

## CASE REPORT

# Pseudoaneurysm formation following ligation of a patent ductus arteriosus

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### INTRODUCTION

The patent ductus arteriosus (PDA) is a vascular connection that connects the proximal descending aorta to the roof of the main pulmonary artery. Since its introduction in 1938, surgical treatment of PDA has been a safe procedure with minimal complications. However, aneurysm formation following surgical ligation of PDA is a rare but fatal complication with only a few case reports in the literature. Even surgical treatment carries a high mortality rate. We present a case of ductal pseudoaneurysm formation 3 weeks after ventricular septal defect (VSD) closure and PDA ligation. The patient underwent successful repair with aneurysmal excision and pericardial patch aortoplasty.

**Keywords:** Pseudoaneurysm, patent ductus arteriosus, thoracotomy, cardiopulmonary bypass

### CASE REPORT

A 4-month-old boy was evaluated at routine post operative echo visit and was diagnosed with a large pseudoaneurysm at the PDA ligation site. Three weeks ago, he had undergone peri-membranous VSD closure and PDA ligation. The PDA, which measured 5mm, had been ligated with no.2 black silk. He had moderate left pleural effusion that required intercostal tube insertion on post operative day 02, but his post operative course was otherwise uneventful. He was asymptomatic until this presentation. The physical examination was normal. No murmur was detected.

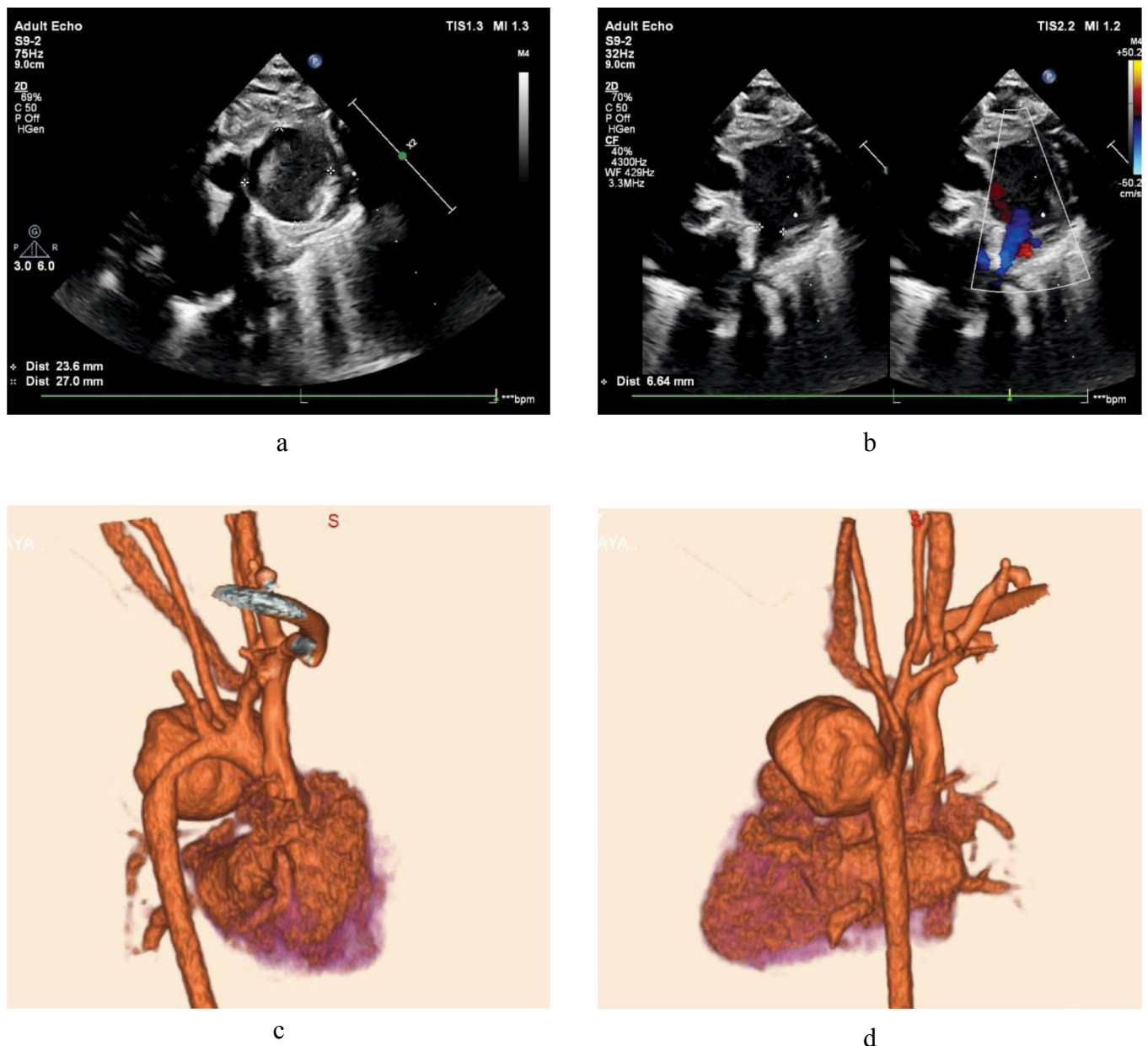
The chest x ray was normal. Echocardiography revealed a 23\*27mm pseudoaneurysm in the PDA region with an opening of 6.6mm into the aorta (Figure 1. a and b). It was compressing the aortic arch, main pulmonary artery (MPA) and left pulmonary artery (LPA). Computed tomography angiography (CTA) confirmed the pseudoaneurysm measuring 4\*3.4\*3cm (Figure 1.c and d). Further investigations showed a high CRP level (80 mg/dl) and high WBC count (15600/uL). He developed intermittent low-grade fever spikes while awaiting surgery.

The patient underwent revision surgery 3 weeks after the initial surgery to re-intervene the pseudoaneurysm. A median sternotomy and aorto-bicaval cannulation were performed to establish cardiopulmonary bypass. The heart was arrested using antegrade Del Nido cardioplegia and circulation was arrested at 22°C with adequate cerebral protection. The pseudoaneurysmal sac was completely excised and sent for culture, revealing a 7mm opening communicating with the aorta. An autologous pericardial patch aortoplasty was performed using 6/0 polypropylene. Cardiopulmonary bypass, aortic cross-clamping, and circulatory arrest times were 134, 60, and 20 minutes, respectively.

Coliform growth was detected in the culture and the patient received piperacillin tazobactam for 2 weeks as per sensitivity. His post operative course was complicated by sternal dehiscence and sternal rewiring was performed on post operative day 10. He was discharged on day 21. One month after being discharged, the patient had a normal echocardiogram and showed no signs of neurological dysfunction.

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**Figure 1.** Post-operative pseudoaneurysm formation following PDA ligation. Echocardiographic appearances (a and b) and 3D reconstructed CT images (c and d). Note the giant size of the pseudoaneurysm which significantly compresses the aortic arch, MPA and LPA.

## DISCUSSION

PDA is a condition where the ductus arteriosus, a fetal structure connecting the aorta and pulmonary artery, fails to close after birth. It develops from the distal portion of the left sixth embryonic aortic arch. PDA aneurysms are rare but life-threatening. Most of them are pseudoaneurysms. They can be spontaneous or acquired, affecting both children and adults. There have been reports of pseudoaneurysm formation after transcatheter PDA device closure, treated successfully with endovascular stenting<sup>1</sup>. The mortality rate for untreated pseudoaneurysms is about 91%, while surgery reduces it to 26%<sup>2</sup>.

A pseudoaneurysm may progressively enlarge within weeks or years compressing the adjacent structures, resulting in symptoms such as back pain, dysphagia and dyspnoea. Other potential complications include infection, thromboembolism, tracheobronchial fistula formation and eventual rupture leading to life-threatening situations.

The exact aetiopathogenesis remains unclear. Several hypotheses have been proposed and recanalization appears to be an essential step. The ligatures may cut through the vessel wall re-establishing the flow through it. This may lead to hematoma formation within the vessel wall and subsequent pseudoaneurysm formation. Another hypothesis is post-stenotic dilatation of the ductus distal to a stenotic aortic end

ligature. This occurs when the pulmonary side ligature is more effective in occluding the PDA than the aortic side ligature, resulting in systemic flow through it.

In a case series by Ross RS et al. bacterial endarteritis was found to be commonly associated with postoperative aneurysm formation<sup>3</sup>. *Staphylococcus aureus* was the most common organism. Recanalized PDAs can easily become infected during bacteremic episodes, leading to the formation of mycotic aneurysms. These can release septic emboli causing pulmonary infarctions. Bahnson et al. postulated that intraoperative surgical contamination or contaminated ligatures could be the cause of infection<sup>4</sup>. However, with current prophylactic antibiotic strategies and sterile surgical environments, the likelihood of intraoperative contamination is highly unlikely. Pre-operative pulmonary hypertension is known to make PDA friable and may be another contributing factor to aneurysm formation. In our patient, preoperative high inflammatory markers and positive culture may suggest infective origin of the pseudoaneurysm.

To prevent pseudoaneurysm formation, measures such as meticulous surgical technique, reducing blood flow during PDA ligation, and dividing and suturing the ductus can be taken. However, it can rarely occur after division and suturing. The diagnosis is typically achieved through transthoracic echocardiography, with CT or magnetic resonance imaging providing improved anatomical delineation.

The surgical approach can be either a median sternotomy or a left lateral thoracotomy, utilizing moderate to deep hypothermia with or without circulatory arrest. Thoracic endovascular aortic repair (TEVAR) has been demonstrated in adults in selected case reports. In left thoracotomy approach, lower body perfusion after distal aortic clamping can be done with right axillary to femoral bypass or femoro-femoral bypass. While left thoracotomy provides easier access to the descending aorta, extensive lung adhesions and friable aneurysms can often result in rupture and exsanguination. Median sternotomy approach using total circulatory arrest has been found to yield better immediate and long-term outcomes<sup>5</sup>. Its main drawback is the limited access to the descending aorta and hilum, but

the aneurysm dissection can be done without dissecting adhered lung and pleura.

In conclusion, large pseudoaneurysm at the PDA ligation site can be repaired with midline sternotomy approach and hypothermic circulatory arrest with good success.

### Learning points

- Even seemingly simple surgeries like PDA ligation can result in rare but fatal complications such as pseudoaneurysm formation.
- In the pediatric surgical setting, it is crucial to pay close attention to even the smallest details, such as the tension of sutures. Overlooking these details can result in undesirable outcomes.

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