Introduction

Bi-directional Glenn shunt (BDGS) is an operative procedure performed for congenital cyanotic heart diseases where total correction is not feasible and leads to abnormal ventricular physiology. This surgical procedure consists of end-to-side anastomosis between the superior vena cava (SVC) and the right or left main branches of the pulmonary artery. BDGS decreases the ventricle volume overload and effectively improves arterial blood oxygen saturation (1).

The BDGS procedure is usually established using cardiopulmonary bypass (CPB) with satisfactory early and late results. However, the use of CPB may have some haemodynamic and inflammatory deleterious effects. The BDGS procedure can be performed without CPB by decompression of the SVC during performing the anastomosis between the SVC and the pulmonary artery (2, 3).

One of the major problems of performing BDGS off pump without a temporary veno-venous shunt to decompress the SVC is the increase in intracranial tension, cerebral oedema and cerebral hypoperfusion and the increased risk of neurological injury due to increased SVC pressure during its clamping (4).

Abstract

**Objective:** The aim of the present study was to evaluate the efficacy of extrathoracic jugulo-femoral venous shunt (JFVS) in avoiding the need for cardiopulmonary bypass (CPB) during the bi-directional Glenn shunt (BDGS) procedure.

**Methods:** A total of 40 patients aged between 9 and 36 months were enrolled in the present study. The patients were classified into two groups, group A (n=20) in which the patients had no veno-venous shunt and group B (n=20) in which the patients had extrathoracic JFVS. Patients requiring CPB, arterial oxygen saturation (SaO\textsubscript{2}), heart rate, mean arterial pressure (MAP) and central venous pressure (CVP) were recorded during surgery. Postoperative time of intubation, intensive care unit (ICU) and hospital length of stays and neurological complications were also recorded.

**Results:** The number of patients who needed urgent CPB was greater in group A than in group B. Intraoperative MAP was significantly lower in group A than in group B 10 min after clamping of the superior vena cava (SVC) and 30 min after declamping of the SVC. The CVP was significantly lower, and arterial SaO\textsubscript{2} was significantly higher in group B than in group A 10 min after clamping of the SVC. The duration of postoperative intubation was significantly shorter in group B than in group A, and the ICU length of stay was shorter in group B than in group A. The hospital length of stay was similar in both groups. Postoperative neurological deficits were comparable in both groups.

**Conclusion:** The use of extrathoracic JFVS during the BDGS procedure avoided the use of CPB, maintained MAP and prevented any significant increase in SVC pressure.

**Keywords:** Cardiopulmonary bypass, cyanotic congenital heart diseases, jugulo-femoral venous shunt, superior vena cava
The present study hypothesised that extrathoracic jugulo-femoral venous shunt (JFVS) can improve the patient’s haemodynamics and may avoid the use of CPB during clamping of the SVC. The primary goal of the present study was to evaluate the efficacy of extrathoracic JFVS in reducing the need of CPB in BDGS. The secondary goal was to evaluate its effect on intraoperative patient haemodynamics and postoperative neurological deficits.

Methods

After approval of the local ethics committee of the Faculty of Medicine, Mansoura University (R117/2015) and written consent from patient’s parents or their guardian, this prospective randomised study was conducted in Mansoura University Children’s Hospital from August 2015 to December 2018. A total of 40 patients of either sex aged between 9 and 36 months, suffering from congenital cyanotic heart diseases (hypoplastic right ventricle, pulmonary or tricuspid atresia) were enrolled in the present study.

Echocardiography and cardiac catheterisation were performed for all patients. Patients with left SVC, heart failure, pre-existing thrombocytopenia and severe renal or hepatic diseases were excluded from the study. Each patient received intramuscular midazolam 0.1 mg kg⁻¹ and ketamine 3 mg kg⁻¹ 15 min before induction of anaesthesia. On arrival to the operating room, a peripheral venous access was secured with monitoring the patients with 5-lead electrocardiography, pulse oximetry, non-invasive blood pressure, nasopharyngeal temperature and capnography.

Anaesthetic technique and surgical team were standardised for all patients. Anaesthesia was induced with fentanyl 5 µg kg⁻¹, sevoflurane and rocuronium 0.9 mg kg⁻¹ and maintained with sevoflurane in oxygen air mixture, rocuronium infusion 0.3 mg kg⁻¹ hr⁻¹ and fentanyl 5 µg mg⁻¹ kg⁻¹ hr⁻¹. Ventilation was adjusted to maintain end-tidal CO₂ 30–35 mmHg, peak airway pressure <20 mm Hg and positive end-expiratory pressure <5 mmHg. A radial artery was cannulated with a 22-gauge catheter for invasive arterial pressure monitoring, and a central venous catheter was inserted into the left femoral vein with complete aseptic precautions. Another central venous catheter was inserted into the subclavian vein for monitoring central venous pressure (CVP). The patients were randomly classified into two groups as follows:

Group A (n=20): patients without extrathoracic shunt
Group B (n=20): patients with extrathoracic JFVS.

Extrathoracic JFVS was established by inserting a 16–18-gauge cannula into both right internal jugular and right femoral veins guided by ultrasound. Both cannulae were connected via a venous extension line after its filling with heparinised normal saline, with three-way stopcock in it close to the internal jugular cannula. This shunt was used after clamping of the SVC to drain the high-pressure venous blood from the internal jugular vein into the low-pressure femoral vein. Active decompression of the SVC was used by applying negative pressure to the internal jugular cannula. Heparin 1–2 mg kg⁻¹ was administered via a central vein to achieve an activated clotting time of ≥200 s. The SVC was clamped, and the distal end of the SVC was anastomosed end-to-side to the right pulmonary artery. Blood from the right internal jugular vein was aspirated using a 20 ml syringe and re-injected into the right femoral vein under complete aseptic precautions with great care to avoid air injection. Syringe injection was repeated 10–20 times min⁻¹ aiming to maintain a CVP <20 mmHg.

Methyl prednisolone 30 mg kg⁻¹ was given to all patients, and the head of the operating table was elevated 30° upwards to allow better venous drainage by gravity through the collateral pathways and to minimise brain congestion. The CPB machine was available as standby for any significant haemodynamic disturbances that might occur, so we could complete the procedure on bypass safely. Dobutamine 5–10 µg kg⁻¹ min⁻¹ was infused before clamping of the SVC, and a volume load, 5 mL kg⁻¹, of a colloid (fresh frozen plasma or blood, according to the haemoglobin level) was given to elevate the mean arterial pressure (MAP) to maintain an adequate cerebral perfusion.

Surgery was conducted through a standard median sternotomy. Mild systemic hypothermia was allowed by maintaining the nasopharyngeal temperature between 33°C and 35°C through the low operating room temperature to reduce brain metabolism. During the procedure, haemodynamic instability was managed by infusions of epinephrine up to 0.2 µg kg⁻¹min⁻¹, norepinephrine up to 0.2 µg kg⁻¹min⁻¹ and colloid (hydroxy ethyl starch) infusion. Urgent CPB was established if haemodynamic instability was persistent during clamping of the SVC. After establishment of anastomosis, the right pulmonary artery and SVC clamps were released.

All patients were transferred to the intensive care unit (ICU) and mechanically ventilated with continuous monitoring of SVC pressure in addition to routine monitoring. The internal jugular venous and femoral cannulae were removed when the patients became haemodynamically stable. The head of the patients was kept up to 45° by keeping the intensive care bed in an anti-Trendelenburg position. The patients were warmed, adequately hydrated and extubated as soon as possible. Patients who developed postoperative neurological dysfunction were scanned via computed tomography (CT) of the brain.
Collected data
The intraoperative parameters were recorded after induction of anaesthesia (T₀), after sternotomy and before clamping of the SVC (T₁), 10 min after clamping of the SVC (T₂) and 30 min after declamping of the SVC (T₃), and these included arterial oxygen saturation (SaO₂), heart rate (HR), MAP and CVP. Intraoperative type and dose of vasoactive agents (epinephrine and norepinephrine), number of cases requiring urgent CPB and duration of surgery were also recorded. Postoperative variables (time to extubation, ICU and hospital length of stays and the presence of any renal or neurological deficits) were recorded. Renal dysfunction was indicated by elevated serum creatinine. Neurological deficits were recognised clinically by the occurrence of fits and confirmed radiologically by CT for the presence of cerebral infarction.

Statistical analysis
Sample size was done based on a pilot study in which the primary outcome was the number of patients requiring CPB. Using the t-test for comparison and setting alpha to 0.05, we need a minimum of 36 cases to detect a similar difference with 90% power. A dropout of 10% of cases will be expected. Therefore, a total number of 40 cases (20 cases per group) will be needed. Calculations were done using G power software Windows version 3.0.10 (Franz Faul, Christian-Albrechts-Universität Kiel, Kiel, Germany).

IBM Statistical Package for the Social Sciences statistical software for Windows version 25 (IBM SPSS Corp.; Armonk, NY, USA) was used for statistical analysis of the collected data. Shapiro–Wilk test was used to check the normality of the data distribution in continuous variables. Continuous variables were expressed as mean±SD, whereas categorical variables were expressed as number (n) and percentage (%). Independent samples t-test and Mann–Whitney independent samples test were used to compare normally and non-normally distributed continuous variables with no follow-up readings, respectively. Repeated measures ANOVA model with Bonferroni post hoc test and 95% confidence interval (CI) were used to compare the follow-up values of continuous data. Fisher’s exact test was used for intergroup comparison of nominal and ordinal data using the crosstabs function. Comparison of follow-up and basal values (intragroup) was conducted using Wilcoxon signed-rank test and McNemar test for ordinal and nominal data, respectively. All tests were conducted with 95% CI. Charts were generated using SPSS chart builder. A p (probability) value <0.05 was considered statistically significant.

Results
There were no statistically significant differences with regard to patient’s characteristics and the type of cardiac anomalies in both groups (group A, n=20, patients without extrathoracic shunt and group B, n=20, patients with extrathoracic JFVS) (Table 1).

Table 2 shows the types and average doses of intraoperative vasoactive drugs infusion (2 µg kg⁻¹ min⁻¹), patients requiring CPB (n, %) and duration of surgery (h). All patients in both groups required epinephrine, but its dose was significantly higher in group A (0.156±0.078 µg kg⁻¹ min⁻¹) than in group B (0.086±0.038 µg kg⁻¹ min⁻¹). With regard to norepinephrine requirements, the number of patients who required norepinephrine infusion was significantly higher in group A (9, 45%) than in group B (2, 10%), and its dose was similar with no significant difference between both groups. The number of patients who needed urgent CPB was significantly greater in group A (7, 35%) than in group B (0, 0%). There were no statistically significant differences between the studied groups regarding the duration of surgery.

The intraoperative MAP was significantly lower in group A than in group B 10 min after clamping of the SVC (T₁) and 30 min after declamping of the SVC (T₃) (Figure 1). The intraoperative HR was comparable in both groups (Figure 2), whereas the intraoperative CVP was significantly lower, and SaO₂ was significantly higher in group B than in group A 10 min after clamping of the SVC (T₃) as shown in Figures 3 and 4, respectively.

| Variables | Group A (n=20) | Group B (n=20) | p |
|-----------|----------------|----------------|---|
| Age (months) | 17.6±6.7 | 15.2±5.7 | 0.821 |
| Gender (male/female) | 13/7 | 12/8 | 0.873 |
| Weight (kg) | 12.18±6.92 | 13.42±5.35 | 0.762 |
| Height (cm) | 92.37±13.14 | 89.29±14.75 | 0.912 |
| BSA (m²) | 0.53 (0.35–0.51) | 0.54 (0.37–0.54) | 0.911 |
| Cardiac anomalies (n, %) | | | |
| Hypoplastic right ventricle | 8 (40%) | 7 (35%) | ns |
| Pulmonary atresia | 5 (25%) | 7 (35%) | ns |
| Tricuspid atresia | 7 (35%) | 6 (30%) | ns |

Data are expressed as mean±SD, number (n) and percentage (%). ns: non-significant; BSA: body surface area.
Table 3 shows the duration of postoperative intubation (h), ICU length of stay (h), hospital length of stay (days) and postoperative complications. The duration of postoperative intubation was significantly shorter in group B (11±5.8 h) than in group A (18.3±9.4 h), and the ICU length of stay was also shorter in group B (38.2±11.7 h) than in group A (53.5±19.6 h). The hospital length of stay was similar in both groups. The incidence of postoperative renal dysfunction was not statistically significant in group B compared to group A. Two patients in group A and one patient in group B developed postoperative renal dysfunction (elevated serum creatinine)
Table 3. Duration of postoperative intubation (h), ICU stay (h), hospital stay (days) and postoperative complications in the studied groups

| Variables                        | Group A (n=20) | Group B (n=20) | p    |
|----------------------------------|----------------|----------------|------|
| Duration of intubation (h)       | 18.3±9.4       | 11±5.8*        | 0.018|
| ICU stay (h)                     | 53.5±19.6      | 38.2±11.7*     | 0.031|
| Hospital stay (days)             | 7.2±3.1        | 6.8±2.4        | 0.82 |
| Post op renal dysfunction (n, %) | 2 (10)         | 1 (5)          | 0.421|
| Post op neurological deficit     |                |                |      |
| Fits (n, %)                      | 2 (10)         | 0 (0)          | ns   |
| Cerebral infarction (n, %)       | 0 (0)          | 0 (0)          | ns   |

Data are expressed as mean±SD, number (n) and percentage (%). *p<0.05 is considered significant when compared to group A. CPB: cardiopulmonary bypass

Discussion

The primary aim of the present study was to determine whether the use of temporary extrathoracic JFVS can avoid the need for CPB or not in patients requiring BDGS. The main results of the present study showed that the use of temporary extrathoracic JFVS significantly decreased the need of CPB, maintained MAP and SaO\textsubscript{2}, prevented marked CVP elevation and decreased the need for vasoactive drugs.

Several previous studies established temporary extracardiac venoatrial shunts (1, 5, 6) to decompress the SVC during its clamping, but its flow capacity was limited. The current extrathoracic JFVS had the advantages over the venoatrial shunt of being extrathoracic and actively drained the SVC. Kandakure et al. (1) found that the use of veno-venous shunt between the innominate vein and the right atrium during off-pump BDGS avoids the need for CPB and its adverse effects and is associated with minimal haemodynamic instability with maintaining SaO\textsubscript{2} >70%. Ayik et al. (7) in their retrospective randomised study that included 83 patients reported that the use of venoatrial shunt during the BDGS procedures is associated with avoiding CPB in all patients, no haemodynamic instability and SaO\textsubscript{2} is maintained >65%.

Patients in group A required higher doses of vasoactive drugs during SVC clamping due to the marked decrease in MAP as venous return from the SVC is responsible for 50% of cardiac output in children. Seven patients in group A developed severe hypotension despite the maximum doses of epinephrine and norepinephrine, and these patients required urgent CPB. Lee et al. (8) reported that SVC cross-clamping results in decreased right ventricular preload with subsequent impairment of cardiac output and hypotension.

With regard to postoperative neurological deficits, there were no significant differences between both groups, but two patients in group A developed fits. This may be attributed to the high intracranial tension and cerebral oedema resulting from clamping of the SVC without its decompression. Lee et al. (8) reported increased intracranial tension and development of brain oedema as a result of SVC cross-clamping. In this study, CVP was significantly high in patients without JFVS resulting in decreased cerebral perfusion pressure that is defined as the difference between MAP and CVP. Cerebral perfusion pressure affects many physiological factors, such as hypercapnia, which causes cerebral vasodilatation and increased cerebral blood flow (9). The use of temporary veno-venous shunt during the BDGS procedures prevents significant and harmful elevation of SVC pressure and maintains cerebral perfusion (10). The use of vasoactive drugs, adequate volume replacement and high transcranial pressure gradients contributes to maintain cerebral blood flow. In the current study, many strategies were used for brain protection and to decrease cerebral oedema that included giving all patients methyl prednisolone 30 mg kg\textsuperscript{-1} and elevation of the head of the operating table by 30° upwards, together with mild hypothermia.

The current study showed that the duration of postoperative intubation and ICU lengths of stays were shorter in group B than in group A with similar hospital length of stay in both groups. The use of JFVS in the current study minimised the degree of brain oedema as it effectively decompressed the SVC. In addition, all patients with JFVS did not require CPB, so its haemodynamic and inflammatory consequences were avoided, and all of these factors contributed to the prolonged postoperative mechanical ventilation and ICU admission.

Levi et al. (11) in their retrospective study that included 35 BDGS shunts performed without CPB using venoatrial shunt

with no significant difference between the studied groups. There were no statistically significant differences between the studied groups with regard to postoperative neurological deficits (fits and cerebral infarction). Two patients in group A developed fits on postoperative day 2 but were neurologically free without any radiological findings at the time of discharge from the hospital.
and 30 BDGS shunts performed using CPB found that there is no significant difference in postoperative ventilation (intubation) time, duration of ICU admission and hospital length of stay between on and off-pump patients.

Establishment of temporary extrathoracic JFVS is an easy procedure that can be performed totally by experienced anesthesiologist. It allows good exposure of the surgical field as the shunt is totally extrathoracic. Extreme cautions have to be taken to avoid infection, by strict aseptic precautions, and to avoid entry of any air bubble during injection of aspirated blood into the femoral vein. It is cost effective as it avoids the use of CPB and its adverse effects, so its use may be suitable in developing countries.

Study limitations
The use of JFVS requires the proper case selection with regard to patient weight and the anatomy of pulmonary vessels. The present study included only patients aged >9 months, so further studies are required to perform JFVS in young patients below this age.

Conclusion
The use of JFVS during the BDGS procedure avoided the use of CPB, maintained MAP, prevented any significant increase in SVC pressure and decreased the needs for vasoactive drugs.

Ethics Committee Approval: Ethics committee approval was received for this study from the ethics committee of Mansoura University (Approval number R117/2015).

Informed Consent: Written informed consent was obtained from patients’ parents or guardians who participated in this study.

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