# [ CASE REPORT ]

# Treating an Intractable Jejunocutaneous Fistula by **Endoscopic Metallic Stent Placement: A Case Report of** Successful Palliative Endoscopic Treatment in a Case **Demonstrating Peritoneal Dissemination with Terminal Stage Gastric Cancer**

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#### **Abstract:**

A 74-year-old woman with recurrent gastric cancer underwent laparotomy for peritoneal dissemination, and the damaged jejunum formed a jejunocutaneous fistula. Because conservative treatment alone could not cure the fistula, we performed an endoscopic placement of a partially covered self-expandable metallic stent (SEMS) to cover the fistula. After the procedure, the contrast medium no longer leaked from the intestinal lumen. One month after stent placement, the cutaneous opening had closed. This case report demonstrates the potential for using partially covered SEMS to treat intractable jejunocutaneous fistula in patients with terminal-stage malignant tumors.

Key words: jejunocutaneous fistula, self-expandable metallic stent, terminal stage, palliative endoscopic treatment

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# Introduction

Endoscopic self-expandable metallic stent (SEMS) placement is a commonly used palliative endoscopic treatment that is safe and effective for patients with malignant gastrointestinal obstruction (1-4). SEMSs are also used to close the leakage of gastrointestinal fistulas or perforations (5, 6). We previously reported two related cases: namely, a successful closure of esophageal perforations, occurring due to endoscopic submucosal dissection and spontaneous esophageal rupture, respectively, by endoscopic covered SEMS placement (7, 8).

A jejunocutaneous fistula is a condition in which the contents of the small intestine leak through an opening in the

skin. The persistent leakage of intestinal contents results in intractable fistulas, which can lead to malnutrition, electrolyte abnormalities, dehydration, and skin disorders (9). Inflammatory bowel disease, poor suturing during intestinal surgery, and malignancy are the common causes of enterocutaneous fistulas.

We herein report a case of intractable jejunocutaneous fistula treated successfully with the endoscopic placement of a covered SEMS in a patient demonstrating peritoneal dissemination with terminal gastric cancer. We propose this treatment as an option to minimize both surgical risk and invasiveness for terminal cancer patients having a low risk of complications.

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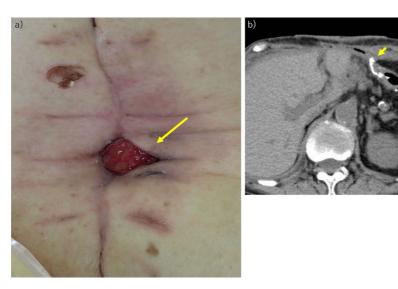


Figure 1. a) Cutaneous opening of the jejunocutaneous fistula (yellow arrow). b) Computed tomography image of the jejunocutaneous fistula (yellow arrow).

**Table.** Comparison of the Blood Test Results Obtained before and One Month after SEMS Placement.

Investigation, unit	Before SEMS placement	One month after SEMS placement
White blood cell count, /μL	4,870	3,500
Red blood cell count, ×10 <sup>4</sup> /μL	319	444
Hemoglobin, g/dL	8.0*	12.5
Platelet count, ×10 <sup>3</sup> /μL	235	171
Total protein, g/dL	5.5*	6.6*
Albumin, g/dL	1.5*	2.9*
Na+, mEq/L	139	139
K+, mEq/L	3.4*	4.4
Cl-, mEq/L	98*	104
C-reactive protein, mg/dL	6.89*	0.20*

<sup>\*</sup>Outside the reference range.

All tests were performed at the Osaka Medical and Pharmaceutical University Hospital.

SEMS: self-expandable metallic stent

## **Case Report**

A 74-year-old woman, who had previously undergone total gastrectomy for advanced gastric cancer 10 years previously, underwent three laparotomies: an intestinal resection, bypass, and ileostomy for peritoneal dissemination caused by a recurrence of gastric cancer. During the total gastrectomy performed 10 years previously, the gastric cancer was found to have penetrated the serosa, but no regional lymph node or distant metastasis was observed. In addition, the patient had received S-1 chemotherapy (tegafur/gimeracil/ oteracil tablets, 100 mg/day) for 1 year as postoperative adjuvant chemotherapy. After the third laparotomy, an abnormal connection developed between the damaged part of the elevated jejunum and the abdominal wall at the site of the skin incision, resulting in a jejunocutaneous fistula (Fig. 1). Conservative treatment, such as fasting and drainage, were attempted, but the leakage of intestinal contents continued resulting in the development of an intractable fistula. In addition, the patient became emaciated and developed electrolyte abnormalities, thereby indicating the limitations of conservative treatment. Due to her condition, she was deemed ineligible for surgery (Table). It was considered that if the fistula was not closed by performing a minimally-invasive procedure, her malnutrition would worsen, possibly leading to her death.

She was transferred to our hospital for endoscopy. Peroral intestinal endoscopy revealed a stricture, 5 cm from the anal side of the esophageal-jejunal anastomosis, through which the scope could not pass (Fig. 2a). Endoscopy performed after administering contrast (amidotrizoic acid; Gastrografin®, Bayer Yakuhin, Osaka, Japan) revealed a fistula on the anal side of the stricture and contrast leakage from its cutaneous opening was also confirmed (Fig. 2b). We decided to perform the endoscopic placement of a partially covered SEMS (Niti-S Duodenal ComVi Stent; diameter, 20 mm; length, 8 cm; Century Medical, Tokyo, Japan) to cover the fistula

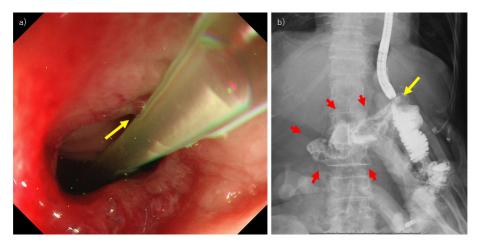


Figure 2. a) Peroral intestinal endoscopy revealed a stricture 5 cm from the anal side of the esophageal-jejunal anastomosis through which the scope could not pass; a yellow arrow indicates the fistula. b) Endoscopy performed after contrast administration revealed a fistula on the anal side of the stricture and contrast leakage was confirmed from the cutaneous opening. The yellow and red arrows indicate the fistula and the leakage of the contrast medium, respectively.

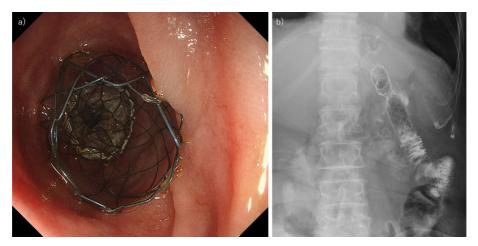


Figure 3. a) A partially covered self-expandable metallic stent was endoscopically placed to cover the fistula. b) After the procedure, the leakage of the contrast medium from the intestinal lumen was found to have stopped.

(Fig. 3). The leakage of contrast medium from the intestinal lumen stopped after the procedure. The oral intake of gruel was started one week after the procedure, and there was no further outflow of residue from the fistula.

One month after stent placement, the cutaneous opening had closed (Fig. 4). An improvement in chronic inflammation, electrolyte abnormalities, and malnutrition was observed on laboratory investigations (Table). She later developed small bowel obstruction owing to adhesion formation and a re-exacerbation of peritoneal dissemination. Her condition was considered stable enough to tolerate surgery and she therefore underwent reoperation to relieve the obstruction by the dissection of adhesions. SEMS was removed at the time of reoperation because it had migrated to the anal side as it had become loosened due to bowel dilatation proximal to the site of obstruction. The natural disease progression of terminal stage gastric cancer led to the patient's death three months later.

### **Discussion**

This report describes the effectiveness of using a partially covered SEMS for treating intractable jejunocutaneous fistula in a patient with a terminal-stage malignant tumor. Although SEMSs have previously been reported to be useful in treating leakage from esophageal or intestinal fistulas or perforations, using SEMSs for intractable jejunocutaneous fistula has not yet been previously discussed (5-8).

Patients with jejunocutaneous fistulas are difficult to manage due to the continuous drainage of intestinal contents, a worsening nutritional status, and electrolyte imbalance. A previous Japanese report described the endoscopic treatment of jejunocutaneous fistula using an over-the-scope clip (OTSC) (10). We consider OTSC placement to be an effective treatment for intractable jejunocutaneous fistula in patients without intestinal strictures. However, the present case

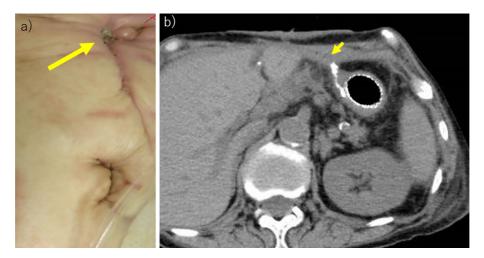


Figure 4. a) One month after the stent placement, the cutaneous opening of the fistula had closed. The yellow arrow indicates the scar at the site of the cutaneous opening. b) Computed tomography image showing the remaining jejunocutaneous fistula (yellow arrow). Compared to Fig. 1b, the presence of subcutaneous air had disappeared.

was complicated by the presence of stricture at the oral side of the fistula; therefore, OTSC was not deemed to be a viable option for our patient. There are two case reports of the successful endoscopic closure of anastomo-cutaneous and intrathoracic esophageal fistulas using polyglycolic acid sheets (11, 12). The clinical features of these cases are comparable to those of the present case and usage of polyglycolic acid sheets would likely have been effective in the absence of gastrointestinal stenosis. Non-invasive treatment using negative pressure wound therapy and fistuloclysis has previously been reported to be effective for the treatment of intractable jejunocutaneous fistulas (13). However, these procedures take time to completely heal the fistula and thus may not be suitable for patients with carcinoma and a limited life expectancy, as in the present case. Although reoperation may be an effective option, it was unsuitable in the present case due to the possibility of another fistula formation after re-operation due to her low nutritional status. The preoperative administration of oral arginine and glutamine may be valuable in patients with enterocutaneous fistulas (14). However, it may not be suitable for such patients because the persistent leakage of digestive juices may extend the time needed to improve their nutritional status.

Although endoscopic SEMS placement is a relatively safe procedure, the risk of stent migration remains. When there is an intestinal stricture near the fistula, as in the present case, the operator should ensure that the stent remains firmly in place to reduce the migration risk. This procedure may also be preferred to others in terms of improving the patient's quality of life in the terminal stage of cancer because oral intake can be resumed early after this procedure. In this case, the fistula closed spontaneously after resuming oral intake; subsequently, the patient's nutritional status improved.

In conclusion, our approach of using a partially covered SEMS for intractable jejunocutaneous fistula in patients with terminal-stage malignant tumors can serve as an effective palliative endoscopic treatment. Such patients can expect a better quality of life due to a faster recovery and return to an oral diet. However, this procedure may be associated with a few complications and its suitability should therefore be determined on a case-by-case basis.

The authors state that they have no Conflict of Interest (COI).

### References

- Yukimoto T, Morisaki T, Komukai S, et al. The palliative effect of endoscopic uncovered self-expandable metallic stent placement versus gastrojejunostomy on malignant gastric outlet obstruction: a pilot study with a retrospective chart review in Saga, Japan. Intern Med 57: 1517-1521, 2018.
- Hamada T, Hakuta R, Takahara N, et al. Covered versus uncovered metal stents for malignant gastric outlet obstruction: systematic review and meta-analysis. Dig Endosc 29: 259-271, 2017.
- 3. Kim CG, Choi IJ, Lee JY, et al. Covered versus uncovered self-expandable metallic stents for palliation of malignant pyloric obstruction in gastric cancer patients: a randomized, prospective study. Gastrointest Endosc 72: 25-32, 2010.
- Tringali A, Didden P, Repici A, et al. Endoscopic treatment of malignant gastric and duodenal strictures: a prospective, multicenter study. Gastrointest Endosc 79: 66-75, 2014.
- 5. El-Sourani N, Miftode S, Bockhorn M, Arlt A, Meinhardt C. Endoscopic management of anastomotic leakage after esophageal surgery: ten year analysis in a Tertiary University Center. Clin Endosc 55: 58-66, 2022.
- Lee JH, Kedia P, Stavropoulos SN, Carr-Locke D. AGA clinical practice update on endoscopic management of perforations in gastrointestinal tract: expert review. Clin Gastroenterol Hepatol 19: 2252-2261.e2, 2021.
- Ota K, Takeuchi T, Ozaki H, Harada S, Kojima Y, Higuchi K. Temporary insertion of a covered self-expandable metal stent to treat esophageal perforation due to endoscopic submucosal dissection. Intern Med 54: 1049-1052, 2015.
- **8.** Ota K, Takeuchi T, Higuchi K. Temporary insertion of a covered self-expandable metal stent for spontaneous esophageal rupture. Dig Endosc **26**: 607-608, 2014.

- Niu DG, Yang F, Tian WL, et al. A technique to establish fistuloclysis for high-output jejunocutaneous fistula through percutaneous enterostomy: a case report. Medicine (Baltimore) 98: e14653, 2019.
- 10. Nishiwaki S, Baba A, Wakayama T, et al. A case of persistent je-junocutaneous fistula successfully treated using an over-the-scope clip with guidance by a guidewire. Gastroenterol Endosc 59: 2719-2724, 2017 (in Japanese).
- 11. Kawabata H, Okazaki Y, Inoue N, et al. Endoscopic closure of an anastomo-cutaneous fistula: filling and shielding using polygly-colic acid sheets and fibrin glue with easily deliverable technique. Endosc Int Open 6: E994-E997, 2018.
- 12. Kida A, Mizukami A, Matsuda K, Matsuda M, Sakai A. Successful treatment of pleural empyema caused by an intrathoracic esophageal fistula after iatrogenic esophageal perforation with

- polyglycolic acid sheets. Gastrointest Endosc 89: 639-640, 2019.
- 13. Hirose W, Murakami K, Usuda M, et al. A case of intractable jejunocutaneous fistula treated by negative-pressure wound therapy and fistuloclysis. Geka To Taisha Eiyou (Jpn J Surg Metab Nutr) 54: 92-96, 2020 (in Japanese).
- 14. Martinez JL, Bosco-Garate I, Souza-Gallardo LM, et al. Effect of preoperative administration of oral arginine and glutamine in patients with enterocutaneous fistula submitted to definitive surgery: a prospective randomized trial. J Gastrointest Surg 24: 426-434, 2020.

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