

Case Report

Cytoreductive Surgery Followed by Hyperthermic Intraperitoneal Chemotherapy for Recurrent Ovarian Cancer with Incidental Bochdalek Hernia and Postoperative Bilateral Thalamic Infarct: A Case Report

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Keywords

Bochdalek hernia · Hyperthermic intraperitoneal chemotherapy · Thalamic infarction · Chest tube

Abstract

Congenital Bochdalek hernia is a defect of the diaphragm and very rare in adults. Only around 100 cases have been reported in the literature. Herein, we present a case with a recurrent ovarian cancer who underwent secondary cytoreductive surgery and hyperthermic intraperitoneal chemotherapy. An oval defect with dimensions of 3 × 4 cm was seen in the left posterolateral site of the diaphragm during surgical exploration. In addition, a 6 × 3 cm iatrogenic right-sided diaphragmatic defect was found and repaired. In the early postoperative period, a bilateral thalamic infarction occurred.

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Introduction

Congenital Bochdalek hernia (CBH) develops as a result of the inappropriate fusion of the pleuroperitoneal fold and septum transversum at the site of the posterolateral lumbo-costal trigone at the sixth week of gestation [1]. This entity was first described by McCauley in 1754 and then by Vincent Alexander Bochdalek in 1848. The incidence of this congenital defect is 1 in 2,500 births. Generally, the defect is left-sided (85%) [1]. This type of diaphragmatic hernia is considered a perinatal disease that presents a few hours after birth, or in early infancy, and it is associated with significant morbidity and mortality [2]. CBH in adults is very rare; it remains asymptomatic until adulthood and generally presents with thoracic and/or abdominal symptoms aggravated in supine position [3].

We present a case of left-sided congenital Bochdalek hernia that remained asymptomatic and was discovered incidentally during secondary cytoreductive surgery for epithelial ovarian cancer; in this patient, a iatrogenic diaphragmatic defect was found, hyperthermic intraperitoneal chemotherapy (HIPEC) was performed and early postoperative bilateral anterior thalamic infarction developed.

Case Report

A 46-year-old female patient with a BMI of 25 was referred to our tertiary center (Cerrahpasa Medical Faculty, Gynecological Oncology Unit) in August 2016 with the diagnosis of platinum-resistant ovarian cancer. She had undergone primary cytoreductive surgery 6 months ago. Her medical history was unremarkable except for treatment for epithelial ovarian carcinoma. Positron emission tomography and computed tomography (CT) revealed implants on the right diaphragmatic surface after first-line adjuvant chemotherapy. She has never had any thoracic or abdominal symptoms and no history of major trauma. Initial preoperative evaluation of the patient revealed no significant sign of diaphragmatic hernia. Her physical examination was normal, including chest auscultation sounds and chest X-ray. Thoracic CT did not reveal any abnormal image that could be evaluated as a diaphragmatic hernia. Secondary cytoreductive surgery and HIPEC were planned. She was placed in flank position, and, under general anesthesia, laparotomy was performed. An oval defect with the dimensions of 3 × 4 cm was seen in the left posterolateral site of the diaphragm (Fig. 1). The most distal portion of the inferior lobe of the left lung was visible through the defect during inspiration and expiration movements. There was no herniation or strangulation of abdominal components. In addition, the right diaphragm was completely stripped because of a tumor implant. Meanwhile, a 6 × 3 cm iatrogenic diaphragmatic defect was detected (Fig. 2). Drainage chest tubes were inserted bilaterally via the 6th intercostal spaces to avoid leakage of the chemotherapeutic agents across repaired diaphragm and postoperative massive pleural effusion. The tissue around the congenital left-sided defect was excised circularly and the defect itself was repaired with interrupted nonabsorbable monofilament sutures. The iatrogenic defect was also repaired with interrupted nonabsorbable monofilament sutures. After complete cytoreduction had been achieved, hyperthermic intraperitoneal chemotherapy with doxorubicin 35 mg/m² and paclitaxel 175 mg/m² for 60 min at 42.5°C was delivered. No leakage via drainage tubes from the thoracic cavity was noted during the procedure. During the surgery, the patient was managed by standard monitoring, which involved continuous monitoring of electrocardiography, invasive blood pressure, central venous pressure, pulse oximetry, body temperature and hourly urine output. Arterial samples were drawn at

regular periods for the determination of electrolytes, acid base status and hematocrit. Finally, the operation lasted 4 h. At the end of the surgery, the patient remained intubated and was transferred to the intensive care unit for monitoring ongoing resuscitation along with fluid and electrolyte management. The patient was extubated 10 h after completion of surgery, and was transferred to the service on the 2nd postoperative day. Prophylactic enoxaparin 4,000 IU was started 6 h after surgery. On the 3rd and 4th postoperative day, excessive sleepiness throughout the day and dyspraxia was noted. Bilateral anterior thalamic infarction caused by emboli that block the thalamosubthalamic artery was demonstrated on brain CT (Fig. 3). The dose of enoxaparin was changed from 4,000 IU once a day to 800 IU twice a day. The patient was discharged on the 12th day after surgery. The patient's symptoms improved within 3 weeks. Follow-up at 4 months after surgery showed no evidence of recurrence and no symptom regarding thalamic infarct.

Discussion

CBH generally presents in newborns with severe respiratory failure or in infancy due to herniation and/or strangulation of the abdominal contents [1]. The presentation of this defect in adulthood is rare. One of the best studies concerning the incidence of the defect in adults was carried out by Mullins et al. [4], who reviewed 13,138 CT scans. Their study revealed an incidence of 0.17%, with 68% being right-sided defects and 77% of the patients being female [4]. Another study, done by Brown et al. [1], produced different results. This was ascribed to differences in the reviewed cases. Since the second study summarized symptomatic cases, demographics identified were different than those in asymptomatic ones [1]. This difference showed that more cases of right-sided CBH in females may remain silent than left-sided cases. These data were supported by several case reports [5]. Nevertheless, in our case, the defect was noted on the left side. Also, no herniation of intestinal loops was noted, and the patient has never had any thoracic or abdominal symptom that might be related to diaphragmatic hernia.

Diagnosis of the disease might be carried out by chest CT when there are clinically unexplained thoracic and abdominal symptoms. A chest CT can show the diaphragmatic defect and the herniated organs [6]. The chest CT of our patient did not reveal any of the above-mentioned patterns. There are several cases concerning surgical repair of the defect – both laparoscopic, via laparotomy, and through the thoracic cavity [7–9]. We performed upper midline incision for laparotomy, and there was no need for another access to carry out the repair of the diaphragmatic defect. Repair of diaphragmatic defect can be performed by various techniques, using absorbable or nonabsorbable sutures in an interrupted or continuous fashion. From our point of view, surgeons may choose any of them provided that they are comfortable with the procedure.

Thoracic drainage using chest tubes may not always be necessary. The main reason for using bilateral chest tubes was to prevent massive pleural effusion due to HIPEC. In addition, intra-abdominal pressure increases during HIPEC, which may subsequently cause a cephalad shift of the diaphragm to increase peak airway pressures and reduce residual capacity. These changes may adversely affect oxygenation and cardiac output. Chest drains which were inserted prior to HIPEC reduce the increase in peak airway pressure, and by this way, the length of the intubation period and hospital stay can be decreased [10].

Postoperative bilateral thalamic infarct is a very rare complication of surgery. Malignancy itself, HIPEC, central venous cannulation, the position of the patient's neck, sudden blood loss and a long duration of the operation were noted as possible risk factors in our case [11].

In conclusion, although congenital Bochdalek hernia commonly occurs in the newborn and infancy period, it must not be excluded in the case of unexplained abdominal and/or thoracic symptoms in adults. Despite the fact that this kind of rare condition presents with pain, obstruction symptoms or pulmonary symptoms, it can be silent until discovered incidentally. Moreover, chest CT cannot always exclude this type of hernia. Iatrogenic diaphragmatic defects may occur in order to provide complete cytoreduction. HIPEC can be used after the diaphragmatic defects have been repaired. Thoracic tubes may be helpful to prevent pleural effusion. Secondly, cerebral embolism should be suspected in the presence of any neurological symptom in postoperative patients.

Statement of Ethics

Written informed consent was obtained from the patient for publication.

Disclosure Statement

The authors declare that they have no conflicts of interest.

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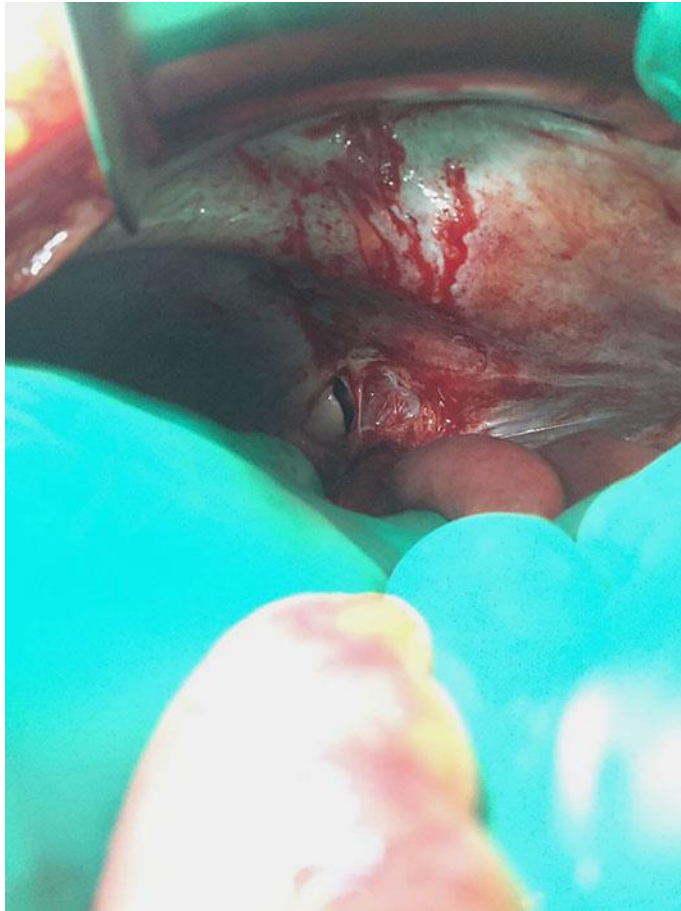


Fig. 1. Intraoperative photo of the left diaphragmatic defect.

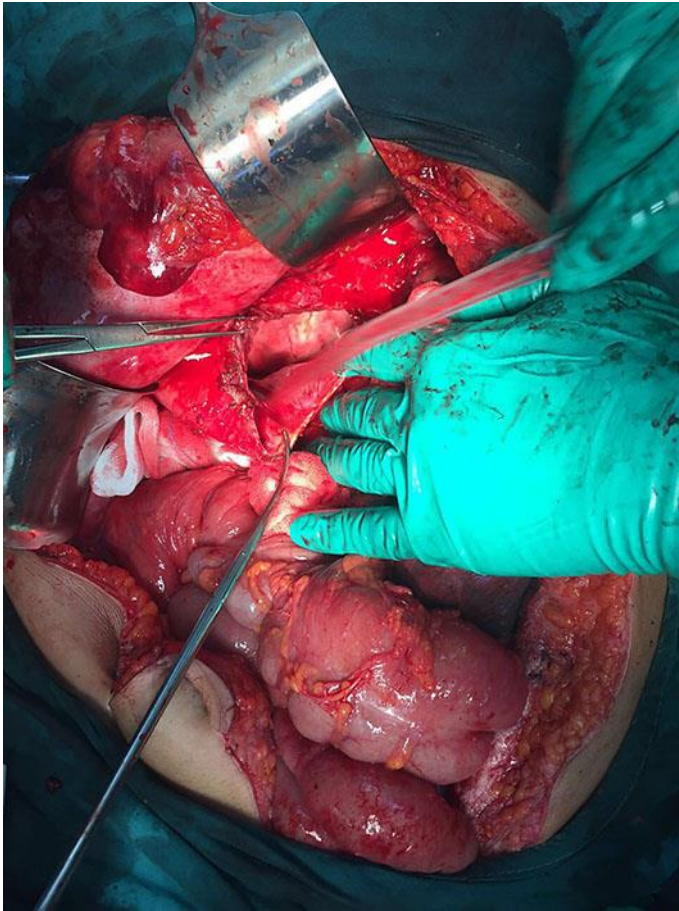


Fig. 2. Intraoperative photo of the right diaphragmatic injury.

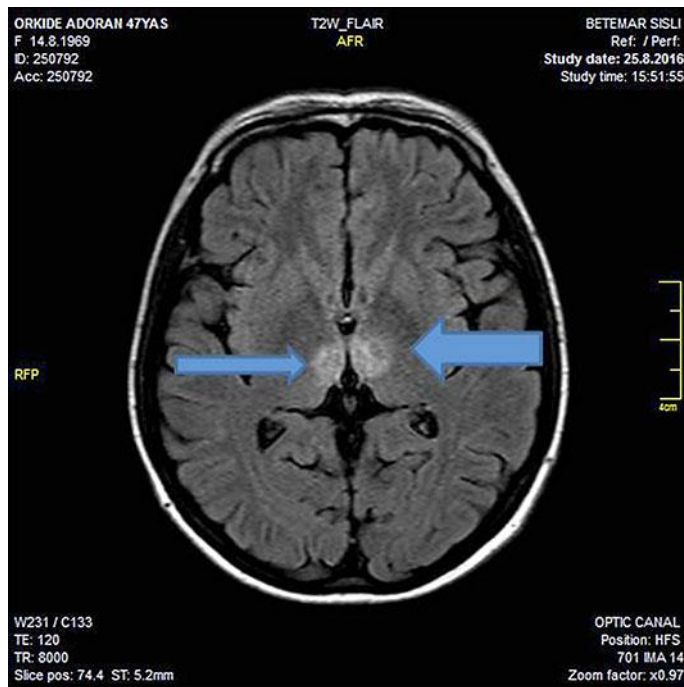


Fig. 3. An axial T2 FLAIR image (both arrows) shows bilateral subacute infarct of the thalamus.