Severe delayed diaphragmatic hernia with total gastric migration after cytoreductive surgery, a rare complication: A case report

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Abstract. Complete cytoreductive surgery is the cornerstone of advanced ovarian cancer management. Diaphragmatic resection may be necessary to remove any residual disease, a procedure that rarely leads to diaphragmatic hernia, a potentially life-threatening complication. The present case report describes a woman with high-grade serous ovarian adenocarcinoma treated with neoadjuvant chemotherapy followed by interval cytoreductive surgery that included bilateral diaphragmatic peritoneal stripping. At 4 months postoperatively, the patient presented with a first sub-occlusive syndrome that was managed with medical treatment. After 3 days, the patient presented with complete bowel obstruction, multiorgan failure and shock. Imaging showed a left diaphragmatic hernia with intrathoracic gastric migration. Emergency surgery confirmed the diagnosis of diaphragmatic hernia and corrected the problem. Though the patient did not experience a recurrence of her diaphragmatic hernia, they did however present with a disease relapse and succumbed 1 year later. Delayed diaphragmatic hernia is a rare, but severe complication of cytoreductive surgery. Computed tomography imaging is key to diagnosis. Surgical repair remains the mainstay of treatment. Known risk factors include diaphragmatic resection, hyperthermic intraperitoneal chemotherapy and bevacizumab, none of which were present in the present case report. This case underscores the importance of recognizing malnutrition as a major risk factor for delayed diaphragmatic hernia following cytoreductive surgery.

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Abbreviations: ACVBP, Adriamycin cyclophosphamide, vindesine, bleomycin, prednisone; HIPEC, hyperthermic intraperitoneal chemotherapy

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Introduction

Complete cytoreductive surgery with no residual disease remains the gold standard for managing patients with ovarian cancer, as it is associated with improved prognosis (1-4). To achieve complete cytoreduction, diaphragmatic resection is occasionally recommended (5-9). However, this procedure carries a relatively rare risk of developing diaphragmatic hernia (10), with left-sided hernias being more frequent than the right-sided ones (11). The incidence of left diaphragmatic herniation in the literature following left diaphragm peritonectomy or resection is 2.2% (12). Although uncommon, diaphragmatic hernia can lead to severe clinical manifestations and requires adequate management. The present case report details a delayed left diaphragmatic hernia, 4 months after interval cytoreductive surgery in a patient with advanced ovarian cancer.

Case report

A 57-year-old woman had a history of mediastinal large B-cell non-Hodgkin lymphoma treated with Adriamycin-cyclophosphamide-vindesine-bleomycin and prednisone (ACVBP) chemotherapy alone. The patient was diagnosed with high-grade papillary serous ovarian adenocarcinoma, with extensive peritoneal carcinomatosis involving the peritoneum and mesentery and associated with ascites (PCI 34/39). The patient was managed at the Gustave Roussy Institute (Paris, France) in August 2009. At diagnosis, the CA125 level of the patient was 12,000 and the CA19.9 level was 157. Preliminary tests also included gastroscopy and colonoscopy, both of which were normal.

Neoadjuvant chemotherapy was initiated with a carboplatin area under the free carboplatin plasma concentration vs. time curve value of 5 and weekly paclitaxel (80 mg/m^2). Paclitaxel was further adjusted during the third cycle of chemotherapy to 60 mg/m^2 due to grade 3 hepatic toxicity. After three cycles, the CA125 level of the patients decreased to 162 and laparoscopic re-evaluation showed a reduction in tumor burden. The patient then underwent an interval cytoreduction that included an en-bloc peritoneal stripping of both diaphragmatic domes and Morrison's pouch, without diaphragmatic or pleural injury, along with omento-splenectomy, cholecystectomy, transverse and left colectomy, posterior pelvic exenteration, fulguration of mesenteric lesions and bilateral pelvic and para-aortic lymphadenectomy.

A complete resection of the peritoneum of the left diaphragmatic dome was performed due to its extensive involvement. Pathological examination confirmed the persistence of extensive residual disease. The patient made a full recovery ≤1 month and resumed chemotherapy for five additional cycles.

The patient developed an asymptomatic lymphocele that did not require intervention. Computed tomography (CT) imaging at 2 months postoperatively still showed moderate lymphocele, and a CT scan 4 days prior to hernia surgery also demonstrated persistent lymphatic effusion (Fig. 1). This patient presented with malnutrition and prior to cytoreductive surgery, the patient reported marked anorexia. The patients also lost >10% of their body weight ≤3 months postoperatively and their serum albumin level (measured 2 days after cytoreduction) was 1.3 g/dl. A few days before hernia surgery, serum albumin level of the patient had increased to 3.1 g/dl.

At 4 months after cytoreductive surgery, the patient presented with subocclusive symptoms. CT scan did not reveal a digestive occlusion, but showed instead gastric distension with horizontal displacement of the stomach, occupying the lower half of the left hemithorax, with ascites (Fig. 2A). Chest X-ray revealed elevation of the left hemidiaphragm, marked gastric distension and partial atelectasis of the left lower lobe (Fig. 2B). The clinical evolution was favorable with medical treatment, allowing the discharge of the patient. However, 3 days later, the patient was readmitted to the intensive care unit for complete bowel obstruction and multiorgan failure, as well as shock. Chest imaging suggested a left pneumothorax (Fig. 2C and D). The patient was intubated for hypercapnic respiratory failure, followed by post-intubation collapse requiring resuscitation. Infectious tests were negative.

Following the diagnosis of a left diaphragmatic hernia with intrathoracic gastric migration, surgical exploration confirmed a 4 cm defect in the left diaphragm through which the entire stomach had herniated. The gastric hernia was reduced without signs of ischemia. Gastric serosal sutures were further added for reinforcement. Diaphragmatic defect was closed using U-shaped stitches and non-absorbable running sutures (Fig. 2E-H).

The immediate postoperative period was complicated by multiple reinterventions (Fig. 1). At 4 days after surgery, the patient developed severe nausea and vomiting. An upper gastrointestinal endoscopy revealed extensive necrosis of the gastric mucosa. Exploratory laparotomy demonstrated a markedly distended stomach without evidence of ischemia or perforation. At 4 days after this reoperation, a deglobulization decrease in hemoglobin levels prompted a thoraco-abdominopelvic CT scan, which revealed hemoperitoneum. An emergency laparotomy was therefore indicated. No active bleeding source was identified. Careful exploration of the entire digestive tract revealed a pre-perforative area in the ileum. This lesion required resection of a 10-cm ileal segment, followed by the creation of a double-barrel ileostomy in the left iliac fossa. The patient did not have a recurrence of their diaphragmatic hernia, but experienced a relapse of their underlying disease 1 year later, which ultimately caused mortality.

Discussion

Delayed diaphragmatic hernia is a rare, but serious postoperative complication that can occur weeks to a range of 3 to 18 months after major cytoreductive surgery. In studies involving patients who underwent hyperthermic intraperitoneal chemotherapy (HIPEC), the interval is much shorter, 2 to 4 weeks (13,14). Among patients treated with bevacizumab, the delay ranges between 6 and 12 weeks (13,14). Similar to the case described in the present case report, the majority of cases reported in literature report signs of intestinal obstruction as initial symptoms (14-18). The respiratory failure experienced by this patient might be the result of a delayed diagnosis, leading to a more complex clinical presentation. Notably, to the best of our knowledge, only one similar case has been described in the literature, involving a delayed diagnosis and postoperative complications after hernia repair (15). This warrants particular attention because of its diagnostic complexity and potential therapeutic consequences (15). Clinical presentation may be non-specific at first: Vomiting and non-specific abdominal discomfort, before progressing to more pronounced gastrointestinal or respiratory symptoms, including hemodynamic instability. Several differential diagnoses should be considered in such cases: Pleural effusion, pneumonia or pneumothorax. In the present case, chest imaging initially suggested a left pneumothorax (Fig. 2C and D), the patient presented with hypercapnic respiratory failure.

The initial CT scan demonstrated gastric migration into the thoracic cavity; however, this finding was interpreted as a subocclusive syndrome with marked gastric distension rather than a diaphragmatic hernia. Because the patient showed clinical improvement under conservative medical management, the index of suspicion for diaphragmatic hernia was low, and the possibility of a hernia was not reconsidered during follow-up. This diagnostic oversight illustrates the intrinsic difficulty in recognizing this rare complication, which can closely mimic more common postoperative conditions, such as a bowel obstruction, as aforementioned in the present case report. The case therefore highlights the importance of maintaining a high index of suspicion and contributes considerably to the existing literature by reinforcing the need for heightened diagnostic vigilance.

Awareness of this condition is essential for early recognition and intervention, before the onset of severe complications (19). CT is considered the gold standard for diagnosis, as it allows precise evaluation of the size, location and contents of the diaphragmatic defect (20). Despite its limitations, standard chest X-ray remains a reasonable screening tool. The most common radiographic finding is an opacity of the hemithorax, often associated with dilated bowel loops and the presence of intrathoracic air-fluid levels (21).

Surgery is the treatment of choice in symptomatic patients. For small diaphragmatic defects, primary closure with non-absorbable sutures is the fundamental principle. It is essential to ensure a tension-free closure with good vascularization of the margins. A synthetic or biological xenogenic mesh may be used depending on the size of the hernia orifice (20). Since the defect measured ~4 cm in diameter, tension free closure was possible without the use of synthetic mesh. Hernia defect closure was performed using a series of simple Vicryl 1 'U-shaped' stitch sutures.

The most common cause of delayed diaphragmatic hernia is partial or complete diaphragmatic resection during cytoreduction, particularly when combined with HIPEC.

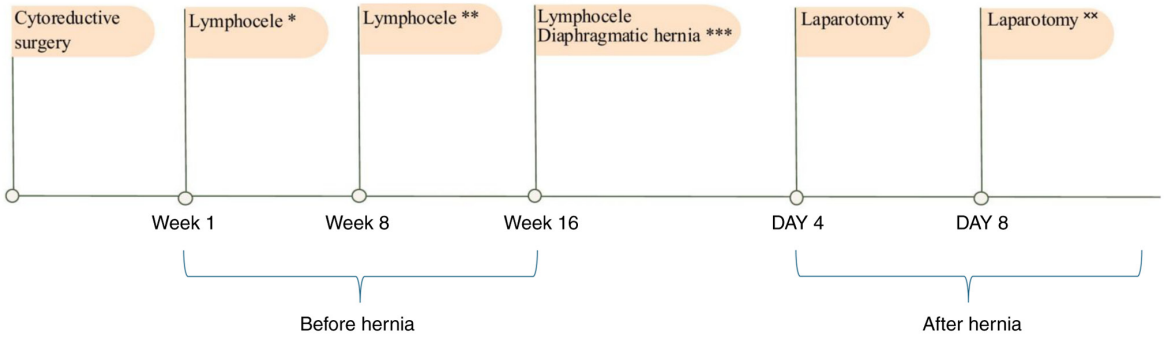

Figure 1. Timeline of the events. *Asymptomatic, discovered during CT scan; **asymptomatic, discovered on follow-up CT scan; ***persistent asymptomatic lymphocele, discovered during CT scan performed for abdominal pain; ˣsuspicion of gastric necrosis; ˣˣhemoperitoneum.

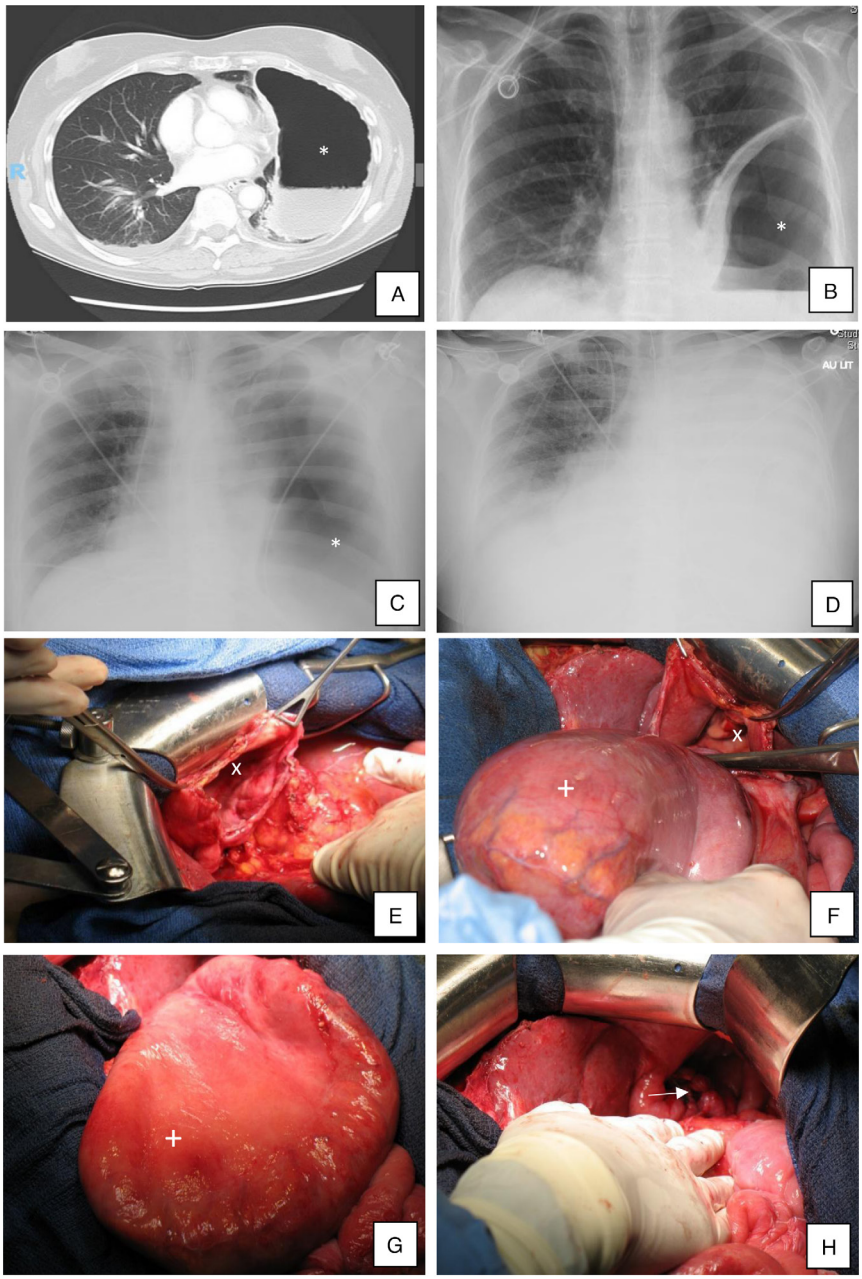

Figure 2. Preoperative imaging. (A) Computed tomography scan of the patient (day 1, at onset of sub-occlusive symptoms). (B) Chest X-ray of the patient (day 1, at onset of sub-occlusive symptoms). (C) Chest X-ray of the patient (day 4, with sub-occlusive symptoms; day 1 of multiorgan failure and hypercapnic respiratory failure). (D) Chest X-ray of the patient (day 5, with sub-occlusive symptoms; day 2 of multiorgan failure and hypercapnic respiratory failure). Intraoperative images of the patient's hernia. (E) Lateral view of the hernia orifice. (F) Inferior view of the hernia orifice. (G) Stomach. (H) Inferior view of the hernia orifice suture. *, gastric bubble in thorax; x, diaphragmatic defect; +, stomach; →, Hernia orifice suture.

Mechanical trauma from surgery, along with the thermal effects of HIPEC, can compromise the structural integrity of the diaphragm (13-17). Other contributing factors may include pharmacologic agents such as bevacizumab, an anti-angiogenic drug used in different types of gynecological cancer. By inhibiting neovascularization, bevacizumab may interfere with tissue regeneration and increase tissue fragility, thereby increasing the risk of diaphragmatic rupture (22-24).

In the present case, the patient developed a delayed diaphragmatic hernia without undergoing HIPEC or receiving bevacizumab, reported in only two previous cases in the literature (15,16). Adjuvant chemotherapy alone has rarely been associated with delayed hernia formation, although some reports suggest it could impair healing (10,15).

Risk factors for diaphragmatic hernia include advanced age, obesity, smoking, malnutrition and emergency surgery. It is also important to consider the prevalence of comorbidities that could lead to increased intra-abdominal pressure such as constipation and ascites (24). The present case study observed persistent lymphocele and, most importantly, malnutrition, which seemed to be the main risk factor. To the best of our knowledge, this has not yet been researched in the literature on post-cytoreductive diaphragmatic hernias.

To the best of our knowledge, this is the first reported case of delayed diaphragmatic hernia after cytoreductive surgery in a patient with a history of mediastinal large B-cell non-Hodgkin's lymphoma, treated with ACVBP. Although this case occurred in 2009, subsequent changes in surgical practice and perioperative management do not affect the interpretation of the present case report.

High-risk patients should undergo systematic nutritional assessment, optimization when needed and closer post-operative monitoring. In patients with atypical gastrointestinal or respiratory symptoms, maintaining a high index of suspicion and considering prompt imaging may facilitate early diagnosis and prevent severe complications.

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Availability of data and materials

The data generated in the present study may be requested from the corresponding author.

Authors' contributions

SG, HEH and PM contributed to the conception and design of the present case report. MS, SG and PP contributed to the acquisition of the data. MS, PP, SG, PM and HEH contributed to analysis and interpretation of data. MS, PP, SG, PM and HEH contributed to manuscript revision. All authors have read and approved the final manuscript. MS and HEH confirm the authenticity of all the raw data.

Ethics approval and consent to participate

The study conformed to the French ethical standards, and the 2008 Helsinki Declaration. According to our institutional policy, ethics committee approval is not required for case reports.

Patient consent for publication

The patient signed a consent form during their first consultation, agreeing to receive care and authorizing the use of their anonymized clinical data, including images and videos for medical, academic and research purposes.

Competing interests

The authors declare that they have no competing interests.

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