

CASE REPORT

맹장-S결장루로 인한 세균과다증식과 관련된 대장가성폐쇄 1예

정경면, 임성욱, 홍형주, 박선영, 박창환, 김현수, 최성규, 류종선
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A Case of Colonic Pseudoobstruction Related to Bacterial Overgrowth Due to a Sigmoidocecal Fistula

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Colocolic fistulas are usually a complication of an inflammatory or neoplastic process. Development of these abnormal bowel communications may lead to bacterial overgrowth. We report on a 71-year-old man with a one-year history of recurrent abdominal distension and irregular bowel habits. Abdominal X-rays and computed tomography showed multiple air-fluid levels and loops of distended bowel without evidence of mechanical obstruction or diverticulitis. Colonoscopy showed a fistulous tract between the sigmoid colon and cecum. Results of a lactulose breath test showed high fasting breath CH₄ levels, which were thought to be the result of intestinal bacterial overgrowth. The patient was diagnosed with a colonic pseudo-obstruction associated with bacterial overgrowth due to a sigmoidocecal fistula. We recommended surgical correction of the sigmoidocecal fistula; however, the patient requested medical treatment. After antibiotic therapy, the patient still had mild symptoms but no acute exacerbations. (*Korean J Gastroenterol* 2014;63:125-128)

Key Words: Colonic pseudo-obstruction; Intestinal fistula; Diverticulum; Colon

INTRODUCTION

Colonic diverticulosis is a common disease, and its incidence increases with age. Development of diverticulitis as a result of infection and inflammation of a diverticulum has been reported in approximately 20% of patients with colonic diverticulosis.¹ Complications of diverticulitis include stricture, bleeding, perforation, and fistula formation.¹ The most common type of fistula is a colovesical fistula arising from the sigmoid colon.² Although rare, development of a colocolic fistula can occasionally occur.² We report on a rare case of a sigmoidocecal fistula leading to bacterial overgrowth with

colonic pseudo-obstruction.

CASE REPORT

A 71-year-old man with a one-year history of recurrent abdominal distension and alternating loose and hard stools was admitted with abdominal distension and diarrhea. Five years previously, a sigmoid diverticulum was discovered during a screening sigmoidoscopy. He was asymptomatic at the time of the screening examination.

At the time of admission, the patient's blood pressure was 110/70 mmHg, heart rate was 88 beats/min, and body tem-

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Fig. 1. Plain abdominal radiography in erect position. Distended intestine, especially the large intestine, and multiple air-fluid levels are seen.

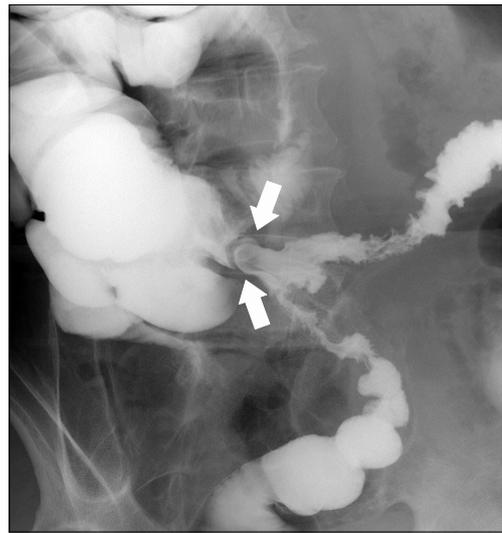


Fig. 3. Barium enema study. A definitive connection of the sigmoid colon and cecum is confirmed (white arrows).

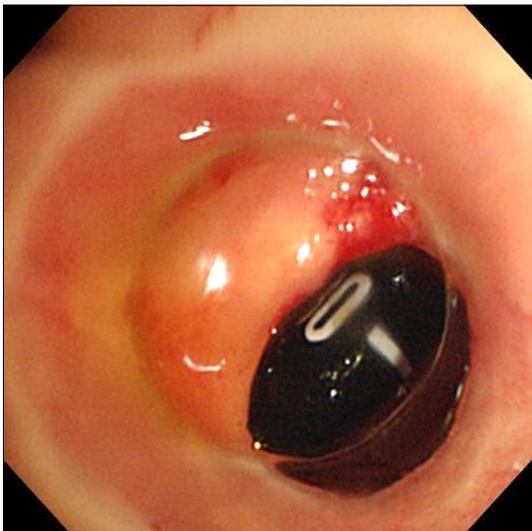


Fig. 2. Colonoscopic findings. White numbers corresponding to the insertion lengths of the colonoscope could be seen through the fistulous opening located in the cecum. The appendiceal orifice was normal.

perature was 36.9°C. He was alert but appeared to be acutely ill. An abdominal examination detected hypoactive bowel sounds, abdominal distension, and diffuse tenderness without peritoneal signs. Laboratory findings included a leukocyte count of 16,100/mm³, hemoglobin of 13.2 g/dL, aspartate aminotransferase of 71 IU/L, alanine aminotransferase of 98 IU/L, and C-reactive protein of 23.1 mg/dL.

Erect abdominal X-rays showed multiple air-fluid levels and distended loops of large and small bowel (Fig. 1).

Abdominal computed tomography showed dilatation of the colon in the absence of any obstructive lesion and no evidence of diverticulitis. The patient received conservative treatment with hydration, bowel rest, and placement of a rectal tube. His abdominal distension disappeared gradually over a five-day period. After the patient's ileus showed clinical improvement, we performed a colonoscopy, which revealed a small fistulous opening in the sigmoid colon. When the colonoscope reached the cecum, white numbers corresponding to the insertion lengths of the colonoscope could be seen through the fistulous opening located in the cecum (Fig. 2). The appendiceal orifice was normal. Barium contrast radiography within two weeks of the colonoscopy showed a definite communication between the sigmoid colon and cecum (Fig. 3). A lactulose breath test was performed using a DP Quintron gas chromatograph (QuinTron Instrument Co., Milwaukee, WI, USA) for determination of whether bacterial overgrowth was the cause of the colonic pseudo-obstruction. The patient's fasting breath CH₄ levels increased to 120 ppm (Fig. 4A), which was considered indicative of intestinal bacterial overgrowth.³ We recommended surgical correction of the sigmoidocecal fistula; however, he requested medical treatment. Therefore, he was treated with antibiotics (rifaximin, 400 mg twice daily) for 14 days, which resulted in clinical improvement of the abdominal distension and diarrhea and a decrease in intestinal gas. He subsequently began taking probiotics capsules (*Bacillus subtilis* 100 million

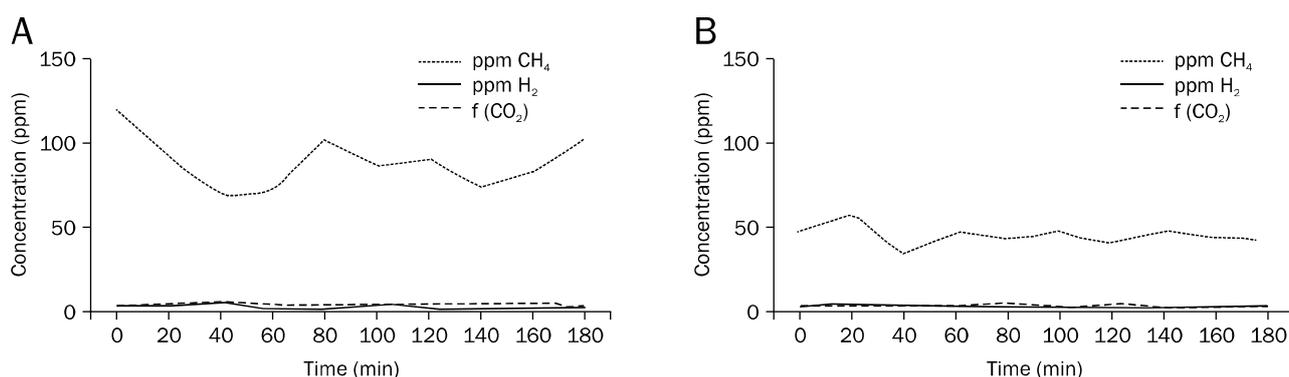


Fig. 4. Lactulose hydrogen breath test. (A) Before treatment, increased methane gas is estimated using a gas chromatograph. (B) After treatment, fasting breath CH₄ concentration levels decline markedly but remain above the normal range. f, fraction.

U, *Streptococcus faecium* 900 million spores) three times per day. At a six-month follow-up examination, he reported the persistence of mild symptoms, but no acute exacerbation of symptoms. Follow-up abdominal X-rays demonstrated the persistence of distended loops of large bowel. The fasting breath CH₄ concentration showed a marked decrease but remained above the normal range (Fig. 4B).

DISCUSSION

Colocolic fistulas are usually a complication of an inflammatory or neoplastic process. This patient had a past history of a sigmoid diverticulum. Diverticulitis is a common cause of colonic fistula, with the fistula most often communicating with the urinary bladder.⁴ A relatively rare finding in diverticulitis is a fistulous tract that parallels the colonic lumen, representing a localized form of colocolic fistula, which has been termed "double tracking." Not surprisingly, fistulous formation of the sigmoid colon predominates in diverticular disease; however, other colonic segments are occasionally involved.⁴ In a thorough search of the English literature, only a few cases of sigmoidocecal fistula due to sigmoid diverticulitis or inflammatory bowel disease were found.^{5,6} These abnormal bowel communications, including colocolic fistula, gastrocolic fistula, or jejunocolic fistula, may lead to bacterial overgrowth.⁷

Most authors consider a bacterial count $\geq 10^5$ bacteria (i.e., colony forming units) per mL of proximal jejunal aspiration to be diagnostic of bacterial overgrowth.⁸ Aspiration and direct culture of jejunal contents are considered the gold standard for diagnosis of bacterial overgrowth.⁹ These proce-

dures have some limitations, including invasiveness, possible contamination by oropharyngeal bacteria, low reproducibility, and the presence of nonculturable bacteria. For this reason, noninvasive tests such as the breath test are commonly used for diagnosis of bacterial overgrowth. These tests are based on production of hydrogen and methane by bacteria as a consequence of carbohydrate fermentation.⁹ Most intestinal gas is produced in the right colon by anaerobic fermentation of unabsorbed carbohydrates.¹⁰ Upon reaching the cecum, unabsorbed carbohydrates are rapidly fermented to short-chain fatty acids by anaerobic colonic bacteria, liberating carbon dioxide, hydrogen, and, in some individuals, methane, which diffuses rapidly into the blood stream and is then exhaled. Bacteria represent the sole source of gut hydrogen and methane, therefore, fasting-breath hydrogen and methane have been used as markers of colonic fermentation.¹¹ Because approximately 20% of colonic methane is excreted via the breath, breath methane analysis has been used as a method for indirect assessment of intracolonic bacterial metabolism.¹² One method of diagnosing small intestinal bacterial overgrowth is the lactulose breath test, where overgrowth is considered present if an increase in concentration of the breath of greater than 20 ppm of hydrogen or methane is observed within 90 min of oral administration of lactulose.¹³ In this case, the patient's fasting breath CH₄ levels increased to 120 ppm, which was considered indicative of intestinal bacterial overgrowth.

Methanobrevibacter smithii, which preferentially colonizes the left colon, is the predominant methanogenic bacterium found in the human.¹⁴ The prevalence of methanogenic flora is higher in constipated patients with slower colonic

transit,¹⁵ diverticulosis,¹⁶ or encopresis. In recent animal studies, methane was reported to cause a decrease in peristaltic velocity and a marked increase in the contraction amplitude of the small bowel and colon.¹⁷ Therefore, bacterial overgrowth (especially, methane production) associated with a sigmoidoceleal fistula might also be associated with slower colon transit and abundant gas formation, thus resulting in colonic pseudo-obstruction in our patient.

Resection of the fistula and re-establishment of the continuity of the gastrointestinal tract is the treatment of choice for abnormal bowel communication.¹⁸ Therefore, we recommended surgical correction for the anatomical abnormality; however, the patient requested medical treatment. Therapy using an antibiotic such as rifaximin followed by probiotics has been reported to be effective for reduction of symptoms in patients with bacterial overgrowth.^{19,20} Detection of methane in association with colonic pseudo-obstruction may suggest potential therapeutic options. Therefore, in this case, we selected antibiotics for acute management and probiotics for maintenance.

In conclusion, colonic pseudo-obstruction associated with bacterial overgrowth due to a sigmoidoceleal fistula is a rare complication of colonic diverticulitis. Although the treatment of choice for a fistula is surgery, administration of an antibiotic such as rifaximin followed by probiotics is a therapeutic alternative.

REFERENCES

1. Stollman NH, Raskin JB. Diagnosis and management of diverticular disease of the colon in adults. Ad Hoc Practice Parameters Committee of the American College of Gastroenterology. *Am J Gastroenterol* 1999;94:3110-3121.
2. Woods RJ, Lavery IC, Fazio VW, Jagelman DG, Weakley FL. Internal fistulas in diverticular disease. *Dis Colon Rectum* 1988;31:591-596.
3. Walters B, Vanner SJ. Detection of bacterial overgrowth in IBS using the lactulose H₂ breath test: comparison with 14C-D-xylose and healthy controls. *Am J Gastroenterol* 2005;100:1566-1570.
4. Pickhardt PJ, Bhalla S, Balfe DM. Acquired gastrointestinal fistulas: classification, etiologies, and imaging evaluation. *Radiology* 2002;224:9-23.
5. Sanowski RA, Costello JL. Sigmoidoceleal fistula. An unusual complication of acute sigmoid diverticulitis. *Am J Gastroenterol* 1967;48:56-62.
6. Hyun YS, Han DS, Kim TY, Eun CS, Jeon YC, Sohn JH. Education and imaging. Gastrointestinal: sigmoidoceleal fistula diagnosed with colonoscopy. *J Gastroenterol Hepatol* 2011;26:208.
7. Husebye E. The pathogenesis of gastrointestinal bacterial overgrowth. *Chemotherapy* 2005;51(Suppl 1):1-22.
8. Khoshini R, Dai SC, Lezcano S, Pimentel M. A systematic review of diagnostic tests for small intestinal bacterial overgrowth. *Dig Dis Sci* 2008;53:1443-1454.
9. Romagnuolo J, Schiller D, Bailey RJ. Using breath tests wisely in a gastroenterology practice: an evidence-based review of indications and pitfalls in interpretation. *Am J Gastroenterol* 2002;97:1113-1126.
10. King TS, Elia M, Hunter JO. Abnormal colonic fermentation in irritable bowel syndrome. *Lancet* 1998;352:1187-1189.
11. Perman JA, Modler S, Barr RG, Rosenthal P. Fasting breath hydrogen concentration: normal values and clinical application. *Gastroenterology* 1984;87:1358-1363.
12. Bond JH Jr, Engel RR, Levitt MD. Factors influencing pulmonary methane excretion in man. An indirect method of studying the in situ metabolism of the methane-producing colonic bacteria. *J Exp Med* 1971;133:572-588.
13. Joseph F Jr, Rosenberg AJ. Breath hydrogen testing: diseased versus normal patients. *J Pediatr Gastroenterol Nutr* 1988;7:787-788.
14. Kajs TM, Fitzgerald JA, Buckner RY, et al. Influence of a methanogenic flora on the breath H₂ and symptom response to ingestion of sorbitol or oat fiber. *Am J Gastroenterol* 1997;92:89-94.
15. Attaluri A, Jackson M, Valestin J, Rao SS. Methanogenic flora is associated with altered colonic transit but not stool characteristics in constipation without IBS. *Am J Gastroenterol* 2010;105:1407-1411.
16. Weaver GA, Krause JA, Miller TL, Wolin MJ. Incidence of methanogenic bacteria in a sigmoidoscopy population: an association of methanogenic bacteria and diverticulosis. *Gut* 1986;27:698-704.
17. Jahng J, Jung IS, Choi EJ, Conklin JL, Park H. The effects of methane and hydrogen gases produced by enteric bacteria on ileal motility and colonic transit time. *Neurogastroenterol Motil* 2012;24:185-190, e92.
18. Schechter WP, Hirshberg A, Chang DS, et al. Enteric fistulas: principles of management. *J Am Coll Surg* 2009;209:484-491.
19. Majewski M, Reddymasu SC, Sostarich S, Foran P, McCallum RW. Efficacy of rifaximin, a nonabsorbed oral antibiotic, in the treatment of small intestinal bacterial overgrowth. *Am J Med Sci* 2007;333:266-270.
20. Barrett JS, Canale KE, Geary RB, Irving PM, Gibson PR. Probiotic effects on intestinal fermentation patterns in patients with irritable bowel syndrome. *World J Gastroenterol* 2008;14:5020-5024.