

A Rare Case of Double Outlet Right Ventricle with Subaortic VSD Complicated by Pneumonia, Renal Failure, and Tracheomalacia in Infancy: A Case Report

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KEYWORDS

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ABSTRACT:

Introduction: Double Outlet Right Ventricle (DORV) with Subaortic VSD is a high-risk congenital heart disease that potentially leads to direct complications due to its anatomical abnormalities or prolonged treatment in the intensive care unit. Congestive heart failure (CHF) with pulmonary oedema, recurrent pneumonia, and acute kidney injury (AKI) are some of the complications that can occur. **Objectives:** This study aimed to report a 9-month-old infant who was admitted to the ICU with DORV and Subaortic VSD complicated with CHF, AKI, and pneumonia. **Case Report:** The patient received prolonged treatment in the ICU with suboptimum conservative treatment due to congenital heart defects and required palliative surgery, namely pulmonary artery banding. Handling complex cases of the infant with congenital heart defects and their complications requires a multidisciplinary approach. Collaboration among healthcare providers can effectively support patients with similar conditions.

1. Introduction

Double Outlet Right Ventricle (DORV) is a congenital heart disease that requires intensive care due to the disorder itself or other complications that arise. The International Society for Nomenclature of Pediatric and Congenital Heart Disease defined DORV as an abnormally structured heart where both main arteries are mostly connected to the right ventricle. [1]. This disease is a complex heart disease with conotruncal anomaly that comprises less than 1% of all congenital heart defects [2].

DORV patients with subaortic ventricular septal defect (VSD) type are divided into two categories based on pulmonary stenosis. Patients without pulmonary stenosis have a clinical appearance similar to pure VSD patients with L to R shunt. These patients will have excessive pulmonary blood flow (PBF) and signs of congestive heart failure because PVR decreases after birth. Babies born with these congenital heart defects (CHD) often experience complications. Defects of the ventricle septum and malnutrition are identified as a risk factor for recurrent pneumonia. Ventricular septal defect patients carry the potential of having recurrent pneumonia as big as 1.551 times and malnutrition 2.591 times compared to the normal population [3]. The patient requires a multidisciplinary approach to prevent respiratory complications.

Data providing the number and risk factors of death of children with CHD who are treated for pneumonia in Indonesia are still limited. Most studies only reveal the risk factors for pneumonia in children with CHD. In addition, the incidence of pneumonia in children with CHD is relatively high at Dr. Soetomo General Academic Hospital and is at risk of becoming severe pneumonia, leading to death. Patients are at risk for experiencing prolonged treatment in the ICU and using mechanical ventilation to provide respiratory assistance to patients. Multidisciplinary management is needed for the treatment of congenital heart disease patients with complex complications similar to this case.

2. Objectives

In this paper, we discuss a male infant patient who was treated in the ICU with congenital heart disease DORV accompanied by complications of congestive heart failure (CHF), pneumonia, and acute kidney injury (AKI).

3. Case Presentation

A 9-month-old male infant with a weight of 4.6 kg and length of 65cm was admitted to the hospital on May 7th, 2023. The indication of intensive care was respiratory distress. The patient had a history of previous hemodialysis. The patient was immediately intubated and connected to a mechanical ventilation machine to support his breathing. The patient also had a fever, with the highest temperature of 39.1°C.

Based on the history taking, the patient was previously admitted to Benowo General Hospital for 1.5 months in PICU before being referred to Dr Soetomo General Hospital. The patient was consulted for ENT due to suspicion of tracheomalacia. The patient also had a history of outpatient care and received Sildenafil 3 x 2 mg, Lisinopril 1 x 0.4 mg, Furosemide 1 x 4 mg, Spironolactone 1 x 4mg, and Infantrini 8 x 88mg for nutrition improvement.

A free airway was obtained from the physical examination, a respiratory rate of 60 x/min, a vesicular breathing sound with rales on both lungs, subcostal retraction (+), and oxygen saturation of 89% (with a simple mask). The patient presented with warm extremities, CRT < 3 seconds, BP 70/40 mmHg, HR 151x/minute, single heart sounds, and murmur (+). The patient showed weak movement and crying. A urine catheter was installed with little production. There were no abnormalities in the bowel and extremities.

Laboratory examination on May 14th, 2023 revealed Hb 10.3, Hematocrit 31.9, Leukocytes 11.65, Platelets 227, Neutrophils 87.2, Lymphocytes 9.2, BUN 55.6, Creatinine 0.9, Uric acid 11.2, Sodium 140, Potassium 4.7, Chloride 98, Calcium 9.8, Magnesium 2.6, Phosphate 6.48. Echocardiography on May 11th, 2023, concluded the diagnosis of DORV + VSD subaortic-inlet + ASD + Non-restrictive pulmonary blood flow. Urology ultrasonography and renal Doppler examination on May 12th, 2023, revealed bilateral parenchymal kidney disease and no abnormalities in the bladder and prostate. Thoracal x-ray with AP projection on May 13th, 2023, showed suspected pneumonia accompanied by pulmonary oedema and cardiomegaly (see Figure 1).

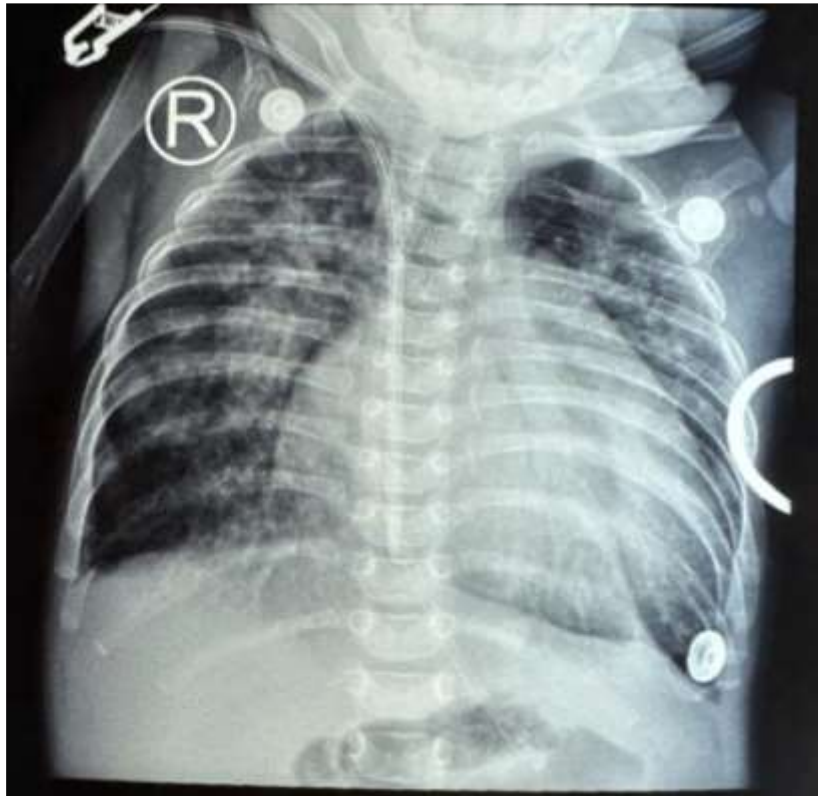


Figure 1: Chest x-ray revealed cardiomegaly, lung infiltrates, and lung oedema

Finally, the patient was diagnosed as DORV + subaortic VSD with non-restrictive pulmonary blood flow + ASD + CHF+ lung oedema + Pneumonia + AKI on failure stage + post-hemodialysis + severely underweight + stunted + severely wasted + respiratory failure. The patient was then intubated and treated in the ICU. In the ICU, the patient received initial therapy with O₂ ventilator setting PSIMV Pins 15, rate 40 PEEP 5, FiO₂ 40 %, intravenous fluid D5 1/4 NS 25 ml/24 hours, E1-E2 D5 30ml, E3- E8 modified F100 50 ml, oral Spironolactone 5mg /8 hours, oral Furosemide 5mg /8 hours, oral Allopurinol 200mg/24 hours, IV Cefoperazone sulbactam 300 mg/8 hour, IV Omeprazole 5mg /12 hours, IV Metoclopramide 1mg /8 hours, IV Paracetamol 75 mg/24 hours, IV Midazolam 1mg/mL, Blood sugar check in every 24 hours, normal saline nebulisation in every 6 hours, airway suction as indicated, mobilisation every 12 hours, bathing/perineal hygiene/12 hours, oral hygiene/12 hours, and head up 30°.

On the second day in the ICU, the patient was still short of breath. The patient was planned to undergo a second hemodialysis (HD). The highest temperature was 39.1⁰, and the respiratory rate was 50x/min, oxygen saturation was 85 % (with ventilator), rales were on both lungs, murmur +, and there was an increased need for airway suction. Important laboratory findings were leukocytes 24,660, BUN 67 (post-HD was 45), Serum creatinine 0.8 (post-HD was 0.7), and uric acid 9.8 (post-HD was 6). From the chest X-ray examination, an increased infiltrate was obtained compared to the previous X-ray, cardiomegaly, which was then concluded as pneumonia with pulmonary oedema and cardiomegaly. The patient was diagnosed with HAP-related pneumonia. The added therapy was antibiotic IV Cefoperazone sulbactam, which was administered 300 mg/ 8 hours.

On the 12th day in the ICU (May 25th, 2023), the patient experienced difficulty in mechanical ventilation weaning. When the endotracheal tube was dislodged, pink, frothy sputum was found. It was later found out that the patient was admitted to the PICU in 2022 for 1.5 months due to difficulty in mechanical ventilation weaning off. The patient was suspected of having tracheomalacia, so it was decided to perform a tracheostomy (as recommended by the pediatric surgeon).

On the 13th day in the ICU (May 23rd, 2023), the patient was short of breath and still on ventilator support; rales and murmurs were found in the auscultation and increased need for sputum suction. The results of important laboratory tests were leukocytes 21,590, creatinine 0.7, BUN 55.9, and procalcitonin 16.95. Blood culture results on May 13th, 2023, revealed *Staphylococcus coagulase-negative Vancomycin-sensitive*; culture of ETT found *Pseudomonas aeruginosa*: sensitive to Ceftazidime. The patient was then diagnosed with VAP-related pneumonia. The patient received therapy of IV fluid D5 1/4 NS 25 ml/24 hours, Infantrini 8x 60 kcal (25 mL), oral Spironolactone 5 mg /8 hours, oral Furosemide 5mg /8 hours, oral Allopurinol 200mg/ 24 hours, Paracetamol syrup 75 mg/ 6 hours when hyperthermia, IV Fluconazole 30 mg/24 hours, IV Ceftazidime 1x250 mg / 24 hours, IV Vancomycin 1x50 mg, IV Omeprazole 5mg /12 hours, IV Metoclopramide 1.5 mg /8 hours, IV Dobutamine (5mg/ml, 0.18 cc/hour), IV Midazolam (1mg/ml, 0.1 mL/hour).

On the 18th day in the ICU (May 31st, 2023), the patient was still short of breath, respiratory rate of >50 x / minute with an oxygen tracheostomy mask of 0.5 lpm. Blood pressure was 92/53 mmHg on dobutamine three gamma. The patient underwent 6th hemodialysis, with laboratory results (pre → post-HD), Leukocytes 13.55 → 16.93, Bun 47.9 → 12.2, SCr: 0.5 → 0.4. A multidisciplinary discussion on May 30th, 2023, concluded that the patient would undergo palliative surgery of Urgent Pulmonary Artery Banding on May 31st, 2023. Besides pulmonary artery banding, a quite large patent ductus arteriosus was concomitantly found, and ligation was performed.

4. Discussion

Double Outlet Right Ventricle (DORV) is defined as both the aorta and pulmonary artery (PA) arising from >150% of the Right ventricle (RV), 100% of one great artery arising from the RV, and more than 50% of the other great artery originating from the Left ventricle (LV) [4]. The physiology of DORV can result in clinical presentations ranging from pulmonary overcirculation and congestive heart failure to cyanosis and pulmonary hypoperfusion.

Types of DORV and clinical presentations are: (1) DORV with subaortic VSD where the great vessels are usually associated; (2) DORV with subpulmonary VSD; (3) DORV with doubly committed VSD; and (4) DORV with noncommitted VSD. Two-thirds of patients with DORV and subaortic VSD also have PS to varying degrees. DORV, VSD subaortic without PS appear with physiology-type VSD. Patients with this diagnosis will have excessive pulmonary blood flow and signs of congestive heart failure because the PVR decreases after birth [5]. Infants with this disorder will have excessive pulmonary blood flow and symptomatic congestive failure in the first week of life, worsening as the PVR decreases. Examination may reveal tachypnea, tachycardia, a holosystolic murmur, and a mid-diastolic rumble (mitral flow murmur). Prominent cardiomegaly and pulmonary vascularity are seen on chest radiography. The mainstay of therapy is non-operative, anti-congestive management, including diuretics and decreasing afterload, mechanical ventilation, and lung vasoconstriction strategies

such as permissive hypercapnia and low inspired- O_2 concentration. When conservative measures fail, surgical management may include palliative PA banding or definitive surgical repair [6]. DORV, subaortic VSD with PS, presents with TOF-type physiology and signs of decreased PBF due to PS. The degree of PS determines the severity of symptoms and clinical signs. The infant will present with a systolic murmur and varying degrees of cyanosis depending on the degree of PS. If pulmonary stenosis is severe or near critical, supplemental flow from a patent ductus arteriosus (PDA) may be required to ensure adequate PBF.

Additional examinations that can be found include chest radiography with cardiomegaly with increased lung vascularisation on DORV without PS or normal to small heart size and reduced lung vascularisation on DORV with PS. Electrocardiography may present right axis deviation and RVH in most DORV or left axis deviation and RVH when associated with AVSD. Echocardiography is needed to analyse heart anatomy segments systematically and sequentially, assess ventricular size to determine the adequacy of biventricular repair, size and location of VSD, measure distance from tricuspid and pulmonary valves, location of great arteries, estimate PA pressure, and coronary artery anatomy when possible. CT and MRI may be required in many cases to evaluate associated anomalies, especially anomalies of the aortic arch, pulmonary veins, and airways. Heart catheterisation and angiography are not routine but may be performed to measure R_p and perform PA wedge angiogram when Eisenmenger reaction is suspected [4]. In the patient we presented, it was found CHF, cardiomegaly on chest x-ray, and diagnosis of DORV subaortic VSD was confirmed on Echocardiography.

In DORV VSD-type (Double-outlet right ventricle, subaortic VSD without pulmonary stenosis), these patients will have excessive pulmonary blood flow (PBF) and signs of congestive heart failure because PVR gradually decreases after birth. If the VSD is subaortic and there is no PS, pulmonary blood flow will be determined by the relationship of pulmonary vascular resistance (R_p) to systemic vascular resistance (R_s). When, as is usually the case after the first few weeks of life, R_p is less than R_s , pulmonary blood flow will be greater than systemic flow, with pulmonary artery oxygen saturation higher than usual. Because R_p is undoubtedly increased as the response to abnormal volume (and pressure) burden, pulmonary blood flow is also reduced. Eventually, if left untreated, this usually leads to obstructive pulmonary vascular disease with severe and irreversible R_p increase, causing progressive cyanosis and early death [7]. Early pulmonary hypertension (PH), which happens on L to R shunt VSD, will cause high flow PH; however, if this continues, it will cause damage to the tunica intima and media of the lung tissue and will be replaced by fibrosis, which will cause high PH resistance that has a poor prognosis [3].

In DORV with increased pulmonary blood flow, signs of CHF may be present. Pulmonary overcirculation is reduced by adjusting the inspired O_2 concentration to maintain a systemic oxygen saturation of 80% to 85%. If mechanical ventilation is required, high FiO_2 and hyperventilation must truly be avoided, and $PaCO_2$ must maintained at 40 mm Hg. This may require sedation and paralysis of the patient. The goal of therapy is to maintain a balanced pulmonary-to-systemic blood flow ratio ($Q_p:Q_s = 1$). As with other lesions, CHF due to increased pulmonary blood flow can also be managed with diuretics, inotropic agents, and systemic afterload-reducing agents [7]. On this patient, signs and symptoms of CHF were observed: shortness of breath, tachypnea, tachycardia, rales +, murmur +, chest x-ray concluded suspected pneumonia accompanied by pulmonary oedema, cardiomegaly. Then,

the patient received Spironolactone (diuretic), Furosemide (diuretic), and IV. Midazolam (sedation), dobutamine (inotropic agent; systemic afterload reducer); O₂ saturation of 85%-92% on mechanical ventilation with FiO₂ 30; hyperventilation with respiratory rate of 60-70; PaCO₂ 38-66.

Kidney injury commonly occurs in patients with heart failure. Reduced cardiac function coupled with renal dysfunction has been termed CRS (Cardio Renal Syndrome). Ronco et al. divided CRS into five types depending on whether the symptoms are triggered by the kidney or heart and whether it is acute or chronic. The most common types seen in pediatric intensive care are Acute CRS (CRS type I – acute decompensated heart failure causes acute kidney failure) and secondary CRS (CRS type V – systemic disease causes organ dysfunction simultaneously). In any setting, declined urine output and fluid retention can worsen symptoms of heart failure and contribute to clinical deterioration [8]. AKI in children is associated with increased duration of mechanical ventilation, inotropes, more extended ICU stay, and increased mortality, even among patients with only minor changes in creatinine. Detection of AKI based on pRIFLE criteria is more sensitive in identifying milder cases of AKI [9].

Indications for hemodialysis procedures usually include: fluid retention with effusion (pleural effusion, ascites) and edema (lungs, brain), hyperkalemia ($K > 6.5$ mmol/L or symptomatic), cardiac volume overload (CVP > 18 mmHg), anuria and AKI > 2 days, increasing metabolic acidosis (pH < 7.0), urea > 200 mg/dL (> 35 mmol/L), rhabdomyolysis (myoglobin $> 5,000$ – $10,000$ ng/mL), and oliguria [10]. This patient was admitted with kidney function tests: BUN: 117.7 and SCr: 3.8. Normal paediatrics GFR Values (6-12 months) = 49 - 157 mL/min/1.73 m², eGFR (Black method): $[(0.413 \times \text{Height (cm)}) / \text{SCr (mg/dL)}]$, $(0.413 \times 65) / 3.8 = 7.06$, pRIFLE / AKIN / KDIGO = AKI stage 3 (Failure), and the patient underwent HD.

One of the significant and problematic morbidities in children with CHD is infection, which often interacts with malnutrition, causing complex health problems. Patients with acute infections, such as acute respiratory infections (ARI), will experience anorexia, malabsorption, or metabolic disorders. Inadequate calorie intake can cause a declining body weight, stunting, immunity disturbance, and mucosal damage. Changes in pulmonary circulation can cause structural abnormalities accompanied by decreased local cellular immunity, making children more susceptible to ARI. Children with CHD also experience disturbance in ventricle function, which furthermore increases lung pressure so that capillary leakage and lung oedema will occur. Recurrent ISPA is often seen in CHD patients, especially in infants under one year of age [11].

Hospital-acquired pneumonia (HAP) and Ventilator-associated pneumonia (VAP) are acquired health conditions with a significant risk of morbidity and mortality. HAP is pneumonia that develops at least 48 hours after hospital admission. VAP is HAP that develops at least 48 hours after endotracheal intubation [12]. In this patient, pneumonia occurred on May 15th, 2023, at least 48 hours after admission to the hospital. Pneumonia occurred after the patient was intubated and received mechanical ventilation for more than 48 hours. This patient experienced fever, shortness of breath, rales +. The patient needed an increased frequency of airway suction. Laboratory examination on May 15th, 2023 revealed: Leukocytes 24.66; on May 26th, Leukocytes was 21.59; on May 27th, Procalcitonin was 16.95. Chest x-ray on May 14th, 2023, showed increased lung infiltrates; conclusion: pneumonia accompanied by pulmonary oedema and cardiomegaly. Right Blood Culture results on May 18th, 2023, revealed

Staphylococcus coagulase-negative sensitive to Vancomycin, while culture of ETT showed *Pseudomonas aeruginosa*: sensitive to Ceftazidime. The patient was then treated with IV Cefoperazone sulbactam 300 mg/ 8 hours, IV Ceftazidime 1x250 mg / 24 hours, and IV Vancomycin 1x50 mg.

Congenital heart disease is a structural problem of the walls, valves, chambers, and arteries or veins adjacent to the heart that present at birth. These defects interfere with normal hemodynamics, leading to pathophysiology that is responsible for inadequate nutrient intake, inadequate nutrient absorption, and increased metabolic demands. Total energy requirements in the neonate consist of the energy required to maintain metabolism and the energy required for growth. Neonates with CHD are at risk for hypermetabolism, decreased mesenteric perfusion, delayed enteral feeding, and congenital difficulty in estimating their nutritional needs. Optimal nutritional provision can improve surgery outcomes, providing a solid foundation to improve or alleviate the pathophysiology associated with CHD. In addition, postoperative nutritional management that provides adequate protein, fat, and electrolytes can help speed recovery, reduce hospital costs, and prevent the long-term progression of malnutrition [13]. Nutritional status is assessed using Z scores, and interventions are given based on SAM management. This patient weighed 4.6 kg and was 65 cm long. The patient received modified F100 therapy. Therefore, this patient was assessed as severely underweight, stunted, and *severely wasted*.

Tracheomalacia is a developmental disorder of the airways that can result in the inability to open the airways mechanically [14]. The clinical presentation of Tracheomalacia (TM)/Tracheobronchomalacia (TBM) includes a variety of nonspecific airway symptoms, depending on the location, extent, and severity of the airway collapse. Many children with TM/TBM are asymptomatic until 2-3 months of age. However, in long-segment TM/TBM, symptoms may present at birth. In some patients, TM/TBM develops immediately after esophageal atresia / tracheal-esophageal fistula (EA/TEF) repair if the patient is not extubated. Most patients with TM/TBM may have a barking cough with expiratory grunts or inspiratory stridor. Severe airway collapse can lead to ineffective cough and decreased clearance of secretions. As a result, patients with TM/TBM are at increased risk for frequent respiratory tract infections, prolonged recovery, and recurrent or persistent pneumonia. Besides, airway obstruction often causes inadequate ventilation, which may cause exercise intolerance and hypoxic or apnea episodes. Symptoms may be aggravated by any activity or condition that increases the patient's intrathoracic pressure and respiratory effort, including activities such as coughing, crying, Valsalva manoeuvre, eating, forced expiration, or breathing while lying supine [15].

There are no definite standard guidelines for the diagnosis and evaluation of TM/TBM. Diagnosis must be suspected with a history and signs and symptoms that indicate TM/TBM, including barking cough, wheezing, recurrent pneumonia, persistent pulmonary infections, feeding difficulty with dyspnea, cough, and aspiration, and transient dyspnea, hypertension, oxygen dependence, ventilator dependence, cyanotic periods, and apparent life-threatening events (ALTE). Patients with apneic events require careful cardiac and neurologic evaluation to rule out these causes. Oesophageal abnormalities, including strictures, tracheoesophageal fistula, and gastroesophageal reflux, must be excluded. Definitive diagnosis of TM/TBM requires direct visualisation, which is achieved with flexible and rigid endoscopes, including laryngoscopy, tracheoscopy, and bronchoscopy [15]. This

patient had repetitive pneumonia, a history of PICU stay due to tracheomalacia, and weaning difficulty, and then the patient underwent a tracheostomy.

When conservative measures fail, operative management may include palliative PA banding or definitive surgical repair. Palliation in infancy (systemic shunt placement to lungs or PA banding) may be suitable for some patients, especially on a very small baby whose anatomy needs Fontan-type surgery. Corrective DORV surgery with increased pulmonary flow must be done during the first 3 months of life to minimise the morbidity of heart failure and avoid the risk of developing pulmonary vascular disease. In patients with DORV subaortic VSD type (without pulmonary stenosis), PA banding may be required initially to reduce pulmonary blood flow so that the patient can grow and maintain the pulmonary vascular bed until definitive repair can be performed [7]. Preoperative management of patients with DORV depends primarily on the patient's age and presenting signs. Medical management is temporary, and all symptomatic patients require surgical correction or palliation.

5. Conclusion

We report a case of a 9-month-old male infant who was admitted to the ICU due to respiratory distress with the diagnosis of DORV + VSD sub Aortic + CHF+ Pulmonary Edema + Pneumonia + AKI stage Failure + malnutrition + respiratory distress. The patient was intubated and received mechanical ventilation. During treatment, the patient experienced recurrent pneumonia, which improved with antibiotics. The patient was also given therapy related to CHF and AKI. Moreover, the patient had undergone several hemodialysis sessions during the course. The patient underwent a tracheostomy related to difficult weaning and suspected tracheomalacia. The patient's complex problems require multidisciplinary care. Conservative treatment that did not provide satisfactory results required Pulmonary artery banding surgery to improve the patient's clinical condition. During the operation, a PDA was found, and then PDA ligation was performed.

Conflicts of Interest

The author states that they do not have any personal or financial conflicts that could influence their work.

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