Bochdalek Hernia in Adults: A Report of Two Cases with a Review of the Japanese Literature

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Abstract

We describe 2 challenging adult cases of Bochdalek hernia. The first case is that of a 22-year-old man who presented with left upper abdominal pain. The second case is that of a 36-year-old man who reported coughing after eating. In both cases, computed tomography revealed a defect in the left posterior attachment of the diaphragm. There was no vesical hernia, but parts of the greater omentum had prolapsed into the thoracic cavity in the first case, and parts of stomach, small intestine, colon, kidney, pancreas, and spleen had prolapsed into the thoracic cavity in the second case. In the first case, we were able to close the hernia defect with sutures by means of thoracoscopic surgery, and in the other case, that of a massive hernia, closure by means of laparoscopic surgery was difficult. We converted to laparotomy to perform suture closure, and we added a mesh patch.

Bochdalek hernia is a congenital diaphragmatic defect that is usually apparent in the neonatal period. Discovery of a Bochdalek hernia in an adult is rare. Surgical treatment is necessary, but there are different approaches. Selection of the appropriate procedure depends on the advantage to be gained by one approach over another in each particular case. The number of reports on thoracoscopic and laparoscopic surgery for treatment of Bochdalek hernia have increased in recent years. We think that endoscopic surgery for Bochdalek hernia will increase in popularity due to improvements in techniques and devices. We describe our 2 cases in detail and review our experience in light of the available Japanese literature.

Key Words

Bochdalek hernia, adult, surgical treatment

Introduction

Bochdalek hernia is the most common type of congenital diaphragmatic hernia, and most cases are discovered during the neonatal period. Patients with this condition present with severe respiratory and circulatory disorders. Presentation in adulthood is rare. Reduction of the prolapsed organs and closure of the hernial orifice are important during treatment, but there are no established approaches or repair methods. There have been reports of cases treated by means of thoracoscopic or laparoscopic surgery due to the increased popularity of endoscopic surgery in recent years. We treated 2 adult cases of Bochdalek hernia surgically in our department, from 1998 through 2015. We describe our 2 cases in detail and review our experience in light of the available Japanese literature.

Cases

Case 1

The patient was a 22-year-old man. His chief complaint was intermittent pain in the left upper quadrant of the abdomen. Abnormal shadows were noted in the thoracic cavity on computed tomography (CT) images, and he was referred to our hospital for...
examination. The patient reported no particular prior illness. Upon physical examination, he measured 169.8 cm in height, weighed 68.2 kg, and had a BMI of 23.7. Abdominal tenderness and abdominal distention were noted. Results of blood tests were normal. CT revealed that that greater omentum had prolapsed into the thoracic cavity through the left diaphragm (Fig. 1), and Bochdalek hernia was diagnosed. Thoracoscopic surgery was performed with the patient in the right lateral position and under differential lung ventilation after pneumothorax at 6 mmHg was established. Surgery was performed via 3 ports: a 12-mm port in the 5th intercostal space along the midaxillary line, a 5-mm port in the 4th intercostal space along the posterior axillary line, and a 15-mm port in the 6th intercostal space along the anterior axillary line. Upon examination of the thoracic cavity, no pulmonary adhesions were observed, and a 4 x 3-cm left posterior diaphragmatic defect was found. There was no hernial sac, and it was clear that the greater omentum had prolapsed into the thoracic cavity (Fig. 2a). The greater omentum was withdrawn into the abdominal cavity with the use of forceps, and the diaphragmatic defect was closed with 2-0 absorbable interrupted sutures. It was difficult to manipulate the forceps in the lateral part of the diaphragmatic defect, so we sutured the diaphragm and thoracic wall percutaneously with 2-0 suture using an Endo Closesutting device(Covidien) (Fig. 2b). The surgery lasted 227 minutes, and the blood loss volume was 33 mL. The postoperative course was uneventful, and the patient was discharged in good condition on postoperative day 7. Four years and 7 months have elapsed since the surgery, and there has been no recurrence.

Case 2

The patient was a 36-year-old man. For 2 years, he had been coughing after eating, and 6 months before presenting to us, he began experiencing respiratory difficulty when climbing and descending stairs. He was referred to our hospital after a contrast upper gastrointestinal tract series during a routine check-up revealed abnormal shadows. The patient’s medical history included cough-variant asthma, which was di-

Figure 1. Thoracoabdominal CT image obtained in Case 1 soon after the patient was referred to us. Prolapse of the greater omentum into the left thoracic cavity was noted.

Figure 2. Intraoperative image obtained upon thoracoscopy.

a. A 4 x 3-cm diaphragmatic defect was observed in the left posterior diaphragm along with omentum that had prolapsed into the thoracic cavity.

b. The hernial orifice was closed with interrupted sutures.

c. The diaphragm and thoracic wall were sutured percutaneously.
Diagnosed when he was 22 years of age and was resolved with oral medication. Upon physical examination, he measured 168.6 cm in height, weighed 71 kg, and had a BMI of 24.9. Abdominal tenderness and abdominal distention were noted. His blood test results were normal. A contrast upper gastrointestinal tract series showed that the major portion of the stomach protruded into the thoracic cavity (Fig. 3). CT revealed that the stomach, small intestine, colon, pancreas, spleen, and left kidney had prolapsed into the left thoracic cavity, causing significant displacement of the left lung (Fig. 4a, b). Bochdalek hernia was diagnosed. There was no luminal structure, such as small intestine or colon, within the right thoracic cavity, and we suspected a concomitant universal mesentery (Fig. 4c). We performed laparoscopic surgery with the patient in the supine position. We inserted a 10-mm camera port at the umbilicus, a 5-mm port into the right upper abdomen, a 12-mm port into the right abdomen, and a 5-mm port into the left abdomen. Pneumoperitoneum was established at 10 mmHg, and when we examined the intraabdominal cavity, we observed a large hernial orifice in the left diaphragm but no hernial sac. The majority of the

**Figure 3.** Upper gastrointestinal tract contrast radiograph obtained in Case 2 soon after the patient was referred to us. Most of the stomach was repositioned in the thoracic cavity.

**Figure 4.** Abdominal CT images obtained before surgery. Coronal view (a) and axial view (b) images, clearly depicted prolapse of the stomach, small intestine, colon, pancreas, spleen, and left kidney into the left thoracic cavity, causing significant displacement of the left lung. Axial view (c), there was no luminal structure, such as small intestine or colon, within the right thoracic cavity, and we suspected a universal mesentery.
stomach, and the small intestine, ascending colon, spleen, pancreas, greater omentum, and superior pole of the left kidney protruded into the thoracic cavity (Fig. 5a, b). A universal mesentery was also observed, there was no ligament of Treitz, and fixation of the ascending colon was completely absent. The area from the jejunum to the ascending colon had a common mesenteric membrane. No adhesions were observed on the prolapsed organs, and no vascular obstruction was observed in any of the organs. We attempted to return the prolapsed organs to the abdominal cavity by means of laparoscopic surgery, but the spleen was entirely contained within the thoracic cavity, and reduction into the abdominal cavity was difficult. Furthermore, due to the universal mesentery, a segment of bowel, extending from the small intestine to the ascending colon, was present within the left thoracic cavity, and even though we placed the patient in the reverse Trendelenburg position, the gastrointestinal tract that had been reduced into the abdominal cavity with forceps returned to the left epigastric region, and it became difficult to expand the visual field. We inserted a 12-mm port into the 7th intercostal space along the midaxillary line and then attempted manipulation using a thoracoscope to view the thoracic cavity. However, reduction of the prolapsed organs proved too difficult, and we converted to laparotomy. We made an epigastric midline incision, and after returning the prolapsed organs to the abdominal cavity, we separated the left kidney, which was in direct contact with the posterior hernial orifice, from the retroperitoneum and fully exposed the hernial orifice (Fig. 6a). Because the hernial orifice was 13 x 10 cm, and because the diaphragm around the posterior hernial orifice was somewhat fragile, we first closed the hernia using 2-0 absorbable suture (Fig. 6b). We then placed and fixed a Composix E/X Mesh (Bard) over the defect (Fig. 6c). We returned the malpositioned intraabdominal organs to their correct physiologic positions, fixed the stomach in position with 3 sutures, and fixed the ascending colon to the retroperitoneum, also with 3 sutures. Surgery lasted 383 minutes, and the blood loss volume was 1379 mL. An intravascular catheter-related infection was detected on postoperative day 5, but this improved readily with antibiotic treatment. The patient was discharged in good condition on postoperative day 14. Four months have elapsed since the surgery, and there has been no recurrence (Fig. 7).

Discussion

Bochdalek hernia, which is a type of congenital diaphragmatic hernia, involves prolapse of abdominal organs into the thoracic cavity through a diaphragmatic defect that arises due to failed closure of the pleuroperitoneal opening during the fetal period (1-3). The condition was first reported in 1848 by the Czech anatomist Victor Alexander Bochdalek and accounts for 75–85% of all congenital diaphragmatic hernias (3). The condition is frequently associated with other malformations such as pulmonary hypoplasia, cardiac malformation, and intestinal malrotation. The abdominal organs enter the thoracic cavity during an early embryonic stage and thus become compressed. Bochdalek hernia is usually diagnosed during the neonatal period. It presents with severe respiratory and circulatory disorders, so the mortality rate is high (1-3). The currently reported prevalence of congenital dia-
Intraoperative images obtained upon laparotomy.

a. Upon complete exposure, the hernial orifice was found to be 13 x 10 cm.
b. The hernial orifice was closed with interrupted sutures.
c. The sutured site was reinforced with ComposixE/X Mesh (Bard).

Figure 6. Thoracoabdominal CT image obtained 4 months after surgery.
The hernia repair remained intact.

Adult Bochdalek hernia commonly presents as gastrointestinal symptoms such as abdominal pain, abdominal distension, and vomiting due to obstruction of the prolapsed gastrointestinal tract. However, who have reached adulthood before Bochdalek hernia is diagnosed have sufficiently formed lungs; few present with respiratory and circulatory disorders. Abdominal symptoms are more common. Herniation occurs on the left side in 70–90% of patients, and this is believed to be due to late failure of pleuroperitoneal opening closure on the left. The right side is considered to be protected by the liver. In 62–90% of patients, there is no hernial sac, resulting in a false hernia. No hernial sac was observed in either of our cases.

Wiseman et al. reported that diagnosis in adulthood is usually precipitated by a state of increased intraabdominal pressure, such as that induced by pregnancy or abdominal surgery. Our second patient began abdominal muscle exercises 1 year prior to presentation, and this may have been the factor precipitating the increased intraabdominal pressure. Chui et al. reported that when a hernial orifice remains in the diaphragm due to failed suture closure, the orifice is obstructed by the hernial sac in cases in which a sac is present and by the peritoneum in cases in which no sac is present. The ensuing clinical course is asymptomatic. Prolapse then occurs when there is peritoneal rupture or when an organ that is obstructing the orifice is displaced due to increased intraabdominal pressure. Findings in the 2 cases we report suggest that the hernial orifice may have been obstructed by the omentum in the first case and by the spleen in the second case.

Adult Bochdalek hernia commonly presents as gastrointestinal symptoms such as abdominal pain, abdominal distension, and vomiting due to obstruction of the prolapsed gastrointestinal tract. However,
there are adult cases that present as thoracic symptoms such as chest pain and respiratory difficulty. There are also cases that are asymptomatic and discovered incidentally during routine health examinations. When the prolapsed gastrointestinal tract becomes impacted or undergoes necrosis or perforation, patients present with severe symptoms, and the condition may be fatal. Our second patient experienced respiratory symptoms including coughing and breathing difficulty, but these symptoms were mild. The abnormal findings during a routine health check were what prompted an examination. Despite the fact that examination showed that a large proportion of the gastrointestinal organs, namely the stomach, colon, and small intestine, had prolapsed into the thoracic cavity, the patient reported no gastrointestinal symptoms.

Many cases of adult Bochdalek hernia are diagnosed after abnormal shadows are seen in the lower lung fields, and characteristic findings include intrathoracic bowel gas and deviation of the mediastinum to the unaffected side. Prolapsed organs in the chest are confirmed on CT images, and coronal and sagittal views obtained by multiple-row detector CT are useful for identifying the hernial orifice, confirming the course of the diaphragm, and confirming the positional relationships of the prolapsed organs. In our first case, prolapse of the greater omentum into the thoracic cavity was observed on CT images, and in our second case prolapse of the stomach into the thoracic cavity was noted in a contrast upper gastrointestinal tract series.

In treating adult Bochdalek hernia, it is important to perform surgical reduction of the prolapsed organs and closure of the hernial orifice as soon as the diagnosis is made. Transthoracic, transabdominal, and combined thoracoabdominal routes are optional approaches. The transthoracic approach is beneficial in terms of the ease of examination and repair of the hernial orifice because direct visualization is possible, and adhesiolysis of prolapsed organs in the thoracic cavity is easily achieved. The transabdominal approach is beneficial in terms of the ease of intraabdominal manipulation of reduced abdominal organs, observation of perfusion disorders in prolapsed organs, and management of injury to the reduced organs. It is also possible to perform reduction and fixation of malrotated intestine, which occurs as a frequent complication. In addition, the abdominal approach is less surgically invasive than the thoracic approach because differential lung ventilation is not required. There are no established guidelines for selecting an approach, but the method that will confer the greatest benefit should be selected on a case-by-case basis.

The number of reports of thoracoscopic and laparoscopic surgery for treatment of Bochdalek hernia have increased in recent years due to technical advances in endoscopic surgery. There are also reports of use of combined thoracoscopic and laparoscopic surgery. The advantages of endoscopic surgery include minimal invasion, improved cosmesis, and reduced hospital stay. We selected a thoracoscopically guided transthoracic approach in the first case because CT images showed that the hernial orifice was small, approximately 4 cm, and that the hernia contained only the greater omentum. In the second case, CT images showed a huge hernial orifice, ≥10 cm, and several prolapsed organs, so we chose a transabdominal approach to ensure safe, certain surgical manipulation from the abdominal cavity and to facilitate the necessary reduction and fixation of the concomitant universal mesenteric membrane. Laparoscopic surgery was initiated, but because the spleen had prolapsed into the thoracic cavity, reduction was difficult, and because expansion of the visual field around the hernial orifice was hindered by the presence of gastrointestinal tract components in the left epigastric reason resulting from the universal mesentery, we converted the procedure to laparotomy. A universal mesentery is a form of intestinal malrotation in which the small intestine and colon have a common mesentery, so the colon retains mobility. The most common form, which we encountered in our second case, involves the cecum and ascending colon and their non-attachment to the retroperitoneum. There are no fixed opinions regarding whether the organs should be fixed during the treatment of universal mesentery, but we performed intestinal fixation in our second case to prevent torsion.

To the best of our knowledge, in Japan there have been 21 reported cases of adult Bochdalek hernia treated thoracoscopically or laparoscopically. These 17 cases plus our 2 cases are shown in Table 1. The number of reports of thoracoscopic and laparoscopic surgery for treatment of Bochdalek hernia have increased in recent years due to technical advances in endoscopic surgery.
(4 cases), and hand-assisted laparoscopic (1 case). A shift to thoracotomy or laparotomy was required in 6 cases and to hand-assisted laparoscopic surgery in 1 case. The hernia orifice was closed with suture (8 cases), mesh (8 cases), and suture and mesh (3 cases). Conversion to an alternative approach was necessary when reduction of the exposed organs proved difficult, when securing a good view was not possible, or when extensive adhesions were encountered. Most of the cases that required conversion involved a large number of exposed organs or large amount of exposed tissue and/or a high BMI. In our Case 2, likewise, a large number of exposed organs were found. We first used a laparoscope and thoracoscope, but because of a poor surgical view, we converted to laparotomy. Going forward, we think that endoscopic surgery for Bochdalek hernia will increase in popularity due to improvements in techniques and devices, but when there are many exposed organs or when the patient is obese, it is necessary to consider thoracotomy or laparotomy.

Suture closure of the hernial orifice during hernia repair is usually performed with nonabsorbable sutures, but when the tissues are fragile or when the hernial orifice is large, it is important to complete the closure with an artificial membrane such as polypropylene mesh or expanded polytetrafluoroethylene (ePTFE) if tension is applied during suture closure. Direct suture was possible in our second case, but because the hernial orifice was large, the diaphragm around the posterior hernial orifice was somewhat fragile. Because tension was applied to the sutures, we added a mesh patch. Generally, suture closure can be applied to cases with small hernial orifices. However, 5 cm is the reported limit for suture closure, and there have been cases of recurrence with hernial orifices of this size. Among the 23 cases shown in Table 1, suture closure was used for most cases of hernia orifices ≤5 cm, while mesh reinforcement was used for most cases of hernia orifices >5 cm.

There are several surgeons who recommended the use of nonabsorbable sutures for suture closure of the hernial orifice; this is thought to prevent recurrence. However, there are recent reports of good outcomes following the use of absorbable sutures. In our Case 1, absorbable suture was used, and there has been no recurrence. For cases in which the hernia orifice will remain tight, it is generally thought possible to use absorbable suture without risking infection. There is no established method of closure of the hernial orifice. Decisions are made by taking the size of the hernial orifice and strength of the diaphragm into consideration.

### Table 1: Reported Cases of Thoracoscopic or Laparoscopic Surgery for Adult Bochdalek Hernia in Japan

<table>
<thead>
<tr>
<th>Case</th>
<th>Age/sex</th>
<th>Defect size (cm)</th>
<th>Herniated organs/tissues</th>
<th>Surgical approach</th>
<th>Conversion</th>
<th>Closure</th>
<th>BMI</th>
<th>Year</th>
<th>Authors</th>
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<td>1</td>
<td>49/F</td>
<td>6.5×5</td>
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<td>2000</td>
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<td>Suture</td>
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<td>Yamaguchi et al.17</td>
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<td>Laparotomy</td>
<td>Mesh</td>
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<td>2003</td>
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<td>64/M</td>
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<td>23</td>
<td>2005</td>
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<td>Suture</td>
<td>18.1</td>
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<td>2009</td>
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<td>Sato et al.30</td>
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<td>Omentum</td>
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Age is shown in years. M: male; F: female; BMI: body mass index; HALS: hand-assisted laparoscopic surgery
Conclusion

Herein, we report 2 adult cases of Bochdalek hernia that we encountered and treated surgically in our department. We predict that thoracoscopic and laparoscopic surgery for the treatment of Bochdalek hernia will increase steadily with continuing advances in endoscopic surgery. For safe and certain surgical treatment, the approach and repair methods should be selected on an individual patient basis, taking the size of the hernial orifice, the degree of organ prolapse, and the presence or absence of intestinal malrotation into consideration.

References
22) Yokoyama T, Watanabe A, Nakagawa K, Inoue T, Migita K, Murogawa T, Ohyama T, Ishikawa H. Laparoscopic surgery for adult Bochdalek’s hernia with difficulties: report of a case [in Japa-


