CASE REPORT

Congenital diaphragmatic hernia with gastric perforation in adult: Intrathoracic gastric perforation

Mehmet Taner Ünlü, M.D.,[1] Serkan Sarı, M.D.,[2] Ozan Çalışkan, M.D.[1]

[1]Department of General Surgery, University of Health Sciences, Şişli Hamidiye Etfal Training and Research Hospital, İstanbul-*Türkiye*
[2]Department of General Surgery, Başakşehir Çam and Sakura City Hospital, İstanbul-*Türkiye*

ABSTRACT

Congenital diaphragmatic hernias are rarely seen and they are usually diagnosed in the neonatal period. Congenital diaphragmatic defect, also known as Bochdalek hernia, usually occurs with the persistence of the pleuroperitoneal canal in the left posterolateral region of the diaphragm in the embryological period. Although it is rarely seen in the adults, conditions such as intestinal volvulus, strangulation, or perforation with congenital diaphragm defect progress with high mortality and morbidity. In this study, we reported our case that we operated for intrathoracic gastric perforation with congenital diaphragmatic defect. When the patient admitted to the hospital, he had an atypical abdominal pain, significant back pain, and suspicious respiratory complaints. Radiological imaging showed that the stomach and the spleen were located in the left hemithorax due to diaphragmatic hernia also stomach was very dilated. Tachycardia, hypotension, and low saturation developed on the 2nd day of the patient's hospitalization. In the control imaging of the patient, in the left hemithorax, stomach was collapsed and the surrounding appearance compatible with hydropneumothorax, after that findings emergency laparotomy was decided. During the operation, as demonstrated by the radiological findings, a diaphragm defect was seen in the left posterolateral region of the diaphragm. The stomach and spleen were herniated to the left hemithorax from this defect. The stomach and spleen were reduced into the abdomen. The left hemithorax was lavaged with 2000 cc isotonic, left tube thoracostomy was applied, and the diaphragm was repaired. The anterior stomach was primarily repaired. In post-operative follow-up, there were no complications other than wound infection and thoracic tube of the patient was removed. The patient who tolerated enteral food was discharged from hospital with full recovery.

Keywords: Bochdalek; diaphragmatic hernia; gastric perforation.

INTRODUCTION

Congenital hernias are usually diagnosed in the newborn or childhood. Bochdalek hernia is also a congenital defect in the left posterolateral region of the diaphragm due to the failure of regression of the pleuroperitoneal canal in the embryological period.[1] Congenital diaphragmatic defect is rarely encountered as a new diagnosis in adults. As Kinoshita et al.[2] stated in their study, the prevalence of these hernias in adulthood can reach up to 20% and most of them are asymptomatic. Bochdalek hernias in adults usually indicate gastrointestinal symptoms and they require to surgical treatment.

Gastric perforation into the thoracic cavity through a diaphragmatic hernia is very rare but, when it occurs, it is associated with high risk of mortality.[3] In this study, we reported a case who came to our clinic with suspicious intestinal symptoms, back pain, and respiratory problems and underwent intrathoracic gastric perforation repair secondary to congenital diaphragmatic hernia by laparotomy.

CASE REPORT

A 24-year-old male patient was consulted after applying to the emergency department due to back and abdominal pain.

Cite this article as: Ünlü MT, Sarı S, Çalışkan O. Congenital diaphragmatic hernia with gastric perforation in adult: Intrathoracic gastric perforation. Ulus Travma Acil Cerrahi Derg 2023;29:00-00

Address for correspondence: Mehmet Taner Ünlü, M.D.
SBÜ Şişli Hamidiye Etfal Eğitim ve Araştırma Hastanesi, Genel Cerrahi Kliniği, İstanbul, Türkiye
Tel: +90 212 - 373 50 00 E-mail: m.taner.unlu@gmail.com

Ulus Travma Acil Cerrahi Derg 2023;29(4):538-542 DOI: 10.14744/tjtes.2022.62289 Submitted: 18.01.2021 Revised: 06.05.2022 Accepted: 023.05.2022

The patient had no known history of chronic disease or chronic medical drug use. In the patient's history or recent emergency call, there was no history of trauma to the thorax or abdomen. The patient described only intermittent shortness of breath during heavy exertion from childhood. He had no history of smoking or alcohol. In the vital signs seen in the emergency department, the patient was normotensive and normocardic, and there was no fever. During physical examination, there was no defense or rebound on the abdomen of patient. In routine blood samples, the patient's amylase value was 188 U/L (28–100), lipase value was 434 U/L (13–60), white blood cell (WBC) was 15.73 10^9/L (3.91–10.9), and C-reactive protein (C-RP) was 16.3 mg/L (0-5). There was not any pathology in the other hemogram and biochemistry values of the patient. In the thoracoabdominal computed tomography (CT) images of the patient, it was seen that the stomach was dilated and herniated to the left hemithorax and the left lung was minimally inflated due to compression atelectasis. Due to the patient had no trauma history, he was diagnosed with bochdalek hernia based on the presence of hernia by the left posterolateral side of diaphragma. After the tomography findings were seen, a nasogastric tube was placed in the patient for decompression of the dilated stomach. When the nasogastric tube was inserted, it was filled with approximately 400 cc (cubic centimeter) of gastric fluid. However, on the 1st day, there was no evidence about acute complication of diaphragma hernia, so the patient, who had high levels of amylase and lipase and described back pain and abdominal pain, was hospitalized with the pre-diagnosis of acute pancreatitis. On the first night of hospitalization, the patient removed the nasogastric tube by himself due to discomfort. Afterward, no nasogastric tube could inserted again due to the absence of nausea or vomiting and the patient's intolerance.

A sudden deterioration of the patient's general condition was observed on the 2nd day of the patient's hospitalization. The patient, who did not have any abdominal defense or rebound on physical examination, continued to have back pain and he had newly developed vomiting complaints. The WBC value was 19.71 10^9/L and C-RP value was 39 mg/L on this day. During the follow-up of the patient, on 2nd day, tachycardia and hypotension have developed. A control thoracoabdominal CT was examined when his saturation measured by the pulse-oximeter was 79. On control CT, there were findings of hydropneumothorax in the left hemi thorax and it was observed that the herniated stomach was collapsed in the left hemi thorax, too. There was no sign of pneumoperitoneum. Although there was no sign of acute abdomen in the abdominal examination of the patient, an emergency operation was decided due to deterioration of general condition, increased acute phase reactants, and unstable vital signs. The patient was urgently taken to the operating room from the tomography room. Two milligrams of ampicillin sulbactam had administered prophylactically before the operation began. Laparotomy was performed with a median incision. In the operation, a diaphragm defect of approximately 5×5 cm was observed in the left posterolateral of the diaphragm as shown in the radiological findings (Fig. 1a, b). The stomach and spleen were herniated to the left hemithorax through this defect. In the exploration, no intra-abdominal pathology was found except herniation of the stomach and spleen to the thorax, and when the herniated organs were reduced into the abdomen, a 2×3 cm necrotic defect was observed in the anterior part of the stomach corpus. Approximately 400 cc stomach contents were aspirated from the left hemithorax. The left hemithorax was washed with 2000 cc isotonic until there was no sign of infection in thoracic cavity. Then left tube thoracostomy was applied and the diaphragm was repaired with non-absorbable

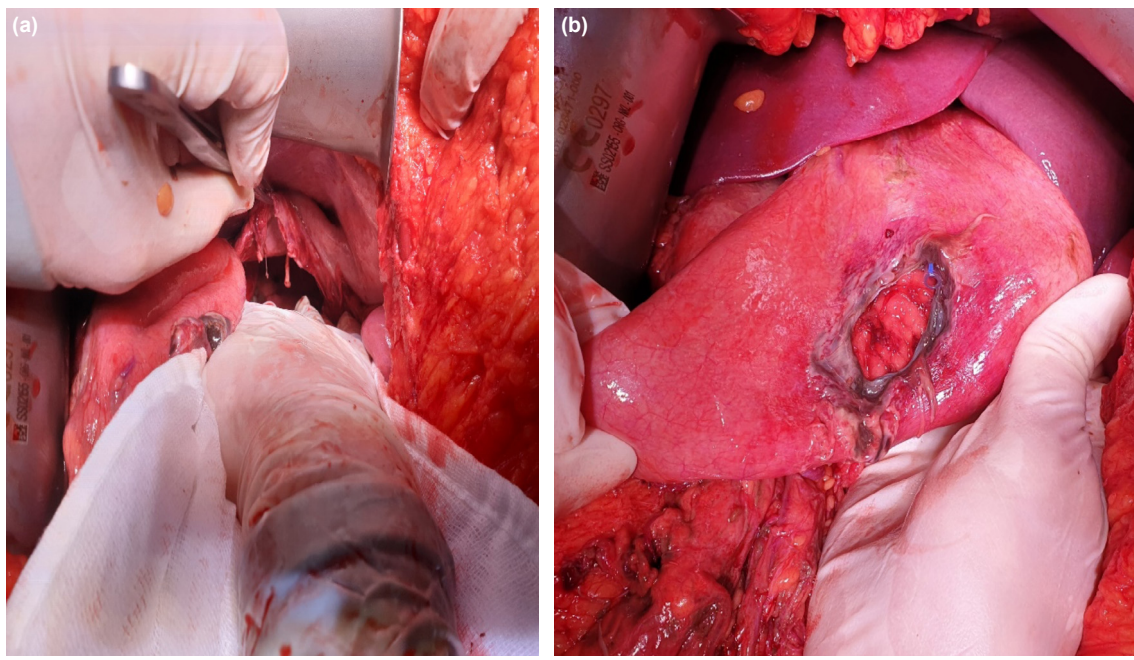

Figure 1. (a) Diaphragmatic hernia, **(b)** perforation in the herniated stomach.

suture. Synthetic mesh was not used in diaphragm repair due to contamination with gastric contents. Defect on the anterior surface of the stomach was repaired primarily as one layer, after debriding the necrotic areas on the edges. It was confirmed that there was no circulatory disorder in the spleen and the operation was completed.

In post-operative follow-up, there was no complication except wound infection on the incision area, and thoracic tube was removed post-operative 2nd day. The patient's oral food intake was gradually opened in post-operative period and he tolerated oral normal food on the post-operative 4th day. Then, the patient was discharged from hospital on 5th day with full recovery.

The patient did not have any intestinal or respiratory symptoms in the post-operative 4th month and in the follow-up. The patient's consent was obtained for this study.

DISCUSSION

Diaphragmatic hernias can be classified as congenital or acquired according to their etiology. The most common cause of diaphragm hernias in adults is acquired diaphragm rupture secondary to trauma.[3] Congenital diaphragmatic hernias are less common in adulthood than childhood period and they are usually caused by the persistence of the pleuroperitoneal canal that occurs between the 4th and 20th weeks of gestation. Although these hernias are usually diagnosed during the neonatal or perinatal period, they may rarely be asymptomatic until adulthood.[4] The most common types of congenital hernias are Morgagni defect which located anteriorly and Bochdalek defect which located posterolaterally.[5]

Bochdalek hernia is a well-known congenital diaphragmatic hernia in the perinatal period due to pulmonary hypoplasia and pulmonary hypertension findings.[6] Studies have shown that congenital diaphragmatic hernias in adults are mostly asymptomatic and not recognized in the pre-operative period. Symptoms of diaphragmatic herniation can be very variable according to the defect region. Most common symptoms are thoracic and abdominal pain, respiratory stress, and mechanic intestinal obstruction if the patient is symptomatic.[7] In the previous studies, commonly Bochdalek hernia was described with the symptoms of chest and/or abdominal pain and symptoms of ileus.[8]

In our case, there was no history of any abdominal or thoracic trauma. The diaphragm defect was located in the left posterolateral region and was compatible with congenital Bochdalek hernia.

Diaphragmatic hernias presenting with strangulation and perforation of luminal organs are rare but life-threatening conditions.[3] According to the review by Gedik et al.,[9] it can be fatal on the 32% of patients present with severe symptoms, due to visceral strangulation, perforation, and due to intrathoracic complications. Although most of them are asymptomatic, there is a case in the literature diagnosed through hematemesis due to bleeding from the herniated fundus.[10] In another one, diaphragmatic hernia has been diagnosed by the splenic vein thrombus.[11] As seen in these reports, diaphragmatic hernias may be presented with unusual symptoms.

Diaphragmatic hernias, which are symptomatic in adulthood, are usually diagnosed by radiological imaging after presenting with complaints of abdominal pain, nausea, vomiting, and shortness of breath. In our case, the patient presented with symptoms of atypical abdominal pain and persistent severe back pain. Furthermore, computer tomography images showed that the stomach was dilated in thoracic cavity and the patient was diagnosed as diaphragm hernia. It has been shown in the literature that gastric strangulation or gastric perforation extending into the thoracic cavity with diaphragmatic hernia is associated with high morbidity and mortality rates.[12] The presented case is a very rare situation that the patient has gastric perforation due to diaphragmatic herniation does not have any acute abdomen symptom and stable at applying hospital. This rare situation has been described in case reports by Vinnicombe et al.[3] and Lim and Kostin.[13] Despite the gastric perforation, it was unusual that acute symptoms did not develop and no findings of acute abdomen were found on physical examination in our patient. Although the symptoms of the patient were ambiguous, a rapid deterioration was observed in general condition on the 2nd day of hospitalization, acute phase reactants increased rapidly, and vital signs became unstable. The patient was taken into an emergency operation due to the sudden onset of hypotension, tachycardia, and low saturation.

The definitive treatment for bochdalek hernias in adults is surgery.[14] Although strangulation or perforation with diaphragmatic hernias is rare, if there is any doubt, early diagnosis, and proper treatment are essential due to the high morbidity and mortality rates.[8] The possibility of strangulation and perforation should not be forgotten in patients who present with vague abdominal pain, shortness of breath, and back pain, and whose radiological imaging shows diaphragmatic hernia. As in the operated case, these conditions may not produce any acute symptoms or signs of acute abdomen. Detailed anamnesis, detailed physical examination, radiological imaging findings, and general condition in follow-up and vital signs in follow-up are very important in these patients. Although there is no severe abdominal symptom and acute abdominal finding in the follow-up of the patients, changes in the general condition and vital signs follow-up may guide the decision of emergency operation. There is still no consensus on the absolute indication and timing of surgery, but due to the high mortality and morbidity rates, emergency surgery may be mandatory in patients with complicated diaphragmatic hernia.[15]

Emergency operations for diaphragmatic hernia could be performed by laparoscopy, thoracotomy and laparotomy.[15] In our case, laparotomy was performed with a median incision. The stomach and spleen were herniated to the left hemithorax. Furthermore, it was observed that there was a necrotic defect on the anterior side of the stomach corpus after stomach and spleen had been reduced into abdomen. Approximately 400 cc stomach contents were aspirated from thorax. Left tube thoracostomy was applied and the diaphragm was repaired with non-absorbable suture. The synthetic mesh was not used due to the contamination of gastric contents. The stomach was primarily repaired. It was confirmed that there was no circulatory disorder in the spleen and the operation was completed.

Morbidity and mortality rates may be higher in hernia with repairs performed in emergency cases compared to elective cases.[16] However, emergency operations should be performed immediately in cases that can be mortal, such as gastric strangulation and gastric perforation.[15]

Conclusion

As a result, patients diagnosed with diaphragmatic hernia should be operated under elective conditions. However, as in the case presented in this study, intestinal perforations due to diaphragmatic hernia, although rare, should be kept in the minds of surgeons and it should not be forgotten that they may require emergency surgery.

Informed Consent: Written informed consent was obtained from the patient for the publication of the case report and the accompanying images.

Peer-review: Externally peer-reviewed.

Authorship Contributions: Concept: M.T.Ü., O.Ç.; Design: M.T.Ü., S.S.; Supervision: S.S.; Data: M.T.Ü.; Analysis: M.T.Ü., O.Ç., S.S.; Literature search: M.T.Ü.; Writing: M.T.Ü., O.Ç.; Critical revision: M.T.Ü., S.S.

Conflict of Interest: None declared.

Financial Disclosure: The authors declared that this study has received no financial support.

REFERENCES

1. La Ganga V, Rossi G, Montobbio A, Gaione M. Bochdalek's diaphragmatic hernia in adults. Description of a clinical case. Minerva Chir 1999;54:443–5.
2. Kinoshita F, Ishiyama M, Honda S, Matsuzako M, Oikado K, Kinoshita T, et al. Late-presenting posterior transdiaphragmatic (Bochdalek) hernia in adults: Prevalence and MDCT characteristics. J Thorac Imaging 2009;24:17–22. [CrossRef]
3. Vinnicombe Z, Little M, Wan A. An unusual diaphragmatic hernia with gastric perforation and sub-acute presentation. Ann R Coll Surg Engl 2016;98:e181–383. [CrossRef]
4. Mullins ME, Stein J, Saini SS, Mueller PR. Prevalence of incidental Bochdalek's hernia in a large adult population. AJR Am J Roentgenol 2001;177:363–6. [CrossRef]
5. Veenma DC, de Klein A, Tibboel D. Developmental and genetic aspects of congenital diaphragmatic hernia. Pediatr Pulmonol 2012;47:534–45.
6. Akita M, Yamasaki N, Miyake T, Mimura K, Maeda E, Nishimura T, et al. Bochdalek hernia in an adult: Two case reports and a review of perioperative cardiopulmonary complications. Surg Case Rep 2020;6:6–11.
7. Mejri A, Trigui E. Bochdalek hernia in an adult: Two case reports and a review of perioperative cardiopulmonary complications. Medicine 2015;94:e2197. [CrossRef]
8. Brown SR, Horton JD, Trivette E, Hofmann LJ, Johnson JM. Bochdalek hernia in the adult: Demographics, presentation, and surgical management. Hernia 2011;15:23–30. [CrossRef]
9. Gedik E, Tuncer MC, Onat S, Avcı A, Tacyıldız I, Bac B. A review of Morgagni and Bochdalek hernias in adults. Folia Morphol (Warsz) 2011;70:5–12.
10. Wani AM, Al Qurashi T, Rehman SA, Al Harbi ZS, Sabbag AR, Al Ahdal M. Massive haematemesis due to strangulated gangrenous gastric herniation as the delayed presentation of post-traumatic diaphragmatic rupture. BMJ Case Rep 2010;2010:bcr0420102874. [CrossRef]
11. Hariharan D, Singhal R, Kinra S, Chilton A. Post traumatic intra thoracic spleen presenting with upper GI bleed!--a case report. BMC Gastroenterol 2006;6:38. [CrossRef]
12. Chau B, Dufel S. Gastric volvulus. Emerg Med J 2007;24:446–7. [CrossRef]
13. Lim D, Kostin R. The rare and unusual presentation of a gastric perforation in the setting of a large diaphragmatic hernia: A case report and literature review. J Surg Case Rep 2018;9:rjy238. [CrossRef]
14. Arya S, Shahab S, Kumar R, Garg PK. Adult Bochdalek hernia with organo-axial gastric volvulus: Misdiagnosed as hydropneumothorax. Acta Medica (Hradec Kralove) 2018;61:108–10. [CrossRef]
15. Testini M, Girardi A, Isernia RM, De Palma A, Catalano G, Pezzolla A, et al. Emergency surgery due to diaphragmatic hernia: Case series and review. World J Emerg Surg 2017;12:23. [CrossRef]
16. Dunn CP, Patel TA, Bildzukewicz NA, Henning JR, Lipham JC. Which hiatal hernia's need to be fixed? Large, small or none? Ann Laparosc Endosc Surg 2020;5:29. [CrossRef]

OLGU SUNUMU - *ÖZ*

Erişkinde konjenital diafram hernisi ile birlikte mide perforasyonu: İntratorasik mide perforasyonu

Dr. Mehmet Taner Ünlü,[1] Dr. Serkan Sarı,[2] Dr. Ozan Çalışkan[1]

[1]Sağlık Bilimleri Üniversitesi, Şişli Hamidiye Etfal Eğitim ve Araştırma Hastanesi, Genel Cerrahi Kliniği, İstanbul
[2]Başakşehir Çam ve Sakura Şehir Hastanesi, Genel Cerrahi Kliniği, İstanbul

Konjenital diafram hernileri nadirdir ve çoğunlukla neonatal dönemde tanı alır. Bochdalek hernisi olarak da bilinen konjenital diafram defekti genellikle diafram sol posterolateral bölgesinde embriyolojik dönemde bulunan plöroperitoneal kanalın persistan olarak kalmasıyla ortaya çıkar. Erişkinde nadir olarak görülse de konjenital diafram defektiyle birlikte olan intestinal volvulus, strangülasyon veya perforasyon gibi tablolar yüksek morbidite ve mortalite ile seyreder. Bu çalışmada konjenital diafram hernisi ile birlikte intratorasik mide perforasyonu sebebiyle opere ettiğimiz olgumuzu raporladık. Hastanın hastane başvurusunda atipik bir karın ağrısı, belirgin sırt ağrısı ve şüpheli respiratuvar şikayetleri bulunmaktaydı. Radyolojik görüntülemelerde diafram hernisine bağlı midenin dilate vaziyette ve dalağın sol hemitoraksta bulunduğu görülmekteydi. Hastanın servise internasyonu sonrası yatışının ikinci gününde taşikardi ve hipotansiyon gelişmesi saturasyon düşüklüğü sonrası yapılan kontrol görüntülemelerinde sol hemitoraksta; midenin kollabe ve etrafında hidropnömotoraks ile uyumlu görünüm olması üzerine acil laparotomi kararı verildi. Operasyonda radyolojik bulguların da gösterdiği gibi diafram sol posterolateralinde diafram defekti görüldü ve bu defektten mide ve dalak sol hemitoraksa herniye olmuştu. Mide ve dalak karın içine redükte edildi. Sol hemitoraks 2000 cc izotonik ile yıkandı, sol tüp torakostomi uygulandı ve diafram onarıldı. Mide ön yüzü primer onarıldı. Ameliyat sonrası takiplerinde yara yeri enfeksiyonu dışında bir komplikasyon görülmeyen hastanın toraks tüpü çekildi. Enteral gıdayı tolere eden hasta şifa ile taburcu edildi.

Anahtar sözcükler: Diafram hernisi; Bochdalek; mide perforasyonu.

Ulus Travma Acil Cerrahi Derg 2023;29(4):538-542 doi: 10.14744/tjtes.2022.62289