Duct occluder in the management of persistent postoperative pleural effusion after bidirectional Glenn's shunt

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The bidirectional Glenn's shunt (BDG) is the first step in the systematic, staged approach to a Fontan operation for patients with univentricular hearts. For the BDG to function well, the flow of blood through the pulmonary circulation must be free from significant impediments so that systemic venous pressure does not reach physiologically unacceptable levels. High systemic venous pressures are associated with high morbidity because of persistent bilateral pleural effusions and pericardial effusions, low oxygenation, increased plasma transfusion requirements, albumin infusions to maintain plasma protein levels, and prolonged ICU stay. We present a case of BDG complicated by prolonged pleural effusions in the immediate postoperative period, which was managed successfully using a percutaneous catheter-based approach, and thereby avoiding the complications of a major redo cardiac surgery.

Keywords:
bidirectional Glenn's shunt, duct occlude, postoperative pleural effusion

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Introduction

The bidirectional Glenn's shunt (BDG) is the intermediate step of the systematic, staged approach to the Fontan operation for patients with univentricular hearts. The staged approach helps to reduce the volume load of the ventricle and minimize changes in ventricular geometry and diastolic function that accompany primary Fontan. In BDG, the superior vena cava (SVC) is divided and anastomosed to the undivided pulmonary arteries (PAs), creating a bidirectional cavopulmonary (Glenn) shunt that supplies blood to both lungs. For the BDG to function well, the flow of blood through the pulmonary circulation must be free from significant impediments so that systemic venous pressure does not reach physiologically unacceptable levels [1]. Systemic venous pressures of 16 mmHg or less are generally tolerated without significant sequelae, whereas those greater than 20 mmHg are associated with a variety of morbidities, especially pleural effusions. Such pleural effusions are often highly resistant to all forms of treatment and increase the length of ICU stay significantly.

After obtaining written and informed consent we present a case of BDG complicated by prolonged pleural effusions in the immediate postoperative period, which was managed successfully by a percutaneous catheter-based approach using a duct occluder (conventionally used for patent ductus arteriosus device closure), and thereby avoiding the complications of a major redo cardiac surgery.

Case history

A female child 1 year 11 months of age was admitted with complaints of gradually increasing cyanosis and dyspnea on exertion since the age of 4 months. On examination, the heart rate was 118/min; respiratory rate was 36/min; blood pressure was 106/65 mmHg; and room air oxygen saturation (SpO_2) was 72%, with systolic murmur in the left parasternal region, and no hepatosplenomegaly or ascites. ECG showed normal sinus rhythm with biventricular hypertrophy. Chest radiography showed cardiomegaly with oligemic lung fields. Echocardiography showed situs solitus, levocardia, normally related great arteries, double-outlet right ventricle, atrioventricular concordance, unbalanced atrioventricular canal defect, severe pulmonary stenosis with good confluent PA anatomy, bilateral SVC, normal pulmonary venous drainage, and normal atrioventricular valve function. Preoperative laboratory investigations indicated that hemoglobin was 12.5 g% and total leucocyte count (TLC) was 8400/cm^2, with normal platelet count, coagulation screen and liver function, and serum creatinine of 0.5 mg/dl.

A bilateral BDG was planned for the child, which would be followed by a Fontan's operation later on. The bilateral BDG surgery was uneventful and inotropic support of dobutamine (5 mcg/kg/min) was required to achieve good hemodynamics in the postoperative period. The child was extubated within 2 h of shifting to ICU. Postextubation arterial blood gases were satisfactory (FiO_2: 0.21, pH: 7.42, PaO_2:

57, $PaCO_2$: 34.5, saturation: 90%, SBE: -4, lactate: 2.6 mmol/l).

The early postoperative period was also uneventful. Inotropic support could be tapered off and mediastinal drain was removed on first postoperative day (POD). The hemodynamics was monitored closely and remained stable. Thus, the invasive lines and urinary catheter were removed on second POD. The right pleural drainage was 220 and 106 ml on POD 1 and 2, respectively. On POD 3, the right intercostal drain (ICD) was removed as drainage was minimal. The child remained comfortable.

However, from POD 4, the child developed tachypnea and oxygen saturation decreased. Chest radiography showed moderate, bilateral pleural effusion. Echocardiography showed patent bilateral Glenn's shunt with laminar, antegrade flow in the main pulmonary artery (MPA), normal ventricular function, mild atrioventricular valve regurgitation, large bilateral pleural effusion, and mild pericardial effusion. Bilateral ICDs were inserted and continuous pleural drainage made the child comfortable again. The nature of pleural fluid was serosanguinous initially and changed to chylous by day 8. The child was treated conservatively with a low-fat, high-protein diet rich in medium-chain triglycerides. As the serum albumin levels were decreasing, the child required albumin infusions to maintain plasma oncotic pressure. The daily pleural drainage ranged from 100 to 150 ml on left and 150 to 200 ml on the right side. On day 10, the child developed puffiness of the face, neck, and upper extremities, suggesting obstruction of SVC drainage.

In view of the persistent pleural effusion unresponsive to conservative measures and development of SVC obstruction, it was decided to occlude the MPA with a catheter-based device to decrease the accessory pulmonary blood flow (APBF). This catheter-based approach has been described by some previous authors and, if successful, can result in avoidance of another major surgical intervention.

The child was transported to the cardiac catheterization laboratory and carefully induced with ketamine (10 mg) and fentanyl (20 mcg) intravenously. Vecuronium (1 mg) was used for tracheal intubation. Care was taken to intubate without trauma with a 4-mm uncuffed polyvinyl chloride endotracheal tube. The previous surgical records showed that a 4-mm uncuffed tube was used. Thus, we chose the same tube with a 3.5-mm tube ready in case we encountered any difficulty. Neema *et al.* [2], in a similar case report, had reported the possibility of difficult intubation because of edema of the airways and tongue. Anesthesia was maintained with oxygen and air (FiO_2: 0.5), with 1–2 minimal alveolar concentration (MAC) sevoflurane and intermittent doses of fentanyl and vecuronium.

Right and left internal jugular venous (RIJV and LIJV) access was obtained. A 4-Fr sheath was placed in the RIJV and 5 Fr in the LIJV. Through the LIJV, a 6 × 2 cm Opta balloon Cordis Corporation,USA over a 0.014 inch wire was used to occlude the ventriculopulmonary artery communication (Fig. 1).

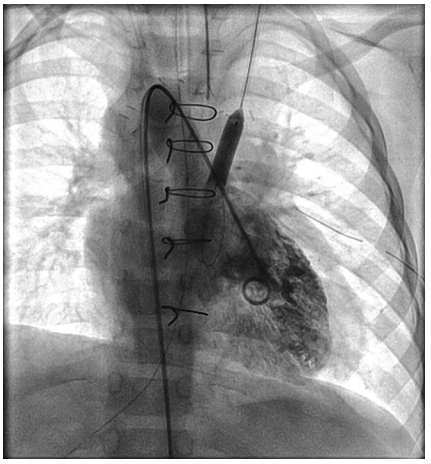
After balloon occlusion, the PA pressure was monitored with the RIJV reduced from 22 to 17 mmHg. In view of this reduction, it was decided to occlude the ventriculopulmonary artery communication. A 6-Fr Mullin's sheath was introduced through the LIJV and was placed in the MPA. An 8´6-mm Lifetech Duct Occluder Lifetech Scientific, China was deployed under fluoroscopic and angiographic guidance in the MPA across the area of supravalvar constriction. The device was released after confirming the position on angiography (Fig. 2).

After placement of the device, the PA pressures were checked and found to decrease significantly. The child could be extubated after 2 h. The pleural drainage became trace and the ICDs were removed after 2 days. The child was discharged after 2 days when chest radiography showed that there were no pleural effusions and the child was comfortable.

Discussion

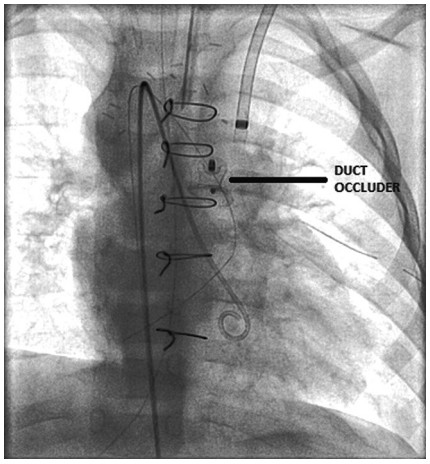
APBF following a BDG may be derived from systemic to PA shunts, aortopulmonary collaterals, or antegrade pulmonary blood across the stenosed right ventricular

Figure 1



Angiogram showing balloon occlusion of the ventriculopulmonary artery communication.

Figure 2



Angiogram showing the duct occluder placed at the ventriculopulmonary artery junction.

outflow tract (RVOT). Controversy exists on whether APBF should be left at the time of BDG to augment systemic saturation or be eliminated to reduce volume load of the ventricle. APBF is considered to improve oxygenation, decrease the incidence of pulmonary arteriovenous fistula formation by providing hepatic factor, and promoting the growth of the PAs. However, it may result in an increase in the BDG pressure, interfering with the flow from the SVC to the PAs. In addition, it is a form of left-to-right shunt that may place an additional volume load on the single ventricle. Thus, the need for an additional source of pulmonary blood flow has to be individualized. Mainwaring *et al.* [3], in their retrospective study, showed that the elimination of APBF at the time of BDG may confer a long-term advantage for patients with a functional single ventricle.

When the APBF across the RVOT leads to elevated venous pressures and dysfunction of BDG, the standard approach for the management of this condition would be PA ligation or division. These children present with features of elevated SVC pressures such as edema of the head and neck, airways, tongue, and upper extremities. There is cerebral congestion and prolonged circulation time. Large bilateral pleural effusions are also present, necessitating ICD insertion. Percutaneous catheter-based management would be a much safer option compared with redo surgeries in these extremely sick children.

The major complications associated with the procedure are local hematoma formation in the groin or neck during vascular access and sheath insertion. The incidences of such complications are similar to all other percutaneous interventions. There may be distal embolization of the device that may necessitate open surgical device retrieval. Utmost care must be taken to avoid device embolization by carefully sizing the device. The procedure should be performed under general anesthesia to avoid sudden movements and bucking, and also to help maintain strict control over the already compromised airway and the hemodynamic parameters. Delayed complication includes thrombosis in the PAs and can be prevented by placing the patient on low-dose aspirin.

There are a few previous similar case reports that have reported the use of devices to occlude the RVOT. Pilla *et al.* [4] used an Amplatzer duct occluder in the setting of a postoperative BDG to obliterate the competitive forward flow from the ventricle to the pulmonary circulation in a 7-month-old boy (weight 6.5 kg) with complete transposition of the great arteries and multiple ventricular septal defects.

Thanopoulos *et al.* [5] reported a case of a 12-month-old infant with a double-outlet right ventricle and pulmonary stenosis who presented with signs of SVC syndrome secondary to a dysfunctioning BDG. The patient was treated successfully with transcatheter obstruction of an APBF using the Amplatzer muscular ventricular septal defect occluder.

Ebeid *et al.* [6] reported the successful use of an Amplatzer duct occluder in a child with a balanced single-ventricle physiology of a double-inlet left ventricle, D-malposition of the great vessels, and subpulmonic stenosis. At the age of 1 year, she underwent a BDG procedure. Angiography indicated the presence of a moderate amount of residual flow from the single left ventricle to the BDG with elevated BDG pressure precluding a Fontan-type repair. The patient was referred for catheter closure of the residual flow from the ventricle to BDG.

Conclusion

The dysfunctional BDG because of persistent pulsatile blood flow in the RVOT can give rise to high morbidity because of persistent bilateral pleural effusions and pericardial effusions, low oxygenation, increased plasma transfusion requirements, albumin infusions to maintain plasma protein levels, and prolonged ICU stay. The percutaneous catheter-based management reduces the morbidity and also avoids a redo surgery for MPA ligation in these sick children with SVC syndrome.

Acknowledgements

Conflicts of interest

There are no conflicts of interest.

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