

A CHILD WITH LATE-PRESENTING CONGENITAL DIAPHRAGMATIC HERNIA ONCE INCORRECTLY SUSPECTED OF PNEUMOTHORAX

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Abstract

A 2-year-old girl suddenly vomited and complained of abdominal pain. A chest and abdominal X-ray showed air and air-fluid level in her left thoracic cavity, leading to incorrect diagnosis of left tension pneumothorax. She was transported to our hospital. Pneumothorax could not be ruled out by X-ray, but respiratory symptoms were poor. Computed tomography (CT) and upper gastrointestinal series indicated her as congenital diaphragmatic hernia (CDH) and gastric dilatation due to gastric volvulus. During her emergency surgery, the intraoperative findings showed the prolapse of the stomach, transverse colon and spleen into the left thoracic cavity through the defect of the left-posteolateral diaphragm.

The late-presenting CDH is considered that many cases are mild cases, and prognosis is considered to be good. But the symptoms are various, therefore it is difficult to perform accurate-initial treatment and preoperative diagnosis of this disease.

Key words : late-presenting congenital diaphragmatic hernia, gastric volvulus, tension pneumothorax

Introduction

Late-presenting congenital diaphragmatic hernia (CDH), accounts for 10% of all CDH and is defined as a diaphragmatic hernia that is detected after 30 days of birth^{1,2)}. Although its prognosis is generally favorable, it does not always lead to the manifestation of respiratory symptoms and this might delay its diagnosis. We treated a case of late-presenting CDH that was initially diag-

nosed as a pneumothorax, resulting in difficulty with the initial treatment.

Case Report

A 2 years and 10 months old girl suddenly vomited food residues and gastric fluid-like vomitus and had abdominal pain in the morning. She previously had wheezing when she had an upper respiratory infection and was diagnosed with asthmatic bronchitis but had not undergone chest radiography. She was examined by her local physician that morning and was diagnosed with gastroenteritis. After undergoing colonic irrigation, she was sent home. However, the abdominal pain and nausea persisted ; therefore, she was taken to another clinic in the afternoon of the same day. A chest radiograph

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showed abnormal shadows in the left thoracic cavity. A diagnosis of left tension pneumothorax with pleural effusion was made; therefore, she was transferred with an ambulance to the pediatric department of our hospital and referred to our department to undergo pleural drainage. The patient was 88 cm tall and weighed 13.8 kg. Despite being diagnosed with a pneumothorax, she had no respiratory discomfort, her SpO₂ was 99% in room air, and her Jugular veins were not distended. Although the patient complained of upper left abdominal pain, an abdominal examination revealed that the abdomen was flat and soft with no clear areas of tenderness. She was nauseous but did not vomit. A chest radiograph showed a large amount of air in the left thoracic cavity with the presence of air-fluid levels in the standing position, and deviation of the mediastinum to the right (Fig. 1). However, the abnormal shadow also appeared like a gastric bubble or an intestinal structure. As the main symptoms were in the abdomen, a tension pneumothorax appeared unlikely. Moreover, the patient's general status was stable; therefore, a thoracoabdominal contrast-enhanced computed tomography (CT) was performed. The CT showed that the stomach and spleen protruded into the thoracic cavity (Fig. 2). Since there was no his-

tory of trauma, the patient was diagnosed with a late-presenting CDH. Gastric volvulus and acute gastric distension were also seen. To avoid gastric perforation, a gastric tube was fluoroscopically inserted to reduce the pressure. The stomach had a reversed- α shape and was rotated along the mesenteric axis (short axis) (Fig. 3). After making a diagnosis of a late-presenting CDH with acute gastric volvulus, emergency surgery was performed on the same day. A laparotomy was performed with an upper left transverse abdominal incision, and a defect measuring 4 × 3 cm was found in the posterolateral part of the left diaphragm (Fig. 4). It was further identified to be a non-saccular Bochdalek hernia. The stomach, spleen, and a segment of the colon between the left transverse colon to the splenic flexure had all protruded into the thoracic cavity. Although the stomach was rotated along the mesenteric axis, blood flow was not obstructed and the colon had a normal color. The protruded organs were gently reduced into the abdominal cavity, and the hernial orifice was closed. To prevent recurrence of a gastric volvulus, the fundus of the stomach was fixed to the inferior surface of the diaphragm. The postoperative course was good, the thoracic drain was removed, and oral intake was commenced on postoperative

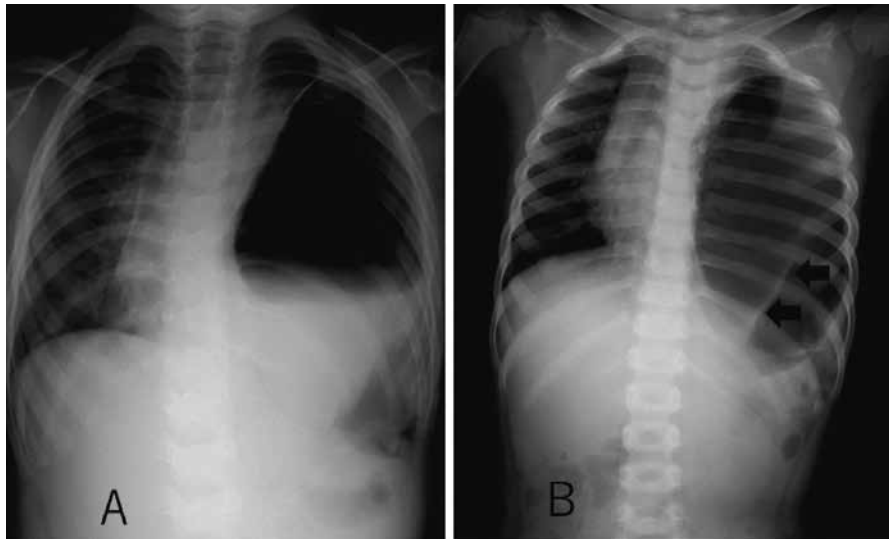


Fig. 1. Thoracoabdominal X-ray on admission A: Standing position, air and air-fluid level formation observed in the left thoracic cavity. B: Prone position: A septum-like structure is observed (arrow), which was believed to be the gut wall.



Fig. 2. Thoracoabdominal CT A : A distended stomach and spleen is observed protruded into the left thoracic cavity. B : A gastric bubble is observed on the abdominal side of A.

day 6. Though gastric distension after meals persisted, no digestive symptoms were observed, and the patient was discharged on postoperative day 10.

Discussion

Late-presenting CDH can be classified into two types³⁾. In the first type, abdominal organs that are nor-



Fig. 3. A transnasal gastric tube was inserted. The stomach presented a reverse-contrast image.

mally in the abdominal cavity suddenly protrude into the thoracic cavity due to rupture of the hernial sac or other reasons. In the second type, strangulation or perforation of previously herniated abdominal organs occur. While we cannot determine whether our patient suffered from the first or second type of CDH as the patient had no previous medical history nor plain chest radiograph images, the lack of adhesion between the hernial orifice and the herniated organs suggests that it was the former. However, it could have been the latter type if the condition that was previously diagnosed as asthmatic bronchitis was a respiratory symptom due to compression of the lungs by the eviscerated organs. Generally, late-presenting CDH is often mild, and the prognosis is usually favorable. This is because late-presenting CDH often involves fewer coexisting malformations compared to prenatally or neonatally diagnosed CDH, and pulmonary hypoplasia is usually mild or absent in such cases. CDH within 24 h of birth most often manifest as respiratory symptoms, while the first symptoms of late-presenting CDH can also consist of digestive symptoms, such as abdominal pain and nausea, symptoms may be varied³⁾. This can make diagnosis difficult and then result in delayed or inappropriate treatments. Gastric volvulus is a

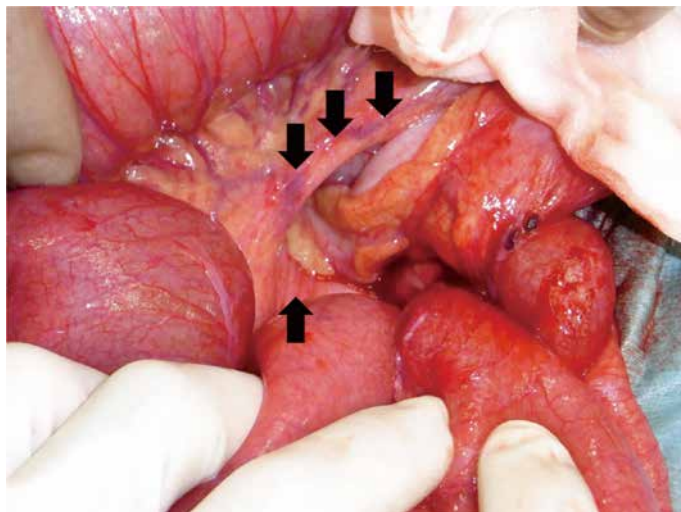


Fig. 4. Intraoperative photograph : A hernial orifice (arrow) was observed on the posterolateral side of the left diaphragm.

general term that refers to rotation of the entire or part of the stomach beyond its physiological range due to abnormal laxity of the gastric ligaments and can induce gastric obstruction or vascular insufficiency. It is classified according to the rotational axis into the organo-axial, mesentero-axial or combined volvulus types⁴⁾, and also into primary or secondary etiological subtypes. The former has no organic cause, and the latter involves abnormalities of nearby organs, such as CDH, diaphragm eventration, hiatal hernia, and wandering spleen. Cribbs⁴⁾, reported that 70% of cases of gastric volvulus are actually secondary and occur in relation to abnormalities of the diaphragm, such as CDH, diaphragm eventration, and traumatic diaphragmatic hernia. Results of imaging tests often lead to misdiagnosis of a late-presenting CDH with acute gastric volvulus as a pneumothorax, pulmonary cyst, pneumonia, pleural effusion, or a diaphragmatic tumor. According to a review by Berman¹⁾, out of 26 pediatric cases of late-presenting CDH with acute gastric volvulus, 16 cases were diagnosed with pulmonary cyst or pneumothorax, and in all 4 cases that were misdiagnosed as a pneumothorax, a chest drain was placed inside the stomach. A rapid increase in intra-abdominal pressure due to acute gastric volvulus causes gastric strangulation, ischemia, perforation, pancreatitis, peritonitis, and shock, with a mortality rate of 80% ac-

ording to Kim *et al*⁵⁾. In another case, gastric fluid that leaked due to an intrathoracic gastric perforation led to a pneumothorax because of the corrosive effects of gastric fluid⁶⁾. These cases highlight the importance of early and accurate treatment for late-presenting CDH. Our patient might have suffered from gastric perforation if gastric tube insertion had been delayed, and an iatrogenic gastric perforation may have occurred during thoracocentesis if we had not doubted the initial diagnosis of tension pneumothorax that the patient was referred with. This could have resulted in a fatal course for the patient.

Correct and early diagnosis and treatment can be life-saving for patients with this condition. However, as was the case of our patient, providing a definitive diagnosis swiftly is difficult and incorrect treatment can be fatal. In cases with digestive symptoms in which a discrepancy is observed between imaging findings and respiratory symptoms, late-presenting CDH should be among the differential, and CT and contrast-enhanced imaging of the digestive tract and other tests should be performed actively to enable a prompt and appropriate intervention.

Conflict of interest

The authors have no conflict of interest to declare.

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