Crohn’s Disease Associated with Colo-Bronchial Fistula

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An 18-year-old female patient with Crohn’s disease presented with left lower lobe pneumonia and pleural effusion which were resistant to treatment with antibiotics. Colo-bronchial fistula had not been recognized until she coughed up yellow sputa with feculent odor and developed acute respiratory distress syndrome. This type of fistula is a rare complication of Crohn’s disease, but the present case certainly alerts physicians to search for a fistula between the bronchus and gastrointestinal tract when encountering patients with Crohn’s disease accompanied by antibiotic-resistant chronic pneumonia.

(Keywords: pneumonia, acute respiratory distress syndrome, colo-jejunal fistula, colo-cutaneous fistula)

Introduction

Crohn’s disease has been recognized as an idiopathic inflammatory bowel disease which is characterized by the formation of internal and external fistulas between diseased bowel and adjacent adherent viscera (1). The frequent types are ileo-sigmoid, ileo-cecal, ileo-rectal, colo-vesical, intestino-vaginal, and entero-cutaneous fistulas (1). However, a fistula between the colon and respiratory tract is a rare complication of Crohn’s disease (2).

In this report, we describe a patient with a colo-bronchial fistula presenting as a drug-resistant pleuropneumonia and acute respiratory distress syndrome (ARDS) in the course of Crohn’s disease.

Case Report

An 18-year-old female patient with Crohn’s disease was hospitalized because of fever, cough, diarrhea, and weight loss. She had been well until three years prior to admission when she developed intermittent high fever of unknown etiology. Two years later, she additionally complained of frequent diarrhea and a diagnosis of Crohn’s disease was established on the basis of the criteria proposed by the Japanese Society of Gastroenterology. She was symptomatically treated, but intermittent fever and diarrhea continued for three months before hospitalization.

On admission, she was 157cm tall and weighed 28kg. She had a temperature of 37.7°C, heart rate of 124 beats/min, and blood pressure of 94/50 mmHg. Laboratory data showed white blood cell count of 5,200/mm³ (73% stabs, 10% segmented neutrophils, 11% lymphocytes, 6% monocytes), red blood cell count of 236 x 10⁶/mm³, hemoglobin 5.8g/dl, hematocrit 21.2%, and platelet counts 34.4 x 10⁴/mm³. Serum protein was low (6.0 g/dl) and C-reactive protein was positive (8.7 mg/dl). The serum iron was 14 mg/ml (normal range 70 to 160) and total iron binding capacity was 139 (normal range 270 to 470). An arterial blood gas analysis with room air revealed pH 7.46, PCO₂ of 34.5 mmHg, and PaO₂ of 76.3 mmHg. Stools were positive for occult blood on two successive occasions. A tuberculin skin test was negative. Pseudomonas aeruginosa was isolated from sputum, and Pseudomonas aeruginosa and Escherichia coli from fecal cultures. Chest X-rays and computerized tomography (CT) showed pneumonia in the left lower lobe with pleural effusion (Figs. 1, 2). An enema study of the large bowel showed colo-jejunal, colo-subcutaneous and colo-retroperitoneal fistulas, but a colo-bronchial fistula was not demonstrated (Fig. 3). Histopathologic examinations of the mucosal specimens obtained from the colon by the fiber optic colonoscopy revealed chronic inflammatory infiltrates. No granuloma was found. She was considered to be in an active stage based on the assessment of International Organization for the Study of Inflammatory Bowel Diseases (IOIBD). She was treated with antibiotics (Minocycline and Cefotiam), parenteral nutrition via a central venous catheter, and several units of packed red blood cells. On twenty-second day of admission, she suddenly coughed up yellow...
sputa with a feculent odor and developed severe hypoxemia (PaO₂ 45 mmHg, PCO₂ 30 mmHg, pH 7.45, BE-1.0 in room air). The chest X-rays showed diffuse bilateral infiltrates (Fig. 4). An enema study through a colo-subcutaneous fistula showed the instant passage of water soluble enema from the splenic flexure of large bowel into the left lower lobe bronchi (Fig. 5).
Colo-Bronchial Fistula in Crohn's Disease

Figure 5. An enema study through a colo-subcutaneous fistula (A) showing the instant passage of water soluble enema from the splenic flexure of the large bowel (B) into the left lower bronchi (C).

The patient was treated with continuous drainage of intestinal digestive fluid, corticosteroid (Methyl-prednisolone 500 mg/day for three days, and thereafter tapered), gabexate mesilate (2,000 mg/day for 12 days), and antibiotics (Ceftazidime and Clindamycin). Her symptoms and chest X-ray findings were remarkably improved two weeks after treatment. The drainage of intestinal digestive fluid and parenteral nutrition via a central venous catheter were continued. An enema study performed two months after initiating treatment showed the closure of colo-jejunal and colo-bronchial fistulas.

Discussion

In this report, we have described a patient with Crohn's disease who developed a colo-bronchial fistula and acute respiratory distress syndrome in the course of the disease. The patient showed pneumonia in the left lower lobe and pleural effusion which were resistant to treatment with antibiotics, but the presence of a colo-bronchial fistula as an underlying condition was not recognized on admission. Fistula formation is a hallmark of Crohn's disease and approximately one-third of the patients develop internal and external fistulas in any portions of the gastrointestinal tract. However, a colo-bronchial fistula is an unusual complication of the disease and, to our knowledge, only one case was reported previously (2).

It is well known that symptomatic or latent involvements of the lung such as bronchopulmonary suppuration (3, 4), granulomatous lung disease (5, 6), lymphocytic alveolitis (7-9), or pulmonary function abnormalities (3, 8-10) frequently occur in patients with Crohn's disease. Since bronchoalveolar lavage and transbronchial lung biopsy were not carried out in the present case, it is not known whether this case was also associated with granulomatous lung disease or lymphocytic alveolitis. Such a diffuse pulmonary involvement may not be directly connected with the fistula formation between the bowel and the bronchus.

The colo-bronchial fistula was not recognized until the twentieth day of hospitalization when she developed acute respiratory failure with diffuse bilateral infiltrates on a chest radiograph. The clue which led us to find the fistula in the present case was the expectoration of yellow sputum with a feculent odor. Domej and colleagues reported that microscopic sputum examinations and the isolation of numerous gram-negative and gram-positive bacteria from sputa such as Clostridium innocuum, Streptococcus viridans, Proteus mirabilis, Klebsiella pneumoniae, Staphylococcus aureus, and bifidus bacteria may also be clues in finding the fistulas (2). In the present case, however, only Pseudomonas aeruginosa was isolated from both sputum and fecal cultures as a pathogenic organism probably because various antibiotics with a wide antimicrobial spectrum had been used for treatment of pneumonia before hospitalization. This organism has been frequently isolated from the sputa of patients with chronic respiratory infections without fistulas. Thus, the isolation of gram-negative bacilli such as Pseudomonas aeruginosa may not always help to identify a fistula between the bowel and respiratory tract.

Once the abnormal connections between the bowel and respiratory tract are suspected, an enema study using water-soluble contrast medium certainly helps to confirm the presence of fistulas (2). In the present case, a contrast study through a colo-subcutaneous fistula demonstrated the passage of contrast medium from the splenic flexure of the large bowel into the left lower lobe bronchi.

The acute respiratory failure which developed soon after she coughed up sputa with a feculent odor may have been due to the aspiration of intestinal digestive fluid containing various proteinases which reached the left lower bronchi through the fistula. This explanation for the development of ARDS seems reasonable since an enema study on admission had already demonstrated a colo-jejunal fistula at the splenic flexure of the colon.

In the present case, surgical therapy was not chosen because we could not obtain her approval for surgical treatment. Domej and colleagues reported that their patient made a full recovery after a prolonged and complicated postoperative course (2). Thus, surgical therapy may be carried out in patients with Crohn's disease accompanied by a colo-bronchial fistula with drug-resistant pneumonia.

In conclusion, this case report alerts physicians to the need to search for a fistula between the lung and the gastrointestinal tract when encountering patients with Crohn's disease and...
unexplained chronic pneumonia.

References


