

Adult Bochdalek hernia: an analysis of eight patients

Yetişkin Bochdalek hernisi: Sekiz hastanın analizi

Fatih Meteroğlu, Atalay Şahin, Menduh Oruç, Serdar Onat

Department of Thoracic Surgery, Medical Faculty of Dicle University, Diyarbakır, Turkey

ABSTRACT

Background: This study aims to analyze congenital diaphragmatic hernias in adult age groups.

Methods: We retrospectively studied files of eight patients (3 males, 5 females; mean age 31.4 years; range 18 to 53 years) of Bochdalek hernias of advanced age who were operated in our clinic between January 2005 and June 2013. Patients' age, sex, associated diseases, symptoms, surgical access, abdominal organs in the thorax, postoperative morbidity and mortality rates, and duration of hospital stay were evaluated.

Results: Cough, chest pain, and dyspnea were the most common symptoms. Intestinal sounds in the thorax were present in six patients on auscultation. Pulmonary function tests, biplane chest X-rays, and thoracic computed tomography were performed. Bochdalek hernia was located on the left side in seven patients and on the right side in one patient. Posterolateral thoracotomy + laparotomy were performed in one patient, while posterolateral thoracotomy was performed in the other seven patients. No postoperative morbidity or mortality was observed. The mean duration of hospital stay was 8.75 days (range 4-25 days). Patients were followed up for a mean of 28.13 months (range 3-60 months).

Conclusion: Although rarely, congenital diaphragmatic hernias may be seen in the older age groups. Life-threatening complications may develop in asymptomatic patients over time. Surgical treatment is essential upon diagnosis.

Keywords: Adult; Bochdalek; diaphragmatic; hernia.

Posterolateral congenital diaphragmatic hernias (CDHs) were first described by Bochdalek in 1848. Bochdalek hernias (BHs) cause the herniation of intraabdominal organs into the thoracic cavity

ÖZ

Amaç: Bu çalışmada yetişkin yaş gruplarında doğuştan diyafragma hernili olgular irdelendi.

Çalışma planı: Ocak 2005 - Haziran 2013 tarihleri arasında kliniğimizde ameliyat edilen ileri yaş Bochdalek hernisi olan sekiz hastanın (3 erkek, 5 kadın; ort. yaş 31.4 yıl; dağılım 18-53 yıl) dosyaları geriye dönük olarak incelendi. Hastaların yaşı, cinsiyeti, eşlik eden hastalıkları, semptomları, cerrahi yaklaşım yeri, toraksta bulunan batin organları, ameliyat sonrası morbidite ve mortalite oranları ve hastanede kalış süreleri değerlendirildi.

Bulgular: Öksürük, göğüs ağrısı ve nefes darlığı en yaygın semptomlar idi. Oskültasyonda altı hastada toraksta bağırsak sesleri vardı. Solunum fonksiyon testleri, iki düzlemlilik akciğer grafisi ve toraks bilgisayarlı tomografi uygulandı. Bochdalek hernisi yedi hastada sol, bir hastada sağ tarafta yerleşimli idi. Bir hastaya posterolateral torakotomi + laparotomi, diğer yedi hastaya ise posterolateral torakotomi uygulandı. Ameliyat sonrası morbidite ve mortalite gözlenmedi. Ortalama hastanede kalış süresi 8.75 gün (dağılım 4-25 gün) idi. Hastalar ortalama 28.13 ay (3-60 ay) takip edildi.

Sonuç: Doğuştan diyafragma hernisi nadir de olsa ileri yaş gruplarında görülebilir. Semptomatik olmayan hastalarda zamanla yaşamı tehdit eden komplikasyonlar gelişebilir. Tanı konulduktan sonra cerrahi tedavi gereklidir.

Anahtar sözcükler: Yetişkin; Bochdalek; diyafragma; herni.

through a defect in the posterolateral diaphragm due to the failed closure of the pleuroperitoneal ducts. These ducts form a primitive communication system between the pleural and abdominal cavities during



Available online at
www.tgkdc.dergisi.org
doi: 10.5606/tgkdc.dergisi.2015.10277
QR (Quick Response) Code

Received: May 12, 2014 Accepted: August 12, 2014

Correspondence: Fatih Meteroğlu, M.D. Dicle Üniversitesi Tıp Fakültesi Göğüs Cerrahisi Anabilim Dalı, 21280 Diyarbakır, Turkey.

Tel: +90 412 - 248 80 01 e-mail: drfatihmeteroglu@hotmail.com

the intrauterine period.^[1,2] The size of the defects may be small enough to contain retroperitoneal fat or large enough to allow for the herniation of abdominal viscera. Therefore, BHs usually present with severe respiratory distress following birth or in the early years of life. However, some patients remain asymptomatic until they are older. The diagnosis of BHs can be easily made during the prenatal period via the use of fetal ultrasonography (USG), but their late presentation in adults makes diagnosing them more difficult. Hence, a careful physical examination combined with the use of imaging studies, such as multidetector computed tomography (MDCT), are needed to reach a correct diagnosis in patients suspected of having a BH.

PATIENTS AND METHODS

Between January 2005 and June 2013, we studied eight patients (3 males, 5 females; mean age 31.38 years; range 18-53 years) in our clinic who had undergone surgical repair for diaphragma. Those who had recently experienced a major trauma were excluded from the study. Biplane chest X-rays and CT were used to make a proper diagnosis, and the patients were evaluated according to age, gender, their symptoms, BH location, surgical procedure, length of hospital stays, and morbidities. A posterolateral thoracotomy with double-lumen endotracheal intubation was the preferred choice of treatment for most patients, and when needed, a laparotomy was also performed. We accessed the patients through either the seventh or eighth intercostal spaces. Synthetic mesh (polypropylene) was used for diaphragmatic defect repair in patients when the diaphragm was not primarily sutured, but nonabsorbable materials were used in the other cases.

RESULTS

The patients' symptoms were a cough, chest pain, dyspnea, abdominal pain, and discomfort. A physical examination revealed decreased pulmonary sounds in seven of the patients on the left side and in one on the right side in addition to the existence of bowel sounds in the thorax. Other examinations yielded nothing abnormal, and the patients had no history of major trauma or previous operations. The diagnoses were made based on imaging and test results based on their complaints. Magnetic resonance imaging (MRI) and multislice CT were the primary radiological instruments used following the chest X-rays (Figure 1, 2a, b, 3).

We electively operated on all the patients, with a posterolateral thoracotomy being performed on seven patients (87.5%) and a posterolateral

thoracotomy + a laparotomy were performed on one (12.5%). The seventh intercostal space was used for access in seven of the patients while the eighth intercostal space was used in the other. During the procedure, the spleen, colon, stomach, kidneys, small intestine, and omentum, which were protruding the thorax, were moved back into the abdomen using extreme caution (Table 1). We also performed an appendectomy on one patient who had a herniated small intestine, transverse colon, and cecum. In four cases, the diaphragm was only present anteriorly, and polypropylene mesh was used for repair. In the remaining cases, the diaphragm was sutured with nonabsorbable materials. Additionally, the left lower lobe was hypoplastic in two of the patients, and in one, the lung could not fill the apex, so the aseptic space persisted postoperatively. However, at the one-year follow-up, this space had disappeared. Moreover, a prolonged air leak and wound infection were observed in one patient with pulmonary hypoplasia, but no mortality occurred in any of the study participants. Furthermore, the mean postoperative length of hospital stay was 8.75 days (range 4-25 days), and the patients were followed up for a mean of 28.13 months (range 3-60 months).

DISCUSSION

A BH is one of the most common type of congenital anomalies. The incidence rate in newborns is 1/5000,^[3]



Figure 1. Posterior-anterior chest X-ray of a Bochdalek hernia.

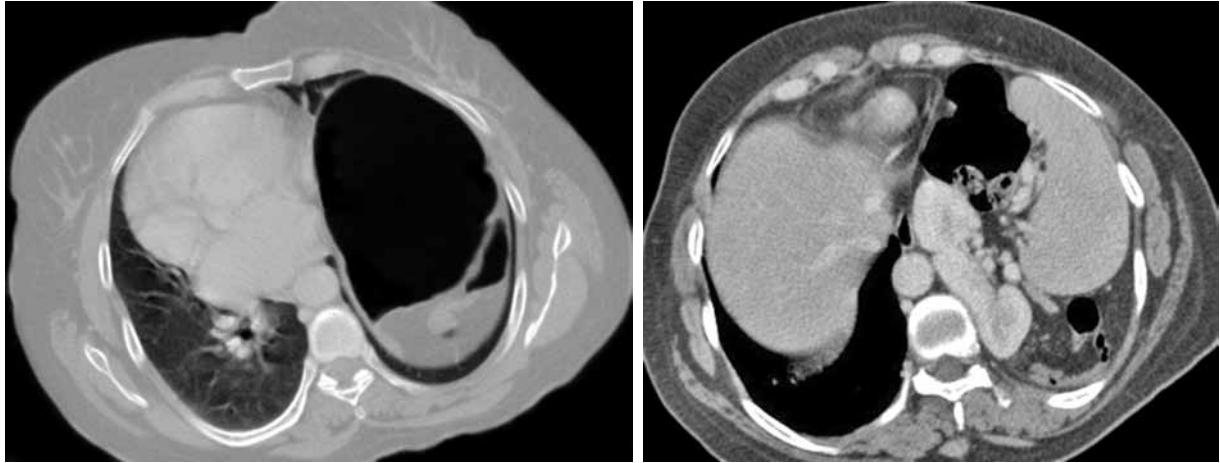


Figure 2. Tomographic scan of a patient with a Bochdalek hernia.

but it is rare in adults. Establishing an accurate prevalence rate for the general population is difficult because the frequency of BHs varies within different populations. However, the incidence of asymptomatic BHs in adults has been reported to be between 0.17% and 12.7% in various studies.^[4-9] The recent widespread use of contemporary imaging tools in asymptomatic patients has yielded higher incidence rates. A BH is normally found in infants by chance,^[10] with the diagnosis generally being made by either a chest X-ray, a gastrointestinal barium swallow, MRI, or scintigraphy. Their lungs may be hypoplastic,

and respiratory and abdominal symptoms have been reported in younger infants.^[11] Previously unnoticed BHs in adult patients are often diagnosed incidentally by routine chest X-rays because of a suspicious soft tissue mass of variable size bulging upward via the posterior part of the diaphragm along with a shift of the heart and mediastinum to the right side. Symptoms in adults vary and include chest pain, difficulty in breathing, abdominal pain, and dyspepsia. Congenital herniation, blunt or penetrating trauma, physical exertion (e.g., during labor and delivery), sneezing, coughing, and even the ingestion of a heavy meal have been put forth as potential reasons for late-presenting hernias, but traumatic causes are difficult to rule out in many cases of BHs diagnosed in adults. Diagnosis of BH can be made after a minor trauma; for example, we diagnosed a BH via a chest X-ray of a boy who had fallen from a bike. Some authors have suggested that hernias in adults can develop from a small diaphragmatic defect that has enlarged over time secondary to increased intraabdominal pressure from physical exertions.^[12] Symptomatic BHs in adults are relatively rare and are often pulmonary or gastrointestinal in nature, but they may lead to intestinal incarceration, visceral dysfunction, or severe pulmonary disease.^[1,6,7] The incidence rate for asymptomatic BHs in adults has been estimated to range anywhere from 1/2,000 to 1/7,000 based on autopsy studies^[2,4] to as high as 6% in early CT findings.^[5,7,8] Although rare, BH repairs have been reported on patients with dyspnea, abdominal distention, pain, and constipation,^[13] and the literature includes case reports of BH in advanced age groups.^[14-17] Our patient group included patients ranging in age from 18 to 53 years old, and respiratory compromise was the most common symptom. The BHs were especially common



Figure 3. Magnetic resonance imaging scan of patient with a Bochdalek hernia.

Table 1. Characteristics of the patients with Bochdalek hernias

Patient number	Age/gender	Complaint	Side	Organs in the thorax	Operation
1	53/F	Chest pain, dyspnea	Left	Spleen	PLT at the 8 th intercostal space
2	18/F	Cough	Left	Colon	PLT at the 7 th intercostal space
3	42/F	Abdominal pain, dyspnea	Left	Colon, omentum	Laparotomy + PLT at the 7 th intercostal space
4	37/F	Dyspnea	Left	Small intestine, transverse colon, cecum	PLT at the 7 th intercostal space
5	18/M	Chest pain	Left	Stomach, colon	PLT at the 7 th intercostal space
6	29/F	Dyspnea	Left	Stomach, colon	PLT at the 7 th intercostal space
7	43/M	Chest pain	Left	Spleen, stomach	PLT at the 8 th intercostal space
8	21/M	Abdominal pain, dyspnea	Right	Colon, kidney	PLT at the 8 th intercostal space

PLT: Posterolateral thoracotomy.

on the left side (87.5%), allowing for the abdominal organs to protrude into the chest. Left-sided BHs may contain the colon, stomach, spleen, small intestine, omentum, and adrenal gland, whereas right-sided BHs typically include the liver, kidney, and omentum. However, the right pleuroperitoneal channel can be easily closed when the liver is present.^[12] A BH is associated with intestinal malformations, cardiac anomalies, and pulmonary hypoplasia,^[9,18,19] and in two of our patients (25%), the left lower lobe was hypoplastic. A right-sided BH presenting with a right-sided pneumothorax has also been reported^[20] as well as one case involving an acute gastric dilatation in a 25-year-old.^[21] In addition, two other studies included some older asymptomatic patients.^[22,23] Imaging studies are not only performed to assess the hernia contents, but they can be used to evaluate the presence of associated abnormalities. A BH may be confused with other thoracic pathologies, such as lingular collapse, consolidation, a pericardial fat pad, sequestration of the lung, mediastinal lipoma, or a mediastinal mass due to the low sensitivity of plain chest X-rays.^[13,20] On the other hand, chest CT can more accurately visualize focal defects in the diaphragm and definitively diagnose herniation. The management of BHs consists of reducing the abdominal contents and repairing the defect via a laparotomy or thoracotomy,^[18,19,24] and any diaphragmatic defects that are present can be primarily closed without tension in most cases. Nonabsorbable mesh is used for repairs when primary suturing is not possible. Successful laparoscopic and thoracoscopic repairs of BH have both been reported,^[25] but if the bowel does not display signs of strangulation, a transthoracic approach should be considered because it is technically feasible. Hernia recurrence in adults is possible after the initial repair is performed; thus, patient follow-ups are important.

When hernia symptoms and acid reflux become severe and chronic, surgery may be the only appropriate course of action. However, our patients experienced no gastroesophageal reflux disease or any other morbidities.^[26] The outcome of symptomatic patients with a BH is related to the clinical presentation. Furthermore, delays in the diagnosis of a BH can cause significant morbidity, and preventing respiratory and abdominal complications is particularly crucial.

In conclusion, although rare, BHs have the potential to be misdiagnosed; therefore, they should be recognized, examined, and treated appropriately to avoid life-threatening complications. Moreover, patients presenting with symptoms of intestinal and pulmonary conditions should be evaluated with BHs in mind.

Declaration of conflicting interests

The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

Funding

The authors received no financial support for the research and/or authorship of this article.

REFERENCES

1. Reynolds M. Congenital posterolateral diaphragmatic hernias and other less common hernias of the diaphragm in infants and children. In: Shields TW, LoCicero J III, Ponn RB, Rusch VW, editors. *General thoracic surgery*. Philadelphia: Lippincott Williams & Wilkins; 2005. p. 761-71.
2. Karamustafaoglu YA, Yoruk Y, Kuzucuoglu M, Yanik F. Massive Bochdalek diaphragmatic hernia in adult with hypoplastic lung. *Asian Cardiovasc Thorac Ann* 2015;23:461-3.
3. Deprest JA, Gratacos E, Nicolaides K, Done E, Van Mieghem T, Gucciardo L, et al. Changing perspectives on the perinatal

- management of isolated congenital diaphragmatic hernia in Europe. *Clin Perinatol* 2009;36:329-47.
4. 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.
 5. Sener RN, Tugran C, Yorulmaz I, Dagdeviren A, Orguc S. Bilateral large Bochdalek hernias in an adult. CT demonstration. *Clin Imaging* 1995;19:40-2.
 6. Gale ME. Bochdalek hernia: prevalence and CT characteristics. *Radiology* 1985;156:449-52.
 7. 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.
 8. Temizöz O, Gençhellaç H, Yekeler E, Umit H, Unlü E, Ozdemir H, et al. Prevalence and MDCT characteristics of asymptomatic Bochdalek hernia in adult population. *Diagn Interv Radiol* 2010;16:52-5.
 9. 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.
 10. Mei-Zahav M, Solomon M, Trachsel D, Langer JC. Bochdalek diaphragmatic hernia: not only a neonatal disease. *Arch Dis Child* 2003;88:532-5.
 11. Newman BM, Afshani E, Karp MP, Jewett TC Jr, Cooney DR. Presentation of congenital diaphragmatic hernia past the neonatal period. *Arch Surg* 1986;121:813-6.
 12. Salaçin S, Alper B, Cekin N, Gülmen MK. Bochdalek hernia in adulthood: a review and an autopsy case report. *J Forensic Sci* 1994;39:1112-6.
 13. Sathyanarayana N, Rao RM, Rai SB. An adult recurrent diaphragmatic hernia with a near complete defect: a rare scenario. *J Clin Diagn Res* 2012;6:1574-6.
 14. Hamid KS, Rai SS, Rodriguez JA. Symptomatic Bochdalek hernia in an adult. *JLS* 2010;14:279-81.
 15. Laaksonen E, Silvasti S, Hakala T. Right-sided Bochdalek hernia in an adult: a case report. *J Med Case Rep* 2009;3:9291.
 16. Heaton ND, Adam G, Howard ER. The late presentation of postero-lateral congenital diaphragmatic hernias. *Postgrad Med J* 1992;68:445-8.
 17. Fine R, Borrero E, Stone A. Bochdalek hernia in adulthood. *N Y State J Med* 1987;87:516-8.
 18. Wenzel-Smith G. Posterolateral diaphragmatic hernia with small-bowel incarceration in an adult. *S Afr J Surg* 2013;51:73-4.
 19. Choi YK, Ahn JH, Kim KC, Won TH. An Adult Right-sided Bochdalek Hernia Accompanied with Hepatic Hypoplasia and Inguinal Hernia. *Korean J Thorac Cardiovasc Surg* 2012;45:348-50.
 20. Zenda T, Kaizaki C, Mori Y, Miyamoto S, Horichi Y, Nakashima A. Adult right-sided Bochdalek hernia facilitated by coexistent hepatic hypoplasia. *Abdom Imaging* 2000;25:394-6.
 21. Quah BS, Hashim I, Simpson H. Bochdalek diaphragmatic hernia presenting with acute gastric dilatation. *J Pediatr Surg* 1999;34:512-4.
 22. Prieto Nieto I, Perez Robledo JP, Hardisson D, Granado de la Fuente A. Bochdalek hernia in an adult. *Scand Cardiovasc J* 1998;32:113-4.
 23. 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. [Abstract]
 24. John PH, Thanakumar J, Krishnan A. Reduced port laparoscopic repair of Bochdalek hernia in an adult: A first report. *J Minim Access Surg* 2012;8:158-60.
 25. Yamaguchi M, Kuwano H, Hashizume M, Sugio K, Sugimachi K, Hyoudou Y. Thoracoscopic treatment of Bochdalek hernia in the adult: report of a case. *Ann Thorac Cardiovasc Surg* 2002;8:106-8.
 26. Rajput MZ, Fisichella PM. An 81-year-old gentleman with symptomatic Bochdalek hernia. *World J Gastrointest Surg* 2013;5:222-3.