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Recurrent pneumonia and colobronchial fistula from Crohn's disease: Infliximab alters and simplifies surgical management

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Abstract

We report a rare case of right-sided colobronchial fistula in a 47-year-old, severely malnourished male with a history of regional enteritis and recurrent right lower and middle lobe pneumonias medically managed with the addition of the immunomodulator infliximab prior to surgery. On admission, evaluation of sputum cultures and chest radiograph pattern of pneumonia led to the suspicion of colobronchial fistula. This diagnosis was confirmed by abdominal CT enteroclysis. This patient's pneumonia was initially treated with empiric antibiotics, then focused antibiotics based on culture results. The treatment for the regional enteritis and the secondary colobronchial fistula consisted of immunosuppression with infliximab, bowel rest, and total parenteral nutrition. The patient was discharged on a limited course of prednisone and received maintenance therapy with 3mg/kg IV infliximab infusions for four additional treatments with dramatic improvement in his clinical condition. Surgical therapy consisted of only bowel resection; no thoracic surgery or lung resection was necessary. The patient has had a dramatic improvement in his clinical condition and is currently disease-free on no maintenance therapy. The use of TNF-blocking agents such as infliximab may simplify the surgical approach in patients with complicated fistulous Crohn's disease.

Keywords Crohn's disease, infliximab, colobronchial fistula

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Introduction

Crohn's disease is a chronic condition of unknown etiology with a North American prevalence of 26-199 per 100,000 individuals [1]. It is characterized by full thickness intramural inflammation of the gastrointestinal (GI) tract most commonly affecting the small bowel and colon. Complications, such as recurrence of enteric and enterocutaneous fistulas, are pathognomonic of this disease, and can occur in up to 50% of patients [2]. Fistulous disease commonly presents as enteroenteric, enterocolic, enterocutaneous, but perianal fistulas are most common [3]. Less common are gastrointestinal fistulas to the urinary tract. Since 1950, only eight cases of colobronchial fistulas, typically left-sided, a rare complication of Crohn's disease, have been reported. We report a rare case of colobronchial fistula resulting in recurrent right-sided pneumonias in a young man with uncontrolled regional enteritis and the surgical management that led to successful long-term recovery.

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Conflict of Interest: None

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Case report

After referral from a local hospital in late 2010, a 45-year-old male patient with history of Crohn's disease and recurrent pneumonias for almost two years, presented with enterocutaneous abdominal fistula and one week of progressive pleuritic right chest pain radiating to the right shoulder, fever, and productive cough with foul smelling brownish sputum. He had a 30 pound weight loss. Past medical history included a twenty pack-year smoking history, five-year history of regional enteritis, and multiple operations including enterocutaneous fistula takedowns and right hemicolectomy in 2004.

Prior to admission, he reported symptoms of loose stools. The patient had failed to follow up with Gastroenterology for treatment of his regional enteritis and had not been adherent to an immunosuppressive therapy regimen.

On admission, vital signs were: temperature 38.3 °C, heart rate 84, blood pressure 116/76, and O_2 saturation of

94%. He was cachectic. Pulmonary exam was notable for coarse rhonchi at the base of the right chest and mildly labored breathing. Pertinent laboratory data demonstrated WBC 3,700/dL, hemoglobin 9 g/dL, erythrocyte sedimentation rate 66 mm/h, and pre-albumin 9 mg/dL. Initial chest radiograph showed bilateral basilar airspace opacities worse on the right and blunting of the right costophrenic angle (Fig. 1). Bronchoscopy and sputum cultures grew polymicrobial enteric organisms and abdominal CT enterography was the diagnostic test that confirmed a right colobronchial fistula (Fig. 2) and also multiple other colonic fistulas. The duodenum and small bowel showed no evidence of mucosal distortion.

Initially, the pneumonia was treated with empiric antibiotics, and Crohn's disease was treated with IV ciprofloxacin, metronidazole, bowel rest and total parenteral nutrition. Gastroenterology had previously recommended delaying infliximab until pneumonia was well controlled. He refused upper endoscopy and upper GI series was done showing normal gastric mucosa and confirming the normal duodenal findings of the CT enteroclysis. After antibiotics were started, we started immunosuppressive therapy with azathiprine and budesonide. He did not tolerate azathioprine. Infliximab (3 mg/kg) with acetaminophen premedication was initiated after five days of antibiotics. After discharge, outpatient maintenance therapy and remission of Crohn's disease was attained with four-weekly infliximab infusions (3 mg/kg) for a total of five doses, prednisone (did not tolerate budesonide), and suppressive amoxicillin/clavulanate. Significant improvement was evident after a twenty pound weight gain, mild intermittent non-productive cough, no recurrence of fevers and malaise, and normalization of chest radiograph (Fig. 3). Antibiotics, and total parenteral nutrition (TPN) were stopped and steroids were weaned off.

Exploratory laparotomy revealed multiple colocolonic

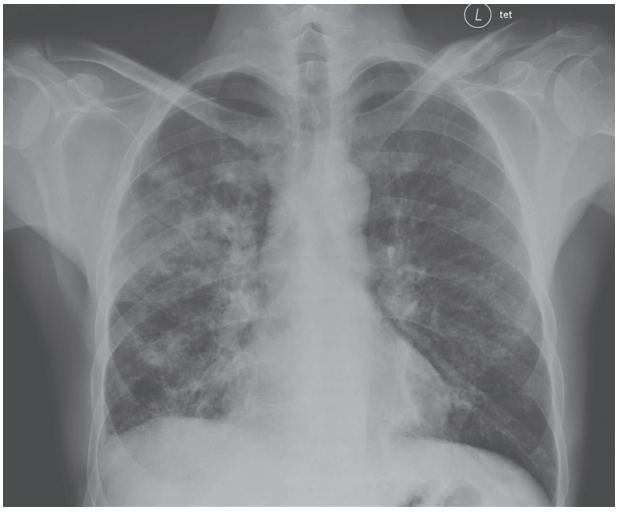

Figure 1 Initial chest radiograph. Increasing areas of confluent airspace disease involving the right middle and lower lobes and stable patchy airspace opacities involving both lungs with stable reactive lymphadenopathy

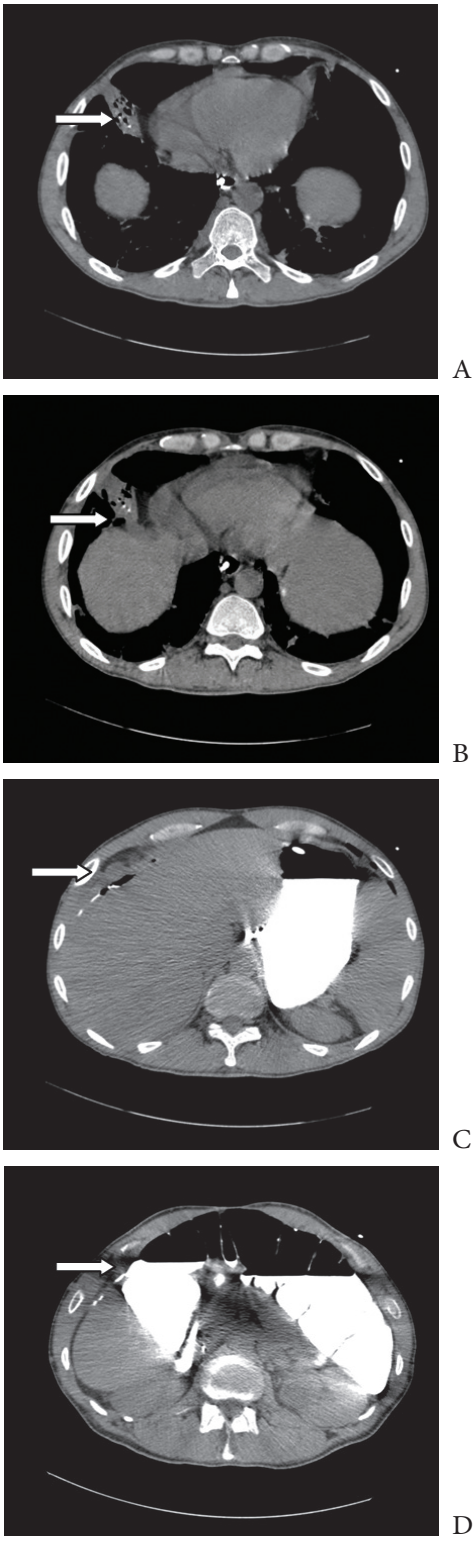

Figure 2 CT enterography. Colobronchial fistula tract (arrows) coursing from hepatic flexure and extending on the liver anteriorly and eventually communicating to right lower lobe bronchus with surrounding lung consolidation. Figures A and B demonstrate gastrointestinal contrast in the lung parenchyma. Figures C and D show fistula tract containing contrast from colon over the right lobe of the liver

fistulas and a colonic fistula at the hepatic flexure that led into a sinus tract going up over the liver and to the diaphragm. The small bowel was normal as was the rectum and colon on flexible sigmoidoscopy to 40 cm. Total abdominal colectomy was performed with subsequent debridement of granulation tissue within the fistula tract. Surgery concluded with a side-to-side ileoproctostomy. No major complications developed in the post-operative period. Pathology of the resected colon revealed no active Crohn's disease at either the proximal small bowel margin or the distal rectal margin. He remains non-compliant with follow-up and has failed to keep his gastroenterology appointments.

Discussion

Although colobronchial fistulas are a rare complication of Crohn's disease, they should be highly suspected in patients with chronic respiratory symptoms and poorly controlled Crohn's disease. Diagnostic tests to determine pneumonia secondary to enteric fistula should include a chest radiograph, bronchoscopy, and sputum cultures consistent with enteric organisms. In this case, suspected colobronchial fistula was confirmed by abdominal CT enterography, an excellent tool for imaging complex Crohn's disease patients [4,5]. Half of the reported cases of visualized colobronchial fistulas used barium or gastrographin enema [6-9]; the remainder visualized them using abdominal CT [10-13]. The majority of fistulas are to the left chest, presumably because the liver protects the right diaphragm from the inflammatory process.

In all previous eight cases of colobronchial fistula, surgery was the standard of care and included bowel resection of the segment involved in the fistula trajectory and often diaphragmatic and pulmonary wedge resection [6-13]. One case did attempt, but failed, to close colobronchial fistula medically with one month of 6-mercapropurine [6]. In our case, because of the poor nutritional state and the severity of Crohn's disease, medical management, to include TPN and infliximab, was our initial goal until optimal conditions for surgery were attained. Infliximab and other TNF-blocking agents have shown dramatic results in some patients with Crohn's disease [14-16]. Infliximab appears to have great utility in unusual cases of fistulizing Crohn's disease [17-19], although it does not replace surgery in most cases because of recurrence [20]. The current patient's treatment with infliximab resulted in dramatic clinical improvement and likely changed the type of surgery required. Although the patient's clinical course indicated possible healing of the colobronchial fistula, the additional multiple colocolonic fistulas required a surgical solution. Ultimately, a completion colectomy and debridement of the fistula were simplified by previous medical management with infliximab. No pulmonary, liver, or diaphragmatic resections were needed.

In conclusion, medical management with immunosuppressive therapy using infliximab is a successful pre-operative adjunct to surgical management for treatment of colobronchial fistula in patients with Crohn's disease and recurrent pneumonia. The use of this class of TNF-blocking agents may simplify the surgical approach in patients with complicated fistulous Crohn's disease.

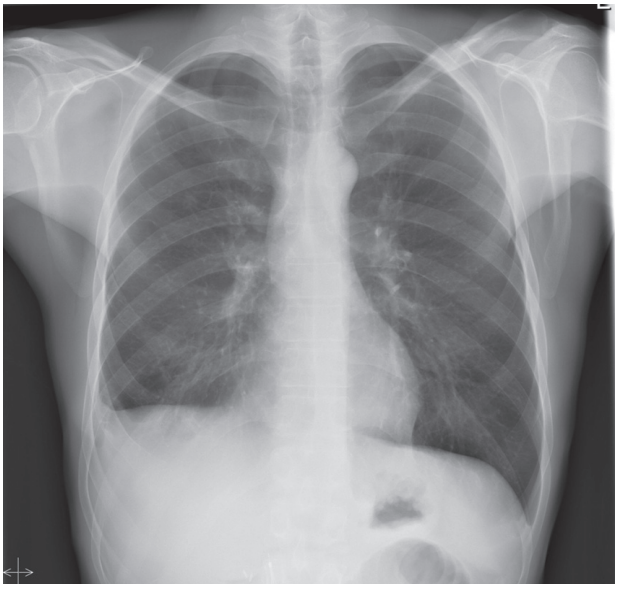

Figure 3 Chest radiograph after recovery

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