

### GASTROCOLIC FISTULIZATION IN CROHN'S DISEASE: A CASE REPORT AND A REVIEW OF THE LITERATURE

M. Priya Khanna, MD; Philip H. Gordon, MD

Internal fistulization is a common complication of Crohn's disease and is characteristically manifested by enteroenteric, ileosigmoid, enterovaginal or enterovesical fistulas. Gastrocolic fistulas, however, are rare in Crohn's disease. Up to 1992, only 31 cases had been reported with no further case reports documented in the literature until 1997, when the first pediatric case was described.<sup>1</sup> We report an additional case of a patient who had a gastrocolic fistula secondary to Crohn's disease and review the literature on the subject.

#### CASE REPORT

A 21-year-old woman had symptoms of crampy abdominal pain, watery diarrhea and feculent vomiting for 1 week. Other clinical manifestations included weight loss over the previous year of 23 kg, a bitter taste in her mouth, a burning sensation in her chest and fecal halitosis. There was no history of fever, chills or bloody stools. An upper gastrointestinal series with small-bowel follow-through revealed mild dilatation of the terminal ileum and a "pinched area in the stomach." On flexible sigmoidoscopy no abnormality was seen.

On admission to hospital, she was afebrile and dehydrated. Her blood pressure was 90/60 mm Hg and her pulse rate was 110/min. Abdominal findings included mild distension, absent bowel sounds and no peritoneal signs. Digital rectal examination revealed green, liquid stool in the rectal ampulla that was negative for occult blood.

Her hemoglobin level was 11.0 g/L, blood urea nitrogen was elevated at 30 mmol/L urea, she had hypoalbuminemia and hypoproteinemia, and cultures of stool specimens were negative for bacteria, ova and parasites. An abdominal series revealed a largely gasless abdomen and a distended stomach. Colonoscopy was normal to 50 cm at which point the mucosa was edematous and friable and the lumen was narrowed. Barium enema examination demonstrated severe changes of granulomatous colitis involving the colon from the hepatic flexure to the sigmoid colon, along with a gastrocolic fistula. The rectum and right hemicolon were not involved. Gastroscopy presented a normal-appearing mucosa except for a puckered area at the site of the presumed attached fistula.

The patient's abdominal pain, vomiting and diarrhea resolved quickly and completely with intravenously administered steroids. She was placed on total parenteral nutrition for 2 weeks in preparation for operation. At laparotomy, the colon was affected with Crohn's disease from the hepatic flexure to the proximal sigmoid colon. There was marked induration and fibrosis in the area of the distal transverse colon where it was attached to the greater curvature of the stomach. The small bowel was normal. A subtotal colectomy was done with an ileosigmoid anastomosis and excision and primary repair of the gastrocolic fistula. The operation and postoperative course were uncomplicated, and the patient was discharged 8 days postoperatively. She remained asymptomatic for 11 years and had 2 uncomplicated pregnancies.

#### DISCUSSION

Historically, gastric fistulas to the colon tended to be of the gastrojejunal type and occurred in the context of marginal ulceration after gastroenterostomy for the treatment of peptic ulcer disease.<sup>2,3</sup> Gastrocolic fistulas unrelated to previous operations for duodenal ulcer are rare, the commonest causes being carcinomas of the stomach and transverse colon.<sup>2,3</sup> Other rare causes of gastrocolic fistulas include trauma, intra-abdominal and pancreatic abscesses,<sup>3</sup> tuberculosis, syphilis, pancreatic carcinoma,<sup>4</sup> renal carcinoma, gastric lymphoma,<sup>5</sup> diverticulitis<sup>6</sup> and carcinoid of the transverse colon.<sup>7</sup> Although the incidence of internal fistulization in Crohn's disease is estimated to be 20% to 35%,<sup>8,9</sup> the fistulas are rarely of the gastrocolic variety. Gastrocolic fistula as a complication of inflammatory bowel disease was first reported in 1937 by Bargen and associates from the Mayo Clinic.<sup>10</sup> At that time, the gastrocolic fistula was thought to be a complication of ulcerative colitis. However, that case in addition to 2 others of gastrocolic fistulas in 1939 and 1957 were reported at a time when granulomatous colitis was not yet considered a separate entity.<sup>11,12</sup> Crohn's disease involving the colon was first identified as a distinct entity in 1960 by Lockhart-Mummery and Morson,<sup>13</sup> so in retrospect it is likely that these fistulas were actually secondary to Crohn's disease. Since then, 29 cases have been reported (Table I<sup>1,3-5,8,10-12,14-29</sup>).

Development of a gastrocolic fistula secondary to Crohn's disease is thought to

*From the Division of Colon and Rectal Surgery, Sir Mortimer B. Davis-Jewish General Hospital and McGill University, Montreal, Que.*

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**Correspondence to:** Dr. Philip H. Gordon, 3755 Côte Ste-Catherine Rd., Montreal QC H3T 1E2; fax 514 340-7560

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**Table 1**

**Treatment and Outcome of Patients in Reported Series of Gastrocolic Fistula in Crohn's Disease**

Series	Age, yr	Sex	Treatment	Outcome
Bargen et al, 1937 <sup>10</sup>	22	M	Medical	No improvement with medical therapy, patient ultimately died
Ormandy and Bargen, 1939 <sup>11</sup>	31	M	Medical	Died 2 mo after admission; gastrocolic fistula diagnosed at autopsy
Altscheck and Summer, 1957 <sup>12</sup>	30	M	Total colectomy and ileostomy; repair of fistula	Uncomplicated recovery
Theony et al, 1960 <sup>14</sup>	NS	NS	2-stage procedure: stage 1—bypass ileosigmoidostomy, stage 2—resection of bypassed bowel and wedge resection of fistula	NS
Leitchling and Garlock, 1962 <sup>15</sup>	50	F	2-stage procedure: stage 1—bypass ileosigmoidostomy, stage 2—resection of bypassed bowel and wedge resection of fistula	Uncomplicated recovery, infrequent abdominal pain
Amlicke and Ponka, 1964 <sup>4</sup>	49	F	Partial colectomy	Died 3 yr postop of hepatitis
Cohen, 1967 <sup>16</sup>	22	M	Gastrojejunostomy and vagotomy, transverse colostomy	Uncomplicated recovery
Haggitt and Meissner, 1973 <sup>17</sup>	NS	NS	Partial gastrectomy and duodenectomy	Died 7 mo postop of acute gastrointestinal hemorrhage
Cody et al, 1975 <sup>3</sup>	29	F	Ileocolostomy and repair of fistula	Persistent primary disease
Wilk and Moqtaderi, 1976 <sup>8</sup>	60	F	2-stage procedure: stage 1—subtotal colectomy and ileostomy, sigmoid mucous fistula, wedge resection of fistula, stage 2—ileosigmoidostomy	Uncomplicated recovery
Metzgher and Ranganath, 1976 <sup>18</sup>	62	M	En bloc resection of terminal ileum, right and transverse colon and antrum; truncal vagotomy	Uncomplicated recovery
Laifer et al, 1977 <sup>5</sup>	31	F	Total colectomy and ileostomy	Postop course complicated by recurrent abscesses; patient ultimately died of extensive Crohn's disease and malnutrition
Kokal et al, 1978 <sup>19</sup>	38	M	Right and transverse colectomy with descending ileocolostomy; partial gastrectomy with Billroth I gastroenterostomy	Brief episodes of diarrhea responsive to medication, otherwise uncomplicated recovery
	34	F	Total proctocolectomy and ileostomy; partial gastrectomy and Billroth II gastroenterostomy	Uncomplicated recovery
Zapolanski and Jagelman, 1980 <sup>20</sup>	33	M	2-stage procedure: stage 1—loop ileostomy, stage 2—proctocolectomy and fistula repair	Uncomplicated recovery
Lawhon, 1980 <sup>21</sup>	19	F	Total colectomy and ileoproctostomy; fistula repair	NS
McDaniel et al, 1982 <sup>22</sup>	26	F	Total colectomy and ileostomy and Hartmann procedure; fistula repair	NS
Broe et al, 1982 <sup>23</sup>	NS	NS	NS	NS
Scully et al, 1983 <sup>24</sup>	39	M	En bloc resection of distal half of stomach and ascending and right transverse colon	Uncomplicated recovery
Jacobson et al, 1985 <sup>25</sup>	26	F	Right and transverse colectomy; wedge resection of fistula	Uncomplicated recovery; no recurrence for 6 yr
	39	M	Hemigastrectomy and vagotomy, Billroth II gastroenterostomy; right and transverse colectomy	Primary healing; no recurrence for 3 yr

**Table 1**

Logio et al, 1987 <sup>26</sup>	42	F	Subtotal colectomy; circumferential sleeve resection of stomach and pyloroplasty	Uncomplicated recovery
Greenstein et al, 1989 <sup>27</sup>	21	M	Initial surgery — gastroenterostomy; re-exploration — subtotal colectomy and ileostomy, mucous fistula and repair of fistula	Initial surgery complicated by recurrent gastrocolic fistula; uncomplicated recovery after re-exploration
	29	F	Total colectomy and ileostomy; wedge resection of fistula	Episode of ileostomy hemorrhage, otherwise uncomplicated recovery
	25	M	Subtotal colectomy and ileosigmoidostomy; sleeve resection of greater curvature of stomach	Uncomplicated recovery
	31	M	Ileostomy and 6-MP postop	Uncomplicated recovery
	49	F	6-MP	Initially improved but relapsed after cessation of therapy; restarted on 6-MP and asymptomatic for 2.5 yr on long-term 6-MP maintenance therapy
	21	M	6-MP	Remission of symptoms and closure of fistula within 3 mo, but fistula reopened and symptoms recurred after cessation of therapy; restarted on 6-MP for 4 yr and remained asymptomatic for 8 yr
	60	F	Subtotal colectomy, mucous fistula; gastroenterostomy and wedge resection of fistula	Uncomplicated recovery
Flueckiger et al, 1990 <sup>28</sup>	29	M	Left hemocolectomy and transverse colosigmoidostomy and fistula repair	Uncomplicated recovery
Pichney et al, 1992 <sup>29</sup>	29	F	Ileocolonic resection and fistula repair	NS
Gidvani and Mizell, 1997 <sup>1</sup>	13	F	Partial colectomy and fistula repair	Re-exploration 3 wk postop because of recurrent symptoms — negative findings; uncomplicated recovery thereafter

NS = not specified, 6-MP = 6-mercaptopurine.

be a result of colitis in the transverse colon with deep ulceration and subsequent fibrosis and fistula formation to the adjacent stomach. This theory is supported by previous reports that consistently show deep ulceration and pseudopolyp formation in the area of the colon where the fistulous tract originates, yet usually without evidence of Crohn's disease in the stomach where the fistula ends. Hence, it has been suggested that the correct term for this fistula would be "colonogastric" fistula.<sup>26</sup>

The classic triad of symptoms in patients with gastrocolic fistulas secondary to Crohn's disease comprises diarrhea, weight loss and fecal halitosis or vomiting but is only present in 30% of the cases.<sup>19</sup> Diarrhea, weight loss and abdominal pain are seen in approximately 50% of patients with Crohn's-related gastrocolic fistulas,<sup>29</sup> but these symptoms can easily be attributed to an uncomplicated exacerbation of the underlying disease process. Hence, it is very difficult to determine the presence of a gastrocolic fistula based on history alone. The presence of feculent vomiting or fecal halitosis, which are pathognomonic of a gastrocolic fistula, may help in the diagnosis.<sup>11,15</sup> The symptoms associated with Crohn's-related gastrocolic fistulas are thought to be secondary to reflux of colonic contents through the fistulous tract with secondary injury to the upper gastrointestinal tract. This retrograde flow of fecal material may result in bacterial overgrowth and be responsible for the debilitating diarrhea and malnutrition that occur with a gastrocolic fistula.<sup>29</sup> Vitamin B<sub>12</sub> malabsorption may occur, but this study was not conducted in our patient.

Physical findings in a patient with a gastrocolic fistula usually include malnourishment and dehydration.<sup>2</sup> Abdominal findings are often nonspecific and minimal; findings may be limited to diffuse abdominal tenderness and distension.<sup>19</sup> Laboratory indices are also nonspecific and are usually limited to electrolyte abnormalities related to the vomiting and diarrhea plus anemia, hypoproteinemia and avitaminoses secondary to the patient's malnourished state.<sup>3,19</sup> Plain films of the abdomen are usually nonspecific; in this case, the abdominal series showed a distended stomach and absence of gas in the distal bowel, features that could be explained by a functional or mechanical obstruction secondary to the inflamma-

tory process in an uncomplicated exacerbation of Crohn's disease.

The diagnosis of a gastrocolic fistula is usually confirmed by a barium enema study, which is reported to have close to 100% diagnostic accuracy in revealing the fistula.<sup>14</sup> Upper gastrointestinal barium study is only effective in disclosing gastrocolic fistulas in approximately one quarter to one third of the cases;<sup>14,21</sup> this is likely because the fistulous opening is usually so small that contrast material preferentially passes through the pyloric channel as the stomach empties by antral contraction.<sup>20</sup> Although esophogastroduodenoscopy and colonoscopy do not demonstrate the fistula well, these examinations are essential preoperatively because they delineate the extent and severity of the disease and hence provide useful information on which the surgeon can base a decision as to the type of resection required.<sup>26</sup>

Medical therapy may play a role in managing Crohn's-related gastrocolic fistulas. Greenstein and associates<sup>27</sup> reported that 2 patients in their series became asymptomatic and showed radiologic closure of the fistulas with prolonged courses of 6-mercaptopurine. However, the potential toxic side effects of this drug, especially when given in the long term, remain a drawback in this setting. These side effects include hypersensitivity to the drug, sore throat, fever, joint pain and pancreatitis.<sup>30</sup>

It is generally accepted that operation is the definitive treatment for a gastrocolic fistula. Although a 2-stage operation was encouraged in the past, improvements in preoperative and postoperative care have allowed a 1-stage procedure to be performed safely in most cases. In the severely debilitated patient, a 2-stage procedure with a proximal diverting stoma has been advised,<sup>18,20,31</sup> but a preoperative course of intravenous hyperalimentation for the severely malnourished patient may obviate this.<sup>18</sup>

In terms of the type of operation, a variety of procedures have been recommended, depending on the extent of disease. Most authors advocate resection of the diseased colon and excision and primary repair of the fistulous opening. In some cases, because of the size of the fistula or significant involvement of the stomach, gastric wedge resection of the fistula or a partial gastrectomy may be required. In 2 instances, because of the presence of disease in the stomach, a sleeve

resection of the stomach was used with resection of the fistula.<sup>26,27</sup> In summary, operation is the mainstay of therapy for gastrocolic fistula in Crohn's disease. Modern-day knowledge with appropriate nutritional support permits a 1-stage operative correction of this rare but debilitating condition.

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