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Archives of Clinical Case Reports

Case Report

Operative, Echocardiographic and Angiocardiographic Details of a Giant, Calcified, Hypertensive Window Ductus Arteriosus in an Adult undergoing Transpulmonary Closure under Normothermic Cardiopulmonary Bypass without Circulatory Arrest

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Received: 30 April 2019; **Accepted:** 06 August 2019; **Published:** 17 August 2019

Citation of this article: Chowdhury, UK., Singh, S., George, N., Sankhyan, LK., Chauhan. V., et al. (2019) Operative, Echocardiographic and Angiocardiographic Details of a Giant, Calcified, Hypertensive Window Ductus Arteriosus in an Adult undergoing Transpulmonary Closure under Normothermic Cardiopulmonary Bypass without Circulatory Arrest. Arch Clin Case Rep, 2(2): 024-029.

ABSTRACT

The short, wide, calcified hypertensive persistent ductus arteriosus in an adult poses extreme hazard with standard closed ligation or division-suture techniques. We report the case of a 27-year-old female patient who underwent transpulmonary closure of ductus with pledgeted sutures under normothermic bypass without circulatory arrest using a Foley catheter for temporary occlusion. Transesophageal echocardiography is crucial in assessing the size of the ductus and confirming the adequacy of repair.

Introduction

Correction of persistent ductus arteriosus remains a surgical challenge in the subset of patients with persistent ductus arteriosus presenting in adolescence and adulthood because of diminished elasticity, friability and/or calcification of vessel wall, previous infection causing ductal endarteritis, unusual anatomic features like giant ductus, wide ductus, short ductus, window ductus, aneurysmal ductus and recurrent ductus [1-14]. It is associated with dreaded complications related to surgical dissection and aortic cross-clamping like injury to the ductal tissue, distended pulmonary arteries, recurrent laryngeal nerve and large lymphatics [4-14]. Here we present the technique and results in one adult patient with a giant, short, calcified, hypertensive, window ductus arteriosus undergoing closure under normothermic cardiopulmonary bypass without circulatory arrest.

Case Report

A 27-year-old female patient was referred to our cardiac clinic with dyspnea on exertion and repeated chest infection for 15 years. Clinically, she had bounding peripheral pulses, cardiomegaly, a systolic-diastolic precordial thrill and a grade 5/6 continuous murmur, loudest at the upper left sternal border. Chest roentgenography revealed cardiomegaly with a cardiothoracic ratio of 0.7, dilated pulmonary arteries and increased pulmonary vascular markings. The electrocardiogram showed atrial fibrillation with signs of left atrial dilatation and biventricular hypertrophy. Transthoracic, two-dimensional color Doppler echocardiography demonstrated a continuous wide flow from the aorta through the ductus arteriosus to the pulmonary artery with a peak flow velocity of 4.8 m/sec, indicating elevated pulmonary artery pressure (Figure 1). The left ventricle was dilated with an end-diastolic diameter of 80 mm and end-systolic diameter of 62 mm. The left ventricular ejection fraction was 0.30. Computed tomography revealed densely calcified, short and wide ductus arteriosus about 18 mm in diameter. Cardiac catheterization revealed systolic pulmonary artery pressure of 100 mmHg and a Qp:Qs of 4:1. The calculated pulmonary vascular resistance index was 7.0 Woods unit/m^2. Reversal of pulmonary vascular resistance index to less than 4.0 Woods unit/m^2 after 100% oxygen and nitric oxide (80 ppm) administration and temporary ductal test occlusion for 10 minutes in the cardiac catheterization lab suggested operability.

Intraoperative transesophageal echocardiography was done with a multiplane probe and color Doppler echocardiography for ductal visualization. The operation was performed under cardiopulmonary bypass with aorto-bicaval cannulation at 34° Celsius via transpulmonary approach. The aorta and pulmonary trunk was dissected for selective aortic cross-clamping. The aorta was individually cross-clamped and the left ventricle was vented through right superior pulmonary vein. Myocardial preservation was achieved by cold blood cardioplegia and topical hypothermia. At the commencement of cardioplegia, 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 distension. Caval snares were tightened. The distal pulmonary arterial trunk was transversely opened in between stay sutures. The pump flow was transiently lowered to 1.5 L/min/m^2 and intracardiac cardiotomy suckers were used for visualization of the pulmonary arterial end of the ductus (Figures 2A-2F). A 20 Fr Foley catheter was inserted through the ductus into the aorta. The balloon was inflated using normal saline and the pump flow was restored to normal.

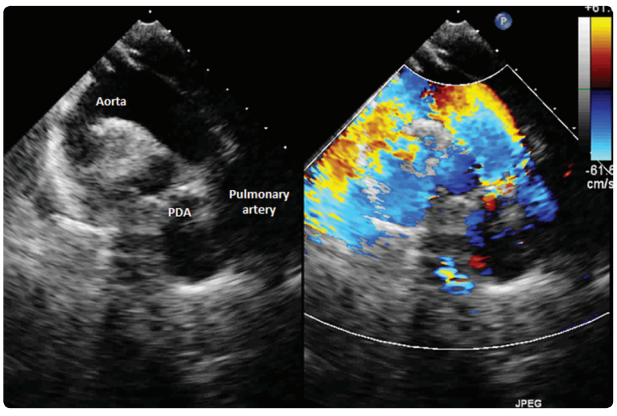

Figure 1: Transthoracic two-dimensional color Doppler echocardiogram showing continuous wide flow from the aorta through the ductus to the pulmonary artery.

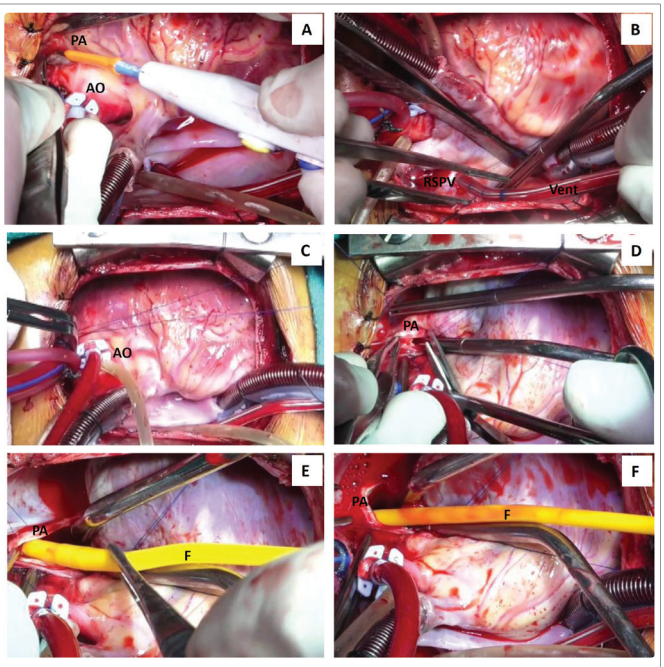

Figure 2a-f: A-C: The aorta and pulmonary trunk were individually dissected for selective aortic clamping. The left heart is vented through right superior pulmonary vein. External digital compression was applied over the pulmonary arterial end of the ductus to prevent over flooding and cardiac distension.

D-F: The pulmonary trunk was transversely opened in between stay sutures. The pump flows were transiently lowered to identify the pulmonary arterial end of the ductal orifice. A 20-Fr Foley catheter was inserted through the ductal orifice into the aorta. The balloon was inflated with normal saline and the pump flows were restored to normal.

Four pledgeted 4-0 polypropylene sutures (Johnson and Johnson Pvt. Ltd, Ethicon, LLC, San Lorenzo, USA) were passed from one side of the defect to the other but not tied. Precaution was taken to advance the Foley catheter little deep into the aorta to avoid balloon rupture during placement of sutures. The pump flow was transiently lowered to 1.0 L/min/m^2 while tying the sutures. The balloon was subsequently deflated and withdrawn. A second layer of continuous polypropylene suture was taken to ensure complete ductal closure (Figures 2G-2N). Aortic cross-clamp was released restoring myocardial blood supply. The pulmonary arteriotomy was closed and patient was weaned from cardiopulmonary bypass (Figures 2O-2R).

The aortic cross-clamp and cardiopulmonary bypass time was 14 min and 38 min respectively. There were no complications due to cardiopulmonary bypass and the balloon occlusion. The systolic pulmonary artery pressure decreased from 100 mmHg to 40 mmHg immediately after surgery. Before chest closure, transesophageal echocardiography was done to ensure that there was no residual flow.

At one year follow-up she was asymptomatic, in New York Heart Association functional class I and in normal sinus rhythm.

Echocardiography ruled out any recanalization of the ductus or pseudoaneurysm formation. Postoperative computerized tomographic angiography confirmed the same (Figure 3 and 4).

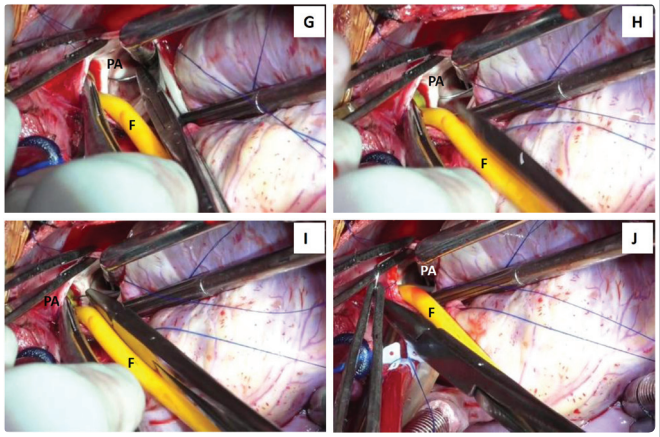

Figure 2g-j: The ductal orifice was closed using multiple interrupted 4-0 polypropylene suture buttressed with Teflon pledgets. The Foley catheter was advanced little deep within the aorta to avoid balloon rupture during suture placement.

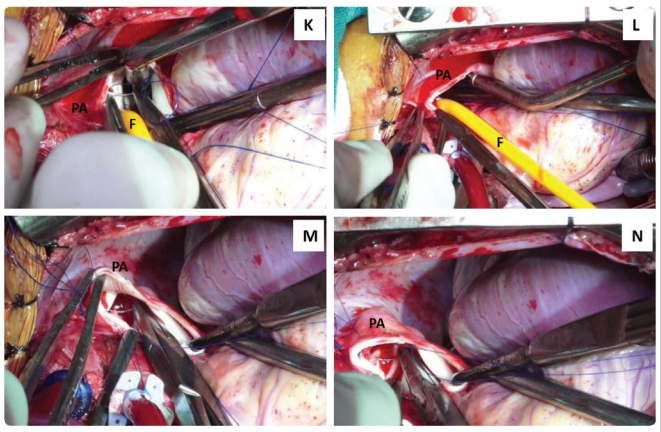

Figure 2k-n: The balloon subsequently was deflated and withdrawn. A second layer of continuous suture was taken to ensure complete ductal interruption.

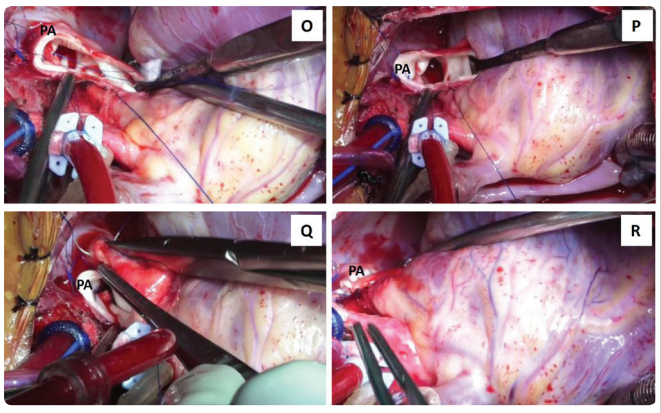

Figure 2o-r: The adequacy of closure was confirmed. The pulmonary artery was closed in 2 layers using 4-0 polypropylene suture.

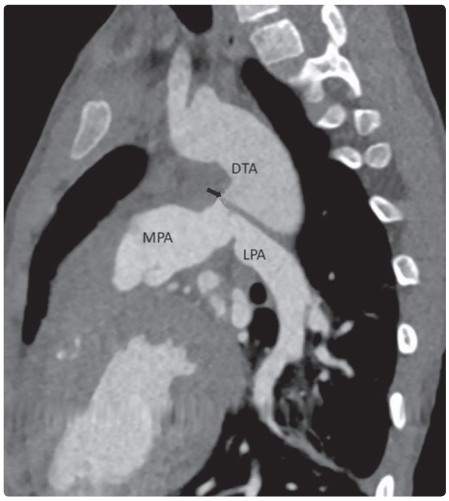

Figure 3: Postoperative post ductal closure: three dimensional thick multiplanar sagittal oblique computerized tomographic image showing the closed ductus arteriosus (arrow) with no flow between the aorta and pulmonary artery. (DTA- Descending thoracic aorta, LPA- Left pulmonary artery, MPA- Main pulmonary artery).

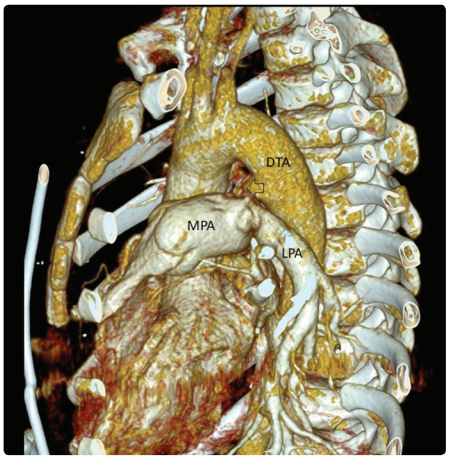

Figure 4: Postoperative post ductal closure: three dimensional volume rendered sagittal oblique computerized tomographic image showing the closed ductus arteriosus (arrow) with no flow between the aorta and pulmonary artery. (DTA- Descending thoracic aorta, LPA- Left pulmonary artery, MPA- Main pulmonary artery).

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 endarteritis, congestive heart failure, pulmonary hypertension with pulmonary vascular disease, aneurysm, calcification and recanalization of the ductus arteriosus [6-15].

Closure of a persistent ductus arteriosus in adults is a surgical challenge, because they are usually large, giant sized or aneurysmal, calcified, hypertensive, has a short and window configuration and sometimes associated with infection [2-6,8-14]. In these situations, simple dissection and closure is not safe if not possible and may lead

to inadvertent ligation of other vital vascular structures [2-6,8-14]. The complication most feared at operation is the uncontrollable haemorrhage from the aortic or distended pulmonary arterial end of the ductus [11-13]. The pulmonary end of the ductus is seldom involved with calcification and the pulmonary arterial wall hold sutures well, lending itself safe for transpulmonary closure of patent ductus in adults.

In 1968, Campbell reported that 20% of patients with untreated PDA died before they reached 30 years of age and they had a 2.5% to 4% annual mortality rate between 4th and 6th decades of life [15]. Only 10% of patients with untreated PDAs lived beyond their 60s [15]. In our review of literature, we found that the oldest male patient to survive without surgical repair was 68 years of age and the oldest female patient was 74 years of age [4,16]. Prolonged survival in PDA is usually, but not necessarily associated with small ductus and it is 3 times more common in females [17]. These patients have only a minimal increase in pulmonary vascular resistance [18]. In order to improve the shortened life expectancy, surgical or interventional closure of the PDA should be considered except in cases of Eisenmenger's syndrome [1-15,19-21].

Clinically, in adults congestive cardiac failure and cardiomegaly are indications for treatment. Atrial flutter and fibrillation related to atrial distension due to left- to- right shunt may contribute to worsening of heart failure [3-10,15]. When atrial fibrillation and cardiac failure responds well to medical treatment, surgical treatment especially in patients over 70 years of age may not be recommended. In our patient, the significant shunt from left-to-right may have contributed to atrial fibrillation due to atrial distension, since sinus rhythm was restored after closure of the ductus [4,14,15].

It is desirable to perform cardiac catheterization in late presenters with ductus arteriosus to evaluate the degree and direction of shunting and pulmonary vascular resistance. Temporary test occlusion with a balloon catheter may provide important information regarding the advisability of closure [19]. But 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 [20]. By convention, patients with a pulmonary vascular resistance of 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 [19,22]. 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 to its use in determining operability [23].

Oldham HN from the Mayo clinic in 1964 defined giant persistent ductus arteriosus as those with a ductal transverse dimension of 15mm and above [11]. Maurice Lev in 1953 defined window ductus as those with a large (more than 15 mm in maximal transverse dimension) ductus and virtually have no length which on external inspection may not be visible [12]. Our patient had a short, giant and window ductus on external inspection, the main pulmonary artery appeared to be directly continuous with the descending thoracic aorta with no visible interposed ductus. The left recurrent laryngeal nerve curled under the aortic arch from left-to-right distal to ductus and not proximal to ductus arteriosus, and hence not beneath the preductal portion of the aortic arch.

Dissection and isolation of the ductus arteriosus in this clinical scenario poses a risk of intraoperative hemorrhage and injury to the recurrent laryngeal nerve, pulmonary artery, phrenic nerve, and chylothorax [1-15]. In an attempt to decrease or eliminate these dreaded complications that are directly related to surgical dissection, diverse surgical techniques have been advanced in the published literature. The techniques include double ligation, division and suture with or without cardiopulmonary bypass, transaortic repair with or without cardiopulmonary bypass, left heart bypass and profound hypothermia and circulatory arrest [24-33].

We preferred to operate on this adult patient via median sternotomy with the aid of cardiopulmonary bypass. There were five major compelling reasons for this preference. First, the ductus was short, calcified, friable and of window configuration; second, the recurrent laryngeal nerve in such clinical situation is susceptible to injury; third, the presence of distended, hypertensive pulmonary arteries; fourth, the presence of a distended, giant, short and wide ductus, and lastly, the pulmonary arterial wall at the end of the short ductus is known to hold sutures well. The unavailability of any large series in adults evaluating various treatment strategies makes it difficult to compare various persistent ductus arteriosus closure techniques [1-14,24-33].

Our technique stems from the technique reported previously by Bhati and colleagues to occlude the ductus in children with concomitant congenital cardiac lesions [14]. We used normothermic bypass and pledgeted sutures in short periods of low-flow state during suture placement and ductal occlusion.

Although profound hypothermia and circulatory arrest or low-flow perfusion was the preferred method for some investigators, the balloon occlusion technique under cardiopulmonary bypass was the easiest way to control the ejecting blood through the defect while placing the sutures [30]. During suture placement, balloon catheter breakage is a probable complication and may cause air embolization if it is inflated without completely removing the air bubbles. On the other hand, reducing the pump flow and pushing the catheter slightly inwards exposes the ductal ostium for correct placement of sutures. Transesophageal echocardiography was also used to confirm the adequacy of repair before weaning from cardiopulmonary bypass.

Due to anatomical window-type configuration, proximity to arch vessels and ductal diameter measuring more than 10mm, video-assisted thoracoscopic ligation, endovascular stent grafting, or device occluders were considered inappropriate in this clinical scenario [34-37].

Conclusions

We conclude that appropriate surgical management of an adult patient with giant calcified and hypertensive window ductus should consist of occlusion with patch or pledgeted sutures on cardiopulmonary bypass via transpulmonary approach using temporary balloon-occlusion. This method is safe, expedient, obviates the need for descending aortic cross-clamping, internal shunting,

profound hypothermia, and circulatory arrest. It avoids dissection in the presence of ductal wall calcification, adhesions, thereby avoiding perioperative injury to the tense pulmonary artery and recurrent laryngeal nerve.

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