

CASE REPORT

크론병으로 인한 십이지장-대장 누공의 Endoloop과 Hemoclip을 이용한 수술 전 내시경적 결찰: 증례보고

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Crohn's Duodeno-colonic Fistula Preoperatively Closed Using a Detachable Endoloop and Hemoclips: A Case Report

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Duodeno-colonic fistula is an enterocolonic fistula that occurs as a complication of Crohn's disease. Symptoms of duodeno-colonic fistula are similar to those of Crohn's disease, such as weight loss and diarrhea. The treatment of choice is surgery, although medical treatment may also be considered. However, surgery is recommended when all available medical therapies have been ineffective. In this case, we report a secondary duodeno-colonic fistula due to Crohn's disease that was temporarily managed by an endoscopic procedure with a detached endoloop and hemoclips as a bridging therapy to final surgical repair. (Korean J Gastroenterol 2013;61:97-102)

Key Words: Crohn disease; Intestinal fistula; Endoscopy

INTRODUCTION

Crohn's disease is an inflammatory bowel disease that involves any portion of the gastrointestinal tract. Fistula, abscess formation, perforation, and intestinal stricture are common complications of Crohn's disease.¹ Up to 50% of patients with Crohn's disease are affected by fistulae² that often severely impair patient quality of life. In a previous study, the cumulative risk of developing a fistula in Crohn's disease was 33% after 10 years and 50% after 20 years, respectively.²

Various types of fistulae induced by Crohn's disease have been reported. In Western study,³ 54.0% of fistulae were perianal (intestine to perianal skin) fistulae, 24.0% were enter-

oenteric (intestine to intestine) fistulae, 9.0% were rectovaginal (intestine to vagina) fistulae, and 13.0% were other fistulae including enterovesical (intestine to bladder) and enterocutaneous (intestine to skin) fistulae. In Korean study,³ perianal fistulas seemed to occur most commonly in 46.8% of patients with Crohn's disease and the cumulative frequency of perianal fistula was 54.3% after 15 years which is in contrast to those of Western studies reported to be 13.0-38.0%.⁴ Duodeno-colonic fistula is an enteroenteric fistula that shows communication between the small and large bowel. In general, the treatment of choice for duodeno-colonic fistula is surgery.⁵ However, surgery is not always safe because of the resultant high morbidity and mortality when a patient is in poor general condition or has a combination of

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serious diseases. As a new treatment modality, we report a case of duodeno-colonic fistula secondary to Crohn's disease temporarily treated with an endoscopic closure and finally cured with surgery after the patient's general condition improved following endoscopic treatment.

CASE REPORT

A 27-year old woman was admitted to Severance Hospital with complaints of refractory diarrhea, nausea, fecal vomiting, and weight loss. She had a medical history of Crohn's disease diagnosed 16 months previously with symptoms of nausea, vomiting, diarrhea and 15 kg of weight loss over 6 months. She was prescribed mesalazine, prednisolone, and azathioprine when her Crohn's disease was first diagnosed. She took mesalazine continuously, tapered prednisolone, but stopped azathioprine due to bone marrow suppression. During the medication period, diarrhea, nausea, and weight loss recurred every 2 or 3 months. Two months before admission, she developed the additional symptom of fecal vomiting and a duodeno-colonic fistula was diagnosed by small bowel series - double contrast image study. Surgery was considered due to persistent symptoms despite medical treatment at another hospital. However, surgery was postponed due to poor nutritional and general condition as assessed by low BMI (12.71 kg/m^2), low white blood cells (WBC) and platelet counts of the patient. Moreover, the patient strongly refused to receive surgical treatment. At that time, she again took azathioprine 25 mg but shortly discontinued this medication due

to the reoccurrence of pancytopenia. Despite medical treatments, the symptoms of diarrhea and weight loss persisted and she was transferred to our hospital for surgery.

At the time of admission to our institution, her blood pressure was 95/56 mmHg, pulse rate 72/min and body temperature was 36.6°C . Physical examination revealed diffuse abdominal tenderness. Her body weight was 37.0 kg and height was 171.0 cm. Initial laboratory findings were as follows: WBC count $2,540/\text{mm}^3$, Hb 9.2 g/dL, platelet count $80,000/\text{mm}^3$, normal liver and kidney function tests, albumin 3.1 g/dL, sodium 141 mmol/L, potassium 3.7 mmol/L, chloride 109 mmol/L, total CO_2 22 mmol/L, c-reactive protein 1.83 mg/dL and erythrocyte sedimentation rate 9 mm/hour. Gastrografin hypotonic duodenography and a computed tomography scan of the abdomen and pelvis revealed fistula formation between the hepatic flexure of the colon and the second portion of the duodenum (Figs. 1, 2). As the patient had symptoms consistent with a fistula, surgery was considered, but the patient refused surgery and the operation was delayed by the surgeon due to poor nutritional condition of the patient. Medical treatment with antibiotics and mesalazine 3 g was started and we decided to attempt an endoscopic closure. Esophagogastroduodenoscopy (EGD) showed a fistula at the second portion of the duodenum (Fig. 3A). First, hemoclippping with Olympus hemoclips (7 mm, HX-610-135L; Amori Olympus Co. Ltd., Okkonoki, Japan) was performed eight times around the fistula orifice and the fistula was closed by a medium sized detachable snare by Endoloop (20 mm, MAJ-340; Amori Olympus Co. Ltd.) (Fig. 3B). After the

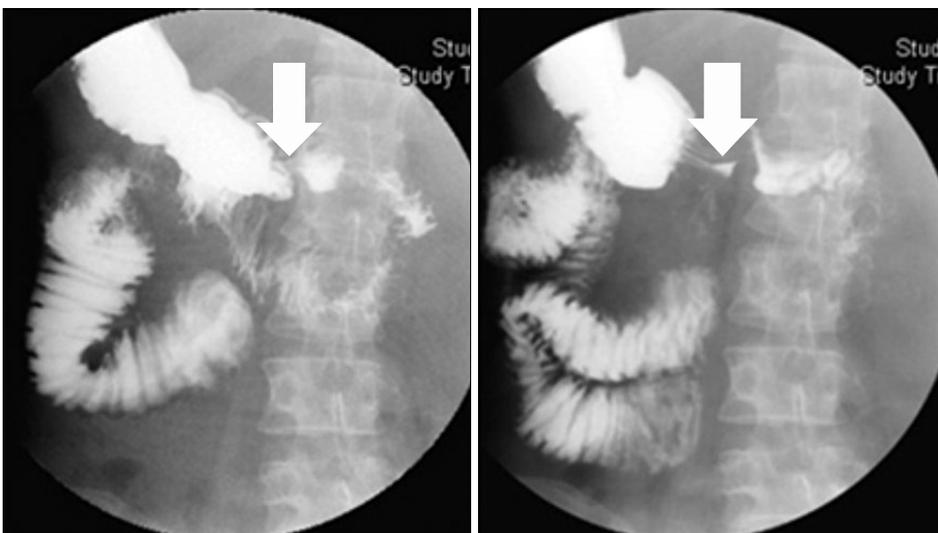


Fig. 1. Gastrografin hypotonic duodenograph at admission. Fistula tract in the second portion of the duodenum was noted (arrows). The size of fistula opening measured approximately 1.5 cm.

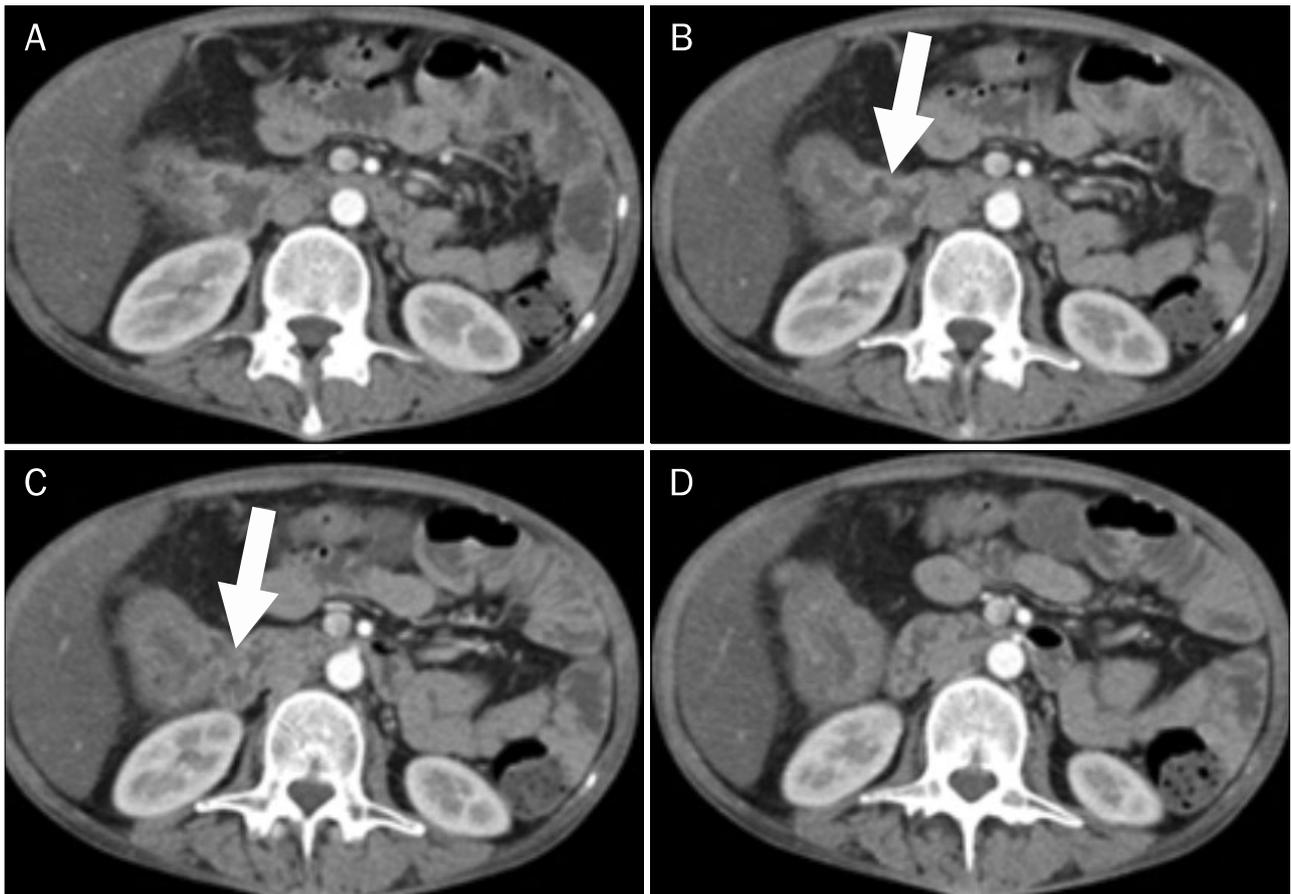


Fig. 2. Computed tomography scan of abdomen and pelvis at admission. Mucosal enhancement and irregular wall thickening was present in the hepatic flexure of colon due to Crohn's disease involvement. Fistula formation between the hepatic flexure of the colon and the duodenal second portion was founded (arrows).

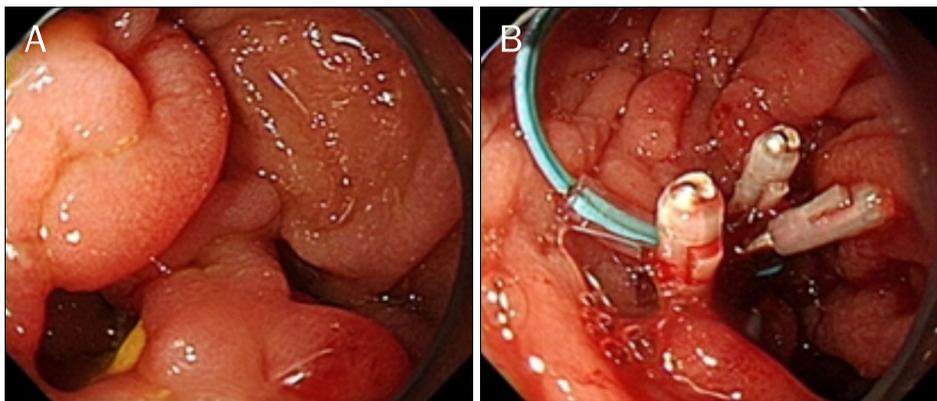


Fig. 3. (A) Esophagogastroduodenoscopy was performed at the first admission. A fistula tract was identified in the duodenal second portion. (B) Endoscopic procedure was performed for closing of fistula. Hemoclipping around the fistula was performed eight times and the fistula was occluded with a medium sized detachable snare.

procedure, the patient was able to tolerate a diet and gained body weight (37.0 to 39.0 kg during 6 day). After the 5th day of procedure, the patient's albumin level increased from 3.1 g/dL to 3.8 g/dL without any replacement, platelet count increased from 80,000/mm³ to 435,000/mm³ and WBC count recovered from 2,540/mm³ to 4,310/mm³ at 10th day

of azathioprine off. She was discharged on the 6th day after the procedure without any specific complications. In the outpatient department clinic, she maintained mesalazine 3 g and continued to refuse surgery.

Five months following endoscopic closure, the patient was readmitted to our hospital due to complaints of poor oral in-

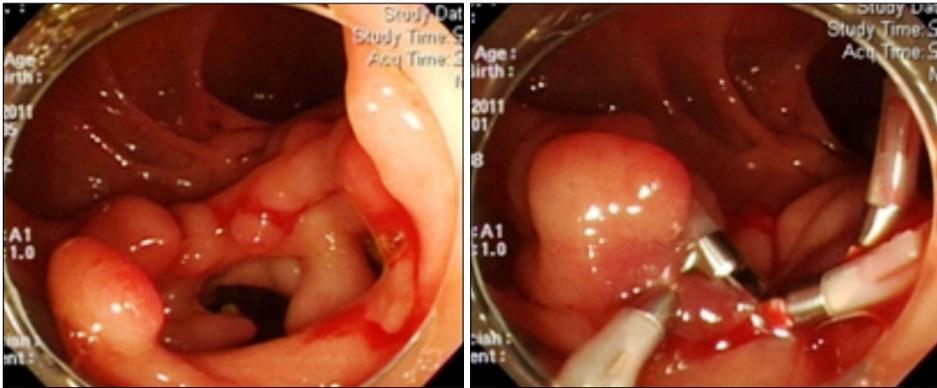


Fig. 4. Endoscopic findings performed at the second admission. Duodeno-colonic fistula was noted recurrently and all of the hemoclips and the detachable snare used in the previous treatment disappeared. The fistula was closed by hemoclippping six times.

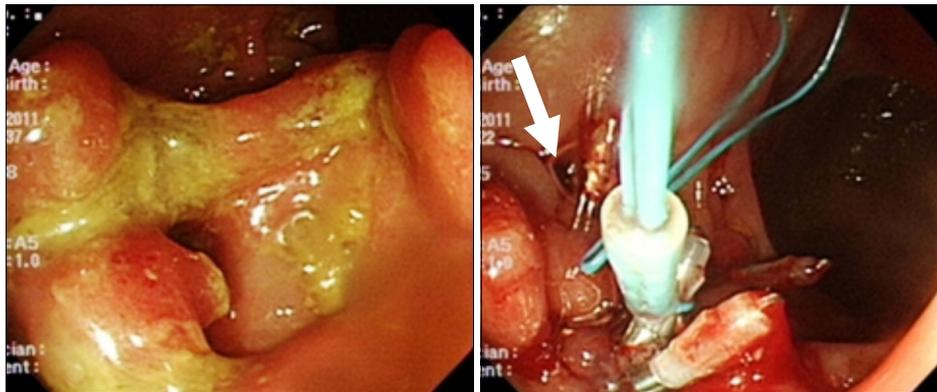


Fig. 5. Endoscopic findings at the third admission. Ten instances of hemoclippping and two-time closure with a detachable snare loop were performed to close a recurrent duodeno-colonic fistula. However, a remnant fistula (arrow) persisted after this procedure, so surgery was considered.

take and weight loss. Before admission, she restarted azathioprine 25 mg for one month but changed with mesalazine 3 g due to leucopenia. An EGD was performed and a recurrent duodeno-colonic fistula was found. Hemoclippping with the same hemoclips as used in the previous procedure was performed six times on the fistula (Fig. 4) resulting in the closure of the fistula. At this time, we did not use an endoloop. After the procedure, her symptoms at admission improved. She continued to be reluctant to undergo surgery and was discharged on the 16th day after the procedure and maintained mesalazine 3 g.

Three months after the second endoscopic procedure, the patient's vomiting, poor oral intake, and general weakness relapsed. On EGD, a fistula was again found. A third endoscopic closure was performed by hemoclippping ten times and was closed twice with a detachable snare loop. However, a remnant fistula was noted (Fig. 5), which indicated that the size of the fistula was reduced, but that it was not perfectly closed. We planned elective surgery after her symptoms partially improved. Ultimately, an extended right hemicolectomy with ileo-transverse colostomy and primary repair of the duodenum was performed. There were no complications during

or after the operation. She was discharged 13 days after surgery.

After surgery, she was followed up at the outpatient department clinic and did not maintain further medications or have further complications or symptoms. Fig. 6 shows the patient's body weight and laboratory changes (albumin, platelet, WBC, and c-reactive protein) during treatment.

DISCUSSION

Fistulae are significant complications of Crohn's disease. The posterior surface of the proximal transverse colon and the anterior surface of the descending duodenum are intimately related, predisposing this area to fistularization.⁶ Duodeno-colonic fistulae are internal fistulae that are difficult to treat due to resistance to medical therapies in most cases, so surgery is usually recommended.^{4,6,7}

Common features of fistulae are weight loss, abdominal pain, and diarrhea, which are similar to the symptoms of Crohn's disease itself. Therefore, it is difficult to differentiate a fistula from Crohn's disease by clinical manifestations.^{1,2,7,8} Also, when fistulae present, a patient's general condition is

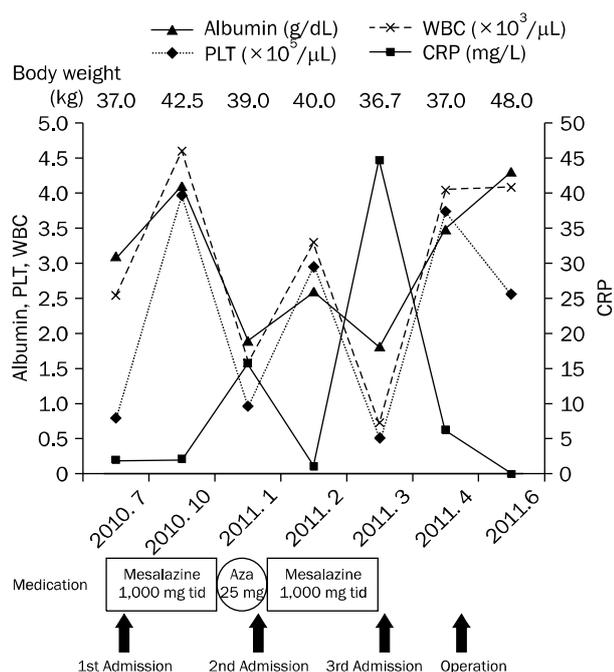


Fig. 6. Patient's medical progress during treatment. WBC, white blood cells; PLT, platelets; Aza, azathioprine

usually worsened. Fecal vomiting is a pathognomic finding in gastro-colonic, but present only in 2% of duodeno-colonic fistulae^{6,9,10} like this case.

Depending on the severity of symptoms, fistula location, number, and complexity of fistula tracts, different medical and surgical therapies are available. Internal fistulae, such as ileoileal or ileocecal fistulae, are mostly asymptomatic and do not require intervention. By contrast, perianal fistulae can be painful and abscesses may develop that require surgical drainage with or without seton placement, transient ileostomy, or in severe cases, proctectomy.² Medical treatment includes traditional medications such as corticosteroids, antibiotics, 5-aminosalicylic acids (mesalazine), and newly emerging medicines like immunosuppressive agents (azathioprine, 6-mercaptopurine), methotrexate, calcineurin inhibitors (tacrolimus), or biologics (infliximab, adalimumab, certolizumab).^{2,11,12} Among these medicines, methotrexate, tacrolimus, mesalazine and corticosteroids are not efficient but thiopurine and infliximab are confirmed to be effective in fistulous Crohn's disease.^{5,12,13} Several studies have reported that external fistulae were more responsive to medical therapies than internal fistulae and internal fistulae were usually refractory to medical treatments.¹¹⁻¹⁴ In this case, im-

munosuppressive agents could not be used due to bone marrow suppression and infliximab could not be used due to the patient's poor general condition, severe leucopenia, and thrombocytopenia. Moreover, infliximab was considered less effective because the lesion was not an active ulcer, and only fibrosis was shown on endoscopic finding. In most cases the most appropriate treatment choice for internal fistula with Crohn's disease is surgical management. However, surgery may be a stressful treatment modality for patients with poor general condition. Therefore, we planned an endoscopic treatment for this patient due to her poor general condition. Endoscopic treatments of fistulae such as occlusion with fibrin glue or clipping have been used when fistula size was small.¹⁵⁻¹⁷ However, in this case this therapeutic modality was not available due to the large fistula size. Instead, we tried to close the fistula with an endoloop and hemoclips for the first time. Fortunately, the result was successful. However, this treatment was temporary, leading to symptom re-occurrence every three or four months. Eventually, surgery was possible after the patient's improved general condition following endoscopic closure. Endoscopic therapy was successful as a bridge therapy, but not as a primary treatment.

Risk factors of post operative recurrence in Crohn's disease include smoking, penetrating disease, previous bowel resection history, perianal involvement, broad small bowel resection.¹² Our patient had one risk factor of penetrating disease, but after the operation, her symptoms of Crohn's disease have not yet recurred. Moreover, she refused to receive any medications. Therefore, we decided to observe the patient cautiously without any maintenance medication and perform a follow-up examination. Because recent guidelines¹² recommend a colonoscopy within 1 year after surgery based on the fact that colonoscopic relapse precedes clinical relapse. We are also planning to perform colonoscopy at 1 year after surgery.

In conclusion, as a new treatment modality, we report a case of duodeno-colonic fistula secondary to Crohn's disease temporarily treated with endoscopic closure and finally resolved with surgery after the patient's general condition improved following endoscopic treatment. This case suggests that endoscopic approach like hemoclippping and looping could be considered as a temporary treatment option with a low risk of complication before initiating curative surgery.

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