

Sclerosing Encapsulating Carcinomatous Peritonitis: A Case Report

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Abstract

Sclerosing encapsulating peritonitis (SEP) is a rare clinical condition characterized by the formation of a thick, fibrous membrane encasing the intestines, which may lead to intestinal obstruction. The pathogenesis is not completely understood, but various risk factors are well established. However, there are only few reported cases of SEP associated with peritoneal carcinomatosis. Herein, we report a case of a 69-year-old male patient who presented clinically with acute intestinal obstruction 2 years after undergoing a resection procedure for gastric cancer. An abdominal computed tomography revealed findings typical of SEP. Consequently, the patient underwent exploratory laparoscopy, which confirmed the diagnosis of SEP and established the etiology as peritoneal metastases. The patient was managed conservatively, and his symptoms showed some improvement. The patient was at an advanced stage of the disease, and thus remained on palliative care and passed away 1 month later. Although very rare, physicians should consider SEP in their differential diagnoses of intestinal obstruction in patients, particularly in those with a history of intra-abdominal malignancies.

Keywords: Gastric cancer, intestinal obstruction, peritoneal carcinomatosis, sclerosing encapsulating peritonitis

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Submitted: 22-Jul-2019 **Revised:** 14-Nov-2019 **Accepted:** 02-Jun-2020 **Published:** 13-Oct-2020

INTRODUCTION

Sclerosing encapsulating peritonitis (SEP) is an unusual etiology of intestinal obstruction characterized by a thick fibro-collagenous membrane encasing the small intestine and colon. This rare clinical entity might be idiopathic or secondary to an underlying condition. Among its various associated causes, peritoneal carcinomatosis is an extremely rare cause, with only a few cases having been reported in the literature.^[1-4]

Herein, we report the case of a 69-year-old male patient with a history of gastric cancer resection who presented with acute intestinal obstruction caused by SEP related to peritoneal metastases of gastric adenocarcinoma. To the best of our knowledge, gastric cancer metastasis to the peritoneum manifesting as SEP after curative resection of primary tumor has not been reported previously.

CASE REPORT

A 69-year-old male presented with a history of dysphagia

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How to cite this article: Alshomimi S, Hassan A, Faisal Z, Mohammed A, Al Dandan O, Alsaif HS. Sclerosing encapsulating carcinomatous peritonitis: A case report. Saudi J Med Med Sci 2021;9:63-6.

Access this article online	
Quick Response Code:	Website: www.sjmms.net
	DOI: 10.4103/sjmms.sjmms_275_19



Figure 1: Axial (a) and coronal (b) contrast-enhanced computed tomographic images showing small loops clustered to the center of the abdominal cavity encased by a thin membrane (arrowheads) and tethered to a central mass (M). Note massive ascites (*) and thick enhancing peritoneal lining (arrow)

and weight loss. Upper gastrointestinal endoscopy revealed a large ulcerative mass involving the gastric cardia, and its biopsy revealed signet-ring cells, suggestive of adenocarcinoma. A staging computed tomography (CT) scan revealed diffuse thickening of the gastric cardia with no enlarged lymph nodes or distant metastases. In addition, tumor markers (carcinoembryonic antigen and carbohydrate antigen 19–9) were within normal limits.

Based on these findings, a multidisciplinary oncology team decided to initiate neoadjuvant chemotherapy that aimed to downstage the tumor after evaluation using diagnostic laparoscopy. Because no evidence of metastatic disease was noted, three cycles of chemotherapy with cisplatin and capecitabine were administered. Repeated CT scan revealed disease regression; hence, surgical management was chosen.

The surgery started laparoscopically but was converted to open owing to dense gastric adhesions to the pancreatic body, which was suspicious for tumor invasion. On exploration, there was no evidence of peritoneal dissemination or distant metastasis. Because of the high body mass index and difficulty in the surgery, total gastrectomy with D1+ instead of D2 lymph node dissection and Roux-en-Y esophagojejunostomy was performed. Histopathological examination of the obtained specimen revealed negative margins and 36 disease-free lymph nodes. Adjuvant chemotherapy in the form of an XP regimen (six cycles of oral capecitabine [1000 mg/m²] twice daily on days 1–14 and intravenous cisplatin [80 mg/m²] on day 1, every 3 weeks) was initiated, and the patient had regular follow-up at the surgical clinic until 1 year after the surgery, with no evidence of disease recurrence.

Two years after the surgery, the patient presented to the emergency department with a 2-day history of abdominal pain, vomiting and constipation, presenting a clinical picture of intestinal obstruction. He reported having similar episodes over the last 2 months, which spontaneously resolved.

Physical examination suggested massive ascites. Laboratory test results, including those of tumor markers and acid-fast stain, were within normal limits. An abdominal CT scan revealed dilated small intestinal loops at the center of the abdomen surrounded by a thin membrane and tethered to a peritoneal mass [Figure 1a]. Furthermore, massive ascites exerted a mass effect on the intestinal loops and liver with diffuse thickening of the parietal and visceral peritoneum [Figure 1b]. Such radiological findings were typical for SEP.

Conservative management with intestinal rest and nasogastric decompression was initiated, which resulted in some improvement. An ultrasound-guided paracentesis was performed, which yielded around 4 L of clear fluid. On analysis, the fluid was negative for any organism, and the cytology results did not reveal abnormalities.

Because the findings on paracentesis were unremarkable and there was no evidence of local recurrence in the surgical bed based on the CT scan examination, the patient was prepared for diagnostic laparoscopy. On exploration, massive ascites was observed along with a thick, white membrane covering the entire small intestine, colon and liver. In addition, there were extensive adhesions within the intestinal loops and peritoneal nodules. Multiple biopsies were obtained from the encasing

Table 1: Types of sclerosing encapsulating peritonitis

Type	Description
1	Partial encapsulation of the small intestine
2	Complete encapsulation of the small intestine
3	Involvement of other organs such as appendix, colon or ovaries
4	Involvement of the entire peritoneal cavity (described in neuroendocrine tumors ^[4])

membrane and peritoneal nodules. Histopathological examination of the peritoneal nodules revealed a metastatic adenocarcinoma, and the resected membrane consisted of dense, hypocellular fibrocollagenous tissue. The tumor cells showed immunohistochemistry positivity for PanCK, CK7 and CK20 and partial positivity for epithelial membrane antigen.

In view of the advanced disease, no further intervention was planned. Our patient remained on palliative care until he passed away 1 month after his initial presentation to the emergency department.

DISCUSSION

SEP was first described by Owtschinnikow in 1907, who termed it as “peritonitis chronica fibrosa encapsulate.”^[5] In 1978, Foo *et al.*^[6] coined the term “abdominal cocoon syndrome” when describing the laparotomy findings of the condition. SEP is classified into different types based on the extent of involvement of the visceral organs by the sclerosing membrane [Table 1].

Although the exact pathogenesis of the condition is still unclear, various risk factors have been implicated. These include peritoneal dialysis, peritoneal shunts, infectious peritonitis, intraperitoneal chemotherapy, sarcoidosis and liver transplantation.^[7] It is presumed that the chronic peritoneal inflammation in these entities induces a fibrotic response, leading to the formation of a thick fibrous sheath around the bowel.^[8]

Malignancy is an extremely rare cause of SEP, with only a few cases being reported in the literature. Such a condition has been described in association with ovarian thecoma and carcinoma,^[9] gastric adenocarcinoma,^[1] pancreatic carcinoma,^[2] renal carcinoma^[4] and, more recently, with advanced midgut neuroendocrine tumors.^[4]

The first case of malignancy associated with SEP was reported in 1976 by Vorhauer *et al.*,^[1] which was due to gastric adenocarcinoma. This association was also reported by Yanagi *et al.*^[10] in a patient with early gastric adenocarcinoma. During laparotomy for distal gastrectomy, it was incidentally discovered that the intestinal loops were

encapsulated in membranes not typical of SEP. However, when the patient needed another laparotomy 2 months later for abdominal symptoms, typical SEP features, such as a thick fibrotic membrane, were present.^[10] In a case series published by Pamo Reyna *et al.*,^[11] there were two cases of gastric adenocarcinoma with SEP. Recently, the association was described again in a young female patient from a developing country. At first, it was suspected to be abdominal tuberculosis, but was later revealed to be SEP, which was the initial presentation of a metastatic gastric cancer.^[12] To the best of our knowledge, our case is the first reported case of SEP as a manifestation of metachronous metastatic gastric cancer.

Several factors may play a role in the development of SEP in patients with abdominal malignancies. For instance, it has been suggested that the mucin production and desmoplastic response in some tumors, such as in cases of signet cell gastric carcinoma, appear to be predisposing factors.^[12] SEP has also been reported to occur after hyperthermic intraperitoneal chemotherapy in a patient with advanced gastric cancer.^[13] Hyperthermic intraperitoneal chemotherapy has been shown to confer a higher rate of chemical peritonitis compared to normothermic chemotherapy among patients with gastric cancer treated with intraperitoneal infusion of mitomycin C.^[14] A clinical trial of the adjuvant use of intraperitoneal cisplatin and 5-fluorouracil in patients with resected gastric cancer showed that the occurrence of SEP was more common among patients who received more cycles of intraperitoneal chemotherapy. They proposed that the alkaline pH of the infused chemotherapy may play a role in the development of chemical peritonitis. In addition, in patients who had undergone cytoreductive surgery and extensive peritonectomy, the traumatized areas remaining without the parietal and visceral peritoneal layers may have developed an inflammatory response and exhibited a wound-healing process. This, in turn, can eventually cause fibrin deposition that may underlie the development of SEP in such patients.^[15] In our case, however, the patient did not receive any kind of intraperitoneal chemotherapy, and limited peritonectomy was performed. Esophagojejunal anastomotic leakage is a common complication following gastrectomy for gastric cancer.^[16] The leakage of luminal content from the surgical connection causes chemical peritonitis, which is another speculation regarding the development of SEP.

The clinical manifestation of SEP is vague and nonspecific, which makes preoperative diagnosis difficult. However,

with the widespread use of radiological imaging, the preoperative diagnosis is possible. CT scan is the diagnostic modality of choice, which may demonstrate clumping of small intestinal loops at the center of the abdomen, encased by a soft-tissue density mantle as well as peritoneal thickening and loculated fluid collections.^[17]

Some other conditions that may give a similar clinical and radiological picture of SEP include tuberculous peritonitis, peritoneal mesothelioma, pseudomyxoma peritonei and congenital peritoneal encapsulation.^[17]

Surgical treatment involving excision of the fibrous membrane and adhesiolysis is the cornerstone in the management of SEP. However, the extensive fibrosis in cases associated with disseminated malignancy might make surgical management unfeasible and has a high complication rate.^[18]

In conclusion, SEP is a rare presentation of peritoneal metastases. Clinicians should have a high index of suspicion for this condition in patients with gastric cancer.

Peer review

This article was peer reviewed by one independent and anonymous reviewer.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient's son has given his consent for his father's images and other clinical information to be reported in the Journal. The patient's son understands that his father's name and initials will not be published, and due efforts will be made to conceal his identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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