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Late-onset congenital diaphragmatic hernia: A case report☆

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ABSTRACT

INTRODUCTION: A Bochdalek hernia is a posterior congenital defect of the diaphragm, usually on the left hemidiafragm, caused by a lack of closure of the pleuroperitoneal canal between the eighth and tenth week of fetal life during the embryonic development. It typically presents in the neonatal period with severe respiratory failure.
PRESENTATION OF CASE: In this paper we present a 35 year old man with a 5-year history of episodes of severe dyspnea who arrived to the emergency room, during his medical work-up we incidentally found an intrathoracic gastric bubble, a laparoscopy was performed, founding a necrotic stomach and for defect correction.
DISCUSSION: This pathology is infrequent in adults, among this age group, there are two different clinical presentations: asymptomatic patients who are diagnosed incidentally when abdominal organs are found in the thorax in a chest X-ray, and symptomatic patients due to side effects of incarceration, strangulation, hemorrhage and visceral perforation in the chest cavity.
CONCLUSION: Diaphragmatic hernias are rare among adult population, and they are usually asymptomatic, in this case we presented a symptomatic patient, diagnosed with a chest X-ray and treated surgically. The surgical approach for the resolution of this pathology is variable and it depends on the presence and severity of visceral complications.
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1. Introduction

The Bochdalek hernia is a posterior congenital defect of the diaphragm caused by a lack of closure of the pleuroperitoneal cavity between the eighth and tenth week of embryonic life. Between 70% and 90% of cases occur on the left.[1] It was first described in 1848 by the Czechoslovakian anatomist, Vicent Alexander Bochdalek.[2] It usually presents during the first hours after birth with severe respiratory failure.[3,4] This condition is infrequent in adults, and there are 130 reported cases in the medical literature.[1]

2. Presentation of case

A 35 year-old male, with a 5-year history of episodes of severe dyspnea for no apparent reason, presented to the emergency room with a 36-h history of sharp, severe left sided chest pain. The pain was intermittent, radiated to the ipsilateral dorsal region, was accompanied by nausea, vomiting and inability to defecate. His vital signs were blood pressure 100/60, heart rate 120 beats per minute, respiratory rate 25 breaths per minute and temperature 37 °C. He was clinically dehydrated, with reduced chest expansion and breath sounds on the left side. He had some mild epigastric tenderness. Laboratory test results showed leucocytosis, elevation of lactate and of the lactate dehydrogenase, normal cardiac enzymes and type I respiratory failure. A postero-anterior and lateral chest X-ray (Fig. 1) showed a prominent gastric bubble and elevation of the left hemi-diaphragm. A subsequent computed tomogram (Fig. 2) demonstrated a left hemi-diaphragmatic defect with herniation of the stomach into the thorax.

An exploratory laparoscopy was performed. There was a 10 cm defect in the left hemi-diaphragm through which the whole stomach and part of the left lobe of the liver protruded. The stomach was completely necrotic, there were multiple adhesions in the abdominal cavity and 200 ml of hemoperitoneum were aspirated. A total gastrectomy and diaphragmatic plasty were performed, as well as a Roux-en-Y esophagojejunal anastomosis with feeding jejunostomy, and placement of an intrapleural catheter. The patient made an uneventful post-operative recovery and was discharged eight days later.

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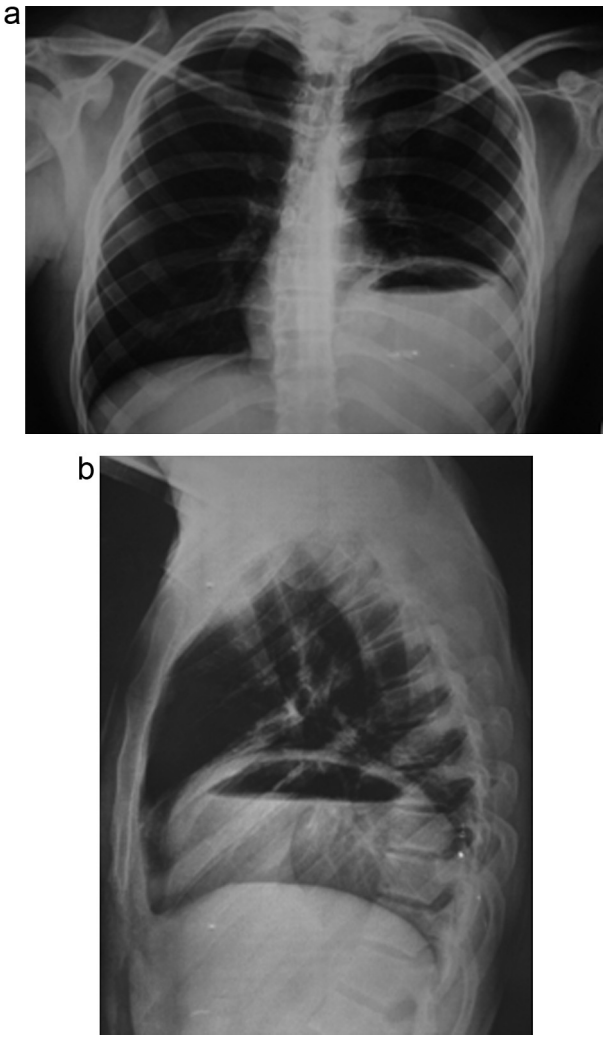

Fig. 1. (a) Antero posterior chest X-ray shows elevation of the left hemi-diaphragm, volume reduction of the left lung and minimal displacement of the mediastinum to the right. (b) Lateral chest X-ray shows elevation of the right hemi-diaphragm and the cardiac silhouette behind the gastric bubble.

3. Discussion

A Bochdalek hernia is a congenital defect of the diaphragm located in the posterior insertion. This is caused by a lack of closing of the pleuroperitoneal cavity by incomplete diaphragmatic development before the intestine returns to the abdomen from the yolk sack between weeks 8 and 10 of gestation. If hernia formation precedes lung development, pulmonary hypoplasia may occur with severe respiratory compromise at birth. In adults, this defect is uncommon, the lung in most cases develops normally and therefore symptoms are rare.[5] In this age group there are two typical clinical presentations: an incidental finding during X-ray examinations performed for symptoms not related to the hernia[6,7] or when symptoms develop as a result of incarceration, strangulation and visceral rupture inside the chest cavity. Symptoms vary according to the affected organ: digestive symptoms include intermittent abdominal pain, vomit and dysphagia while respiratory symptoms include chest pain and dyspnea.[6,8] In the case we presented the patient complained of intermittent dyspnea, vomiting and chest pain. The most frequently displaced organ is the stomach followed by the colon, spleen, small intestine and ureter.[5,9,10] In this case the stomach and a segment of the left lobe of the liver herniated. The diagnosis can be achieved through a simple chest

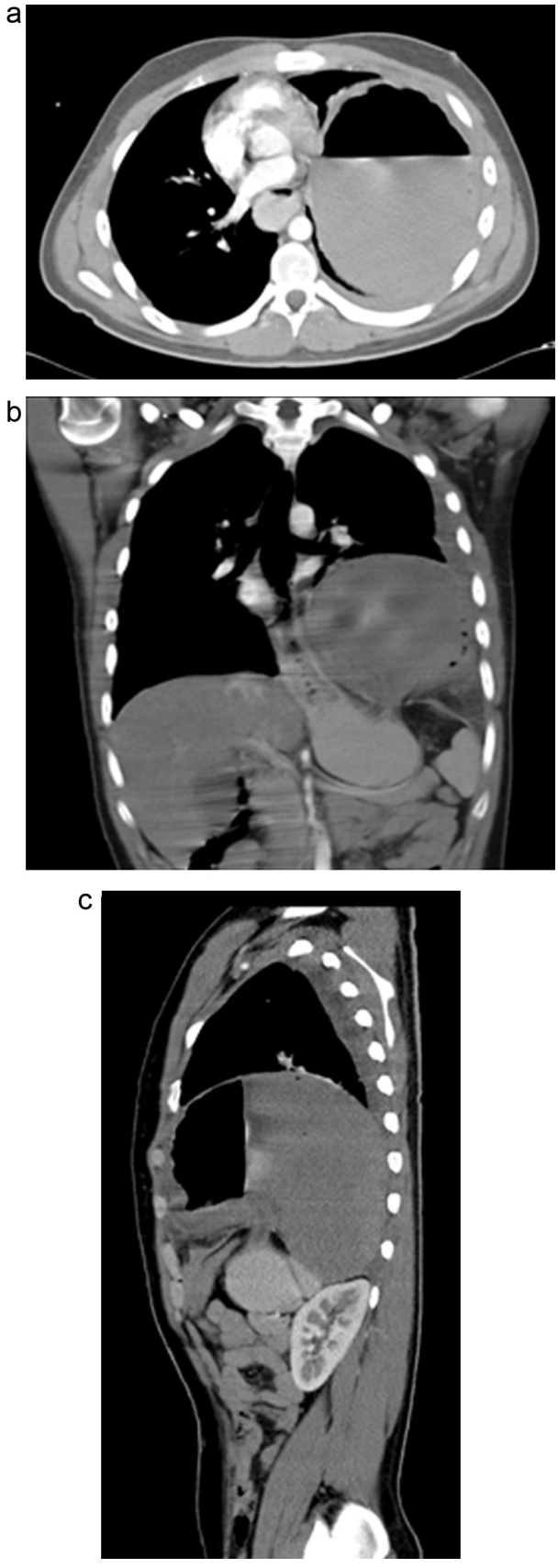

Fig. 2. (a) Transverse thoraco-abdominal CT shows a defect of the left hemi-diaphragm with an intra-thoracic stomach. (b) Longitudinal thoraco-abdominal CT shows stomach protrusion up to the heart level through a diaphragmatic defect, increased peri-gastric fat in the abdominal cavity and esophageal dilatation. (c) Lateral thoraco-abdominal CT showing the elevation of the right hemi-diaphragma, with an intra-thoracic stomach compressing the right lung.

X-ray, computerized axial tomography, MRI or an upper gastrointestinal series.[8,11,12] In the chest X-ray gas and organs are seen over the diaphragm. The typical findings of the computerized axial tomography are the presence of fat or soft tissue over the upper surface of the diaphragm characteristically posterolateral, a mass adjacent to the diaphragmatic defect, and a continuous density over and under the diaphragm's discontinuity.[12] The surgical approach for this pathology depends on the presence of visceral complications. In an elective setting most authors recommend the thoracic approach; on the other hand, when there are septic complications, the abdominal approach is preferred. The current trend is to use minimal invasive surgical techniques such as laparoscopy, and specially thoracoscopy, which has been satisfactorily performed in adults.[4,13–15]

4. Conclusion

Congenital diaphragmatic hernias are an uncommon diagnosis among adult populations because they are mainly recognized in infancy. They can be easily documented with a chest X-ray, in most cases incidentally although some adult patients may present with symptoms due to hernia complications. The knowledge of this anatomic defect presenting among adults is crucial for the identification and management, as it should be surgically corrected to avoid complications or to correct them if they are already present.

Conflicts of interest

The authors declare there are no conflicts of interest.

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None.

Ethical approval

Written informed consent was obtained from the patient for publication of this case report.

Author contributions

Marcel Adalid Tapia Vega – main surgeon of our patient, decided to turn our case into a case report, contributed to the writing.

Regina Darse Herbas Maldonado – surgeon of our patient, contributed with the data collection and writing

Gretty Tatiana Tapia Vega – contributed with the data collection and writing, and with the follow-up and post operative care of the patient.

Alenka Ximena Tapia Vega – contributed with the data collection and selection, and with the follow-up and post operative care of the patient.

Elisafat Arce Liévano – surgeon and first medical contact in the emergency room of our patient, contributed to the writing.

Paulina Moctezuma Velázquez – contributed with the data collection and writing, and with the follow-up and post operative care of the patient.

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