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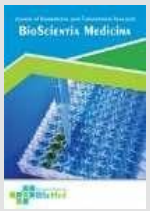

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Delayed Bochdalek Diaphragmatic Hernia Appearance and Coincidence with Pneumonia in One-Year-Old Baby: A Case Report

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A B S T R A C T

Background: Congenital diaphragmatic hernia (CDH) is a developmental defect in the diaphragm leading to protrusion of abdominal contents into the thoracic cavity. The incidence rates showed that this disease happened to one in 2,200-3,000 births. This study aimed to describe a case of delayed Boschdalek diaphragmatic hernia and coincidence with pneumonia in a one-year-old baby. **Case Presentation:** A 12-month-old girl came to the emergency room of Dr. M. Djamil General Hospital with a chief complaint of breathlessness for 2 days before admission to the hospital. She had difficulty gaining weight, only about 200 grams per month. The symptoms are breathlessness, cyanosis, asymmetry of lung sounds, change in the position of the heart sound, and scaphoid abdomen. The chest X-ray showed the opacity loops, which are fluid-filled, suggestive left diaphragmatic hernia. **Conclusion:** Pneumonia-like symptoms and signs could be an initial or coincidence with delayed presentation of Bochdalek hernia diaphragmatic.

1. Introduction

Congenital diaphragmatic hernia (CDH) is a condition resulting from a developmental defect in the diaphragm leading to protrusion of abdominal contents into the thoracic cavity.[1] CDH is a rare disease and is usually found in the neonatal period. The incidence rates showed that this disease happened to one in 2,200-3,000 birth.[2] Some studies showed that the prevalence of males is higher than females, but another study reported that there was no difference between males and females.[1,2]

The etiology of CDH is still unknown. There is more than one factor related to CDH, like genetic, environmental, and nutritional state.[1,3,4] CDH can be an isolated anomaly or associated with anomalies in other organ systems like heart defects, neural tube defects, intestinal anomalies, renal anomalies, and others. The signs and symptoms are different depending on the size of the defect. The symptoms are breathlessness, cyanosis, asymmetry of lung sounds, change in the position of the heart sound, and scaphoid abdomen.[1,4,5] CDH is associated with pulmonary disease in children, mainly pneumonia. This contributes to the increase in pulmonary morbidity and mortality rates. Pneumonia is also associated with the nutritional state. The infection is increasing in malnourished children and is associated with decreased immune function and limited

productivity in the future.[1]

This case was different from the common cases of CDH. The patient was a girl, and she was diagnosed with a congenital diaphragmatic hernia at 12 months of age without any symptoms since birth. This revealed delayed manifestation of CDH which could occur in small defects of the diaphragm. The delayed manifestation of CDH was like mild respiratory distress syndromes, such as pneumonia, recurrent infection symptoms, gastrointestinal disorder, feeding problem, and growth disorders.[1,4,5]

2. Case Presentation

A 12-month-old girl came to the emergency room of Dr. M. Djamil General Hospital with a chief complaint of breathlessness for 2 days before admission to the hospital. She had difficulty gaining weight, only about 200 grams per month. Her birth weight was 3 kg, and her present weight is 5.8 kg. The child is routinely checked by a pediatrician at a private hospital every month because of difficulty gaining weight. She had coughed with phlegm and sniffled 8 days before admission. Fever for 6 days before admission for four days, recurrent, peak temperature was up to 40°C. Breathlessness since 2 days before admission, not influenced by activities nor weather. She got therapy due to a high fever on the second day of fever in the emergency room of a private hospital, antibiotics, cough medicine, and paracetamol. She was referred from a private hospital with a diagnosis of a diaphragmatic hernia based on a chest X-ray. She had never experienced breathlessness before, and there were no family members who suffered from the same disease as her.

The patient is the second child of two siblings from an expected pregnancy. During pregnancy, the mother was healthy, regularly checked up on her pregnancy by healthcare providers, never consumed drugs and herbs, and never suffered from fever or bleeding. The mother had a cesarean section with an indication of a previous cesarean section, and she was assisted by a doctor. The patient was born full-term with a birth weight of 3000 grams and a birth length of 48 cm. She immediately started crying loudly after birth. There was no history of cyanosis, breathlessness, jaundice, nor seizures. Basic immunizations were incomplete. The patient only got HB0, BCG scar on the right hand, and DPT-1. The patient's growth and development are appropriate for her age.

On physical examination, the child looks moderately ill and conscious, with blood pressure 90/60 mmHg, pulse rate 140x/minute, respiratory rate 38x/minute, temperature 36.5ºC, body length 72cm, weight 5,8 kg. The nutritional status of this child was malnourished. Warm skin with no enlarged lymph nodes is palpable. The conjunctiva is not anemic, the sclera is not icteric, the pupil is isochoric with a diameter of 2mm/2mm, and the light reflex is normal. Head circumference is 45 cm (normocephalic). The movement of the left chest was falling behind, with no retraction, pectus carinatum, prominent ribs, no retraction, bronchovesicular, find rales on both lungs, and there was no wheezing. Examination of the heart was within normal limits. Supple abdomen, no distension, liver, and spleen are not palpable. Pubertal status A1M1P1. Warm extremities, capillary refill time <2 seconds, with no oedema.

Laboratory finding hemoglobin 11,2 g/dl (anemia), white blood count 19,320/mm^3, differential count 0/0/5/57/29/7, platelets 357,000/mm^3. Electrolyte concentration was sodium: 136 mmol/L, potassium: 4,1 mmol/L, chloride 104 mmol/L, calcium: 9,3 mg/dl, and albumin level were 4,1 gr/dl. The blood gas analysis examination revealed hypoxemia. Urine and feces within normal limits. The chest X-ray showed the opacity loops, which are fluid-filled, suggestive left diaphragmatic hernia.

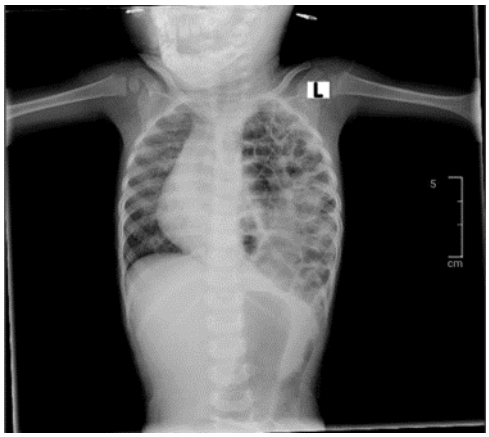

Figure 1. Left hernia diaphragmatic.

Based on those findings, the patient was diagnosed with a left diaphragmatic hernia, bronchopneumonia, severely malnourished, incomplete immunization, and suspected COVID-19. The patient was given breastfed/formula milk 4x50 cc, 4x75 cc, Ceftriaxone 2 x 250 mg, and O_2 via nasal cannula 2 liter/minute. The patient also has undergone a blood culture and plans to repair the hernia through elective surgery.

During 2nd week in warded, the patient performed surgery to repair the hernia (Figure 2). During postoperative treatment, the child was placed in the PICU room and extubated. The child underwent post-*op* chest X-ray imaging revealed left pneumothorax, and post-op laboratory test revealed anemia. After 1-week post-surgery, the patient significantly improved and was discharged in good condition.

Figure 2. Hernia defect on surgery.

3. Discussion

A case of a 12-month-old girl was presented with a diagnosis of congenital diaphragmatic hernia (CDH). CDH is a congenital abnormality of the diaphragm, in which the thoracal cavity is filled by abdominal organs through a defect in the diaphragm.[1] This case was CDH of a girl diagnosed at 12 months old.

Males with CDH cases are higher rather than females, but other studies report no association between CDH and gender.[1,2]

CDH has two main pathological findings; lung hypoplasia and abnormal pulmonary vascular development.[1,5] Lung hypoplasia has decreased in the era of air routes, terminal bronchioles, and alveoli.

Unusual vascular remodeling might happen and cause the thickening of blood vessel walls. The thick wall of the pulmonary vessel will cause pulmonary hypertension (PH). Worsening of PH lead to lung hypoplasia and left ventricular dysfunction. Lung hypoplasia, abnormality of lung vasoreactivity, and PH are the major contributor to morbidity and mortality of CDH.[1,5]

Signs and symptoms of CDH can be found in the neonatal period, depending on the size of the diaphragm's defect. The symptoms are breathlessness, cyanosis, asymmetry of lung sounds, changing of the heart's apex position, and scaphoid abdominal. However, delayed presentation of CDH's symptoms occurs in small defects, usually preceded by mild respiratory disturbances at the beginning. This patient had symptoms at the age of 1 year old, without any symptoms since birth. Patients came with pneumonia symptoms, and the hernia diaphragmatic was revealed through chest X-ray due to pneumonia indication. Leeuwen has reported that the cause for developmental delay in diaphragmatic hernia patients is multifactorial, counting expanded catabolic stretch within the neonatal period, GER, and determined pneumonic impedance. Other literature states that diaphragmatic hernia is ordinarily present within the quick neonatal period with respiratory problems. In any case, postponed introduction as a coincidental finding has been detailed in an 11-month-old boy who was referred from a nearby healing center with the intense onset of the hack.

Clinical signs of late displaying diaphragmatic hernia are so different that gastrointestinal indications (such as heaving and stomach torment) and respiratory side effects (such as dyspnea, hack, and cyanosis) can show alone or in combination.[5] Based on the location of the defect, CDH can be classified into different types, Morgagni hernias, central hernias, and Bochdalek hernias. Morgagni hernias has defect in the anteromedial. The incidence is 20% to 25% of CDH. The incidence of central hernias is about 2% to 5%. Bochdalek hernias result from a deformity within the postero-lateral portion of the stomach (Figure 3).

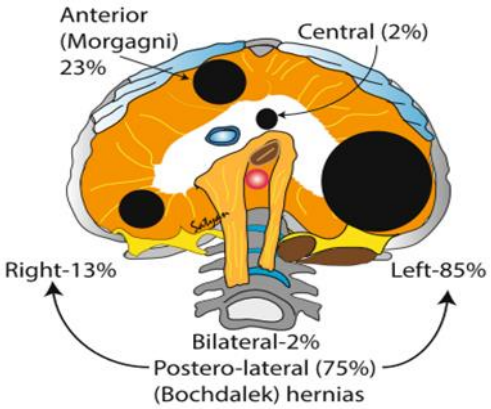

Figure 3. Classification of diaphragmatic hernia based on location.[6]

The congenital diaphragmatic hernia was associated with morbidity and mortality due to pneumonia. It is thought to be due to its inherent imperfections and mechanical ventilation, which are essential to support life.[7] Respiratory complications included constant lung disease, household oxygen prerequisites, aspiration pneumonia, hypertension, and obstructive airline route disease.[1] Lejeune showed that diaphragmatic hernia in children is related to pneumonia, workout restriction, and chest deformation. Pulmonary disorders in children with CDH consist of obstructive conditions (46%) and an irregular cardiopulmonary workout test (66%).[2]

The etiology of pneumonia in the pediatric population can be classified according to age and specific pathogens. Common organisms which are related to spontaneous delivery are the β *streptococcus* group, *Klebsiella*, *Escherichia coli,* and *Listeria monocytogenes.* The common cause of pneumonia in newborns and children between 30 days and 2 years old are viruses. In children 2 to 5 years old, the rise of cases associated with *S. pneumoniae* and *H. influenzae* type B. However, *S. pneumoniae* is still the most common cause.[8,9]

In the era of the pandemic, COVID-19 must also be considered as the etiology of pneumonia in children. Most children appear to have contracted the disease from adults positive for COVID-19, especially from family contact.[10] In this case, the results of the COVID-19 antigen swab examination were negative, and the SARS-CoV-2 PCR swab was negative. It is very difficult to distinguish the causative organism in most cases of pneumonia. Blood cultures are used to isolate the causative organism. However, submission of blood type is low for bacterial pathogens (approximately 25% to 33%) and is unreliable.[11] The culture results in this patient were no growth. However, the patient still got the antibiotics ceftriaxone.

Incomplete immunization and malnourished condition could worsen the outcome. Development retardation was found in 23% of the diaphragmatic hernia survivors. Other components may be related to poor development during the early stages, including the fact that patients with diaphragmatic hernias are sometimes narrow-minded toward enteral nutrition because of cardiorespiratory weakness, deep sedation, or limitations in volume retention. Gastroesophageal reflux, verbal abhorrence, and hypersensitivities can contribute to challenges in enteral nourishing. Moreover, an increment within the vitality prerequisite may be a belittled calculation. Among 59% of patients with diaphragmatic hernia were hypermetabolic with a tall resting vitality consumption, and 30% more vitality was required for perfect body weight pick up. These discoveries demonstrated that expanded vitality prerequisite in newborn children with the diaphragmatic hernia was a significant chance calculate of development hindrance.[12]

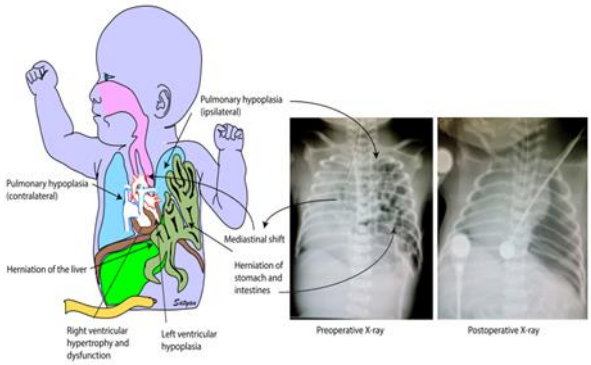

Figure 4. Anatomy and radiologic features of diaphragmatic hernia.[6]

The patient underwent the hernia repair in the second week of hospitalization. Surgical repair is the first-line treatment following the diagnosis of a CDH.[13] Repair of diaphragmatic hernia may be fulfilled through a thoracic or stomach approach and may be performed in an open or negligibly intrusive way. Long-term results depend, perhaps most critically, on the characteristics of diaphragm deformity. Patients with small solid deformities who are approached effectively should have minimal recurrence and complications. There has been an expanding trend toward thoracoscopic repairs, which are thought to play down postoperative torment and scarring and rush recuperation.[6,14]

This patient underwent well recovery and had a good outcome. The outcomes of infants with CDH have improved in recent years. CDH administration has led to advances in ventilation procedures, PH administration, and refinement of surgical strategies. There moved forward administration of CDH over the past few long time. The survival rate CDH is 60% to 70%.[1] The prognosis of isolated diaphragmatic hernia is, for the most part, superior to diaphragmatic hernia complicated by multiple anomalies.[6]

4. Conclusion

Pneumonia-like symptoms and signs could be an initial coincidence with the delayed presentation of Bochdalek hernia diaphragmatic. The appropriate management, such as elective repair surgery, will improve the outcome and the survival of this patient.

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