

CASE REPORT

젊은 남자에서 캡슐내시경으로 진단한 급성 광범위 허혈성 소장염 1예

정우성, 송현주, 나수영, 부선진, 김홍업, 김진석, 최국명¹
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Acute Extensive Ischemic Enteritis in a Young Man Diagnosed with Wireless Capsule Endoscopy: A Case Report

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Ischemic enteritis is caused by either the interruption or significant reduction of arterial inflow to the small intestine. Risk factors are old age, diabetes mellitus and cardiovascular disease. It is very rare in young patients. We experienced a 21-year-old man with recurrent acute ischemic enteritis who was diagnosed with capsule endoscopy. He had previously taken medications for pulmonary hypertension and obstruction of both carotid arteries, and about 20 months earlier, he had been admitted due to hematochezia. Two sessions of angiography did not reveal the cause of hematochezia. At that time, capsule endoscopy showed mucosal edema and erythema in the terminal ileum, suggesting healed ischemic enteritis. The patient was admitted again due to hematochezia. Abdominal computed tomography showed focal celiac trunk stenosis and diffuse wall thickening of the small intestine, suggesting ischemic enteritis. Capsule endoscopy showed multiple active ulcers and severe hemorrhage with exudate, extending from the proximal jejunum to the terminal ileum. Using capsule endoscopy, the patient was diagnosed with acute extensive ischemic enteritis. Because endoscopic images of ischemic enteritis have rarely been reported, we report a case of a 21-year-old man who was diagnosed acute extensive ischemic enteritis with capsule endoscopy. (*Korean J Gastroenterol* 2013;61:160-165)

Key Words: Ischemia; Enteritis; Capsule endoscopy

INTRODUCTION

Ischemic enteritis is caused by either the interruption or significant reduction of arterial inflow to the small intestine.¹ Ischemic enteritis is much rarer than ischemic colitis.² Risk factors for small-bowel ischemia are hypertension, ischemic heart disease, diabetes mellitus, ischemic cerebrovascular disease and atrial fibrillation.³ Diabetes mellitus, lupus erythematosus and sickle-cell anemia are reported to be asso-

ciated with ischemic enteritis in young people.¹ Most patients are older than 60 years. Ischemic enteritis seems to be a rapidly progressing disease with high mortality.⁴ Therefore, its early diagnosis is one of the most important factors determining a patient's prognosis.⁵ Conventional angiography and multidetector CT are essential for the rapid diagnosis of mesenteric ischemia.⁵ Capsule endoscopy is another method that has been widely used for the diagnosis of small-bowel diseases, such as occult gastrointestinal (GI) bleeding, sus-

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pected Crohn's disease, chronic diarrhea, protein-losing enteropathy and ischemic enteritis.⁶ However, capsule endoscopic images of ischemic enteritis have rarely been reported.⁷ We report herein a rare case of a 21-year-old man who was diagnosed with acute extensive ischemic enteritis using capsule endoscopy.

CASE REPORT

A 21-year-old male patient was admitted to the emergency department in Jeju National University Hospital due to sudden upper abdominal pain that had begun in the early morning of the same day. He had eaten seafood, including sushi, sashimi, oysters and squid, during the 5 days prior to this admission. The patient did not usually complain of intermittent abdominal pain after meals. In his medical history, he had suffered from pulmonary artery hypertension and bilateral carotid artery obstruction 5 years earlier and had received medications, such as calcium channel blockers, prostaticin analogues, diuretics and steroids, at a local clinic.

About 20 months before his admission, he had suffered from hematochezia for 4 days, and when angiography was performed twice at another hospital, the exact bleeding site was not found. Gastroscopy and colonoscopy did not show any bleeding sites. However, small amounts of melena were observed in the colon, suggestive of small bowel bleeding. Abdominal CT angiography revealed focal stenosis at celiac trunk, definite causes such as compression of adjacent organs or atherosclerosis were not found. In our hospital, capsule endoscopy showed mucosal edema and hyperemia in the terminal ileum, we thought that this lesion might be ischemic enteritis in the recovery phase (Fig. 1).

The patient was weakly positive for antinuclear antibody (1 : 40), but negative for antineutrophilic cytoplasmic antibodies (< 0.2), anticardiolipin antibody (IgG 0.20, IgM 0.10), and anti-beta 2 glycoprotein antibody (IgG 4.3, IgM 2.1). He showed the normal ranges of antithrombin III (77.3, normal 75-125%), proteins C (85, normal 73-132%) and S activity (76, normal 60-140%), and complements 3 (122, normal 90-180 mg/dL) and 4 (27, normal 10-40 mg/dL).

On admission, his blood pressure was 150/100 mmHg, pulse rate was 73/min, respiratory rate was 22/min, and body temperature was normal. There was direct and rebound tenderness in the epigastric and right upper abdominal



Fig. 1. Capsule endoscopic findings obtained 20 months earlier before admission. Mild mucosal edema with hyperemia in the terminal ileum (yellow arrow) suggested ischemic enteritis in the recovery phase.

areas, and his bowel sounds were normal. Routine blood test results were as follows: hemoglobin 16.9 g/dL, white blood cells 10,400/mm³, platelets 192,000/mm³, and high-sensitivity C-reactive protein 1.09 mg/dL (0.00-0.30 mg/dL). Serum chemical test results were as follows: albumin 3.9 g/dL, total bilirubin 0.7 mg/dL, ALP 298 U/L, AST/ALT 23/21 IU/L, total calcium 8.1 mg/dL, BUN 12.4 mg/dL, creatinine 1.1 mg/dL, amylase 28 IU/L and lipase 23 IU/L.

Abdominal pelvic CT showed focal celiac trunk stenosis and diffuse wall thickening of the small bowel loops and multiple enlarged mesenteric lymph nodes in the abdomen, which were suggestive of ischemic enteritis (Fig. 2). We planned to perform angiography for the evaluation of vascular diseases, but the patient refused the invasive procedure. On the third day, capsule endoscopy revealed diffuse hemorrhagic mucosal desquamation with severe edema, extending from the proximal jejunum to the distal small bowel. Focally scattered normal mucosae were seen. Fresh blood oozing into the lumen of the small bowel was observed. Multiple active ulcers with necrotic exudate were also observed. Capsule stasis occurred due to severe small bowel edema (Fig. 3). Thus, the patient was diagnosed acute extensive ischemic enteritis with capsule endoscopy. His condition improved with conservative treatment, and he was discharged after resuming oral intake on the fifth hospital day.

Four months later, the patient visited our emergency de-

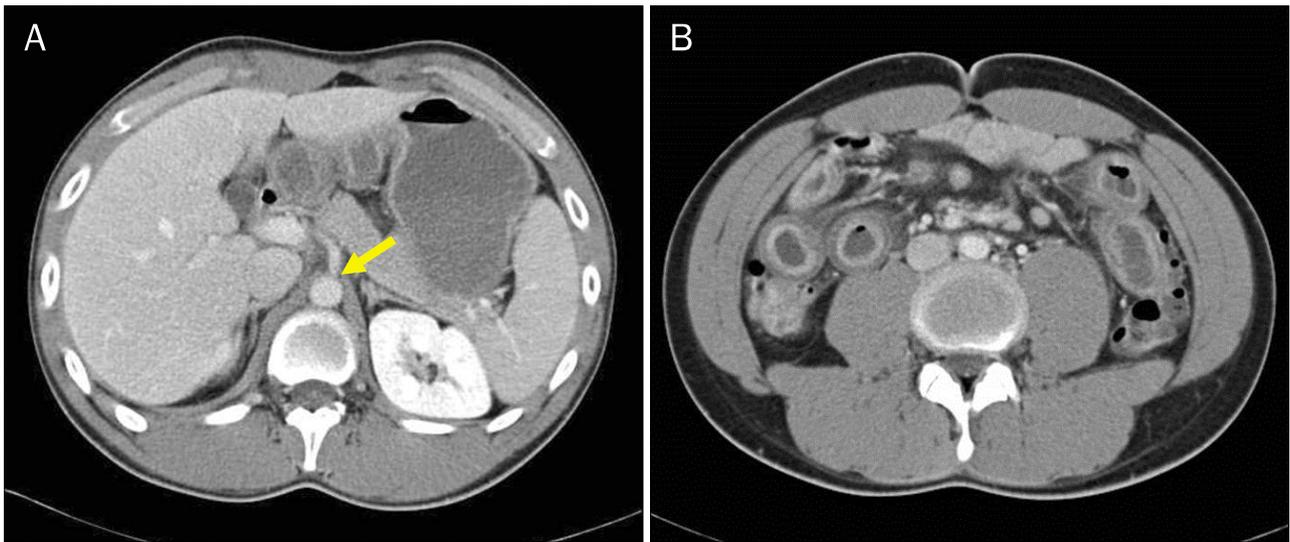


Fig. 2. Abdominal pelvic CT showing celiac trunk stenosis (A) (yellow arrow), diffuse wall thickening of the small bowel (B), which were suggestive of extensive ischemic enteritis.

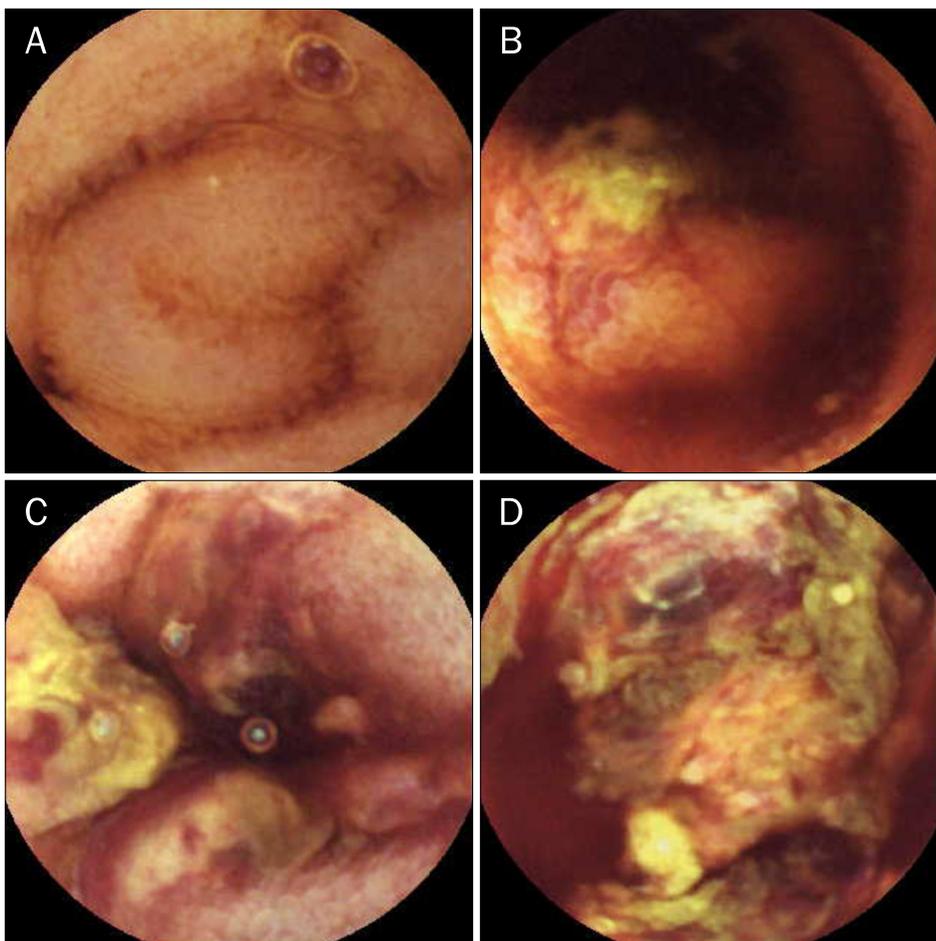


Fig. 3. Capsule endoscopy findings on admission. (A) Diffusely swollen mucosa was seen in the duodenum. (B) Active ulcers with exudates were noted in the proximal jejunum. (C, D) Severe mucosal hemorrhage with desquamation was seen, extending from the proximal jejunum to the terminal ileum.

partment again due to a left kidney infarction with complete renal artery obstruction. Three months thereafter, he was re-admitted due to pulmonary hemorrhage. He was suspected

of having vasculitis based on his multiple vascular problems. However, a definitive diagnosis has not yet been made because his unusually extensive vascular manifestations and

laboratory test results did not meet diagnostic criteria for vasculitis. Biopsy was not performed because the skin and nerves were not involved in our case. During the 13-month follow-up, there were no recurrent attacks of ischemic enteritis. The patient is currently receiving controlled doses of warfarin and steroids.

DISCUSSION

Ischemic enteritis can be classified as either occlusive or nonocclusive.¹ Embolism and thrombosis of the superior mesenteric artery causes this disease entity in 30% and 25% of all patients, respectively. Nonocclusive ischemic enteritis occurs in 25% of all patients.⁸ Small-bowel ischemia has been classified as extensive and segmental. Segmental ischemia is caused by various conditions, including a limited embolism or thrombosis in the context of hypercoagulable state, trauma, strangulation, collagen vascular diseases, vasculitis, radiation injury and drugs.³ Although embolism of the superior mesenteric artery usually occurs in the middle colic artery, small-intestinal branches are also sometimes occluded.¹ The jejunum is affected in 20% of all patient and the ileum in 45-55% of all patients.⁹ In our patient, although angiography was not performed on admission, CT scans showed no signs of vascular obstruction.

Ischemic enteritis usually develops in patients older than 60 years.¹ However, younger patients, especially those with diabetes mellitus, lupus erythematosus or sickle-cell anemia, may also present with ischemic enteritis.¹ Intestinal vasculitis usually occurs secondary to systemic vascular diseases, such as Buerger's disease, Behçet's disease, rheumatoid arthritis, and systemic lupus erythematosus, or in association with primary intestinal diseases such as Crohn's disease.¹⁰ There was no evidence of these diseases in his past medical history or test results. Our patient had been diagnosed with pulmonary artery hypertension and bilateral carotid artery obstruction 5 years earlier. After the ischemic enteritis event, he also had a left renal infarction and pulmonary hemorrhage. These vascular problems may have been associated with his recurrent ischemic enteritis, even though he was very young.

A characteristic symptom of mesenteric ischemia is severe abdominal pain that is inconsistent with physical findings. Laboratory studies are often not helpful in clinical

settings. Findings that support mesenteric ischemia include metabolic (lactate-based) acidosis, leukocytosis, and elevated levels of amylase or seromuscular markers.^{4,5}

Ischemic enteritis is a rapidly progressive disease with high mortality.⁴ Its early diagnosis is one of the most important factors determining the patient's prognosis, and conventional angiography and multidetector CT are useful in the diagnosis of mesenteric ischemia.⁵ The standardized use of angiography in patients with suspected mesenteric ischemia without peritoneal signs is supported by a number of studies that have shown reduced mortality as well as the high sensitivity and specificity of the procedure.¹¹ Multidetector CT angiography is reported to have a sensitivity of 93% and a specificity of 100% in the diagnosis of acute mesenteric ischemia when both vascular occlusions and the consequences of tissue damage are assessed.¹² In our patient, no signs of thrombosis, bleeding, or embolism were identified, when CT angiography was used to evaluate the hematochezia that he had suffered 20 months earlier. Because the patient had already undergone abdominal and pelvic CT, he did not undergo further angiography. Although the previous angiography identified no abnormalities, additional angiography on admission may have been helpful in evaluating the patient.

Capsule endoscopy and double balloon enteroscopy have become the methods of choice for the evaluation of small-bowel disorders, such as obscure GI bleeding, suspected Crohn's disease, chronic diarrhea, and protein-losing enteropathy.^{6,13} Capsule endoscopy has been shown to significantly modify diagnostic and therapeutic workups, to shorten the length of time to a definitive diagnosis, to reduce the number of further examinations, to reduce blood transfusion requirements, and to reduce the length of hospital stay.¹³⁻¹⁶ Although capsule endoscopy cannot treat lesions during the procedure, it is particularly useful when an accurate diagnosis is difficult to make based on imaging study results alone.

Endoscopic images of ischemic small bowel have rarely been reported. There was a report of capsule endoscopic findings of ischemic necrosis in the part of ileum in a 23 year-old male who had malignant hypertension.⁷ He underwent operation due to intramural inflammation, ulceration, and free perforation. Histologic examination of the resected ileum showed thrombosis and fibrinoid necrosis of the small arterioles, resulting in ulceration and necrosis of the mucosa.

Transient ischemic small-bowel ulcers secondary to acute superior mesenteric artery branch thromboembolism diagnosed by double balloon enteroscopy showed many ulcers geographic, linear or coin-like like form.¹⁷ A ischemic enteritis associated with polyarteritis nodosa (PAN) revealed dusky and purple mucosa with hemorrhagic ulcerations in the proximal jejunum.¹⁸ Our case showed diffuse hemorrhagic mucosal desquamation with severe edema with fresh blood oozing, extending from the proximal jejunum to the distal small bowel. Multiple active ulcers with necrotic exudate were also observed, which was compatible with ischemic enteritis. However, our patient was recovered with conservative treatment even though the extent of ischemic enteritis was wider.

Incidences of GI bleeding in patients suffering from systemic vascular diseases, such as PAN,¹⁸ Churg-Strauss syndrome,¹⁹ or systemic lupus erythematous²⁰ have been reported in the literature. In this case, the patient's multiple vascular obstructions and pulmonary vascular hemorrhage gave an important clue to vasculitis. Therefore, we suspected Churg-Strauss syndrome, PAN and Behçet's disease. The patient had a history of asthma and pulmonary infiltrates, but he did not have characteristic clinical manifestations of Churg-Strauss syndrome including peripheral blood eosinophilia, neuropathy, paranasal sinus abnormality or extravascular eosinophils. PAN is a multisystem, necrotizing vasculitis of small and medium-sized muscular arteries in which involvement of the renal and visceral arteries is characteristics.¹⁸ However, PAN does not involve the pulmonary arteries, it was ruled out due to the patient's pulmonary artery hypertension. The patient did not have oral or genital ulcers, eye inflammation, and skin lesions; above findings did not meet the criteria of Behçet's disease. Abdominal CT angiography revealed focal stenosis at celiac trunk, definite causes such as compression of adjacent organs or atherosclerosis were not found. Due to the fact that the patient's ischemic regions are the area supplied by the superior mesenteric artery, it is unlikely that stenosis at celiac trunk caused the ischemic enteritis. Therefore, even though the authors did not make the definite diagnosis, systemic vasculitis accompanied with pulmonary hypertension, internal carotid artery and left renal artery obstruction might cause ischemic enteritis.

We reported a case of a 21-year-old man who was diagnosed with recurrent acute extensive ischemic enteritis associated with systemic vasculitis using capsule endoscopy.

REFERENCES

1. Delikoukos S, Christodoulidis G, Zacharoulis D, Poultisidi A, Hatzitheofilou C. Multiple small bowel ruptures due to ischemic enteritis: a case report. *World J Gastroenterol* 2006;12:4262-4263.
2. Díaz Nieto R, Varcada M, Ogunbiyi OA, Winslet MC. Systematic review on the treatment of ischaemic colitis. *Colorectal Dis* 2011;13:744-747.
3. Nishimura N, Yamamoto H, Yano T, et al. Balloon dilation when using double-balloon enteroscopy for small-bowel strictures associated with ischemic enteritis. *Gastrointest Endosc* 2011;74:1157-1161.
4. Clavien PA. Diagnosis and management of mesenteric infarction. *Br J Surg* 1990;77:601-603.
5. Renner P, Kienle K, Dahlke MH, et al. Intestinal ischemia: current treatment concepts. *Langenbecks Arch Surg* 2011;396:3-11.
6. Yang XY, Chen CX, Zhang BL, et al. Diagnostic effect of capsule endoscopy in 31 cases of subacute small bowel obstruction. *World J Gastroenterol* 2009;15:2401-2405.
7. Liatsos C, Goulas S, Karagiannis S, Patelaros E, Sabaziotis D, Mavrogiannis C. Diagnosis of small-bowel ischemic necrosis by capsule endoscopy. *Gastrointest Endosc* 2005;62:439-440.
8. Levy PJ, Krausz MM, Manny J. Acute mesenteric ischemia: improved results—a retrospective analysis of ninety-two patients. *Surgery* 1990;107:372-380.
9. Kitchens CS. Evolution of our understanding of the pathophysiology of primary mesenteric venous thrombosis. *Am J Surg* 1992;163:346-348.
10. Eryigit E, Hoentjen F, Barbe E, van Meyel JJ. Intestinal ischaemia caused by mesenteric inflammatory veno-occlusive disease. *Neth J Med* 2008;66:486-488.
11. Brandt LJ, Boley SJ. AGA technical review on intestinal ischemia. American Gastrointestinal Association. *Gastroenterology* 2000;118:954-968.
12. Aschoff AJ, Stuber G, Becker BW, et al. Evaluation of acute mesenteric ischemia: accuracy of biphasic mesenteric multi-detector CT angiography. *Abdom Imaging* 2009;34:345-357.
13. Mönkemüller K, Bellutti M, Malfertheiner P. Small-bowel endoscopy. *Endoscopy* 2007;39:978-985.
14. Pennazio M, Santucci R, Rondonotti E, et al. Outcome of patients with obscure gastrointestinal bleeding after capsule endoscopy: report of 100 consecutive cases. *Gastroenterology* 2004;126:643-653.
15. Rondonotti E, Villa F, Mulder CJ, Jacobs MA, de Franchis R. Small bowel capsule endoscopy in 2007: indications, risks and limitations. *World J Gastroenterol* 2007;13:6140-6149.
16. Chan FS, Chu KM. Capsule endoscopy for gastrointestinal bleeding of obscure origin. *Asian J Surg* 2008;31:96-99.
17. Hashimoto Y, Endo Y, Kuroki Y, Yoshikumi H, Yoshiba M. Transient ischemic small-bowel ulcers secondary to acute superior mesenteric artery branch thromboembolism diagnosed by double balloon enteroscopy. *Endoscopy* 2008;40(Suppl 2):E161.
18. Lee KH, Park JM, Chun SW, et al. A case of acute ischemic enteritis caused by polyarteritis nodosa. *Korean J Gastrointest*

Endosc 2006;33:303-306.

19. Park JH, Jung YS, Kim YK, et al. A case of Churg-Strauss syndrome with interstitial perforation. *Tuberc Respir Dis* 2009;66: 374-379.
20. Lin HP, Wang YM, Huo AP. Severe, recurrent lupus enteritis as the initial and only presentation of systemic lupus erythematosus in a middle-aged woman. *J Microbiol Immunol Infect* 2011;44: 152-155.