The Heart Surgery Forum #2015-1449
19 (4), 2016 [Epub August 2016]
doi: 10.1532/hsf.1449

Online address: http://journal.hsforum.com

Unintended Pulmonary Artery Ligation during PDA Ligation

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ABSTRACT

A 10-day-old boy was transferred to our hospital due to tachypnea. Patent ductus arteriosus (PDA), 4.8 mm in diameter, with small ASD was diagnosed on echocardiography. Surgical ligation of the ductus was performed after failure of three cycles of ibuprofen. However, the ductus remained open on routine postoperative echocardiography on the second postoperative day, and chest CT revealed inadvertent ligation of the left pulmonary artery (LPA) rather than the PDA. Emergent operation successfully reopened the clipped LPA and ligated the ductus on the same (second postoperative) day.

Mechanical ventilator support was weaned on postoperative day 21, and the baby was discharged on postoperative day 47 with a normal left lung shadow.

INTRODUCTION

Patent ductus arteriosus (PDA) is a congenital heart disease with various symptoms [Lam 2015]. Large and symptomatic PDAs require surgical intervention. Surgical ligation is recommended if the PDA remains open after conservative treatment. Ductal tearing during surgery can be a fatal complication, but can be detected immediately. However, left pulmonary artery (LPA) ligation during trial ductal ligation can also be fatal, and is difficult to detect both during surgery and immediately afterwards [Orzel 1986; Jaffe 1986]. We report a case of unintended LPA ligation during the course of PDA treatment.

CASE REPORT

A 10-day-old boy was transferred to our NICU due to tachypnea and moaning. The male infant was born at 36 weeks' gestation with a body weight of 3.0 kg, and had ventilator support with heart failure management. Echocardiography revealed PDA (4.8 mm in diameter) with small ASD. Three cycles of ibuprofen were administered for PDA closure, but conservative therapy failed, so surgical closure of the PDA was planned.

A small posterolateral thoracotomy was performed via the fourth intercostal space. Subcutaneous tissue and muscle were severely edematous and the lung parenchyma was stiff. During dissection of the upper part of the descending thoracic aorta, the mediastinal pleural tissue was also found to be severely edematous. We misunderstood hypoplastic isthmic part of the descending aorta to left subclavian artery (LSCA) (Figure 1, A), and misjudged the ductus to proximal descending aorta, and LPA to ductus inadvertently. The left recurrent laryngeal nerve was located on the undersurface of the LPA. This vessel, which was 5 mm in diameter, was double-clipped with large-sized hemoclips, and the immediate postoperative course was stable.

Two days after the operation, routine follow-up echocardiography was performed, but the PDA still appeared to be 5 mm in diameter, and thus chest CT angiography was performed (Figure 1, B). The 2 clips were located on the LPA and no blood flow was observed to the whole left lung parenchyma. Moderate coarctation of the aorta was found with the hypoplastic isthmic portion of the descending thoracic aorta, which was thought to be the LSCA. Emergent reoperation was performed to remove the clips and to ligate the PDA. The left lung was purple in color and stiffer than seen previously. The PDA and ligated LPA were dissected carefully from the proximal descending aorta and arch branches. However, removal of the clips without damaging the vessel was

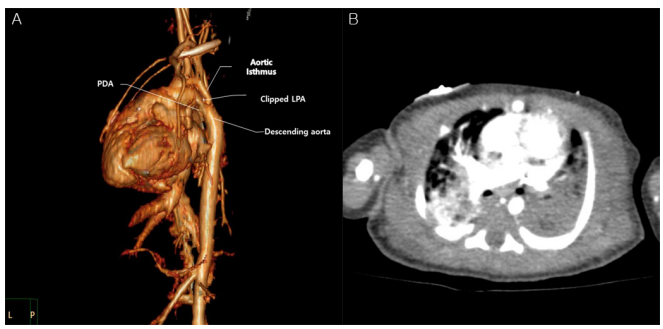

Figure 1. Chest CT angiography on postoperative day (POD) 2 following the initial operation. (A) Reconstructed 3D image showing the clipped LPA and remaining PDA. A narrow aortic isthmus was observed and PDA remained. The left pulmonary artery was clipped. (B) Chest CT showing interruption of left pulmonary artery blood flow. There was no left pulmonary artery blood flow due to inadvertent clipping.

Received November 12, 2015; received in revised form April 14, 2016; accepted May 15, 2016.

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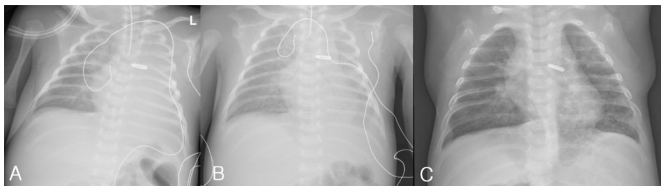

Figure 2. Chest AP during the recovery phase. (A) Chest AP immediately following reoperation. Haziness was observed over the whole left lung field. (B) Chest AP on POD 14. The parenchymal lung shadow was improved on the left side. (C) Chest AP on POD 102. Both lung fields appeared normal and the baby was in good overall health.

challenging. A towel clip was applied to open the hemoclips, which were spread and removed without injury to the LPA. The ductus was successfully ligated with hemoclips. Postoperatively, the whole left lung was hazy on CXR (Figure 2, A) and the color had not changed. Follow-up CXR was performed periodically. The parenchymal shadow improved after postoperative day (POD) 14 (Figure 2, B) and weaning of the ventilator and extubation of the endotracheal tube were done on POD 21. The infant was discharged on POD 47 with laboratory values of white blood cells 10,110 and C-reactive protein 0.34. The patient remained in good condition during the 2-month follow-up period (Figure 2, C) and arterial blood gas values were 7.36 (pH), 36.3 mmHg (PCO_2), 128 mmHg (PO_2), and 20.1 (HCO_3^-).

DISCUSSION

Aortic arch anatomy in premature babies may be complex, and making ligation of PDA, especially combined with uninformed coarctation, could be a challenge.

Unusual aortic arch and neighboring anatomy can make it difficult to differentiate the PDA from the hypoplastic isthmic part of the descending thoracic aorta. Therefore, complete and clear dissection to the arch branches is necessary in all cases of ductal ligation.

After our mistake was detected, prompt diagnosis and emergent removal of the inadvertent LPA ligation with ductal ligation was performed. In other reports, although lung parenchyma recovered after 4 days of LPA ligation, it took 6 months to completely restore lung function [Fleming 1983]. Thus, rapid detection and repair are critical. Pneumonectomy or other lung resection may need to be considered if septic shock or necrotic pneumonitis develops. In this case, vital signs and radiographic images showed gradual improvement, and life-threatening complications did not occur. In our case, it took 3 weeks for a normal shadow to appear on CXR after release of inadvertent LPA ligation, and the condition of the baby was good 3 months after reoperation. However, the long-term respiratory outcomes of this case are not yet clear.

In conclusion, complete and clear dissection to the arch branches is needed in all cases of ductal ligation, especially when anatomic ambiguities are found around the ductus, in order to avoid ligation errors and associated complications.

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