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Endovascular Closure of Aortic Pseudoaneurysm Following Pulmonary Artery Vegetectomy for Residual Patent Ductus Arteriosus Endarteritis- a case report

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Case Report

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Abstract

Background: Residual patent ductus arteriosus (PDA) following surgical ligation is an uncommon occurrence that may predispose to infective endarteritis at the pulmonary artery end. Surgical vegetectomy may be required when antibiotic therapy fails. The development of an aortic pseudoaneurysm at the isthmus–descending thoracic aorta junction following trans-pulmonary artery vegetectomy and residual PDA closure is an extraordinary complication rarely documented in the literature.

Case Presentation: A 2-year-old female underwent surgical ligation of a large PDA via left thoracotomy. Postoperative echocardiography revealed a 1 mm residual shunt. Six months later, she presented with high-grade fever and echocardiography demonstrated a 6 × 8 mm vegetation at the left pulmonary artery end of the residual PDA, which had enlarged to 3 mm. After failure of prolonged intravenous antibiotic therapy, surgical intervention was performed via sternotomy. Due to dense adhesions precluding external re-ligation, vegetectomy and PDA closure were accomplished from within the pulmonary artery using low-flow cardiopulmonary bypass under deep hypothermia. On subsequent follow-up, chest radiography revealed superior mediastinal widening, and CT angiography confirmed a pseudoaneurysm (5.5 × 3.8 × 3.7 cm) with eccentric thrombosis at the aortic isthmus–descending thoracic aorta junction, causing compression of adjacent structures. The pseudoaneurysm was successfully occluded percutaneously using an 8 mm Amplatzer plug via right femoral artery access, with post-deployment angiography confirming complete exclusion of the sac.

Conclusion: Even trivial residual PDA shunts carry a lifelong risk of infective endarteritis. Post-discharge surveillance imaging is essential to detect occult complications following complex re-operations in infected fields, and multidisciplinary collaboration is critical to optimizing outcomes.

INTRODUCTION

Residual Patent Ductus Arteriosus (PDA) following surgical ligation is an uncommon occurrence that may serve as a focal point for potential infective endarteritis at the pulmonary artery terminus [1–3]. These endarteritic vegetations may necessitate a surgical intervention in light of antibiotic failure [3, 4]. The occurrence of an aortic pseudo-aneurysm at the isthmus-descending thoracic aorta junction subsequent to a pulmonary artery vegetectomy and the closure of residual PDA (in light of residual PDA and infective endarteritis) represents an extraordinary complication. To our knowledge, this is the first reported case of aortic pseudoaneurysm at the isthmus-DTA junction following transpulmonary artery vegetectomy for residual PDA endarteritis successfully treated by percutaneous device closure.

CASE

A 2-year-old female child presented with features of heart failure and recurrent lower respiratory tract infections. Transthoracic echocardiography (TTE) was suggestive of a large PDA with a left-to-right shunt and severe pulmonary arterial hypertension. A computed tomography scan (CT scan) was done to

rule out associated co-arctation of the aorta (Fig. 1). The child then underwent surgical closure of large patent ductus arteriosus (PDA) via left thoracotomy. TTE performed on postoperative day 1(POD-1) revealed a residual trivial leak measuring approximately 1 mm. The postoperative course was otherwise uneventful, and the patient was discharged in stable condition (on antibiotic prophylaxis) with the intent of spontaneous closure of a trivial leak via fibrosis.

The child remained on regular outpatient follow-up. Six months after surgery, she presented with an intermittent high-grade fever of one week's duration. TTE demonstrated a vegetation measuring 6 × 8 mm at the LPA end of the residual PDA, an increase in size of the residual ductus from 1 mm to 3 mm, and moderate pulmonary arterial hypertension. She was admitted and started on intravenous antibiotics.

Despite prolonged intravenous antibiotic therapy, the patient showed no clinical improvement, and serial TTE examinations over a month revealed no reduction in vegetation size. Consequently, a surgical approach was deemed necessary. Following sternotomy, dense adhesions were noted in proximity to the residual ductal region, and the surrounding tissue exhibited fragility, precluding the external looping of the patent ductus arteriosus (PDA). Therefore, the decision was made to occlude the PDA from within the pulmonary artery following vegetectomy, utilizing low-flow CPB for a duration of 45 seconds under conditions of deep hypothermia, with intermittent restoration of blood flow. This low-flow CPB cycle of 45 seconds under deep hypothermia was repeated three times with flow restoration in between. The surgical procedure was otherwise unremarkable. Postoperative echocardiography revealed no residual patent ductus arteriosus (PDA) and good biventricular function. Intra-operative cultures were sterile. Intravenous antibiotics were continued for a period of two weeks, after which the patient was discharged with a regimen of oral antibiotics for six weeks.

The patient continued regular follow-up in the outpatient department. After 3 months of follow-up, chest radiography revealed superior mediastinal widening (Fig. 2A). An urgent CT angiography was performed, which demonstrated a pseudoaneurysm with eccentric thrombosis arising at the aortic isthmus–descending thoracic aorta (DTA) junction, measuring 5.5 × 3.8 × 3.7 cm (Fig. 2B and 2C). The pseudoaneurysm caused significant compression of adjacent mediastinal structures, including displacement of the left and main pulmonary arteries and mild rightward deviation of the trachea and mediastinum. Clinically, the patient was asymptomatic.

The patient was then planned for pseudoaneurysm plugging. Right femoral artery access was taken using a 6-French vascular sheath. Catheterization of the DTA was done using the Judkins right coronary catheter (JR). On contrast angiogram, there was a large pseudoaneurysm sac noted arising from the left anterolateral wall of the DTA at the previous PDA repair site. An 8 mm Amplatzer plug was deployed through the JR catheter. Post-plug deployment angiogram showed non-opacification of the pseudoaneurysm sac (Fig. 3) (Video 1). A post-procedure check TTE reported progressive thrombosis of the pseudoaneurysm sac with no gradient in the DTA which was later reconfirmed in the follow up TTE done a month after the procedure.

Video 1 (Supplementary file): Preprocedural 2-dimensional echocardiography demonstrating the pseudoaneurysm, and postprocedural plug angiogram confirming successful closure.

DISCUSSION

This case illustrates a rare cascade of complications following surgical patent ductus arteriosus (PDA) ligation: residual shunt, infective endarteritis refractory to medical therapy, and subsequent aortic pseudoaneurysm formation vegetectomy and residual PDA closure.

Residual patency after surgical PDA ligation is an uncommon but well-recognized phenomenon. In an institutional series of 17 patients, transcatheter closure for previously surgically ligated arterial ducts documents residual duct diameters that range from 1 to 5.7 mm following surgical ligation, presenting from three months up to 40 years after the surgery, and were eventually closed through a transcatheter approach. [1]. The mechanisms underlying residual flow include incomplete ligation of a thick or calcified duct, recanalization through organized thrombus, and ductal wall disease that prevents secure external closure [1], [2]. In our patient, the 1 mm residual shunt detected on POD-1 likely reflected incomplete ligation of the ductal tissue.

PDA-associated infective endarteritis is usually characterized by a protracted fever, continuous murmur, and vegetations limited to the pulmonary artery end of the duct. Most often, the culprit organisms are Staphylococcus aureus and coagulase-negative staphylococci; however, culture-negative cases might occur, as in our case [3]. Surgical indications in PDA endocarditis are persistent bacteremia despite suitable antibiotics, extensive or mobile vegetations, hemodynamic compromise, and local consequences such as pulmonary artery perforation or pseudoaneurysm [4].

When external re-ligation is considered inappropriate due to factors such as inflammation, adhesions, or potential vascular compromise, an intraluminal strategy employing pulmonary arteriotomy during cardiopulmonary bypass (CPB) facilitates direct observation and secure excision of infected tissue. This approach allows for the meticulous extraction of vegetations affixed to the pulmonary artery wall and the final closure of the duct from an internal perspective, consequently alleviating the hazards linked to dissection in a delicate, infected operational setting [5], [6]. In our case, the choice to undertake trans-main pulmonary artery vegetectomy under low-flow CPB with deep hypothermia was influenced by the dimensions and positioning of the vegetation, along with significant involvement of the pulmonary artery.

Aortic pseudoaneurysms in children are rare and typically arise from surgical anastomotic disruption after transplantation (in the ascending aorta), ventricular assist device explantation (in the ascending aorta), blunt trauma (in the DTA), or infection at prior repair sites [7], [8].

The PDA originates from the aortic isthmus immediately distal to the left subclavian artery, and any prior surgical manipulation of this segment including external ligation, internal closure, or adjacent dissection leaves the ductal stump as a potential locus of wall vulnerability [9]. In this case, different mechanisms

may have contributed, like suture line stress at the ductal stump adjacent to the aortic isthmus, residual infection extending to the aortic wall, or a combination of these factors.

The eccentric thrombosis and mass effect causing pulmonary artery compression and tracheal deviation underscore the urgency of intervention. The eccentric, rather than circumferential, distribution of intrasaccular thrombosis on CT angiography and the narrow, well-defined neck of the pseudoaneurysm amenable to plug closure are morphologically consistent with a focal, point-source mural defect at the stump site rather than diffuse aneurysmal dilation, supporting a discrete mechanical disruption as the structural entry point [10].

Transcatheter closure utilizing Amplatzer occluders has become a viable and increasingly favored alternative to traditional surgical repair for aortic pseudoaneurysms in high-risk patients, as exemplified in our case where device closure was chosen due to the challenging reoperative environment, suitable anatomy of the pseudoaneurysm neck, and the aim to minimize CPB-related morbidity in a pediatric patient [11].

Conclusion

This case highlights that even a very small residual patent ductus arteriosus shunt of 1 mm after ligation typically hemodynamically insignificant otherwise can still become a lifelong risk factor for developing infective endocarditis. In line with established American College of Cardiology guidelines, after surgical ligation, a residual patent ductus arteriosus calls for annual echocardiographic follow-up. Follow-up imaging after discharge is crucial to uncover hidden complications that can arise from such complex re-operations in an infected environment. With appropriate care and support, meaningful multidisciplinary collaboration among cardiac surgery, interventional cardiology, and vascular surgery is essential to improve outcomes in these difficult cases.

Abbreviations

PDA	Patent Ductus arteriosus
MPA	Main Pulmonary artery
LPA	Left Pulmonary artery
CPB	Cardiopulmonary Bypass
LRTI	Lower respiratory tract infections
TTE	Trans-thoracic echocardiography
PAH	Pulmonary arterial hypertension
POD-1	Post-operative day 1
CT scan	Computed tomography scan
Ao	Aorta
RV	Right ventricle
LV	Left ventricle
DTA	Descending thoracic aorta
SVC	Superior vena cava
JR	Judkins right coronary catheter

Declarations

Patient's perspective

Patient is a 2 year old child. Here is a brief translated account of her father who happens to be a non English speaker. I was very concerned about my child who had encountered such a rare complication, as explained to me by the author and his team. I was very concerned about the success of the treatment, as otherwise my child could have landed in another surgery. Post-treatment my child is doing well and is in regular follow-up at the author's institute.

1. Consent to Publish Declaration: Consent to publish was obtained from guardian of patient.
2. Funding Declaration: Authors have received no funding.
3. Clinical trial number: not applicable (since it is a case report).
4. Ethics declaration: not applicable (due to retrospective nature of case).

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Figures

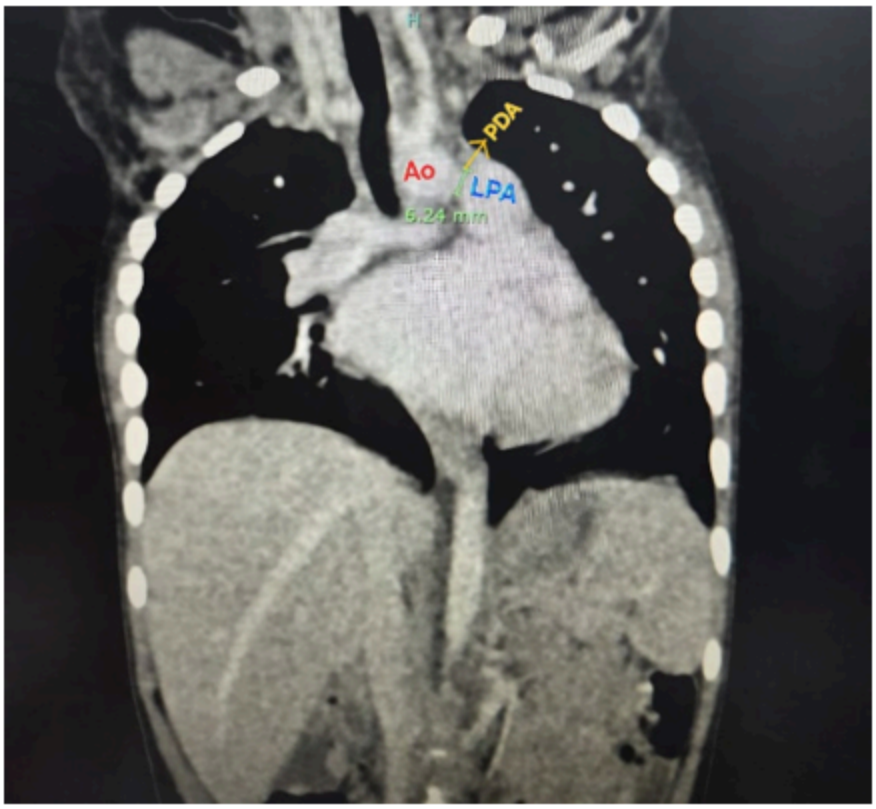


Figure 1

Preoperative computed tomography demonstrating a patent ductus arteriosus (PDA). (Ao, aorta; LPA, left pulmonary artery; PDA, patent ductus arteriosus.)

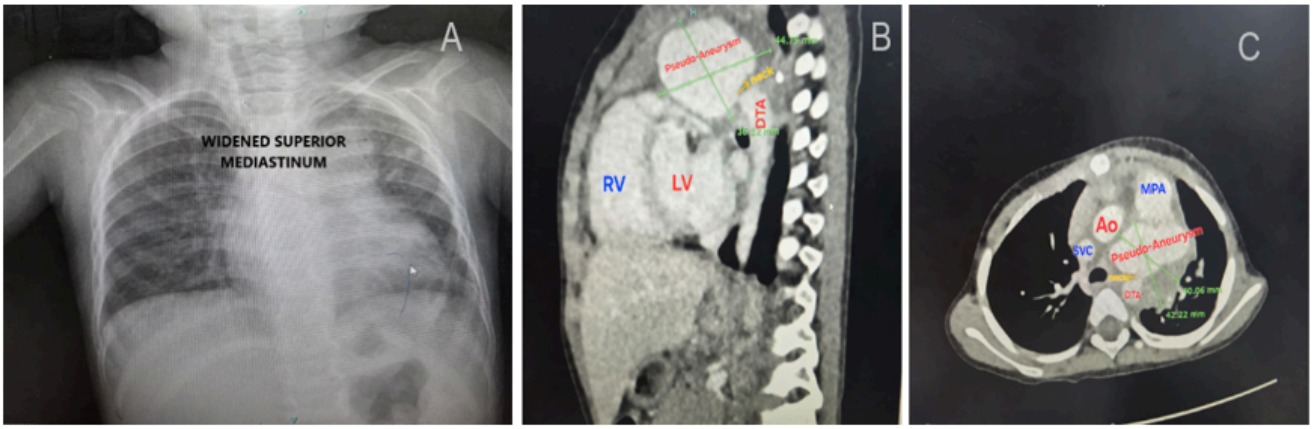


Figure 2

(A) Follow-up chest radiograph post-vegetectomy demonstrating widening of the superior mediastinum with associated opacity in the left upper hemithorax. Computed tomography depicting aortic pseudoaneurysm. [B] Sagittal section, [C] axial section. Ao = Aorta, RV = Right ventricle, LV = Left

ventricle, DTA = Descending thoracic aorta, neck = Neck of pseudoaneurysm, MPA = Main pulmonary artery, and SVC = Superior vena cava. Note the location of neck of pseudoaneurysm near the surgical site.

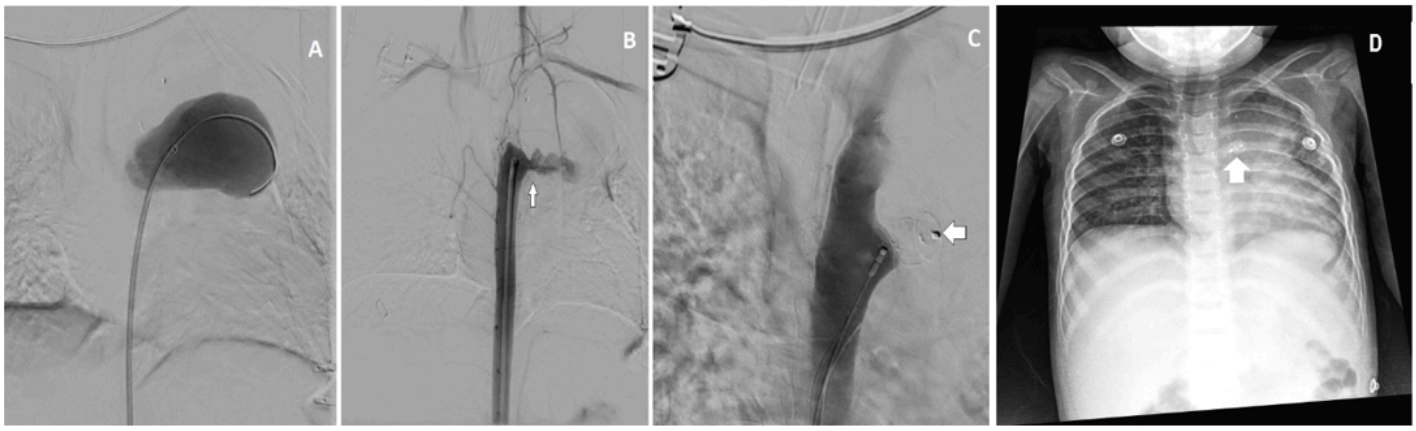


Figure 3

Device closure of pseudoaneurysm. (A) Aortic pseudoaneurysm (B) pseudoaneurysm neck (C) no residual filling of pseudoaneurysm sac post-device closure (D) post-procedure chest x ray showing Amplatzer plug in intended position.

Supplementary Files

This is a list of supplementary files associated with this preprint. Click to download.

- video1BMC.mp4