CASE REPORT

Inflammatory fibroid polyp (Vanek's tumour): my worst nightmare in the emergency department

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SUMMARY

A middle-aged woman presented in shock with bleeding per rectum for 2 days without any significant past medical or surgical illness. Abdominal sonography, oesophagogastroduodenoscopy and sigmoidocolonoscopy were unremarkable. Despite 6 units of blood transfusion, 3000 mL of plasma expanders and ionotropes, the arterial pressure gradually decreased. We had no choice but to undergo life-threatening yet possibly life saving laparotomy. We found a 1×1 cm ileal bleeding mucosal polyp about 20 cm away from the ileocaecal junction. We resected and anastomosed end to end. Perioperatively, the patient was transfused with another 4 units of blood product. Immunohistopathology revealed a large number of CD34 positive spindle cells and eosinophils with extensive thick-walled capillaries compatible with inflammatory fibroid polyp. This case of Vanek's tumour is reported for its atypical site, which presented as massive obscure gastrointestinal haemorrhage without any prior warning signs.

BACKGROUND

Inflammatory fibroid polyp (IFP) is a rare benign lesion of the gastrointestinal (GI) tract, first described by Vanek, in 1949, as submucosal granuloma with eosinophilic infiltration.¹ The most common sites of affection are the gastric antrum (70%) and small intestine (20%).² The large bowel and oesophagus are uncommon sites of affection, rarely reported.² There are no definite signs and symptoms of IFP. The clinical picture depends on the location and size of the tumour. They present with dull aching abdominal pain, dyspepsia, intestinal obstruction and, rarely, massive GI bleeding. We report a case of ileal IFP that presented with near fatal obscure GI bleeding without any prior warning signs.

CASE PRESENTATION

A 45-year-old woman was hospitalised at another centre at 10:00 AM with bleeding per rectum for the last 2 days. Initially, the blood was mixed with faeces followed by frank bleeding per rectum. The patient had no significant medical or surgical history in the past. She had attained menopause 2 years prior. She had used oral contraceptive pills for 5 years. The physical examination revealed temperature of 37.2°C, blood pressure (BP) of 80/60 mm Hg, low volume pulse of 126/min and respiratory rate of 20/min. The patient was pale with no icterus and no cyanosis. Cardiovascular, respiratory and abdominal examination did not reveal any significant findings. Per rectal digital

examination revealed fibrosed external haemorrhoids at the 7 o'clock position. Routine haemogram showed haemoglobin 5 g/dL, total leucocyte count 11 000/cmm, total platelet count 138 000/cmm with no malignant cells and no haemoparasites. Serum creatinine was 1.6 mg/dL, blood urea nitrogen 22 and prothrombin time for test was 19.8, for control 10.4 and for international normalised ratio (INR), 1.84. Stool routine examination showed plenty of RBCs with scanty faecal matter, with no pus cells and no parasites. Both oesophagogastroduodenoscopy and sigmoidocolonoscopy were unremarkable. The patient was treated with intravenous-pantoprazole, tranexamic acid and 3000 mL of crystalloid. She continued to bleed per rectally and the systolic blood pressure dropped to 60 mm Hg. She was shifted to the intensive care unit (ICU), started on vasopressors and transfused with 6 units of fresh whole blood over 12 h. All efforts were futile and by evening the patient complained of retrosternal chest pain with increased perspiration. ECG showed 1.5 mm elevation of ST segment in V1–V4, suggesting antero-septal ischaemia. Then the patient was referred to our hospital.

We received the patient in the emergency department at 1:00 AM lying in a pool of blood. She was severely pale, disoriented and sweating profusely. The monitor showed a heart rate of 160/min with occasional ventricular premature complexes with barely palpable pulse. Manual blood pressure measurement showed systolic blood pressure of 64 mm Hg. Bedside sonography did not reveal any gross abnormality. We were almost sure the site of bleed was in the small intestine, as recent oesophagogastroduodenoscopy and sigmoidocolonoscopy could not pick up any lesion, and the chest pain was probably because of inappropriate coronary circulation because of overall hypovolaemia. We thought of many possibilities for the massive obscure gastrointestinal bleed (vascular causes, tumour, ruptured Meckel's diverticulum, mesenteric ischaemia), but nothing was fitting in this case. We had no choice other than an extremely high risk emergency exploratory laparotomy.

TREATMENT

Within 45 min, the patient was shifted to the operation theatre. Two peripheral lines and one central line were used for continuous infusion of crystalloids and blood products. A mid-midline incision was made. The liver, spleen and other organs were normal, and there was no peritoneal collection. Terminal ileum and colon were discoloured due to

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Figure 1 Intraoperative distal ileum showing actively bleeding polyp after enterotomy.

intraluminal collection of blood. The stomach and rest of the small intestine appeared normal in colour. On milking the small intestine from the duodenojejunal junction towards the ileocaecal junction, we could feel a small intraluminal polyp in the terminal ileum, around 20 cm proximal to the ileocaecal junction. An enterotomy was performed directly over the palpable tumour, which revealed an actively bleeding small intestinal polyp of 1×1 cm (figure 1). Limited resection of the small intestine was carried out with a 1 cm healthy margin. Double layer end-to-end anastomosis of the affected ileal segment was performed and the abdomen was closed layer-wise. Perioperatively, we transfused 4 units of whole blood and two packs of fresh frozen plasma. Postoperatively, we transfused another 4 units of whole blood. The patient was allowed clear liquids orally after 36 h.

OUTCOME AND FOLLOW-UP

The patient's general condition gradually improved postoperatively and she was discharged on the eighth postoperative day, without any complication. Histopathological evaluation of the ileal polyp revealed variable cellularity with a large number of spindle cells, lymphocytes, plasma cells and eosinophils. There were extensive thick-walled capillary vessels compatible with IFP (figures 2 and 3). On immunohistochemistry, the spindle cells were positive for CD34 and negative for CD117,

Figure 2 Photomicrograph shows a submucosal lesion in the small intestine, composed of whorls of plump spindle cells in a myxoid background (H&E, ×40).

Figure 3 Photomicrograph shows oval to spindle cells with inflammatory infiltrate rich in eosinophils (H&E, ×40).

cytokeratins and S-100. The morphological features and the immunohistochemistry were consistent with the diagnosis.

DISCUSSION

IFPs are very uncommon benign submucosal lesions of the GI tract.¹ The first case was reported in 1949.¹ The most common histopathological description is submucosal granuloma associated with eosinophilic infiltration.¹ Small bowel lesions usually present with episodic abdominal pain, obstruction due to intussusception and lower GI bleeding.³ The first case of obscure GI bleeding from an IFP in the ileum was reported in 1992, but that case was found to be associated with Crohn's disease.⁴ Though there are some case reports of GI bleeding from IFPs, they were located in the stomach. Histologically, IFPs appear as proliferation of fibroblasts with plenty of eosinophilic and lymphocytic inflammatory response, with increased vascularity.⁵ Although the term IFP was introduced by Helvis and Renier to indicate the non-neoplastic nature of this lesion, recent studies suggest that these lesions should be considered as platelet derived growth factor driven benign neoplasms.^{6,7} Surgical excision with a healthy margin of 1–2 cm is sufficient and curative treatment. The laparoscopic approach is still controversial due to the high risk of tumour rupture or bleeding, which can result in peritoneal spreading.⁸ Uncomplicated gastric IFPs are usually managed by endoscopic submucosal dissection.⁹

Learning points

- Obscure massive gastrointestinal (GI) bleeding is a surgical emergency.
- Vanek's tumour may present with massive GI bleeding without prior warning symptoms. Though the antrum of the stomach is the most common site, the distal ileum can also be affected.
- Midline exploratory laparotomy should always be preferred in inconclusive aetiologies rather than the time consuming laparoscopic approach.
- Severe hypovolemia may present as anginal chest pain.

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REFERENCES

- 1 Vanek J. Gastric submucosal granuloma with eosinophilia infiltration. *Am J Pathol* 1949;25:397–411.
- 2 Blackshaw AJ, Lewison RA. Eosinophilic infiltrates of the gastrointestinal tract. *J Clin Pathol* 1986;39:1–7.
- 3 Stolte M, Finkenzeller G. Inflammatory fibroid polyp of the stomach. *Endoscopy* 1990;22:203–7.
- 4 Catalá M, Sánchez A, Hinojosa J, *et al.* An inflammatory fibroid polyp (eosinophilic granuloma) and Crohn's disease located in the ileum. *Rev Esp Enferm Dig* 1992;82:193–5.
- 5 Navas-Palacios JJ, Colina-Ruizdelgado F, Sanchez-Larrea MD, *et al.* Inflammatory fibroid polyps of the gastrointestinal tract. An immunohistochemical and electron microscopic study. *Cancer* 1983;51:1682–90.
- 6 Schildaus HU, Cavalar T, Binot E, *et al.* Inflammatory fibroid polyps harbour mutations in platelet-derived growth factor receptor alpha (PDGFRA) gene. *J Pathol* 2008;216:176–82.
- 7 Lasota J, Wang ZF, Sobin LH. Gain-of-function PDGFRA mutations, earlier reported in gastrointestinal stromal tumors, are common in small intestinal inflammatory fibroid polyps. A study of 60 cases. *Mod pathol* 2009;22:1049–56.
- 8 Li CJ, Huang MT, Chen CS, *et al.* Application of laparoscopic techniques for resection of individual gastric submucosal tumors. *Surg Laparosc Endosc Percutan Tech* 2007;17:425–9.
- 9 Abboud B. Vanek's tumor of the small bowel in adults. *World J Gastroenterol* 2015;21:4802–8.

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