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Case Report
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Diagnostic and Management Challenges of Congenital Diaphragmatic Hernia in Preterm Newborn: A Case Report in Remote Area, Asmat, South Papua

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Abstract

Background: Congenital diaphragmatic hernia (CDH) is a rare condition associated with pulmonary complications, as the herniation of abdominal viscera into the chest can impact lung development. The majority of CDH newborns have respiratory issues at delivery or shortly after and require mechanical ventilation, which puts them at risk when they are transferred to a tertiary hospital for more extensive treatment.

Objective: This article presents a case of a preterm infant with low birth weight with CDH managed in the General Hospital, Asmat Regency.

Case Presentation: A male, born preterm at the gestational age of 31 weeks by spontaneous vaginal delivery, with a birth weight of 1505 grams. The patient had respiratory distress and required invasive ventilation shortly after delivery. When the patient showed improved oxygenation with a nasal cannula, we obtained a chest X-ray, which revealed bowel loops in the left hemithorax and a rightward mediastinal shift. CDH treatment must be administered in tertiary hospitals to ensure adherence to standard protocols and multidisciplinary care. Transferring the patient to a tertiary hospital requires continuous ventilation during the full-day transport, but the patient was not transportable; thus, we decided to operate. Significant progress was observed three months following the operation.

Conclusion: CDH requires a CT scan for definitive diagnosis, surgical intervention, and intensive treatment in a tertiary hospital. Despite the limited resources and no referral options, the patient showed clinical improvements.

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INTRODUCTION

Congenital diaphragmatic hernia (CDH) is a rare condition associated with pulmonary complications, as the herniation of abdominal viscera into the chest can impact lung development.[1] The pathophysiologic mechanism and genetic etiology of CDH are yet unknown. However, it is suspected that early pulmonary embryological development abnormalities exist[2]. There are several classifications of CDH, with Bochdalek hernias accounting for around 95% of cases.[2] There are 1 to 4 CDH cases per 10,000 live births[3], with the survival rate for CDH being about 67%.[4]

Symptoms of CDH typically appear during the first few hours of life, with the onset of respiratory distress[5], and chest radiography tends to be the initial imaging test performed.[6] An air-fluid level in the thorax on radiography with worsening breathing misdiagnoses 38% of these cases as tension pneumothorax, hydropneumothorax, hemothorax, empyema, effusion, or pneumonia [7], and that's why a CT scan was required for a definitive diagnosis[4] because it is the most accurate study to see the internal parts and openings of a hernia directly.[8]

Most newborns with CDH have respiratory problems due to pulmonary hyperplasia, hypertension, or both[9], and should be intubated at birth, avoiding bag and valve ventilation.[6] At times, mechanical ventilation is required to treat this condition.[5] This made the patient immobile and ventilator dependent, and in the present case, referring to tertiary care was a dilemma because referral transport requires days on the ship and continued flights to reach the nearest tertiary facility. We present a case of a preterm infant with low birth weight who, despite diagnostic and intervention difficulties, was successfully managed in a remote area in South Papua, Asmat Regency.

CASE PRESENTATION

Patient Information

A male, born preterm at the gestational age of 31 weeks by spontaneous vaginal delivery, with a birth weight of 1505 grams. The patient's mother, in her mid-twenties, came to the hospital when it was time for labor. The patient's mother did not receive any antenatal care and had no history of ultrasonographic examination of the current pregnancy or any associated risk factors.

Clinical Findings

The patient had persistent respiratory distress, thus requiring invasive ventilation shortly after birth resuscitation. We'd like to do an X-ray, but since we don't have a mobile X-ray, the patient who is receiving continuous invasive ventilation while being transported might be at potentially life-threatening risk. We decided to wait until the patient was stable and transportable. On the treatment, we found no gastrointestinal clinical symptoms. The patient received parenteral and enteral nutrition, intravenous antibiotics, and doses of corticosteroids. After several days of treatment, the patient's oxygenation gradually improved, and they were switched to CPAP (Continuous Positive Airway Pressure) and later to a nasal cannula at 25 days of age. We decided the patient was transportable for an X-ray examination. However, after 24 hours with the nasal cannula, the patient immediately showed respiratory distress

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again and was put back on CPAP and then re-intubated afterward.

Diagnostic Assessment

Anteroposterior and lateral chest X-rays showed bowel loops in the left hemithorax and a mediastinal shift to the right hemithorax, which then identified left-sided CDH in Figure 1.

Therapeutic Intervention

At first, we suspected a hyaline membrane disease that caused persistent respiratory distress; thus, the patient received antibiotics and a dose of dexamethasone for bronchopulmonary dysplasia. After a chest X-ray was performed, CDH was identified. Transferring the patient to a tertiary hospital would require continuous ventilation during the full-day transport, but the patient was not transportable, so we decided to perform the operation.

The operation was performed when the patient was 31 days old, with a body weight of 1800 g. We opened the abdomen through the left subcostal incision. After the peritoneum had opened, we found various intra-abdominal organs, consisting of multiple bowel loops, stomach, spleen, and left liver lobe (Figure 2), had been displaced through a diaphragmatic defect into the left thoracic cavity.

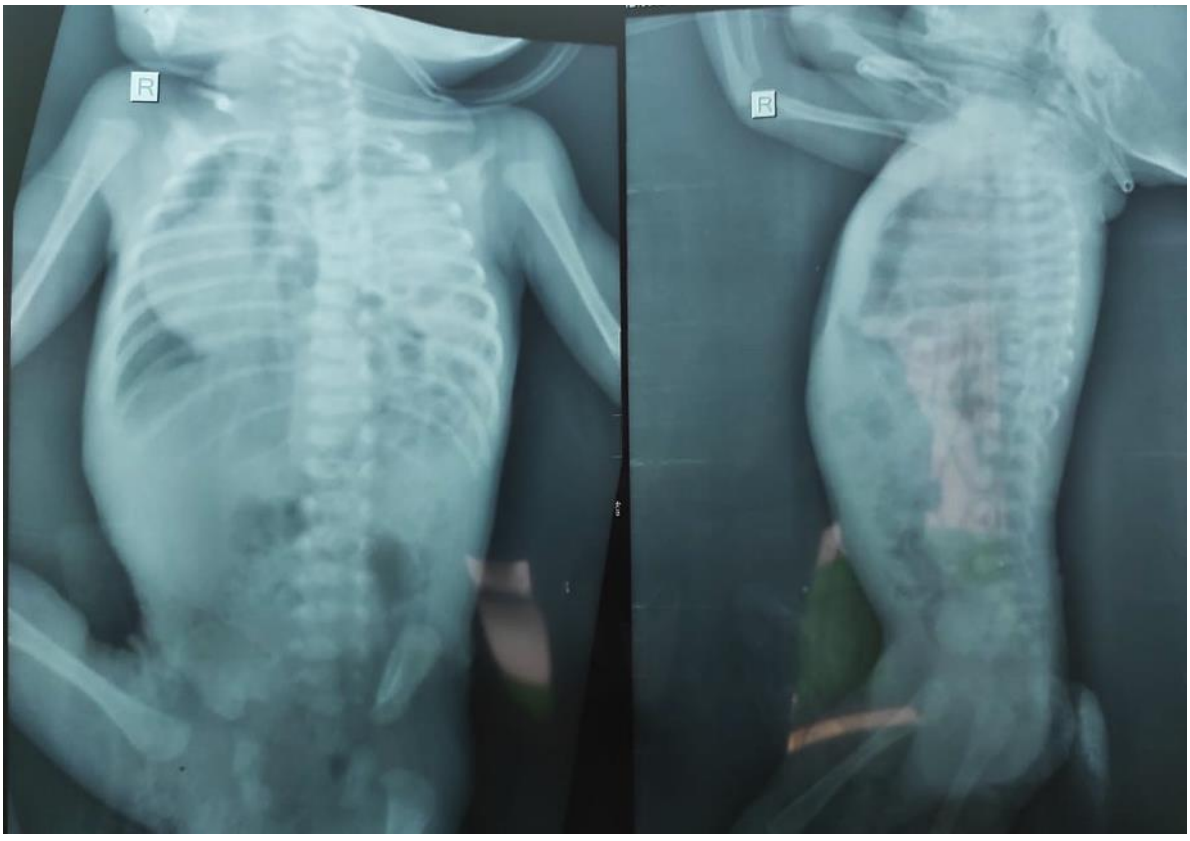

Figure 1. Preoperative anteroposterior (AP) and lateral (L) Chest X-ray.

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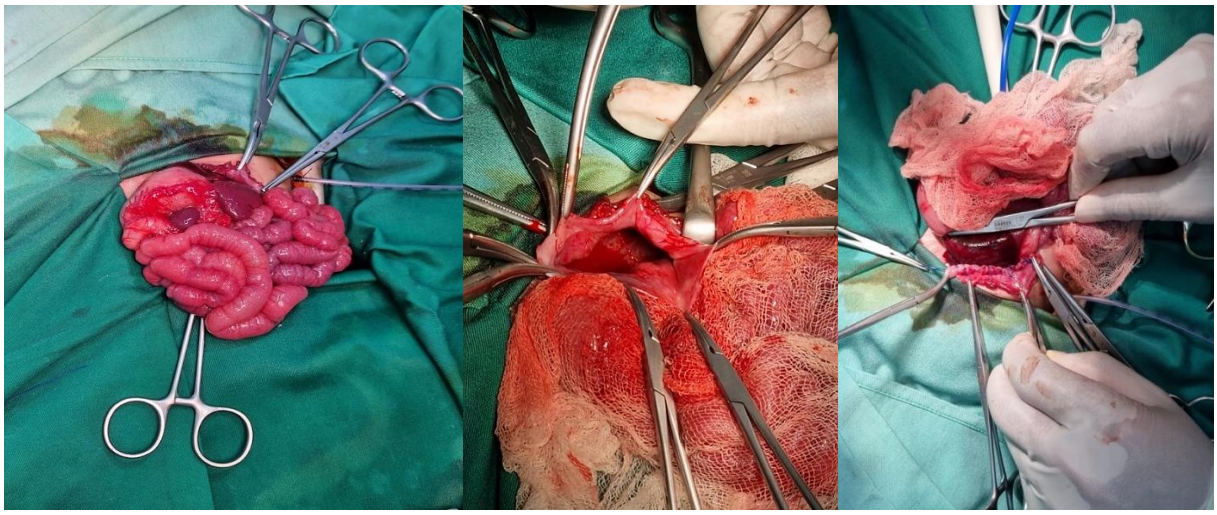

Figure 2. Various intra-abdominal organs, including multiple bowel loops, the stomach, the spleen, and the left liver lobe, have been displaced through a diaphragmatic defect into the left thoracic cavity (left).
Figure 3. A thin transparent membrane defect, sized 6 cm and 3 cm, was found on the left posterolateral side of the diaphragm (center).
Figure 4. The defect was closed with a primary simple interrupted non-absorbable suture. Some portion of the anterior rim of the diaphragm was encircled by the ribs (right).

The intra-abdominal organs were returned to the peritoneal cavity, and we identified a thin, transparent membrane defect on the left posterolateral aspect of the diaphragm, measuring 6 cm by 3 cm in Figure 3. Excision was done at the membrane defect. We evaluated the edge approximation, which did not cause tension or flatten the diaphragm; thus, we decided to close the defect with a primary simple interrupted non-absorbable suture using 3/0 monofilament in Figure 4. Some portions of the anterior rim of the diaphragm were narrow; therefore, we encircled the suture to the ribs, leaving one chest tube drain.

Follow-up & Outcome

The postoperative chest X-ray showed fully inflated lungs bilaterally in Figure 5. The patient experienced paralytic ileus; therefore, the patient continued to receive total parenteral nutrition and nil per os for four days. We initiated oral feeding on the 5th postoperative day, and the patient achieved full oral intake on the 12th day. We extubated the patient on the postoperative 4th day, removed the chest tube on the postoperative 10th day, and discontinued oxygen therapy on the postoperative 15th day, at which point the patient was breathing room air without respiratory distress.

The patient was discharged on the 19th postoperative day, at 50 days of age, with a weight of 2005 g. We had a follow-up at 3 months of age, and no gastrointestinal or respiratory clinical symptoms or neurodevelopmental delays were found, with a body weight of 4300 g in Figure 6.

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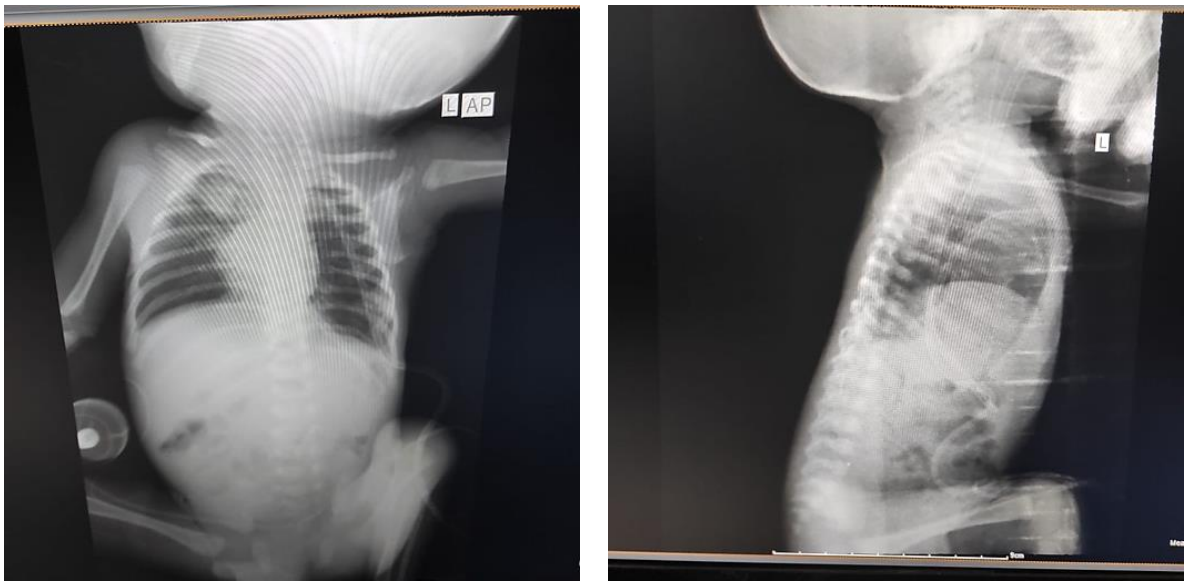

Figure 5. Postoperative anteroposterior (AP) and lateral (L) chest X-ray

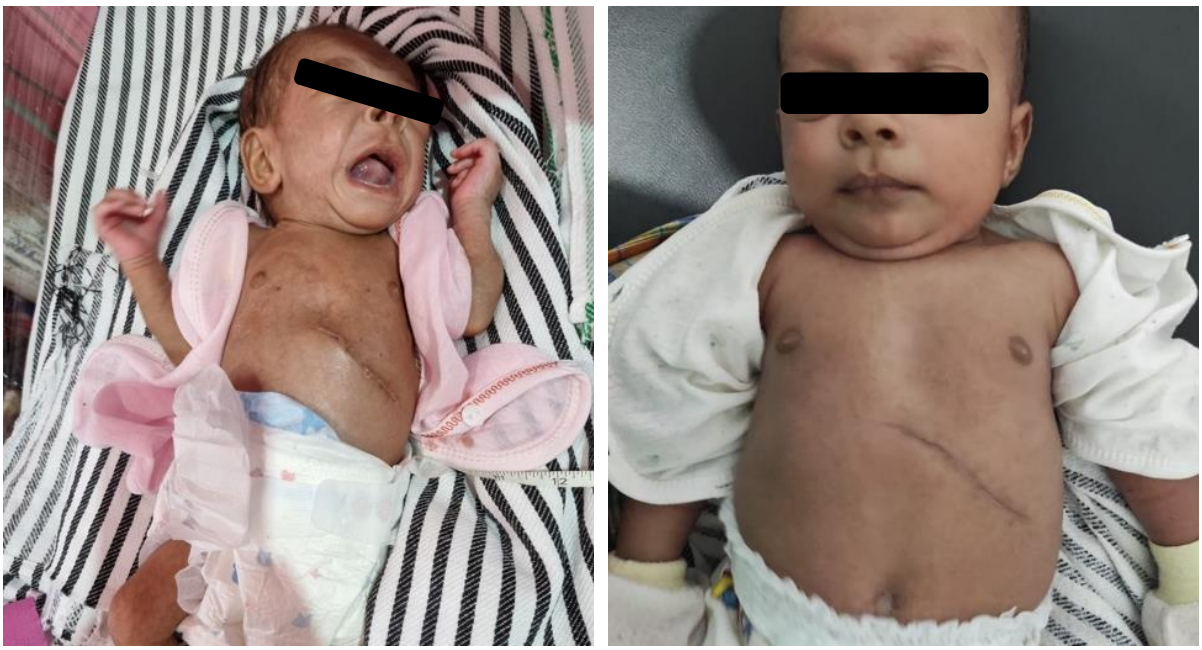

Figure 6. The patient was discharged on postoperative day 19, at 50 days of age (left), and had a follow-up at 3 months of age (right).

DISCUSSION

Ultrasound findings are crucial in prenatal evaluation, counseling about the prognosis, and management of CDH.[10,11] Prenatal ultrasound can detect several risk factors, including liver herniation, stomach herniation, low fetal lung volume, and a low lung area-to-head circumference ratio (LHR), as well as chromosomal abnormalities, concurrent anomalies, and hydrops fetalis.[12,1] In the present case, the patient's mother did not actively pursue any kind of antenatal care or ultrasound testing, which resulted in the delayed detection of CDH.

Usually, neonates affected by CDH experience respiratory distress either immediately after birth or shortly thereafter. This can be at-

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tributed to the presence of pulmonary hypoplasia, pulmonary hypertension, or a combination of both factors.[4,6,9] In this case, it was seen that the patient was having respiratory distress, which was entirely dependent on mechanical ventilation.

For patients who do not have a prior history of trauma and present with respiratory symptoms, it is essential to conduct an initial diagnostic examination in the form of a chest X-ray, including both anteroposterior and lateral views.[10] Even so, we experienced challenges during the diagnostic investigation, as the patient was unable to undergo a chest X-ray due to the unavailability of a mobile X-ray machine in our facility. Additionally, the patient's physical limitations made transportation impossible. Therefore, we decided to do a chest X-ray after being hemodynamically stable at 25 days of age. We identified bowel loops in the left hemithorax, as seen in CDH[1], but we can't confirm it further because we don't have a CT scan as a definitive diagnostic study.[4,8,13]

Management of CDH that follows a standard, multidisciplinary approach, including lung-protective strategies, in tertiary care can achieve a survival rate of more than 85%.[14] However, CDH mortality and morbidity are usually associated with mechanical compression of the herniated viscera on the developing lung, resulting in pulmonary hyperplasia and pulmonary hypertension.[15,16] In this case, we don't have echocardiography as an essential examination to evaluate cardiac anatomy and screen for pulmonary hypertension14, and ECMO (Extracorporeal Life Support) is unavailable if the patient's condition worsens on the ventilator due to severe pulmonary hypertension. [17,16,18]

We still performed the operation because the patient's condition was not transportable. During surgery, we found a small defect in the diaphragm, which the approximation did not make tense or flatten. We decided to do a primary suture, like the majority of cases[15] in which primary suture has been the standard of care for CDH[19] and use mesh for the larger defect[20]. The operative procedure was successful, with improvement in hemodynamics.

We had 3 months of follow-up at the polyclinic and found no respiratory or gastrointestinal issues or delays in neurodevelopmental outcomes. However, long-term follow-up into adolescence is recommended for all patients with CDH because multisystem adverse outcomes are common.[14,15,1]

CONCLUSION

We encountered numerous difficulties in prenatal and postnatal diagnostics and therapeutic interventions. Despite these obstacles, with limited resources and inadequate referral options, we succeeded in managing CDH Bochdalek, and the patient showed clinical improvements. But still, close monitoring and follow-up are required until the patient reaches adolescence. Considering this situation, we hope to share our experience in managing Bochdalek CDHs with surgeons in remote locations.

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