CASE REPORT

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The Unexpected Diagnosis in a Patient with Previous Normal Chest X-rays: Late-Onset Diaphragmatic Hernia

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ABSTRACT Diaphragmatic hernia is a serious condition resulting from a defect in the diaphragm that allows abdominal organs to move into the thoracic cavity, most commonly observed on the left side. It may be detected immediately after birth with severe respiratory distress, or in 10-13% of cases, it may present later in life with respiratory and gastrointestinal complaints or growth retardation. This case report presents a 2-year-old male patient with no findings on 2 previous chest X-rays taken at different stages of his life, who was diagnosed with right-sided Bochdalek-type congenital diaphragmatic hernia after presenting with nausea, vomiting, cough, tachypnea, and fever.

Keywords: Congenital; diaphragmatic; hernia; late onset; right-sided

Diaphragmatic hernia is characterized by the passage of abdominal organs into the thoracic cavity due to a defect in the diaphragm.[1,2] It is primarily congenital but can also develop due to trauma, severe vomiting, and chronic constipation. Bochdalek-type diaphragmatic hernias are most commonly seen and typically occur on the left side; however, they can also be found on the right side in 8-21% of cases.[3] Congenital diaphragmatic hernia is often diagnosed during routine ultrasounds in pregnancy. Patients may present immediately after birth or within hours with symptoms ranging from mild to life-threatening respiratory distress.[4,5] In 10-13% of all congenital diaphragmatic hernias (CDH), diagnosis occurs at older ages.[6] This study presents a case of a right-sided Bochdalek-type congenital diaphragmatic hernia diagnosed in a 2-year-old patient who presented with nausea and vomiting, despite having no findings on 2 previous chest X-rays taken at different times in life.

CASE REPORT

A 2-year-old male patient was admitted to an external center due to sudden onset of nausea and vomiting with food contents, intermittent fever (38.5ºC), cough, and respiratory distress. The patient was diagnosed with pneumonia upon examination, tests, and chest radiography (Figure 1A). After treatment, the patient returned to us 10 days later with persistent complaints. Physical examination revealed: a temperature of 37°C, pulse at 140 beats/min, respiratory rate at 38 breaths/min, oxygen saturation: 88-90% (without oxygen) and >93% (with oxygen), arterial blood pressure of 87/68 mmHg, height 80 cm [(-2.77 sodium dodecyl sulfate (SDS)], and weight 10.5 kg (-2 SDS). Breath sounds were diminished in the right hemithorax. Other system examinations were normal. In laboratory tests, no pathology was detected. Chest radiography (Figure 1B) revealed in-

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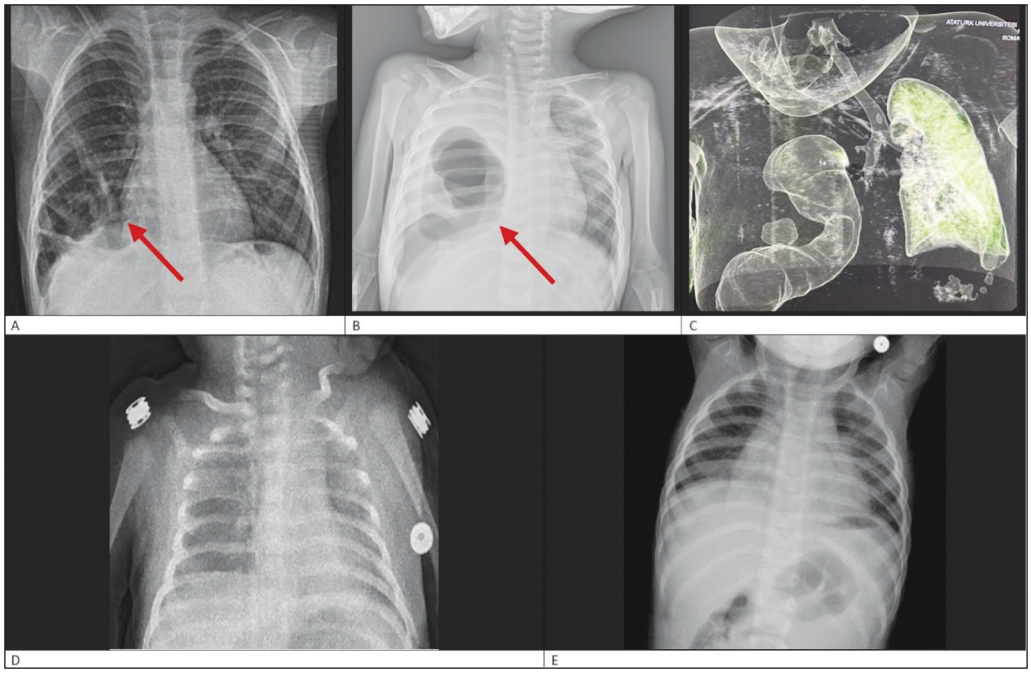

FIGURE 1: In the radiograph taken 10 days before the patient's admission to our institution (**A**), there is a disruption of the diaphragm margin in the right lower lobe at the midline, along with air images of dilated bowel loops and linear opacities associated with the bowel wall in the thorax. Similar findings are present in the radiograph taken at the time of admission (**B**) and in the contrast-enhanced thoracic CT (**C**). In the radiographs taken after the patient suffered a traffic accident at 1 month old (**D**) and another taken for a different reason at 1 year old (**E**), there is no evidence of diaphragmatic hernia, and the radiographs appear normal. However, the radiographs taken before admission and at the time of admission show a pronounced diaphragmatic hernia.

distinct diaphragm borders in the right hemithorax, with images suggesting the presence of bowel loops within the thorax. The contrast-enhanced thoracic computed tomography (CT) (Figure 1C) was interpreted by the radiology clinic as follows: "The heart and mediastinal structures are deviated to the left hemithorax, there is widespread effusion in the right hemithorax, the right lung appears totally atelectatic, there is prominent axial interstitial thickening in the peribronchovascular area of the left lung parenchyma, and ground-glass opacities are present, the diaphragm on the right cannot be clearly delineated, and herniation of colonic loops into the right hemithorax is observed, with an increase in the diameter and leveling of the involved colonic loop, causing separation in the fissural structures of the right upper and lower lobes."

The patient was born via cesarean section as the 2nd living child of a 32-year-old mother, weighing 3,250 grams. He cried immediately after birth without any cyanosis. He did not require follow-up in the neonatal intensive care unit. At 1 month of age, he was involved in a traffic accident and had multiple hospital admissions due to chronic constipation that had persisted since birth. A retrospective review revealed no evidence of diaphragmatic hernia in the chest radiographs taken after the traffic accident (Figure 1D) and at one year of age due to acute upper respiratory infection (Figure 1E). However, the radiograph taken during the admission ten days prior (Figure 1A) showed the presence of this image.

The patient was discussed with the pediatric surgery clinic, and a decision for surgery was made. During the surgery, it was found that the liver was deviated to the left, and there was a defect in the right posterolateral diaphragm (Bochdalek hernia), with colonic loops passing into the thorax along with the omentum through the defect. There was no hernia sac. Since the hernia area was quite small, the trapped bowel was extracted after enlarging the hernia opening slightly, and the defect was repaired (Figure 2).

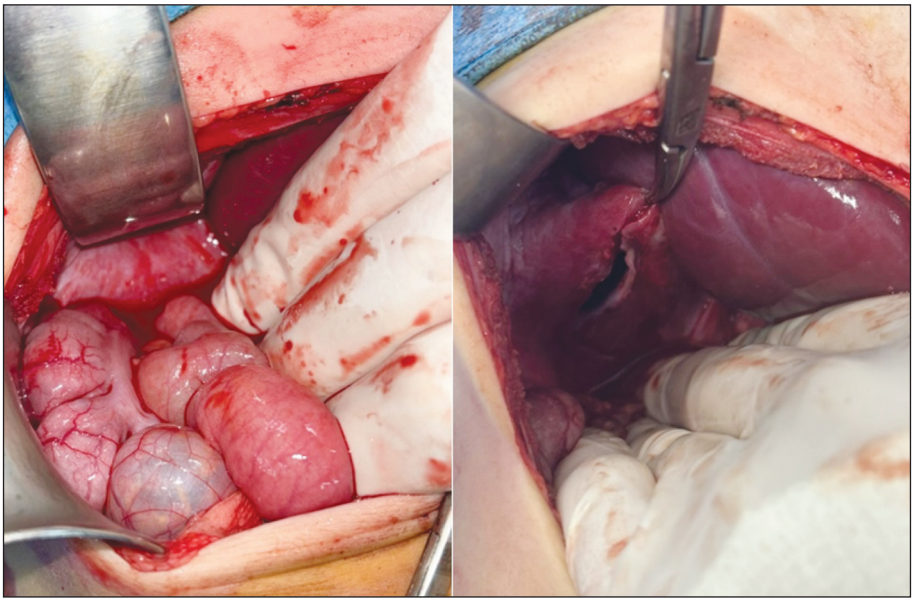

FIGURE 2: Appearance of the defect in the right diaphragm and herniated colonic loops during the operation

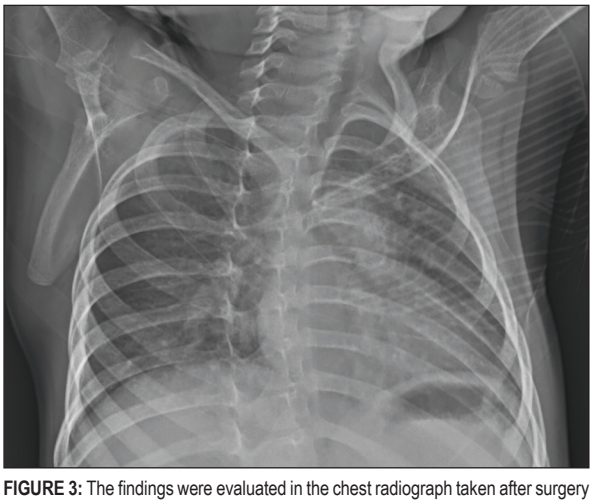

FIGURE 3: The findings were evaluated in the chest radiograph taken after surgery

The patient, who had an uneventful postoperative course, showed rapid improvement in symptoms. The chest radiograph was normal (Figure 3). The patient was discharged on the 4th day after surgery.

An informed consent was obtained from family in terms of publication.

DISCUSSION

CDH has a reported incidence of 2-3/10,000 live births and is frequently diagnosed at birth.[7-9] However, 10-13% of cases are detected later in life.[6] The nonspecific nature of symptoms can lead to misdiagnosis and delayed treatment.[10] Patients with delayed diagnosis may present with nonspecific respiratory and/or gastrointestinal symptoms. Growth retardation is also a common reason for consultation.[11,12] In patients presenting with these symptoms, or if radiographic examinations are performed for other reasons, hernia may be detected.[13] However, due to the rarity of the disease, findings on direct X-rays may be confused with respiratory system diseases such as pneumonia or pleural effusion. Consequently, patients may present with advanced and life-threatening clinical conditions.[13]

Diaphragmatic hernias are typically observed as posterolateral (dorsolateral side of the diaphragm). Posterolateral hernias are called Bochdalek hernias, while those observed on the anterior side (ventral side of the diaphragm) are referred to as Morgagni hernias. In addition to these hernias, hiatal hernias, and diaphragmatic eventrations can also lead to herniation. Bochdalek-type hernias are the most clinically common and generally indicate that the primary cause of the hernia extends back to the congenital period.[14] The chest X-rays taken at different stages of the patient's life suggest that CDH may not present

with classic images in the neonatal and early infancy periods and that typical images may emerge with advancing age (Figures 1B, D). Upon reviewing the literature, no case report with such clinical progression was found. Absence of any finding indicative of a hernia in the patient's previous chest radiographs and the emergence of radiological changes over time that led to the diagnosis is noteworthy. This situation can be explained by several possibilities:

Presence of an Initial Asymptomatic Small Congenital Defect: increased intra-abdominal pressure due to chronic constipation may have enlarged the defect and caused abdominal organs to herniate into the thoracic cavity.

Possible Effects of the Traffic Accident: The patient, who had experienced a traffic accident at 1 month of age, may have developed a small diaphragmatic defect, which could have enlarged over time. However, the location and structure of the hernia assessed during surgery suggested that the diaphragmatic hernia in our case was congenital.

CDH should be treated with a multidisciplinary approach at the time of diagnosis. Prolonged exposure to CDH can lead to pulmonary hypoplasia, pulmonary hypertension, cardiac dysfunction, and pulmonary vascular resistance. Reducing accompanying symptoms and, if there are no underlying factors that pose a surgical risk, surgery can provide dramatic improvement for the patient.[14,15] In this case, we decided that surgical treatment would be appropriate due to the absence of comorbidities that increase surgical risks beyond the hernia. All symptoms of the patient regressed after surgery, and he was discharged on the 4th-day post-operation.

In conclusion, the presented case indicates that in pediatric patients presenting with respiratory and/or gastrointestinal findings accompanied by growth retardation, CDH should be considered even if previous X-rays appear normal. Additionally, systematic evaluation alongside examinations aimed at our differential diagnoses (such as pneumonia) during direct X-rays will increase the likelihood of detecting significant pathologies.

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: *Turgay Aras, Ragıp Afşin Alay;* ***Design:*** *Turgay Aras;* ***Control/Supervision:*** *Dilek Yünlüel;* ***Data Collection and/or Processing:*** *Turgay Aras;* ***Analysis and/or Interpretation:*** *Turgay Aras, Ragıp Afşin Alay;* ***Literature Review:*** *Turgay Aras, Dilek Yünlüel;* ***Writing the Article:*** *Turgay Aras, Ragıp Afşin Alay.*

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