A Rare Cause of Haemorrhage in the Upper Gastrointestinal System

Bochdalek Hernia

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ABSTRACT

Diaphragmatic hernia originates from insufficient closure of the pericardioperitoneal canals and pleuroperitoneal membranes. It is seen in one in every 4000 births. The general finding in the newborn period is respiratory difficulty. Mortality is 40–50%. There may be other accompanying organ anomalies. Congenital diaphragmatic hernias diagnosed after the newborn period are known as late-presenting congenital diaphragmatic hernias. This group is seen at a level of 5–20% and poses difficulty in diagnosis. This report describes a case under observation and receiving treatment for gastrointestinal haemorrhage, diagnosed as Bochdalek hernia.

Keywords: Bochdalek hernia, congenital diaphragmatic hernia, gastrointestinal haemorrhage

Una Causa Rara de la Hemorragia en el Sistema Gastrointestinal Superior

La Hernia de Bochdalek

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RESUMEN

La hernia diafragmática se produce a causa del cierre insuficiente de los canales pericardioperitoneales y las membranas pleuroperitoneales. Se la encuentra en uno de cada 4000 nacimientos. El hallazgo general en el período neonatal es la dificultad respiratoria. La mortalidad es del 40-50%. Pueden presentarse otras anomalías acompañantes en los órganos. Las hernias diafragmáticas congénitas diagnosticadas después del período de recién nacido, se conocen como hernias diafragmáticas congénitas de presentación tardía. Este grupo se observa en un nivel de 5 – 20% y plantea dificultades de diagnóstico. El presente reporte describe un caso bajo observación y tratamiento por hemorragia gastrointestinal, diagnosticada como hernia de Bochdalek.

Palabras claves: Hernia de Bochdalek, hernia diafragmática congénita, haemorragia gastrointestinal

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INTRODUCTION

Bochdalek hernia was first described by Victor Alexander Bochdalek in 1867. Congenital diaphragmatic hernia is seen in one in every 4000 live births. Bochdalek hernia is an emergency condition generally seen in the neonatal period. It is not hard to diagnose in the neonatal period and early infancy, where findings are cyanosis and respiratory difficulty (1, 2).

Late-onset diaphragmatic hernia was first described by Kirkland in 1959 (3). Diagnosis is made outside the neonatal period in approximately 5–20% of cases (4). The reason for delayed treatment of Bochdalek hernia given late findings is difficulty of diagnosis. Clinical findings in Bochdalek hernia producing late findings are generally associated with the respiratory and gastrointestinal systems (4–6).

This report is intended to emphasize that, albeit rarely, Bochdalek hernia may be a cause of upper gastrointestinal haemorrhage.

CASE REPORT

A 6.5-year old girl with spastic tetraparesis but no known previous gastrointestinal complaints presented to the hospital emergency department with haematemesis during the preceding day after being monitored for vomiting for three days at hospital. She was moderately dehydrated and was conscious. Haemorrhagic gastric contents emerged from nasogastric tube. Weight was 17 kg (3–10 percentile), height 105 cm (< 3 per-

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centile), pulse 138/minute, respiratory rate 34/minute, oxygen saturation (SPO_2) at room temperature 97% and arterial blood pressure 90/50 mmHg. There were absent bronchial breath sounds in the right lung at system examination, and dislocation of cardiac sounds. On abdominal examination, the stomach was collapsed, no intestinal sounds were detected, and the entire abdomen was tender to palpation. The liver was 5 cm in the midclavicular line and painful. The spleen could not be palpated. Laboratory findings were white cell count 21 000/mm^3 (86% PNL), haemoglobin (Hb) 12.6 g/dL and thrombocyte 233 000/mm^3. Other biochemistry and haemorrhage profiles were normal.

At imaging, the right lung was collapsed. The intestine in the right haemothorax was filled with loops and there was mediastinal displacement. The oesophagus and stomach were significantly dilated and the stomach extended to the inguinal region. The liver appeared anteriorly displaced and the intrahepatic bile ducts were dilated (Fig. A, B). The patient was taken for emergency surgery with a diagnosis of diaphragmatic hernia. A 9 × 7 × 5 cm defect was determined in the posterolateral aspect of the right diaphragm. All small intestinal and colonic segments had passed to the thorax through this defect, and gastric volvulus and incarceration were present. The position of the intestinal segments and stomach were restored, and surgery was concluded by repairing the diaphragm following establishment of gastric perfusion.

The presence of respiratory difficulty at physical examination, the absence of respiratory sounds in the herniation area, intestinal sounds in pulmonary areas and dislocation of cardiac sounds, together with small and large intestinal herniation at anteroposterior chest X-ray and thoracic CT imaging, suggested congenital diaphragmatic hernia. The location of the hernia in surgery, other sections of the diaphragm being normal and the absence of trauma that might suggest diaphragm perforation in her history, together with clinical and radiological findings all supported a diagnosis of Bochdalek hernia. The patient was monitored in hospital for one week postoperatively and discharged after the gastrointestinal passage returned to normal (Fig. C).

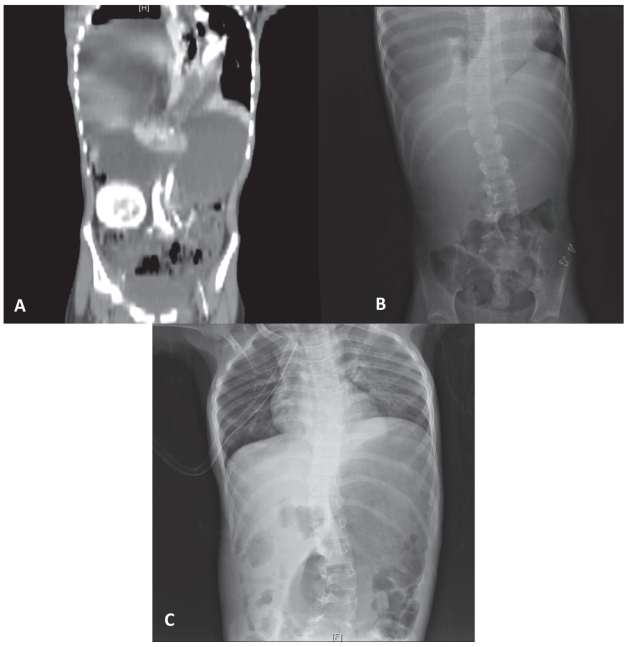

Figure: A) Preoperative longitudinal thoraco-abdominal computed tomography scan; B) Preoperative anteroposterior chest X-ray. The esophagus and stomach are significantly dilated and the stomach extends to the inguinal region. The liver appears anteriorly displaced and the intrahepatic bile ducts are dilated; C) Postoperative anteroposterior chest X-ray: gastrointestinal passage is normal.

DISCUSSION

Congenital defects of the diaphragm develop in the event of impairment in any of the embryological development stages. Bochdalek hernia is the most common form of diaphragmatic hernia. It occurs as a result of failure of the pleuroperitoneal channel to close completely at weeks 8–10 of pregnancy. It is generally (80–90% of cases) left posterolaterally localized and is twice as common in males (1, 7, 8). Late-onset Bochdalek hernia is generally seen in right-sided defects (5). The defect in our case was on the right and there was a large cavity in the diaphragm. The liver being on the right permits diaphragmatic defect closure, albeit partly. Hernias on the left are rare and generally appear in middle or advanced age. Symptoms and prognosis in late-onset cases generally depend on defect location and size and the organs and systems involved. Recurrent pulmonary infections, dyspnoea, wheezing, chest pain, recurrent abdominal pain, dysphagia, lack of appetite, nausea, vomiting and diarrhoea have been reported in the literature. Gastrointestinal haemorrhage and anaemia may develop as a result of diaphragmatic hernia-related gastric volvulus (4, 5, 8, 9). Gastric volvulus-related upper gastrointestinal haemorrhage was present in our case. Misdiagnosis as a result of failure to consider the diagnosis in patients with congenital diaphragmatic hernia means they may be exposed to incorrect procedures. Late detected hernias may occur as a result of increased intra-abdominal pressure, such as trauma, physical strain and obesity. The first-choice imaging technique for assessing diaphragmatic integrity is pulmonary radiography. Bochdalek hernia can be easily diagnosed with direct pulmonary radiography. Emergency CT can be performed when accompanying pathologies are suspected and when the intestinal loops are strangulated. Direct abdominal radiography can be important in the diagnosis of patients with vomiting of uncertain cause (4, 5, 8, 10, 11). Indeed, we learned from her history that she had been monitored for three days in another centre because of vomiting. There is a high risk of complications in large hernias with abdominal organs. Surgical treatment should be considered in these cases (5, 11, 12). Emergency surgery was performed in our case. No complication was seen during or after surgery.

In conclusion, although the great majority of Bochdalek hernias are seen in infants, they can also be seen in other age groups. Suspicion is needed in order to make the diagnosis.

When accurately diagnosed, the condition can be easily corrected with surgery, to the great benefit of the patient.

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