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Duodenal-Pleural Fistula in Crohn's Disease: Successful Long Term Medical Management

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To the editors:

A forty year old Caucasian male was diagnosed with Crohn's disease in 1981. Medical therapy was partially effective until 1987. In January of 1987, an upper endoscopy demonstrated narrowing of the pylorus. He remained on outpatient therapy until August of 1987 when his pyloric obstruction required treatment with a gastrojejunostomy. Changes consistent with mild Crohn's disease activity were observed on the serosal surfaces of the mid and distal small intestine.

A mesenteric abscess required inpatient antibiotic therapy in January 1988. Azathioprine 75 mg daily was added to a slowly tapering corticosteroid treatment. Inpatient treatment for pneumonia was required in August 1989. In December 1989, a jejunal resection (about 100cm) was necessary due to focal obstruction, a result of active Crohn's disease

Medical therapy was continued. Colonoscopy in March 1993 revealed an ascending colon inflammatory stricture as well as active Crohn's in the splenic flexure. A small bowel follow through revealed a stricture and fistula at the ileocecal valve and a second stricture in the mid-small bowel.

A mid-small bowel perforation and septic shock occurred in 1993. Emergency small bowel resection was performed with a two-layered end to end primary anastomosis. A colot-cutaneous fistula and intra-abdominal abscesses complicated the subsequent course, requiring several CT guided drainages. Two further laparotomies resulted in a right transverse end colostomy and long Hartmann's pouch. Repeat laparotomy was required soon thereafter because of necrosis at the colostomy and spillage of bowel contents into the abdominal cavity. In January 1994 an entero-cutaneous fistula in the vicinity of the gastrojejunal anastomosis developed.

Despite azathioprine at doses up to 125 mg daily, and low dose prednisone, symptomatic fistulae and a stricture of the colostomy necessitated repeat surgery in June 1994. The right colon and preexisting gastrojejunostomy were resected and a Roux-en-Y gastrojejunostomy created with an end ileostomy. A subsequent small bowel follow through revealed that contrast appeared in the excluded distal colon suggestive of an enterocolic fistula. Despite ongoing azathioprine and intermittent antibiotics between 1995 and 1997, a jejunal-colic

fistula, a complex colo-cutaneous fistula and an entero-enteric fistula developed as well. However, these were managed rather successfully in the outpatient setting.

In June of 1999 a right lower lobe pneumonia and empyema required hospital admission. A CT scan demonstrated the empyema and a fistula extending through the posterior aspect of the right diaphragm. A small bowel follow through exam revealed a fistula from the duodenum to the right chest (Figure 1). The pneumonia and abscess resolved with drainage and antibiotics. A PPD was borderline positive but serial sputum AFB stains were negative. The mesalamine was discontinued and the azathioprine dose was increased to 225 mg daily (2.5 mg/kg).

Three infliximab infusions were administered in October and early November 1999. Incisional fistula drainage resolved for the first time in at least three years. Azathioprine was continued. Recurrent drainage from the incisional fistulae as well as right lower quadrant discomfort was reported, however in late December 1999. These resolved with another infliximab treatment in early January 2000. Episodic infliximab infusions were planned.

A right empyema recurred in February of 2000. Treatment with chest tube drainage and antibiotics was again successful. Endoscopy was done attempting to identify the origin of the pleural fistula. A gastroenterostomy was seen and one small opening adjacent to the anastomosis was injected with contrast material under fluoroscopy but did not fill.

The pleural infection resolved, azathioprine was continued and regularly scheduled infliximab infusions were resumed. Recurrent drainage of the midline incision fistulae again resolved. Colonoscopy performed in April 2000 revealed diffuse pseudopolypoidosis, minimal active inflammation and a left transverse colon stricture, which could not be traversed, was found about 4 cm distal to the proximal extent of the Hartmann's pouch.

Infliximab infusions now have continued for 10 years every 6 to 7 weeks, along with azathioprine 225 mg daily. There has been no recurrence of empyema or cutaneous fistula drainage. The patient remains well, active and employed.

Whereas clinically significant duodenal Crohn's disease is unusual, Crohn's fistulae involving the duodenum are rare. Previously described duodenal fistulae have been entero-cutaneous or entero-entero, most arising from distal bowel, often from an ileocolic anastomosis, involving the duodenum secondarily. Treatment has generally been surgical.¹⁻³

Crohn's disease fistulae involving the thorax are very rare. A colo-pulmonic fistula⁴ and an esophageal-pulmonary fistula⁵ have been reported. A Crohn's disease duodenal-pleural fistula, however, has not previously been described.

Obstructing duodenal Crohn's disease was often successfully treated surgically in the pre-steroid era. As medical therapy evolved, some patients, certainly not all, could be managed non-operatively with corticosteroids, acid suppression, immunosuppressives and/or antibiotics^{1, 6}. More recently, infliximab has been used successfully to successfully treat obstructing duodenal Crohn's disease⁷.

The rarity of Crohn's duodenal fistulae has not allowed any conclusions to be drawn regarding treatment. Fistulae arising from the distal bowel have generally been successfully treated surgically^{2, 3}. Surgical treatment of two primary duodenal fistulae that failed corticosteroid treatment has been reported. Simple fistula closure was performed in one case and duodeno-jejunostomy in the other. Both were eventually successful, but complicated by

post-operative abscess^{1, 8}. Medical therapy for a primary duodenal fistula has not been previously described.

This case demonstrates: 1. a complication of Crohn's disease not previously reported i.e. a duodenal-pleural fistula and 2. the successful long term (>10 years) non-operative management of complex fistulizing Crohn's disease with the combination of thiopurine and anti-tumor necrosis factor medical therapy.

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FIGURE 1.
Small bowel follow through study with anterior/posterior (A) and lateral (B) views demonstrating a complex duodenal-pleural fistula.