CASE REPORT

Pouch pseudoaneurysm: a rare cause of recurrent haemorrhage following ileal pouch-anal anastomosis

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SUMMARY

Bleeding following restorative proctocolectomy for chronic ulcerative colitis is an uncommon complication. Usually bleeding occurs from the ileal pouch-anal anastomotic site in the early postoperative period. Significant pouch bleeding in the late postoperative course is rarely encountered. A mesenteric artery pseudoaneurysm along the ileal pouch staple line may be a cause of late-presenting massive per-rectal bleed. Although a rare phenomenon, it must be considered to be a differential in such a situation.

BACKGROUND

Total proctocolectomy with ileal pouch-anal anastomosis (IPAA) is a well-established surgical treatment for chronic ulcerative colitis (CUC). Pouch bleeding is one of the uncommon presentations of this procedure. It has been cited in 1.5–3.8% of the cases, a majority of which are reported in the early postoperative period.^{1 2} Common causes of the postoperative bleed are suture line dehiscence, pouchitis or pouch ischaemia. Here, we present a unique case of a mesenteric arterial pseudoaneurysm developing in the ileal pouch and resulting in a delayed, massive per-anal bleed. To the best of our knowledge, a pseudoaneurysm causing pouch bleed has never been reported in the world literature.

CASE PRESENTATION

A 24-year-old boy was referred to us from a tertiary hospital with a diagnosis of refractory ulcerative colitis. He was diagnosed with left-sided ulcerative colitis 3 years back and was on medical therapy since then. However, his symptoms were poorly controlled even with high-dose mesalazine and parenteral corticosteroids. He had never received immunosuppressant therapy or biological agents for his condition because of financial constraints.

Following admission, sigmoidoscopy with minimal air insufflation was performed at our centre, which revealed inflamed and friable rectal and colonic mucosa until splenic flexure. In view of refractory disease, he was planned for definitive surgery. He underwent laparoscopy-assisted total proctocolectomy with stapled ileal 'J' pouch-anal anastomosis with covering ileostomy. The postoperative course was uneventful, and he was discharged on postoperative day 7. He remained well in the follow-up. Ileostomy was closed after 3 months of index surgery, and the patient was discharged in a stable condition. He revisited the

emergency department after 20 days of ileostomy closure with symptoms of intermittent fresh per anal bleed for the past 6 days. On examination, the patient was pale and had tachycardia. Per-anal examination revealed an ileal reservoir filled with bright red blood. He was admitted, resuscitated and transfused with 3 units of packed cells. After resuscitation, the pouch was irrigated with cold saline with epinephrine (1:100 000) and clots evacuated. Pouchoscopy examination was performed, which revealed a pulsatile polypoidal swelling at the lateral wall of the pouch 3 cm above the anal verge along with few oozing spots at the posterior wall at the suture line (figure 1). A CT angiogram revealed a 2 cm round structure adjacent to the anastomosis with fluid and high density contrast that 'washed out' on delayed imaging consistent with a pseudoaneurysm arising from a branch of the superior mesenteric artery (figure 2).

DIFFERENTIAL DIAGNOSIS

Pouchitis, staple line bleed.

TREATMENT

The patient was taken to the operation theatre where bipolar ligation of the pseudoaneurysm and re-enforcement of the suture line were performed by the transanal approach under general anaesthesia.

OUTCOME AND FOLLOW-UP

The postoperative course was uneventful, and the patient was discharged after 7 days of observation. He was followed-up subsequently for 3 years and has not reported any per anal bleeding until now.

DISCUSSION

IPAA is the most attractive continence-preserving surgical alternative for management of CUC. Recent surgical advances have made total or assisted-laparoscopic proctocolectomy with IPAA feasible and safe. The reported mortality rates range between 0 and 1%. However, morbidity following IPAA remains relatively high. In early studies, the reported incidence of major complications was as high as 54%, but with increasing experience, it has declined to ~19%.³

In a series of 3195 patients undergoing IPAA, pouch haemorrhage was reported in 47 (1.5%) cases. The majority (66%) of the bleeding occurred within 7 days of surgery.¹ Factors contributing to early pouch bleeding include inadequate haemostasis,

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Figure 1 On pouchoscopy, a pulsatile polypoidal mass was seen over the lateral wall of the pouch, 3 cm above the anal verge.

misfired staples, underlying bleeding disorders and the use of anticoagulants in the postoperative period. Studies have shown that the stapled technique of IPAA may be associated with a slightly higher rate of anastomotic bleeding, as these stapling devices are not haemostatic.^{4 5} Conservative management with pouchoscopy and epinephrine enemas is usually effective in most of these cases.¹ Late pouch bleed, though rare, is usually secondary to more sinister causes. Non-specific ileal pouch inflammation (pouchitis), cuffitis and ileoanal anastomotic stricture are among the common causes of pouch bleed in the long term. These late complications may result in total pouch failure in 3.8–12.5% of cases, necessitating pouch excision.^{6 7}

Mesenteric artery pseudoaneurysm causing a delayed pouch bleed has not been reported in the literature until now. Such a pseudoaneurysm results from a tear in the vessel wall due to trauma with subsequent periarterial haematoma formation. This is unlike a true aneurysm that involves all layers of the arterial wall, each of which is thinned but intact.

Figure 2 CT angiogram showing a 2 cm round lesion adjacent to the pouch-anal anastomosis, consistent with a pseudoaneurysm arising from the branch of the superior mesenteric artery.

Our patient presented with severe pouch bleeding 3.5 months after total proctocolectomy with IPAA. He required hospitalisation and blood transfusions for this complication. Diagnosis in cases of a delayed pouch bleed requires a detailed history, routine laboratory investigations, including complete blood count, stool examination and pouch endoscopy.

Recurrent episodes of fresh pouch bleed in an otherwise asymptomatic individual (as in our case) may be an indication of a pseudoaneurysm in the pouch. On pouch endoscopy, a pseudoaneurysm may be identified as a pulsatile swelling with evidence of recent bleed. In such cases, CT angiography may prove very helpful in establishing the diagnosis and delineating the exact site of the pseudoaneurysm.

The mechanism of pouch pseudoaneurysm formation could not be ascertained in our patient; probably inclusion of a mesenteric vessel in the staple line might have led to this late complication. One of the fascinating points about this case is that the pseudoaneurysm did not manifest itself early after creation of the pouch, presumably when it would have formed along the staple line. It only manifested soon after the ileostomy closure 3 months later in the form of a recurrent pouch bleed. The authors speculate that the stretching of the pouch following the introduction of an effluent stream after ileostomy closure caused trauma to an established pseudoaneurysm that formed 3 months earlier at the time of pouch creation. For management of late pouch bleeding following IPAA, no standardised protocol is available in the literature. Treatment with epinephrine enema could prove effective in cases of a diffuse pouch bleed, while endoscopic cauterisation or clipping might be more effective when specific bleeding points could be identified.⁸ Until the early 1990s, the only treatment available for pseudoaneurysm was surgical ligation. Since then, ultrasound-guided thrombin injection, coil insertion and fibrin adhesives have been tried with variable success. Covered stents provide another option of excluding the pseudoaneurysm from the circulation. However, the size and rigidity of endostents preclude their placement in the distal tortuous branch and small vessel pseudoaneurysms.⁹

In our patient, the pseudoaneurysm was located close to the anal verge, making its successful management possible through the transanal route. Close follow-up of the patient is mandatory for early detection and management of any recurrence.

Learning points

- ▶ A pseudoaneurysm arising from the mesenteric artery may be a cause of delayed bleed following IPAA.
- ▶ CT angiography may be very useful and should be undertaken when more usual causes of pouch bleeding have been ruled out.
- ▶ Transanal ligation of the pseudoaneurysm may provide an effective remedy when the diagnosis is already established.

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