A rare case of diaphragmatic hernia after cytoreductive surgery and hypertermic intraperitoneal chemotherapy

Lorena Sorrentino, Francesca Cabry, Francesco Serra, Roberta Gelmini

Department of Surgery, Policlinico of Modena, University of Modena and Reggio-Emilia, Italy

Abstract

Cytoreductive surgery (CRS) combined with hyperthermic intraperitoneal chemotherapy (HIPEC) is an effective therapeutic approach for selected patients with gastrointestinal and gynecological malignancies with peritoneal spread. The most frequent postoperative surgical complications are anastomotic leakage, digestive perforations, fistulas, intestinal obstruction, abscess and peripancreatitis. This report presents case of a patient with late postoperative diaphragmatic hernia after CRS and HIPEC. A 50-year-old woman previously treated with CRS and HIPEC for a pseudomyxoma peritonei was admitted to our unit with diagnosis of intestinal obstruction. At the CT scan a left diaphragmatic hernia involving the splenocolic flexure was found. Both stripping of the diaphragmatic peritoneum during CRS, sometimes combined with diaphragmatic resection and the heat of HIPEC might be responsible for such complication. The diaphragmatic hernia is rarely diagnosed after CRS and HIPEC. Surgical techniques for repair can be the direct suture of the defect or closure with synthetic or biological tissue, both are possible surgical techniques for repair with a good long term results.

Correspondence: Roberta Gelmini, Department of Surgery, Policlinico di Modena, University of Modena e Reggio Emilia, Modena, Italy.
Tel.: +39.0594223662 - Fax: +39.0594222046.
E-mail: Roberta.Gelmini@unimore.it

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Introduction

Peritoneal carcinomatosis from colorectal and gynecological malignancy has been regarded in the past as a lethal clinical condition, and considered as an exclusion criteria for resective surgery. Sugarbaker in 1995 suggested a new strategy for treatment of established tumor implants within the abdominal cavity.[1] Cytoreductive approach including parietal and visceral peritonectomy combined with hyper thermic intraperitoneal chemotherapy (HIPEC) is an effective therapeutic approach for selected patients with gastrointestinal and gynecological malignancies with peritoneal spread. The aim of cytoreductive surgery (CRS) is to obtain complete macroscopic cytoreduction (CC-0/1) as a precondition for the application of HIPEC. The residual disease is classified intraoperatively using the completeness of cytoreduction (CCR) score. CCR-0 indicates no visible residual tumor and CCR-1 residual tumor nodules 2.5 mm, CCR-2 and CCR-3 indicate residual tumor nodules between 2.5 mm and 2.5 cm and equal or more than 2.5 cm, respectively. In case of CCR-0/1, HIPEC can be performed with open or closed abdomen technique.[2] A special pump is necessary to circulate the chemotherapy via several catheters placed into the peritoneal cavity. The intraperitoneal temperature is usually 41°C to 43°C. Several drugs can be used for HIPEC (as cisplatinum, mitomycin C, oxaliplatin, irinotecan, *etc.*).[3]

Pseudomyxoma peritonei (PMP) is a rare disease characterized by mucinous ascites, which results from a mucinous neoplasm often originating from the appendix. According to the *redistribution phenomenon*, mucinous tumor cells accumulate at specific sites as the abdominal cavity recesses, sparing the antimesenteric surface of the small bowel and to a lesser extent other parts of the gastrointestinal tract. Due to gravity and the peritoneal fluid absorption, tumor cells accumulate mainly by the greater and lesser omentum and the under-surface of the diaphragm, particularly on the right. The intraabdominal mucus can accumulate in the inguinal canal and by physical examination be indistinguishable from the usual inguinal hernia.[4] As previously said, CRS combined with HIPEC can be the optimal treatment for PMP. Total operating time for complete CRS and HIPEC for extensive PMP is around 10 h and generally involves bilateral parietal and diaphragmatic peritonectomies, right hemicolectomy, and radical greater omentectomy with splenectomy, cholecystectomy and liver capsulectomy, a pelvic peritonectomy with, or without, recto sigmoid resection and bilateral salpingo-oophorectomy with hysterectomy in females.[5]

The most frequent post-operative complications are anastomotic leakage, digestive perforations, fistulas and abscesses. In literature, only three cases of diaphragmatic hernia secondary to CRS are reported.[6,7]

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Case Report

A 50-year-old female patient was diagnosed with pseudomyxoma peritonei originating from the appendix in September 2015 when the patient underwent a total splenectomy after trauma. (Initial staging: pT2, pM1 [peritoneal], G1, Rx). CRS and HIPEC were performed in March 2016 including appendectomy, omentectomy, lesser omentectomy, peritonectomy of the right and left upper quadrant, peritonectomy of the Douglas pouch, cholecystectomy, and peritonectomy of the hepatoduodenal ligament obtaining a complete cytoreduction. She was admitted to the emergency room in June 2017 with symptoms of intestinal obstrucion. Standard X-ray of the abdomen was performed showing dilatation of both small bowel and colon with some air-fluid levels. A CT scan was performed, confirming the conspicuous enlargement of both small and large bowel, especially for the right ascending colon and transverse up to the splenic flexure where a clear transition point were visible. The patient was admitted to our surgical unit after two days of observation in an internal medicine ward. At a second X-ray the colic splenic flexure was clearly herniated in the thorax through a left hemi diaphragmatic defect (Figures 1 and 2). The patient underwent an exploratory laparotomy. Peritoneal washing and samples of peritoneal liquid were taken to exclude a relapse of pseudomyxoma. Adhesiolysis was performed during which a little blunt on small bowel was sutured but still with a little spillage of enteric material. The exploration of the upper left abdomen revealed the splenic flexure herniated through a defect in the posterolateral portion of the left diaphragm. Six inches of strangulated bowel was reducted in abdomen with adjoining mesentery. After reduction the colon was well vascularized with no signs of necrosis. Adhesions between colon and diaphragm were resected. The defect in the diaphragm was closed with non-absorbable polypropylene interrupted stiches. A thoracic drainage was positioned. The decision of using a direct repair technique of the diaphragm instead of using synthetic or biological tissue was made not only for the higher risk of infection and abscess given by the enteric spillage after adhesiolysis, but even after the possibility of disease relapse in the future. Postoperative constipation occurred with bilious vomit treated with prokinetic drugs with gradual relief. In 9th postoperative day a CT abdominal scan was performed because of hyperpyrexia with recent negative chest X-ray. A fluid sub Glisson abscess of 2x2 cm was found. The antibiotic therapy was upgraded from Piperacillin-tazobactam to Vancomycin for the next 5 days. The patient was discharged in 14th postoperative day in good health. A 2-month follow-up CT scan was performed, showing the complete resolution of the sub Glisson abscess. No transition obstacles both in large and small bowel and no relapse of diaphragmatic hernia were found (Figure 3).

Discussion and Conclusions

Regional intraperitoneal cancer spread required palliative systemic chemotherapy in the past. With Sugarbaker CRS and HIPEC became a new way of treatment and opened new horizon for some of these patients not without an increased morbidity. When possible a multimodal therapy concept can be employed, including surgery and chemotherapy. The aim is to perform the widest possible resection of the macroscopic tumor on one hand and destruction of the remaining microscopic tumor by using cytotoxic chemotherapeutic agents and hyperthermia on the other hand.

As shown in several studies complications are not unusual after CRS and HIPEC. Casado-Adam *et al*.[8] analyzed 147 patients with gastrointestinal complications after CRS and HIPEC. Their study showed an overall incidence of gastrointestinal events [grade I-IV, using the Common Toxicity Criteria (version 3.0) of the National Cancer Institute] of 17%. Complications of grade III and IV (intervention/operation necessary) accounted for 8%. Youssef *et al.* described postoperative in-hospital mortality accounted 1.6% and grade 3/4 morbidity of 7% in a cohort of 456 patients treated with CRS and HIPEC after pseudomyxoma peritonei.[9] According to the Kaplan-Meier statistical method, the 5 and 10 years overall survival predicted was of 69% and 57%, respectively. For 289 patients who had complete cytoreduction, 5 and 10-year predicted survival was 87% and 74% while for the 152 who had major tumor debulking, predicted survival was respectively 34% and 23%.[9] Many risk factors have been detected in several studies; Casado-Adams showed that peritoneal cancer index is a certain risk factor for gastrointestinal complications.[8] According to Younan *et al*. male sex (OR 4.2), no previous systemic chemotherapy (OR 3.5), and surgical time over 8.7 h (OR 6.3) are indipendent factors for bowel complications.[10] As Hansson suggested, even if CRS is associated with high morbidity and mortality, the potential benefit indicated by long-term survival, the adverse event from this treatment is considered acceptable.[11] Among all complications observed, most frequent events are anastomotic leakage, fistulas, abscesses and digestive perforations. Diaphragmatic hernia can be assumed as a very rare

Figure 1. Preoperative abdominal standard X-ray showing the left colonic flexure herniation and intestinal enlargement of small bowel and right colon.

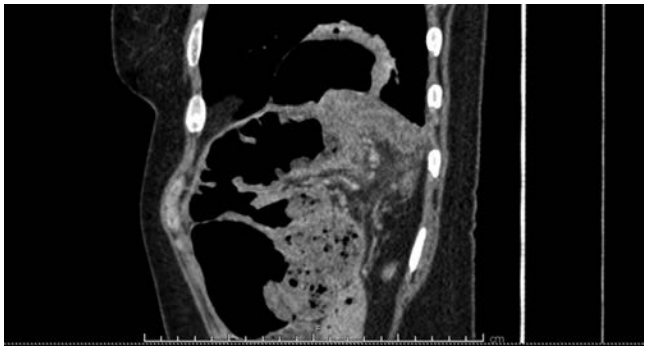

Figure 2. Preoperative abdominal computed tomographic scan showing the left diaphragmatic defect.

complication after CRS and HIPEC. The first description in literature of this type of complications comes in 2009 when Laterza *et al.* described a similar case of a 70-year-old patient diagnosed with malignant mesothelioma of the peritoneum. After CRS and HIPEC the patient developed a colo-bronchial fistula. The occurrence of this fistula was aided by a diaphragmatic defect.[6] In 2014 Lampl *et al*. described two cases of a 36-year-old patient diagnosed with pseudomyxoma peritonei and 65-year-old male patient with peritoneal carcinomatosis after gastric cancer. Both of the patient underwent total CRS and HIPEC. The 36-year-old patient was admitted to the hospital for surgical treatment of diaphragmatic hernia found out after a follow-up CT scan. The defect was closed by direct suture, no synthetic or biological xenogeneic material was used. The 65 years old patient was diagnosed with left-sided pneumonia 2 weeks after CRS and HIPEC and after a CT scan of the thorax and abdomen, a pleural abscess was detected above the left side of the diaphragm. After a CT-guided needle puncture revealing intestinal spillage, the patient was transferred to the OR. Surgery revealed an anastomotic leak of the left colon flexure with connection into the left hemithorax. The diaphragmatic defect was closed using a bovine pericardial patch.[7] A small-unnoticed lesion during diaphragmatic peritonectomy can cause such a defect. Furthermore, electrical power damage must be considered responsible when performing electro surgery for peritonectomy. Parietal peritonectomy in the area of the diaphragm is performed by monopolar cautery (with the ball-tip, 3 mm diameter, electrosurgical hand piece, at 100 W). Muscular tissue is partially removed during this procedure, leading to a thin muscular layer with possible adjacent lesions due to electrocoagulation and hyperthermia with subsequent defect formation. The cytotoxic effect of the HIPEC may contribute to prolonging the healing of the defect. In addition, the elevated abdominal pressure during HIPEC can str*etc*h the diaphragm and possibly foster the development of a hernia.

The treatment of choice for diaphragmatic hernia is surgery. While urgent surgery is frequently needed for the treatment of the symptomatic DHs, the surgical treatment of asymptomatic DHs may be performed days to years later according to patient's status. Small diaphragmatic defects are usually repaired by primary closure with non-absorbable sutures; Polypropylene 2.0 interrupted stiches is the best way to close small defect. It is important to perform a tension free closure with a good blood supply to the margins. If there is a large defect or muscle weakness, the synthetic or biological xenogeneic materials should be used because the primary repair could cause excessive tension. Because adhesions between the synthetic graft and organs can occur after operation, polypropylene mesh should be placed between the layers of the peritoneum, thus in patient that underwent CRS with total diaphragmatic peritonectomy polypropylene mesh should not be used.[12] A bovine pericardial patch can be an adequate option for hernia repair.[7]

When resection of infiltrated muscle layers are performed, prevention of later herniation can be assessed by a direct suture of the thinner areas. Lampl even suggested performing the resection with a linear stapler followed by sewing over the staple line.[7]

Although is a rare complication after CRS and HIPEC, diaphragmatic hernias should be considered especially when peritonectomy of the upper quadrants and HIPEC are performed. The absence of diaphragmatic resections do not reduce the risk of diaphragmatic complications. Optimal surgical repair is possible in both urgency and elective surgery with good long-term results, resolution of symptoms and improvement of life quality.

Figure 3. A) Post operative abdominal standard X-ray showing no relapse of left diaphragmatic hernia. B) Post operative abdominal computed tomographic scan showing no relapse of left diaphragmatic hernia.

Refereces

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