

Endoscopic Fistulotomy Heals a Y-Shaped Entero-Entero-Cutaneous Fistula

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ABSTRACT

Patients may develop fistulas due to Crohn's disease or as a postoperative complication after restorative proctocolectomy with ileal pouch anal anastomosis. Unfortunately, the treatment of fistulas can be challenging. The current standard of care may include medical therapy and/or surgical intervention. However, endoscopic treatment for postoperative pouch complications has emerged as a valid alternative option. We describe a case of persistent drainage from a Y-shaped entero-entero-cutaneous fistula that resolved after endoscopic fistulotomy with needle knife.

INTRODUCTION

Fistulas may form as a complication in patients with Crohn's disease, due to a cryptoglandular abscess, or postoperatively after bowel surgery such as ileal pouch anal anastomosis (IPAA). A tract develops connecting two epithelialized surfaces; for example, connecting the pouch to any adjacent hollow organ or to the skin. Fistulas can be difficult to heal and have persistent drainage, which causes significant morbidity. Medical treatment may involve long-term antibiotics, immunomodulators, and/or biologics, as well as hyperbaric oxygen. Surgical management with resection or revision of the pouch, fistulotomy, seton placement, or diversion of the fecal stream may be necessary. Previously, treatment options for patients who have failed medical therapy or are too high-risk for surgery were limited. In the past decade, endoscopic treatment of postoperative pouch complications like strictures and sinus tracts has been shown to be effective. As for endoscopic treatment of fistulas, injection of various substances such as adipose-derived mesenchymal stem cells, fibrin glue, doxycycline, and dextrose (50%) have been investigated with some success. Both over-the-scope clip (OTSC) systems and endoclips have also been used with moderate success to assist in endoscopic closure of the internal opening of the fistula.¹ Endoscopic fistulotomy via needle-knife therapy can be effective in healing an ileal pouch-to-pouch fistula.

CASE REPORT

A 59-year-old black man presented with a complicated past medical history including extensive ulcerative colitis (UC), for which he underwent total proctocolectomy with the creation of a J-pouch in 2008. His postoperative course was complicated by a pouch outlet stricture and a persistent presacral sinus. He received serial endoscopic balloon dilation for the treatment of the stricture. Endoscopic needle-knife sinusotomy was performed for the treatment of the presacral sinus.

The patient remained well for 2 years, but he subsequently developed a complex Y-shaped fistula that connected the skin surface to the tip of his J pouch and the pouch inlet (Figure 1). He had multiple hospital admissions and was treated with antibiotics, but computed tomography enterography did not show intraabdominal or intrapelvic abscess. He continued to have a low-grade fever and drainage from the cutaneous fistula on his abdomen, which

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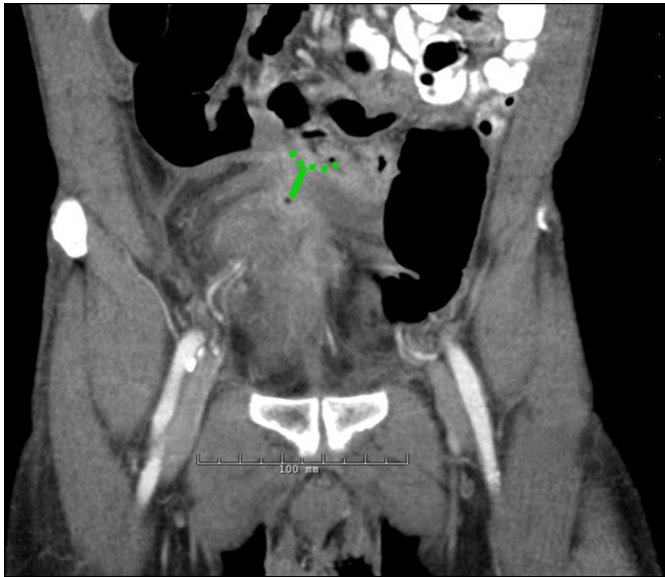


Figure 1. Computed tomography scan of the Y-shaped fistula between pouch lumen and skin. The dotted line indicates the pouch-to-pouch fistula that was later treated with endoscopic fistulotomy. The solid line indicates the cutaneous fistula linked to the pouch-to-pouch fistula.

required the use of a stoma bag. Diverting ileostomy with subsequent pouch excision was recommended, but the patient declined to have surgery. He was maintained on long-term ciprofloxacin and tinidazole.

An endoscopic approach was then attempted. The initial therapy to treat the fistula included spraying the tract with a solution of hydrogen peroxide, doxycycline, dextrose in water (50%), and betadine, followed by deployment of two size-12 over-the-scope clips (OTSC; Ovesco Endoscopy AG, Tübingen, Germany) at either end of the fistula tract openings within the pouch, one at the tip of the “J” area and the other at the pouch inlet. The patient did not experience symptom relief, and the cutaneous fistula persisted. When he returned for repeat pouchoscopy 2 weeks later, both OTSCs were found to be dislodged, and so they were removed (Figure 2). A flexible soft-tip guidewire was used to probe the fistula tract from the pouch inlet to the tip of the “J” (Figure 2). The fistula tract within the pouch was

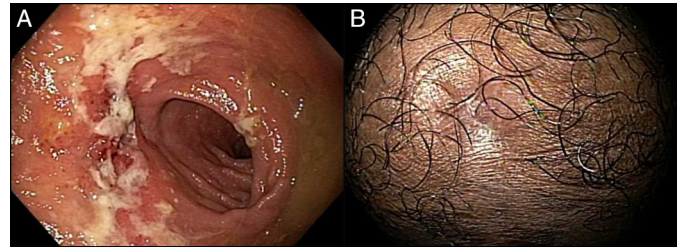


Figure 3. (A) Endoscopic image of the healed fistula two weeks after endoscopic fistulotomy. (B) Healed enterocutaneous fistula on the abdomen.

sprayed with betadine via an endoscopic spray catheter, and betadine was seen flowing out of cutaneous fistula opening. A complete fistulotomy was performed endoscopically using a needle knife (Boston Scientific, Natick, MA) with ERBE electrocauterization (ERBE USA Incorporated, Marietta, GA) on the spray setting to cut through the full thickness of the septum of the proximal pouch to the channel of the pouch-to-pouch fistula (Figure 2). Some purulent fluid was seen draining from the incised area. Hemoclips (Cook Medical, Bloomington, IN) were placed along the mucosal surface of the cut to prevent it from forming another tract as it healed (Figure 2).

The patient tolerated the outpatient procedure well and was discharged after being observed in the endoscopy recovery room for 30 minutes. A repeat pouchoscopy performed 2 weeks later showed an endoscopically normal pouch body and terminal ileum. The proximal pouch-to-pouch fistula was completely healed. Examination of his abdomen showed that the enterocutaneous fistula had also healed (Figure 3). Sustained resolution of the Y-shaped pouch-to-pouch-to-cutaneous fistula was evident on a subsequent pouchoscopy 6 weeks later. The patient tolerated all procedures well with minimal blood loss and no other serious complications.

DISCUSSION

The formation of fistulas can be a late complication of surgery.^{2,3} The patient likely developed the Y-shaped entero-entero-cutaneous fistula as a result of a postoperative complication of IPAA; it was unlikely to be caused by de novo

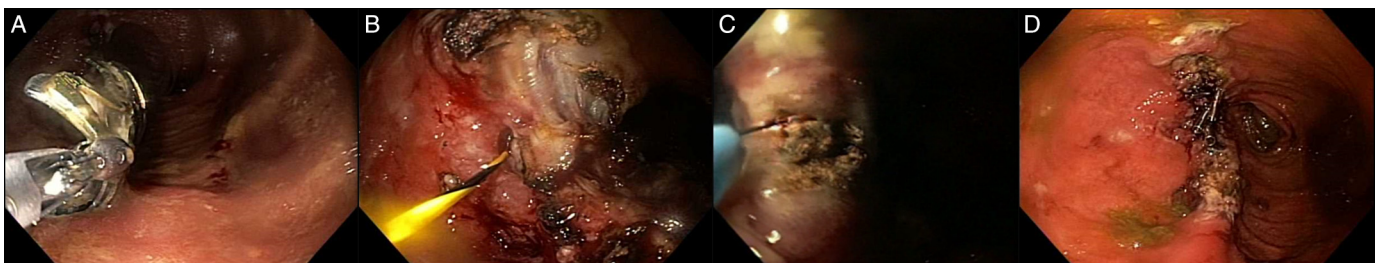


Figure 2. Endoscopic images of (A) the removal of the dislodged over-the-scope clip, (B) the flexible tip guidewire probing the pouch-to-pouch fistula tract, (C) fistulotomy using a needle knife on the ERBE spray setting, and (D) hemoclips placed after the pouch-to-pouch fistulotomy.

Crohn's disease, as he was noted to have completely normal mucosa of the pouch and afferent limb on his pouchoscopy. Given this, medical therapy with immunosuppressants or biological agents would not have been useful. He may have benefited from antibiotics if there was a concern for abscess or pouchitis, but there was no such concern in this case. The current standard of care for patients with an enterocutaneous fistula includes surgical intervention such as diverting the fecal stream, resection of the fistula, and closure of the internal defect.^{4,5}

In recent years, investigation of endoscopic therapy to manage postoperative complications of bowel surgery has yielded some positive results. Endoscopic needle-knife therapy has been used to successfully treat sinus tracts, mucosal bridges, and strictures that occur in the postoperative setting.^{1,6-9} Needle-knife therapy has also been used to perform a complete endoscopic fistulotomy for treatment of an ileal pouch-to-pouch fistula.¹⁰ We report complete resolution of a Y-shaped entero-entero-cutaneous fistula 2 weeks after partial fistulotomy with needle-knife therapy performed at the site of the ileal pouch-to-pouch fistula. Repeat pouchoscopy 8 weeks after the endoscopic fistulotomy showed sustained resolution of the fistula. This type of response to an endoscopic needle knife has not been described in the literature and will need further investigation. Endoscopic management of complications of inflammatory bowel disease, if performed by an experienced endoscopist, has emerged as an alternative option for select patients who are poor surgical candidates or would like to avoid surgery.³

DISCLOSURES

Author contributions: H. Lee wrote the manuscript. B. Shen edited the manuscript and is the article guarantor.

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Informed consent was obtained for this case report.

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