the transcranial pressure greater than 30–40 mmHg was expected to maintain adequate cerebral perfusion.

The general strategies to protect the brain are: (a) establishing bypass, cooling, and low flows when on bypass. (b) Veno-venous [9,13] veno-atrial shunts to drain the SVC when performed off-pump. (c) Minimizing the duration of the clamp.

CPB was proven to increase pulmonary vascular resistance, promote fluid sequestration, and depress myocardial function. All these are hazardous physiologic changes in the postoperative period following the BDG shunt.

The debate of performing off-pump Glenn has been raised for the last 15 years. Lamberti first published his series of seven patients in 1990. Since then, we have been able to identify 17 published series (Table 4), which have reported off-pump BDG. The primary concerns regarding off-pump BDG center around three issues: (a) perform the surgery rapidly to minimize the duration of SVC clamping. (b) The long-term effects of high SVC pressures on the brain. (c) The need to use temporary shunts to decompress the SVC.

Minimizing the duration of the clamp time, so decreasing the possibility of brain insult by doing this technique with an experienced surgeon and by a good selection of cases. After the transaction of SVC, the closure of the right atrial stump is done after the

Table 4 Off-pump bidirectional Glenn publication summary

Study	Years	Number of patients	Temporary shunt
Lamberti *et al*. [3]	1990	7	SVC–RA
Lal and Mahant [4]	1996	6	SVC–RA
Murthy *et al*. [5]	1999	5	SVC–PA
Jahangiri *et al*. [6]	1999	6	No shunt
Villagra *et al*. [7]	2000	5	-
Tireli *et al*. [8]	2003	30	SVC–RA/PA
Maddali *et al*. [9]	2003	2	SVC aspiration
Liu *et al*. [10]	2004	20	SVC–RA/PA
Luo *et al*. [11]	2004	36	SVC–RA
Maeba	2006	18	SVC–RA/PA
Kotani	2006	14	SVC–RA
Hussain *et al*. [14]	2007	22	No shunt
Bhan [15]	2008	37	No shunt
Hussain *et al*. [16]	2009	31	SVC–RA
Murthy *et al*. [17]	2010	168	SVC–RA
Mostafa *et al*. [12]	2014	50	SVC–RA
Deebis *et al*. [18]	2017	30	SVC–RA/no shunt
Total	17 studies	487	

SVC, superior vena cava.

anastomosis to minimize the duration of the SVC clamp. Leaving the azygous vein open during the construction of the anastomosis acted as an internal shunt. CPB apparatus was always kept ready to function in case of emergency.

In our study, we compared the result of the external shunt with the result of the internal shunt (veno-atrial).

In group I, we conducted the procedure, in the same steps as we discussed in the methodology, using the veno-atrial shunt. The results during clamping show that the mean SO_2 was 71.3±1.5, the mean SVC pressure before opening the shunt was 40.4±3.4, while after opening the shunt 28.5±3.8 and the MAP was 79.3±2.5.

The predictors of caval drainage during the reconstruction of the anastomosis are the pressure difference between SVC and right atrium, the central gravity, and the radius and length of venous cannulas that were used. According to the Poiseuille's formula, the most important determinant of flow is the size of the cannula because of the strong inverse relationship between flow and the fourth power of the radius. The size of the cannula depends on the weight and age of the patients. We generally tried to use the same-sized venous cannula (12 F) in SVC, which allows flow up to 400 ml/min and (16 F) as a common atrium venous cannula that allows flow up to 800 ml/min in all patients.

The result in group E was nearly similar to group I, the mean SO_2 was 69.6±3.1, the mean SVC pressure before opening the shunt was 37.6±4.5, while after opening the shunt was 26.4±2.1 and the MAP was 77.5 ±3.5.

With a small central line in the RIJV, increased viscosity of blood in patients with cyanotic heart diseases makes it very difficult for gravity alone to achieve these flows. So, one has to actively aspirate and push the blood. We were able to achieve about 20 times per minute, although the 20 G main lumen of CVL would be sufficient to decompress the SVC.

The rest of the results in the postoperative variables and the postoperative hemodynamics were nearly similar.

By analysis of these data, we found that the external shunt was as efficient as the internal shunt. The external shunt has many advantages such as a simple procedure, short duration, decreasing complications of CPB, and good mobility of SVC because there is no cannula in SVC.

In the present study, the most important limitation was that it did not assess the psychometric or developmental evaluation of the patients, which could reflect any subtle brain damage. Indeed, we need to include more advanced diagnostic tools for such minor brain insults in further studies.

Conclusion

Off-pump BDG can be safely performed by an experienced surgeon in selected patients, either with external or internal shunt avoiding CPB complications.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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