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Upper GI bleeding associated with recurrent pseudomyxoma peritonei: An unusual case report

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Abstract

Pseudomyxoma Peritonei (PMP) presents commonly with symptoms of appendicitis and small bowel obstruction. It has not been reported to cause Upper Gastrointestinal Bleeding (UGIB). In this case report, we present a 69-year-old female who presented to the emergency department with symptoms of UGIB in the setting of recurrent PMP and suffered from subsequent hemorrhagic shock. In addition, there were multiple duodenal ulcers seen during endoscopy. She underwent multiple upper gastrointestinal endoscopy and interventional radiology attempts to control her bleeding but eventually required an urgent open exploratory laparotomy to control the bleeding. We discussed the potential mechanisms for UGIB in PMP and the role of surgical intervention in such cases.

Keywords

Pseudomyxoma peritonei; Upper gastrointestinal bleeding; Duodenal ulcers.

Introduction

Pseudomyxoma Peritonei (PMP) is a rare clinical syndrome characterized by accumulation of diffuse gelatinous fluid collections with mucinous implants on peritoneal surfaces and the omentum [1]. PMP most commonly originates from the appendix as a consequence of a perforated appendiceal mucinous neoplasm, although other sites of origin have been described [2]. It usually presents with gastrointestinal symptoms such as those of appendicitis and small bowel obstruction. It might also present with systemic symptoms including anorexia and weight loss [3,4]. Duodenal ulceration and upper gastrointestinal bleeding (UGIB) have not been previously reported in association with PMP.

The current standard treatment is a combination of cytoreductive surgery and heated intraperitoneal chemotherapy (HIPEC) [5]. We present a case of recurrent PMP associated with an unexpected massive UGIB and hemorrhagic shock which required interventional radiology and endoscopic interventions

initially and ultimately urgent laparotomy for definitive control.

Case Presentation

A 69-year-old female with a known history of PMP underwent cytoreductive surgery and heated intraperitoneal chemotherapy 12 years before the current presentation. Two years ago, the patient was found to have recurrence on CT scan but was asymptomatic at the time. However, over the past year she started having continuous episodes of severe crampy abdominal pain and transient partial bowel obstructions that she self-managed with liquid diet.

In the current encounter, she presented to the emergency department complaining of melena and anemic symptoms. She was admitted and repeat abdominal CT scan showed disseminated multiple cystic lesions with internal calcifications throughout the abdomen including the area around the duodenum, porta hepatis, and the lesser omentum. However, there was no tumor around the stomach and other parts of the small bowel (Figure 1).

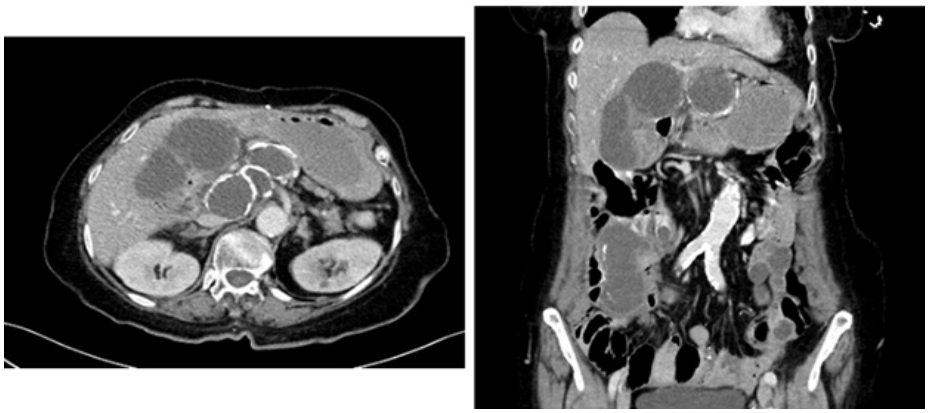

Figure 1: These Computed Tomography images show mucinous masses adjacent to the duodenum and in the right lower quadrant.

Esophagogastroduodenoscopy (EGD) showed gastritis, nodular mucosa in the second part of the duodenum, and three non-bleeding duodenal ulcers. Biopsies showed peptic duodenitis without dysplasia and chronic, inactive gastritis without helicobacter pylori organisms.

The patient continued to have melena which was associated with severe hypotension and hemoglobin that dropped to 4.8 g/dl which required ICU admission, resuscitation and blood transfusion. Subsequently, the patient underwent mesenteric angiography that showed no active extravasation or vascular anomalies, but embolization of the gastroduodenal artery was performed to control the recurrent massive bleeding. Follow up EGD showed three non-bleeding cratered duodenal ulcers with a clean base, two of them were in the duodenal bulb and the third in the second part of the duodenum.

The patient remained stable thereafter until three days later when she had massive hematemesis. She underwent another EGD and one of the ulcers in the postero-lateral aspect of the duodenal bulb was oozing intermittently with a visible vessel which was controlled by epinephrine injection and clipping. The patient stabilized but due to concern for recurrent bleeding from extrinsic pressure of tumor on the duodenum, she underwent exploratory laparotomy which showed hard well-circumscribed masses along

the duodenum in the lesser sac. These masses were opened revealing thick mucinous material compatible with PMP. The decision was made not to proceed with duodenotomy or antrectomy due to increased risk of leak due to abnormal mucinous tissues around the areas and extensive adhesions. In addition, there were multiple hard masses in different areas of the abdomen including adjacent to the ascending colon which were also debulked. There was no injury made to the duodenum while debulking.

Histopathological examination from the periduodenal masses showed extracellular mucin with focal dystrophic calcification, compatible with PMP. In addition, there was organized and partially hyalinized granulation tissue containing extracellular mucin with few strips of neoplastic epithelium.

Postoperatively, the patient was stable and did not develop further UGIB. She developed bilateral lower extremity DVTs which required an IVC filter and heparinization that was later switched to Apixaban. Despite receiving therapeutic anti-coagulation, she did not have any further bleeding.

Discussion

This is an interesting case of recurrent UGIB from duodenal ulcers in a patient with recurrent PMP. The exact mechanism of UGIB in this condition is difficult to ascertain but H.pylori was negative. We hypothesize that the tumor masses adjacent to the duodenum were causing pressure on the duodenal wall which may have contributed to ulcerations and recurrent bleeding. In support of this, UGIB has been reported as a result of extrinsic compression on the esophagus and stomach [6]. In addition, in another case report, melena and mucosal ulceration were reported in association with external compression on the cecum by diffuse large B-cell lymphoma of the colon [7].

Another possible contributing factor in our patient is the recurrent episodes of partial small bowel obstruction which she had over several years before her current admission. Bowel obstruction leads to a rise in intra-duodenal pressure and distention, which subsequently impairs intramural blood flow and results in duodenal ischemia and necrosis. This concept has been suggested in a case report of acute gastric dilatation that caused gastric necrosis and perforation [8]. It has also been reported as a contributing factor in severe UGIB in the setting of small bowel obstruction caused by Superior Mesenteric Artery Syndrome [9].

In this case, multiple endoscopic and IR procedures were used to control the bleeding, but she required surgical debulking of the peri duodenal masses for definitive management. In our case, the decision was made not to proceed with duodenotomy or antrectomy due to increased risk of leak due to abnormal mucinous tissues around the areas and extensive adhesions. However, we suggest that even when duodenotomy, vessel ligation, and other extensive approaches like ulcer excision or bowel resection are possible, it might be enough to debulk the tumor masses around the duodenum in order to stop the bleeding.

Conclusion

In conclusion, herein, we present the first case of UGIB and duodenal ulceration due to recurrent PMP. Furthermore, when bleeding fails to be controlled by less invasive procedures such as endoscopy and IR, surgical exploration is warranted but debulking instead of a more invasive procedure like duodenotomy

may be sufficient to control the bleeding. This principle could be applied to other forms of extrinsic compression associated with UGIB.

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