CASE REPORT

Enterocutaneous fistula 3 years after resection of an advanced gallbladder carcinoma

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SUMMARY

A 71-year-old woman presented to the emergency department with abdominal pain and fever. Her surgical history was significant for gallbladder adenocarcinoma for which she had undergone extensive resection 32 months previously. At that time she underwent cholecystectomy, wedge resection of the liver, pancreatoduodenectomy, right nephrectomy and right hemicolectomy for a locally advanced gallbladder adenocarcinoma. Examination revealed a tender, warm, upper midline abdominal wall mass. A CT scan with oral contrast revealed a fistulous tract extending from the gastrojejunostomy (GJ) into an abscess cavity in the adjacent anterior abdominal wall. She underwent open wound drainage with debridement, and was started on parenteral nutrition and intravenous antibiotics. The patient then underwent surgical repair excision of the fistula and refashioning of the GJ 1 month later. Histological examination of the specimen revealed well-healed suture lines, and no evidence of tumour recurrence.

BACKGROUND

This is an unusually late presentation of a post-operative enterocutaneous fistula originating from the gastrojejunostomy (GJ) anastomosis line without evidence of tumour recurrence.

CASE PRESENTATION

A 71-year-old woman presented to the emergency department with abdominal pain and fever. Prior to presentation, she had not noticed any abdominal masses, and did not report nausea or vomiting. Her medical history was significant for hypertension, discoid lupus erythematosus and dyslipidaemia.

Her surgical history was significant for an extensive resection 32 months earlier for locally advanced poorly differentiated adenocarcinoma of the gallbladder extending to the liver, bile duct, kidney and colon (figure 1). At that time she underwent cholecystectomy, wedge resection of the liver, pancreatoduodenectomy, right nephrectomy and right hemicolectomy. A two-layered end-to-side, duct-to-mucosa pancreaticojejunostomy and a single-layer end-to-side duct-to-mucosa hepaticojejunostomy were performed. The side-to-side GJ consisted of a continuous 3–0 Vicryl (polyglactin 910) inner layer, and an outer layer of 3–0 silk Lembert sutures. The ileocolonic anastomosis was created in a side-to-end two-layered fashion.

Histopathological examination of the specimen revealed a 6.5 cm moderately to poorly differentiated gallbladder adenocarcinoma, with metastatic

Figure 1 Pre-resection abdominal CT scan showing the gallbladder tumour invading multiple abdominal viscera (A, axial view; B, coronal view).

carcinoma seen in one of seven nodes, with local extension to the liver, pancreas, bowel and right kidney. The margins were uninvolved; however, the venous and perineural invasion was present.

Postoperatively she developed a superficial surgical site infection that was treated with drainage, packing, antibiotics and subsequently negative pressure wound therapy for 8 weeks. At the time, a CT scan that showed no intra-abdominal source of infection and she did not develop an abscess or sepsis postoperatively. She underwent adjuvant chemotherapy with gemcitabine and chemoradiation with capecitabine and 5040 cGy in 180 cGy/day fractions.

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Physical examination revealed a fever of 38.6°C, heart rate 97 bpm and blood pressure 134/63 mm Hg. A well-healed laparotomy scar and a tender, erythematous warm swelling in the upper aspect of her scar were noted. No discharge was appreciated. Her physical examination was otherwise unremarkable.

INVESTIGATIONS

Basic haematological tests revealed a normal leucocyte count of 8000/ μ L (normal 4000–11 000/ μ L) and haemoglobin 10.7 g/dL (normal 12–15 g/dL). A CT scan with contrast revealed a fistulous tract extending directly from the GJ into a large (8.6×3.5 cm) anterior abdominal wall fluid collection. There was evidence of indurations, fat stranding and inflammatory changes throughout the adjacent anterior abdominal wall (figure 2).

DIFFERENTIAL DIAGNOSIS

- Malignant enterocutaneous fistulae (ECF) caused by recurrent gallbladder cancer

Figure 2 Abdominal CT scan showing enterocutaneous fistulae arising from the gastrojejunostomy with anterior abdominal wall collection (A, axial view; B, sagittal view).

- Radiation-induced ECF
- ECF caused by a perforated marginal ulcer

TREATMENT

The patient was admitted to the hospital, started on intravenous antibiotics and was then taken to the operating room (OR). In the OR the wound was opened, debrided and packed with sterile packing. An ostomy appliance was applied around the ECF skin opening, which was draining gastrointestinal (GI) contents. She was kept *nil per os* (NPO) and started on parenteral nutrition. One month later, following nutritional optimisation, she underwent an exploratory laparotomy with adhesiolysis, the takedown of the ECF and resection of the old GJ anastomosis from where the ECF had arisen. A Roux-en-Y GJ was then formed to reinstate GI continuity. The resultant abdominal wall defect was closed with the use of an anterior component separation technique.

OUTCOME AND FOLLOW-UP

Histological examination of the GJ specimen revealed small bowel with fistula formation, fibrosis, acute and chronic inflammation and focal ulceration, and stomach with oxyntic mucosa with chronic inflammation and reactive changes. The specimen containing the abdominal wall scar and fistula site revealed chronic inflammation without tumour recurrence. The patient was discharged home on a regular diet on post-operative day 11. At her 2-month postoperative appointment, she had a well-healing wound, without signs of infection or ECF recurrence.

DISCUSSION

Enterocutaneous fistulae (ECF) are a disease entity with a wide variety of aetiologies, but the majority (75–85%) occur following surgery.¹ Risk factors for the development of a postoperative ECF include the presence of infection, and breakdown of an intestinal anastomosis due to ischaemia, anastomotic tension or distal obstruction.² ECF that result from anastomotic breakdown present early, usually within the first postoperative week. Marginal ulceration is a known complication of any GI anastomosis and is seen in 0.6–16% of gastrojejunostomies.^{3–6} A perforated marginal ulcer can lead to peritonitis if associated with gross contamination of the peritoneal cavity and can also be present in a more insidious manner as an ECF or enteroenteric fistula (EEF).^{7–8}

In patients undergoing abdominal or pelvic radiation for cancer treatment, the incidence of radiation enteritis ranges from 2% to 5%.^{9–10} Only 1–2% of these patients will require an operation due to excessive bleeding, perforation, obstruction or fistulisation.¹¹ The intestinal compartments most frequently affected are lower ileum, caecum and rectosigmoid.¹²

Our patient presented with an ECF and a history of advanced, poorly differentiated adenocarcinoma of the gallbladder. Consequently recurrent malignancy disease causing a malignant fistula was foremost in our differential diagnosis, and had to be excluded. Fortunately, the pathological examination revealed no evidence of malignant recurrence. Given that the stomach is one of the less commonly involved organs in radiation enteritis and that the histopathology showed no evidence of such processes (ie, fibrosis, endarteritis and oedema), radiation-induced ECF was less likely in this case. Although the patient denied abdominal pain or any stigmata of upper GI bleeding, we believe that a small marginal ulcer penetrated through the abdominal wall and over time progressed to form an ECF.

Learning points

- ▶ An enterocutaneous fistulae (ECF) can occur for many reasons, the most common of which is surgery.
- ▶ An ECF can present several years after the inciting operation.
- ▶ It is of paramount importance to rule out tumour recurrence in patients with an ECF and a history of malignancy.
- ▶ Marginal ulcers are a cause of morbidity and mortality and can lead to ECF or enteroenteric fistula formation.

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Competing interests None.

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