Successful Percutaneous Thrombectomy of the Left Pulmonary Artery in a Neonate After a Patent Ductus Arteriosus Clipping

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Summary

We report a neonate with a successful percutaneous thrombectomy of a total thrombotic occlusion of the left pulmonary artery (LPA) after a surgical clipping for a patent ductus arteriosus (PDA). We suspected the compression of the LPA by the clipping and postoperative hemodynamic instability caused the LPA obstruction. After the surgical removal of the PDA clip and division of the PDA, we could safely retrieve the LPA thrombus with a non-hydrodynamic thrombectomy catheter for coronary arteries.

Key words: Catheter intervention, Infant, Thrombus, Pulmonary infarction

C lipping for a neonatal patent ductus arteriosus (PDA) has been commonly performed because of the short procedure time and minimally invasive method for neonates. Various complications after clipping the PDA have been reported: intraoperative bleeding, pneumothoraces, phrenic nerve paralysis, injury to the lymph vessels, and left recurrent laryngeal nerve paralysis. However, pulmonary infarctions have never been reported.

A pulmonary artery thrombus could lead to hypoxia and a fatal hemodynamic condition. Therefore, the patient should undergo an urgent thrombus removal by thrombolytic medication, percutaneous thrombectomy, or surgery. In children, percutaneous thrombectomies in the pulmonary artery have been performed after systemic-pulmonary or superior vena cava to pulmonary shunts. There are two percutaneous transcatheter mechanical thrombectomy methods: (1) the mechanical crash method using guidewires, catheters, or balloons, and (2) the hydrodynamic crash and aspiration method. We report on a 28-day-old patient with a successful percutaneous thrombectomy of a total thrombotic occlusion of the left pulmonary artery (LPA) by a non-hydrodynamic thrombectomy catheter for coronary arteries after a PDA clipping.

Case Report

An elective cesarean section delivered an infant weighing 2,870 g due to a cephalopelvic disproportion at the gestational age of 38 weeks. There was no history of collagen disease or coagulation disorders in her family. She was admitted to the neonatal intensive care unit with mild respiratory distress. She had a minor anomaly of an enlargement of her head size, hypertelorism, forehead protrusion, and low-set ears. A patent ductus arteriosus (PDA) was confirmed by echocardiography. She had prolonged respiratory distress and poor weight gain, and those symptoms were refractory to water restrictions, diuretics, and three doses of indomethacin. Then she underwent a surgical clipping of the PDA with a 12 mm Hemoclip on day 28.

Eight hours after the clipping, her systemic mean blood pressure decreased from 50 to 20 mmHg, and her partial pressure of the oxygen (PO2) decreased from 90 to 45 mmHg. This condition was not ameliorated even after using hydrocortisone, dopamine, or a fluid infusion. Echocardiography revealed thrombotic occlusion of the left pulmonary artery (LPA). It was unresolved by a continuous infusion of unfractionated heparin and a tissue plasminogen activator (t-PA, Alteplase 300000 units/kg). The patient was transferred to our hospital for an advanced direct intervention for a left pulmonary thrombotic embolism.

Enhanced thoracic CT (eCT) showed that the PDA clip was placed at the center of the PDA, a proper location, and it was likely that the clip was slightly pressed on the LPA (Figure 1A-C). We suspected that the PDA clip disrupted the anterograde flow of the LPA and formed a thrombus in the LPA. We first requested a surgical thrombectomy procedure but were unable to complete it due to the patient's unstable condition. Therefore, we proceeded with a percutaneous approach.

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Figure 1. Images from the enhanced computed tomography scan. A: Axial view; B: Sagittal view; C: Coronal view. The red asterisk (*) is a thrombus in the left pulmonary artery. The solid arrows show the patent ductus arteriosus (PDA) clip, and the location of the clip appears to be in an appropriate position for the PDA. Ao indicates aorta; LPA, left pulmonary artery; MPA, main pulmonary artery; and RPA, right pulmonary artery.

Figure 2. Angiography of the pulmonary artery before and after the procedure and the removed thrombus. A: Angiography before the thrombectomy. The contrast media does not run to the left pulmonary artery (LPA) from the proximal branch due to an occlusion caused by a thrombus (*). A chest tube is placed in the left thoracic cavity for drainage after a patent ductus arteriosus division and ligation. B: Angiography after the thrombectomy. The LPA is recanalized throughout the proximal side to the distal side. C: This photograph shows parts of a red thrombus sucked out by the device from the left pulmonary artery. LPA indicates left pulmonary artery; and RPA, right pulmonary artery.

Thrombectomy, but our surgeons told us that a surgical thrombectomy of the proximal LPA might be possible, but it may be impossible to remove all the thrombus in the distal LPA. Therefore, removal of the clip, followed by division and ligation of the PDA with a left lateral thoracotomy, was performed 20 hours from the onset of the thrombus formation. Then a percutaneous thrombectomy for the thrombotic occlusion of the LPA was scheduled.

The LPA was totally occluded at the proximal branch (Figure 2A). A 0.035 inch Terumo coated guidewire pene-
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Figure 3. Fluoroscopic images during the procedure. A, B: These fluoroscopic images show the procedure of the thrombectomy. The schemas are shown on the right side of the images, respectively. The red arrows, blue arrows, and black arrows indicate the tip marker of the outer catheter, tip marker of the inner catheter of the Dio™, and 0.0018 inch guidewire, respectively. The thrombus could not be aspirated by suction from the outer catheter (A). The inner catheter was inserted into the outer catheter. The thrombus could be aspirated while digging the thrombus out of the LPA by inserting and pulling back on the inner catheter inside the outer catheter (B). LPA indicates left pulmonary artery.

trated the thrombus to the distal LPA, and then a 5 Fr Dio™ thrombus aspiration catheter (NIPRO Corporation, Osaka, Japan) was advanced along the wire into the distal LPA. However, the thrombus could not be aspirated by this catheter (Figure 3A).

A soft inner Dio™ catheter was then inserted into the outer Dio™ catheter and advanced to the distal LPA. By inserting and pulling the inner and outer catheters back and forth from the proximal to the LPA distal branch, the obstructed thrombus was finally aspirated (Figure 3B). Recanalization of the LPA throughout the proximal branch to the peripheral PA was confirmed by angiography (Figure 2B). The aspirated thrombus was a crushed white and red thrombus (Figure 2C). In total 60 mL of blood, aspirated while attempting to aspirate the thrombus, was given as a transfusion. Heparin was continuously administered after the procedure to prevent thromboses and to achieve thrombolysis.

Her circulatory status and oxygen saturation rapidly improved after those procedures. She was transferred to the referring hospital and was administered warfarin and aspirin until a genetic analysis for protein C excluded coagulopathy. She was diagnosed with Sotos syndrome by her minor anomaly, and it was confirmed by genetic analysis of a nuclear receptor binding set domain protein one deletion (NSD1−: ish del[5]q35q35]). She has been followed up without any recurrence of a thrombus for two years after that.

Discussion

To the best of our knowledge, this is the first report of a successful thrombectomy for occlusion of the LPA in a neonate using a non-hydrodynamic thrombectomy cathe-
The PDA clip was located almost at the center of the ductus arteriosus, which was an optimal position for the PDA closure; however, the eCT suggested the PDA clip was compressing the LPA. The clip compressing the LPA disrupted the blood flow in the LPA and led to the obstruction of the LPA, hypotension, and desaturation. At the time of the surgery or during the acute postoperative period, considerable endothelium damage due to surgical procedures or a PDA clip may result in the formation of a thrombus in the proximal LPA. Also, part of the thrombus in the proximal LPA may crumble and move to the distal part of the LPA. This decrease in the blood flow velocity in the peripheral LPA may further form an LPA thrombus. Therefore, the placement of a PDA clip per se may potentially have the risk of compressing other organs.9)

In other countries, the efficacy of a hydrodynamic thrombectomy catheter device has been reported. Concerning neonates, 2 cases of percutaneous thrombectomies for pulmonary arteries have been reported. First, a thrombectomy of thrombotic occlusion of the LPA with a hydrodynamic catheter in a 1-day-old neonate after surgery for an aortopulmonary shunt was reported with extracorporeal membrane oxygenator support. Although the thrombus was removed, the neonate finally died. Second, a thrombectomy of thrombotic occlusion of an LPA with a hydrodynamic catheter in a 3-day-old neonate after a surgical repair of a total anomalous pulmonary vein drainage was reported. The LPA was recanalized, but ventricular pacing was needed during the procedure due to atrioventricular block.10) However, that device is also reported to cause significant adverse events such as hypotension, hemolysis, hyperhydration, and bradycardia.5,11)

The Dio™ catheter has the feature of excellent back-up support,7) good followability, larger inner diameter, and smaller outer diameter than other thrombectomy catheters in Japan. The inner diameter of the 5 Fr Dio™ is 1.51 mm, which is larger than the 1.11 mm and 1.25 mm inner diameter of the other 6 Fr and 7 Fr thrombectomy catheters, respectively. Another key to this procedure’s success was to dig out the thrombus by inserting and pulling back and forth on the inner and outer catheters.

Conclusion

We have to be reminded that pulmonary embolisms may occur after a PDA clipping. In that instance, a percutaneous thrombectomy with a non-hydrodynamic thrombectomy catheter for the pulmonary artery could be a safe, effective interventional procedure even in neonates.

Disclosure

Conflicts of interest: None.

References

1. Mandalen P, Brown S, Kukkady A, Samarakkody U. Surgical closure of patent ductus arteriosus in preterm low birth weight infants. Congenit Heart Dis 2009; 4: 34-7.
2. Weisz DE, McNamara PJ. Patent ductus arteriosus ligation and adverse outcomes: causality or bias? J Clin Neonatol 2014; 3: 67-75.
3. Matsubara D, Katoaka K, Takahashi H, Minami T, Yamagata T. A patient-specific hollow three-dimensional model for simulating percutaneous occlusion of patent ductus arteriosus. Int Heart J 2019; 60: 100-7.
4. Funakoshi H, Matsui H, Fushimi K, Yasunaga H. Variation in patient backgrounds, practice patterns, and outcomes of high-risk pulmonary embolism in Japan. Int Heart J 2018; 59: 367-71.
5. Fleming GA, Khan M, Janssen D, Doyle T. Angiojet rheolytic thrombectomy in infants following cardiac surgery. Catheter Cardiovasc Interv 2010; 76: 233-40.
6. Qureshi AM, Petit CJ, Crystal MA, Liou A, Khan A, Justino H. Efficacy and safety of catheter-based rheolytic and aspiration thrombectomy in children. Catheter Cardiovasc Interv 2016; 87: 1273-80.
7. Komatsu T, Yaguchi I, Yufa T. Successful percutaneous coronary intervention of an anomalous right coronary artery with high anterior takeoff using a DIO thrombus aspiration catheter. J Invasive Cardiol 2012; 24: e185-7.
8. Tatton-Brown K, Douglas J, Coleman K, et al. Genotype-phenotype associations in Sotos syndrome: an analysis of 266 individuals with NSD1 aberrations. Am J Hum Genet 2005; 77: 193-204.
9. Harris LL, Krishnamurthy R, Browne LP, Morales DL, Friedman EM. Left main bronchus obstruction after patent ductus arteriosus ligation: an unusual complication. Int J Pediatr Otorhinolaryngol 2012; 76: 1855-6.
10. Nicholson GT, Kogon B, Vincent R. AngioJet rheolytic thrombectomy in a neonate with pulmonary artery thrombus. Catheter Cardiovasc Interv 2013; 82: e704-7.
11. Dwarka D, Schwartz SA, Smyth SH, O’Brien MJ. Bradyarrhythmias during use of the AngioJet system. J Vasc Interv Radiol 2006; 17: 1693-5.