Surgical Management of the Adult Ductus, the Giant Ductus, the Recanalized and Calcified Ductus using Normothermic Cardiopulmonary Bypass without Circulatory Arrest: Review of 20 Years’ Experience

Lakshmi Kumari Sankhyan1, Ujjwal Kumar Chowdhury1*, Vasubabu Gudala1, Suruchi Hasija2, Vishwas Malik1 and Priya Jagia2

1Department of Cardiothoracic and Vascular Surgery, All India Institute of Medical Sciences, India
2Department of Cardiac Anaesthesia, All India Institute of Medical Sciences, India
3Department of Cardiac Radiology, All India Institute of Medical Sciences, India

Abstract

Background: Use of trans pulmonary approach under normothermic cardiopulmonary bypass without circulatory arrest has been poorly documented for the giant ductus, adult ductus, recanalized and calcified ductus as an alternative to decrease the high perioperative complication rate associated with division and suture.

Patients and Methods: Between January 1998 and September 2018, 18 consecutive patients underwent closure of persistent ductus arteriosus under normothermic cardiopulmonary bypass without circulatory arrest via trans-pulmonary approach for adult ductus. Age ranged from 26 to 35 years. Indications were adult ductus arteriosus (n=3), calcified ductus (n=6), infected ductus (n=1), window ductus (n=2), short and wide ductus (n=2), giant ductus (n=3) and recanalized ductus (n=1).

Results: There was no early or late death. There was no phrenic or left recurrent laryngeal nerve damage, chylothorax, massive bleeding or recanalization. At a mean follow-up of 12.6 ± 5.8 years, all patients are in NYHA functional class I. Computerized tomographic angiography (n=14) revealed complete ductal interruption with no residual shunt or ductal aneurysms.

Conclusion: On the basis of these results, ductal closure via trans-pulmonary balloon occlusion under normothermic cardiopulmonary bypass without circulatory arrest appears to be a versatile technique for ductal closure presenting in adulthood and eliminates the operative complications associated with ductal division and suture. We believe that the absence of mortality and morbidity resulted from careful operative technique and complete closure guided by intraoperative transesophageal echocardiography. Long-term evaluation will be mandatory to assess the possible occurrence of specific complications encountered with cardiopulmonary bypass.

Keywords: Adult ductus arteriosus; Calcified adult ductus; Recanalized ductus arteriosus; Hypertensive ductus arteriosus; Window ductus arteriosus; Giant ductus arteriosus; Ductal closure under cardiopulmonary bypass

Introduction

Among the variety of surgical options available for persistent ductus arteriosus, standard closed ligation techniques or ductal division and suture are the ideal options [1-4]. However, such a direct approach may be difficult to perform in patients presenting late in adulthood with diminished elasticity and/or calcification, previous infection causing ductal endarteritis, unusual anatomic features like wide, short, giant ductus, window ductus, aneurysmal ductus and recurrent ductus [4-14].

Today, the refinements in extracorporeal circulation and the perfection of the surgical strategy described here-in enable the surgeon to perform surgical closure of the ductus arteriosus under normothermic cardiopulmonary bypass obviating the need for circulatory arrest [15-19].
We present herein our experience with 18 patients in whom transpulmonary ductal closure under normothermic cardiopulmonary bypass without circulatory arrest was performed for a variety of reasons. The indications, techniques, concerns and results are discussed.

**Patients and Methods**

This study conforms to the principles outlined in the declaration of Helsinki. Consent was taken for anonymous analysis of data, and we obtained approval from our institutional ethics committee. The procedures were performed by a single surgeon (corresponding author), thus achieving uniformity in surgical protocol. Medical records were reviewed to obtain demographic characteristics, associated medical conditions, detail of operations, results of the echocardiographic, computerized tomographic examination and cardiac catheterization.

Between January 1998 and September 2018, 18 patients with adult ductus (n=3), giant ductus (n=3), short and wide ductus (n=2), window ductus (n=2), calcified ductus (n=6), infected ductus (n=1) and recanalized ductus (n=1) underwent interruption of the ductus arteriosus via transpulmonary approach under normothermic cardiopulmonary bypass without circulatory arrest. The patient with recanalized ductus had undergone ductal ligation 12 years ago via left posterolateral thoracotomy.

There were 10 (55.5%) male and 8 (44.5%) female patients with a mean age of 28.2 ± 6.0 (range 26 to 35) years. All patients had progressively increasing shortness of breath of New York Heart Association class III (n=10) and class IV (n=8). Four (22.2%) patients complained of angina pectoris. Pertinent physical findings included bounding peripheral pulses, a continuous murmur over the upper left sternal area (machinery), loud pulmonic component of second heart sound and cardiomegaly. Chest radiography revealed cardiomegaly with a cardiothoracic ratio between 0.6 and 0.7 (specifically with signs of left atrial and left ventricular enlargement), dilated pulmonary arteries and increased pulmonary vascular markings. Calcification around the ductus was noted on roentgenogram and computerized tomographic examination in 6 patients. ECG-gated multi-detector computed tomography was used to evaluate the size of the persistent ductus arteriosus and to assess the extent of calcification.

The electrocardiogram revealed atrial fibrillation in 8 and sinus rhythm in 10 patients. All patients had electrocardiographic evidence of bialtrial enlargement and biventricular hypertrophy. Transthoracic two-dimensional color Doppler echocardiography demonstrated a continuous wide flow from the aorta through the ductus arteriosus, dilated pulmonary arteries and increased pulmonary vascular markings. Calcification around the ductus was noted on roentgenogram and computerized tomographic examination in 6 patients. ECG-gated multi-detector computed tomography was used to evaluate the size of the persistent ductus arteriosus and to assess the extent of calcification.

The surgical techniques, intraoperative and postoperative management protocols were uniform throughout the study period. Transesophageal echocardiography was performed preoperatively and intra operatively in every case. All operations were performed under normothermic cardiopulmonary bypass at 34º Celsius, through angled venous cannulas into the superior and inferior caval veins and aortic cannulation.

The aorta and pulmonary trunk were dissected for selective aortic cross-clamping. The left ventricle was vented through right superior pulmonary vein. Four stay sutures were placed on the distal pulmonary trunk near the bifurcation. The aorta was individually cross clamped. Cold hyperkalemic St. Thomas based blood cardioplegia (1:4) and topical hypothermia was used for myocardial preservation.

The aortic arch vessels were not isolated. At the commencement of cardiopulmonary bypass, the head end was lowered, and external digital compression was applied over the pulmonary arterial end of the ductus to prevent over flooding and cardiac distention. Cavae were tightened. Under total cardiopulmonary bypass the distal pulmonary trunk was transversely opened in between stay sutures. The pump flows were transiently lowered to 1.5-2.0 l/min/m² and intra cardiac cardiomyocyte suckers were used for visualization of the pulmonary arterial end of the ductus (Figures 1A-1F). A 18-Fr or 20-Fr Foley catheter was inserted through the ductus into the aorta. The balloon was inflated with normal saline and the pump flows were restored to normal.

The ductus was closed using multiple interrupted 4-0 polypropylene suture buttressed with Teflon pledgets (Johnson and Johnson Pvt. Ltd., Ethicon, LLC, San Lorenzo, USA) with a goal to obtain a perfect hemostasis. Precautions were taken to advance the Foley catheter little deep within the aorta to avoid balloon rupture during suture placement. The balloon was subsequently deflated and withdrawn. The pump flows were temporarily lowered while tying the sutures. A second layer of continuous suture was taken to ensure complete ductal closure (Figures 2A-2F). Aortic cross-
clamp was released restoring myocardial blood supply. After releasing the aortic cross-clamp myocardial perfusion was restored, pulmonary arteriotomy was closed and patients were weaned from cardiopulmonary bypass. Transesophageal echocardiography confirmed no residual shunt before chest closure.

Mean aortic cross-clamp time was 16.4 ± 3.0 minutes (range 11 to 20 minutes) and cardiopulmonary bypass time was 40 ± 16.6 min (range 34 to 46 min). No complications due to cardiopulmonary bypass and the balloon occlusion method were seen. No residual shunt was found on echocardiogram and the value of pulmonary artery pressure decreased significantly from 62.0 ± 18.4 mmHg to 33.6 ± 6.4 mmHg at 6 months after the operation (p<0.01). A follow-up echocardiography was performed annually after the operation. Till date, neither recanalization of the ductus nor pseudo aneurysm has been recognized. Computerized tomographic angiography (n=14) revealed complete ductal interruption with no residual shunt or ductal aneurysms.

Results
Early results

There was no operative death. None of the patients required reoperation for bleeding. These patients were extubated on the first postoperative day. There was no left recurrent laryngeal nerve damage, no residual ductal flow or chylothorax. All patients were routinely commenced on blockers and oral angiotensin-converting enzyme inhibitors on day 1 postoperatively (0.5-1.0µg/kg/every 8 hours), prior to weaning from nitro glycerine and sodium nitroprusside. The average stay in the ICU was 2 days (range 1-4 days).

Late results

There was no late death. All survivors were periodically evaluated every 6 months by institutional cardiologists and surgeons. Their records were reviewed for all pertinent pre-and postoperative information including electrocardiogram and Doppler echocardiogram.

They have been followed-up for periods ranging from 12 months to 20 years (mean 12.6 ± 5.8 years). All patients are in New York Heart Association functional class I at their last follow-up visit. Two-dimensional and Doppler echocardiography at the time of discharge did not reveal any residual ductal shunting or aneurysm formation in any patient. Anti-hypertensive medication was stopped in all patients.

Discussion

Although persistent ductus arteriosus generally presents in infancy and childhood, a significant percentage of patients in developing countries do present in adulthood with complications like endocarditis, congestive heart failure, pulmonary hypertension and pulmonary vascular disease, aneurysm, calcification and recanalization of the ductus arteriosus [5,6,9,15,21].

Patient survival is directly related to the incidence and degree of congestive heart failure. According to Campbell and colleagues, a third of patients with patent ductus arteriosus would have died by the age of forty years and sixty percentages before sixty years of life [21]. However few patients survive with little disability and have a normal life span. In the majority of adults, exercise tolerance is liable to deteriorate rapidly and in them surgical treatment has been of
great benefit, even at 65 years of age. Available information, however, has been based on single case reports with little long-term follow-up. Prolonged survival in patent ductus arteriosus is usually, but not necessarily associated with small ductus and it is three times more common in women [22]. These patients have only a minimal increase in pulmonary vascular resistance [23].

Symptoms and cardiomegaly are indications for treatment. However surgical correction of patent ductus arteriosus is riskier in late presenters due to friable tissues, peculiar morphology, presence of calcification and aneurysmal dilatation. Although surgical treatment usually restores normal exercise tolerance and reduces heart size on a long-term basis, it fails to relieve symptoms in few patients and progressive pulmonary hypertension continues. Myocardial damage secondary to long duration of a left-to-right shunt and progressive pulmonary hypertension may be the factors which determine poor results in these older patients [5,6,15,21,22].

In our review of literature, we found that the oldest male patient to survive surgical repair was 68 years of age and the oldest female patient was 72 years of age [4,24]. Atrial flutter and atrial fibrillation related to atrial distension due to left-to-right shunt may contribute to worsening of heart failure [25]. When atrial fibrillation and heart failure respond well to medical treatment, surgical treatment especially in patients over 70 years of age is not essential and can be avoided.

Cardiac catheterization is particularly important in adult ductus to evaluate the pulmonary vascular resistance and the degree of shunting. Temporary test occlusion with a balloon catheter may provide important information regarding the advisability of closure [26]. Michael Rigby has pointed out that the key to safe management of patients with hypertensive ductus should be based on accurate measurement of pulmonary vascular resistance rather than pulmonary artery pressure with duct occlusion, which is inevitably flawed [8].

By convention, patients with a pulmonary vascular resistance more than 6 Wood units when breathing 100% oxygen are considered unsuitable for repair of congenital heart defects with left-to-right shunts because they have higher perioperative morbidity, mortality, and likelihood of progressive pulmonary vascular resistance despite repair of the defect [26,27].

Bush and colleagues pointed out the importance of accurate measurement of pulmonary vascular resistance. For patients breathing oxygen, its crucial to include values for dissolved oxygen in the calculation of pulmonary vascular resistance. Failure to include dissolved oxygen in the measurement of pulmonary blood flow results in underestimation of pulmonary vascular resistance. Assumption of values from nomograms for measurement of oxygen consumption provides a potential source of inaccuracy in pulmonary vascular resistance measurements [27].

Despite 80 years of experience, surgical treatment of persistent ductus arteriosus remains a technical challenge in the subset of older patients with persistent ductus arteriosus due to diminished elasticity, ductal wall calcification and in those with recurrence [4-16]. These patients are exposed to major operative risks with the use of anatomic techniques involving direct exposure of the persistent ductus arteriosus i.e., double ligation, transfixion and suture ligation, division and suture.

Even today, there are problems involved in the hemodynamic determination of operability in cases of congenital heart diseases with severe pulmonary hypertension. The Heath-Edwards classification, histopathologic criteria in widespread clinical use, can provide only qualitative information about plexogenic pulmonary arteriopathy and consequently has limitations for use in determining operability [28].

In 1973 Dushane and Kirklin reported that a pulmonary vascular resistance index of 14 Wood units/m² or less was an indication for surgery and in 1976 Kirklin and colleagues reported that a value of 10 or less to be an operative indication among older children [29,30]. More recently, Momma and colleagues have reported a pulmonary vascular resistance index of 8 units/m² or less to be an operative indication [31]. Based on results of 23 autopsies and 26 lung biopsies of patients of ventricular septal defect and persistent ductus arteriosus operated on, Yamaki and colleagues demonstrated that regardless of patients’ age, when pulmonary vascular resistance of 8 Wood units/m² or more were obtained, operative indications should be determined on the basis of lung biopsies. The Index of Pulmonary Vascular Disease (IPVD), a composite and quantitative evaluation of the severity of pulmonary vascular disease was introduced to determine the operability. An IPVD of 2.1 without Down’s syndrome was considered as the upper permissible limits for surgical intervention [32]. We have not performed lung biopsy on any of our patients in this study.

In addition, the cases of short, wide ductus and large window ductus that are more common in adults are potentially lethal surgical trap [12,13,20]. In a median sternotomy approach, a very large persistent ductus arteriosus obscures the origin of the left pulmonary artery because it is located directly above the ostium of the left pulmonary artery. The small aortic isthmus which is intra-pericardially located may be mistaken for a persistent ductus arteriosus. Extra-pericardially, the main pulmonary artery may appear to be directly continuous with the descending thoracic aorta, resembling one continuous artery with no visible interposed ductus [12,13,20].

Dissection and isolation of the ductus arteriosus in these clinical scenarios poses a risk of intraoperative hemorrhage and injury to left recurrent laryngeal nerve, phrenic nerve and chylothorax. Lung dysfunction and adhesions due to previous thoracotomy present additional technical challenge in re operative cases.

“...the artery should be taken up at some distance from the diseased part.” An account of Mr. Hunter’s method, communicated in a letter to Dr. Simmons by Mr.Everard Home, Surgeon [33].

In an attempt to decrease or eliminate these dreaded complications that are directly related to surgical dissection and aortic cross-clamping, while performing ductal interruption in late presenters, diverse surgical techniques have been advanced. Published literature documents the following techniques for different kinds of complicated persistent ductus arteriosus.

**Double ligation of the adult ductus arteriosus**

Some investigators have described a technique of double ligation of the ductus arteriosus suturing two Teflon felt pledgets on the superior and inferior surfaces of the ductus and gradual tightening of the two mattress sutures under controlled hypertension [7,34].

**Division and suture without cardiopulmonary bypass**

Thomas and associates described a technique wherein a Potts-
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2018 | Volume 1 | Article 1028

Division and suture without cardiopulmonary bypass

Bell-Thomson and colleagues described a technique of division and suture of adult ductus arteriosus in which the aorta was cross-clamped above and below the persistent ductus arteriosus. A third vascular clamp is applied at the ductus itself. Care should be taken not to include the pulmonary artery within the confines at this clamp. The ductus is divided at the aortic end including a small rim of the aortic wall. Both the aortic and pulmonary ends are sutured using 4-0 polypropylene suture [4].

Trans aortic repair of calcified ductus arteriosus without utilizing cardiopulmonary bypass

The left subclavian artery is isolated and tapes are passed around the same. A shunt is placed between the left subclavian artery and descending thoracic aorta after low dose heparinisation. The aorta is clamped proximal and distal to the persistent ductus arteriosus. A Satinsky clamp is placed over the pulmonary artery about 0.5 cm from the ductal orifice. Great care is taken to avoid injury to the ductus during placement of the clamp. The aorta is opened longitudinally. The aortic end of the ductus is closed using a Dacron patch [35].

Ductal interruption under cardiopulmonary bypass

Morrow and Clarke described a technique of ductal interruption for a large, calcified ductus under cardiopulmonary bypass [36].

Trans aortic patch closure under cardiopulmonary bypass

Two arterial cannulas were placed, one into the left subclavian artery and the other into the left femoral artery and a single venous drainage cannula into the right ventricular outflow tract. Total cardiopulmonary bypass was instituted, the main pulmonary artery, the aorta proximal and distal to the ductus was occluded. An incision was made in the lateral wall of the aorta, an area free of calcification. The aortic end of the ductus was closed using a Teflon fabric and a series of interrupted mattress sutures.

Cardiopulmonary bypass, mild hypothermia, transpulmonary Fogarty balloon occlusion

Bhati and colleagues described a technique of ductal interruption under cardiopulmonary bypass from inside the pulmonary artery after the aortic orifice had been occluded with the help of a venous Fogarty catheter. They described this innovative technique on 3 patients with persistent ductus arteriosus and associated congenital anomalies [15].

Cardiopulmonary bypass, profound hypothermia transpulmonary balloon occlusion

Arbatli and colleagues described the technique of ductal interruption via transpulmonary route, under cardiopulmonary bypass, profound hypothermia, low flow state without circulatory arrest [16].

Profound hypothermia and circulatory arrest

In 1978, O’Donovan and Beck described a method in which ductal closure was done under cardiopulmonary bypass with profound hypothermia and a low-flow state with circulatory arrest and without the benefit of balloon occlusion [37].

Profound hypothermia and circulatory arrest

This technique has been advocated for transpulmonary closure of adult ductus arteriosus by some investigators. These investigators thought that in cases of short, wide ductus, balloon catheter occlusion had a danger of balloon catheter rupture during suture placement [23, 38].

Cardiopulmonary bypass, transpulmonary patch mounted on a Fogarty catheter

Taira and Akita described a method, using a patch mounted on a Fogarty catheter via trans-pulmonary route under cardiopulmonary bypass. The ductus is closed via pulmonary artery [17].

Left heart bypass

Laustela and associates used a Teflon pledgeted suture ligation technique with left heart bypass [39].

Kirklin and Silver were the first to describe a technique to close the ductus through median sternotomy that was laterpopularized by McGoon [40, 41]. However there is a high chance of inadvertent tearing during the manipulation of a tense, wide ductus with high intraluminal pressure [15, 42, 43].

Some investigators in the initial years closed the ductus through the pulmonary artery under circulatory arrest, in a manner described by Kirklin and Devlloo for closure of Potts anastomosis. Adoption of such a policy is indeed time consuming, with the added risks of air embolism, flooding of the pulmonary circulation, cardiac distension and is therefore abandoned for patients undergoing division and suture for ductal interruption. Other investigators including ourselves have employed a C-shaped vascular clamp at the aortic end of the ductus and a right angled vascular clamp at the pulmonary arterial end under controlled hypotension using sodium nitroprusside and division is performed through the aortic tissues which allow for a secure closure of both sides without undue risk of bleeding. In case aorta is cross clamped, it is recommended by Crafoord and Nylin that the cross clamp not be applied longer than 15 min [44, 45].

We believe that with the trans aortic method, there are substantial risks of cerebral hyper perfusion at the time of aortic clamping in a heparinized patient. Extensive dissection in the ductus area necessitating two clamps in the aorta and one in the pulmonary artery in the presence of adhesions and calcification has been another concern.

We used the balloon occlusion technique as reported previously by Bhati and associates to occlude the ductus in children with concomitant congenital cardiac lesions [15]. We focused on transpulmonary approach under normothermic cardiopulmonary bypass without circulatory arrest for the underlying reasons:

1. It is simple and safe as full control of the juxta ductal aorta and pulmonary artery is easily achieved.
2. The ductus is usually calcified and friable in adult patients.
3. It avoids injury to the recurrent laryngeal nerve that is usually susceptible to injury in reoperation.
4. The pulmonary arterial end of the ductus is usually healthy for holding the sutures.
5. Although profound hypothermia and circulatory arrest or low flow perfusion were the preferred method for some investigators [37, 38], the balloon occlusion technique was the easiest way to control
the ejecting blood through the defect while placing the sutures [16,19].

6. This method obviates systemic cooling, profound hypothermia, internal shunting, descending aortic cross-clamping and circulatory arrest.

We avoided balloon catheter breakage during suture placement by reducing the pump flow and advancing the Foley catheter slightly inwards, thereby exposing the ductal ostium for secure placement of the sutures. Intraoperative transesophageal echocardiography was of great assistance to confirm the adequacy of repair prior to weaning from cardiopulmonary bypass.

The damage to the left recurrent laryngeal nerve is uncommon and usually temporary. Some investigators found an incidence of 8.8% of the left vocal cord paralysis related to the difficulty of dissection in underweight children undergoing ligation of persistent ductus arteriosus [46,47]. Liang and associates have reported cases of vocal cord paralysis after percutaneous coil embolization of ductus arteriosus [48]. Jatene MB and colleagues reported 8.8% incidence of vocal cord paralysis and 14.7% incidence of dysphonia in the postoperative period [9].

The possibility remains that recanalization and/or aneurysmal change of persistent ductus arteriosus occurs in the late postoperative period through the pulmonary trunk [4,5]. Neither recanalization nor aneurysmal dilatation of the ductus has been noted in our series. However annual echocardiography will be mandatory for prompt detection of this odious complication.

Interventional PDA closure seems to have a high level of reproducibility. However, intermediate (1 year) residual shunt was present in 5% of patients enrolled in the European Registry [49]. Immediate residual patency however was 41% and it is unknown in how much time complete occlusion occurs. Consequently, concerns about effective complete closure, repeat examinations, duration of follow up, and necessity of long-term antibiotic prophylaxis exist [50]. Superseding the Rashkind double umbrella PDA occluder, the Amplatzer occluder is an alternative to coils in large ductus. Despite the new devices which are available and widely used, interventional PDA closure maintain the disadvantages and potential intra procedural risks such as peripheral vascular injury, device migration, obstruction of the pulmonary arteries or thoracic aorta, late embolization flow disturbance in the left pulmonary artery or descending thoracic aorta from a protruding device, hemolysis from high velocity shunting, distal embolization, thrombosis of the vascular access, infection, severe residual shunting and recanalization [51-53]. The VATS technique is safely applicable in patients with ductus diameters <9mm [54].

Recently multiple cases of endovascular stent grafting of aorta to close a patent PDA have been reported. Presence of arch vessels and lack of proper proximal landing zone, endoleak, delayed cessation of ductus patency and residual shunt limit the use of this technique to a few selective patients who have comorbidities that make them a high risk candidate for surgery [55-57].

Conclusions

We conclude that the transpulmonary balloon occlusion method under normothermic cardiopulmonary bypass is an expedient, safe and effective technique in adult ductus, window ductus, giant ductus, recanalized and calcified ductus. This method facilitated the procedure of ductal interruption, obviating the need for deep hypothermia and circulatory arrest. It avoids dissection in the presence of friable ductal adhesions in redo operations, in the presence of ductal wall calcification, thereby avoiding perioperative injury to the ductal tissue, recurrent laryngeal nerve and large lymphatics. Knowledge of this approach should contribute to the armamentarium of the cardiac surgeon faced with such complex congenital anomalies. Transesophageal echocardiography is crucial in assessing the size of the ductus and confirming adequacy of repair.

References

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