

CASE REPORT

경구 내시경 근절개술을 이용한 내시경점막하박리술에 의해 성공적으로 제거된 십이지장 신경내분비종양 1예

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Successful Endoscopic Submucosal Dissection Using Open Peroral Endoscopic Myotomy for Duodenal Neuroendocrine Tumor

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Duodenal neuroendocrine tumors (NETs) are subepithelial tumors that are difficult to remove endoscopically, particularly when located just beyond the pylorus. This paper reports a case of a successful endoscopic submucosal dissection (ESD) using open gastric peroral endoscopic myotomy (POEM) for a remnant duodenal NET detected after endoscopic mucosal resection (EMR). A 67-year-old male presented with a 5 mm remnant duodenal NET close to the pylorus after EMR for a duodenal polypoid lesion performed four months earlier. Duodenal ESD was performed under conscious sedation using I-type and IT II knives. The tumor adhered to the fibrotic tissue, and the submucosal cushion was insufficient. Open gastric POEM was performed concurrently during ESD, resulting in the complete resection of the NET. This case suggests that while challenging, open gastric POEM can serve as a valuable technique for endoscopic resection in cases of early gastric cancer or duodenal masses located around the pylorus. (*Korean J Gastroenterol* 2024;83:61-64)

Key Words: Neuroendocrine tumors; Endoscopic submucosal dissection; Peroral endoscopic myotomy; Duodenum

INTRODUCTION

Duodenal neuroendocrine tumors (NETs) are rare, accounting for approximately 2-3% of all NETs.^{1,2} The incidence of NETs has increased in recent years, coinciding with the widespread use of upper gastrointestinal endoscopy. In response, the European Neuroendocrine Tumor Society (ENETS) has advocated the removal of small non-functioning duodenal NET lesions, specifically those measuring 5 mm or less, rather than opting for surveillance. Endoscopic mucosal resection

(EMR) and endoscopic submucosal dissection (ESD) are the primary treatment modalities for tumors less than 15 mm in size.¹

The duodenum, characterized by its thin wall and division into four distinct anatomical parts, presents unique challenges owing to the varied anatomy. The complication rate associated with endoscopic resection in the duodenum is relatively high compared to that in the stomach.³ NET, a subepithelial tumor, poses challenges for endoscopic removal, especially when located just beyond the pylorus.

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This paper presents a case of successful ESD using gastric open peroral endoscopic myotomy (POEM) for a remnant duodenal NET detected after EMR.

CASE REPORT

A 67-year-old male was referred to the authors' outpatient clinic. Four months earlier, the patient underwent EMR for a duodenal polypoid lesion. The tumor, measuring 10 mm, was diagnosed histologically as a duodenal NET (grade I) with invasion of the submucosa and mucosa. No evidence of mitosis, lymphatic, or venous invasion was noted. Immunostaining revealed a Ki-67 index of less than 1%.

He also had a history of diabetes mellitus. The laboratory tests indicated normal values with well-controlled blood sugar. The urine 5-HIAA levels were within the normal range. Abdominal computed tomography showed no lymph node metastasis. Esophagogastroduodenoscopy revealed a duodenal NET measuring 5 mm in size just beyond the pylorus (Fig. 1A).

The open gastric POEM was pre-planned before initiating ESD, considering the fibrosis caused by the prior EMR. Duodenal ESD was performed under moderate sedation using

midazolam, propofol, and fentanyl with 3.5 mg, 50 mg, and 100 mcg doses intravenously, respectively. The procedure was performed using an I-type knife (Finemedix, Daegu, Korea) and an II knife (insulated-tip diathermic knife II; Olympus, Tokyo, Japan). The I-type knife was used for marking, producing a round incision, and performing a submucosal dissection (Fig. 1B). Pyloric myotomy was performed using an IT II knife until the pyloric contraction disappeared. The dissection plane was straightened (Fig. 1C). Despite the tumor adhering to some fibrotic tissue and an insufficient submucosal cushion, ESD was conducted successfully using POEM, resulting in complete resection (Fig. 1D). The entire procedure was performed by one skilled endoscopist with experience of more than 5,000 cases of ESD and 20 cases of gastric POEM. The procedure time for ESD and POEM was 20 and 5 minutes, respectively. The resected specimen measured 20 mm (Fig. 2A). The pathology examination revealed a neuroendocrine tumor (grade I) measuring 2 mm without a lymphovascular invasion (Fig. 2B-D).

No complications occurred post-ESD. A follow-up upper series on the second day excluded duodenal leakage, and the patient resumed a regular diet. Subsequent endoscopy performed two months later revealed no recurrence of the NET.

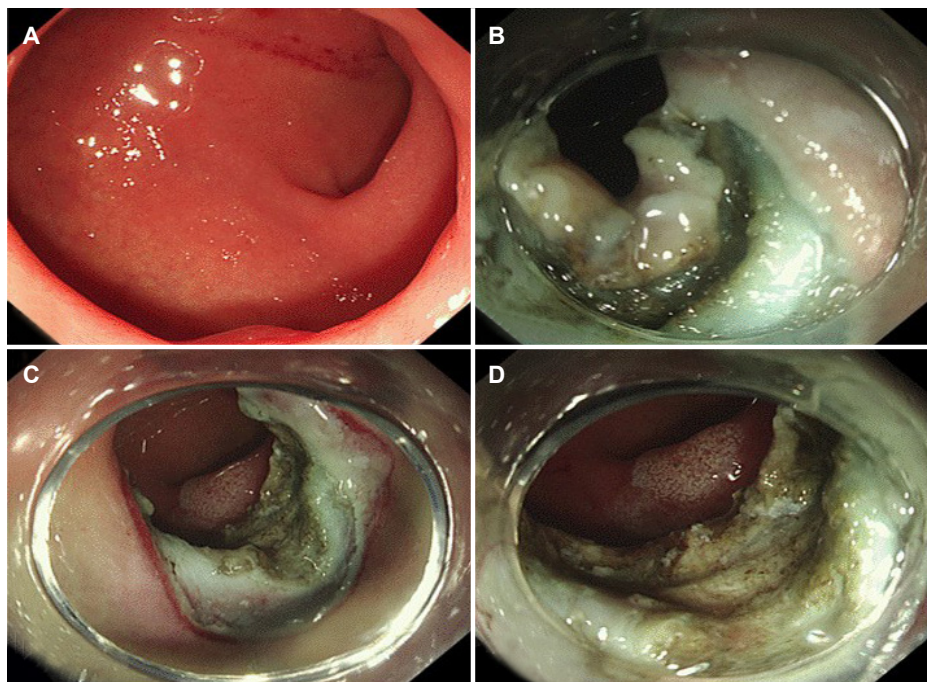


Fig. 1. Endoscopic submucosal dissection (ESD) using gastric open peroral endoscopic myotomy (POEM). (A) Remnant tumor in the duodenum, near the pylorus. (B) Endoscopic submucosal dissection could not proceed to duodenum. (C) Gastric open POEM was achieved. (D) ESD was completed successfully.

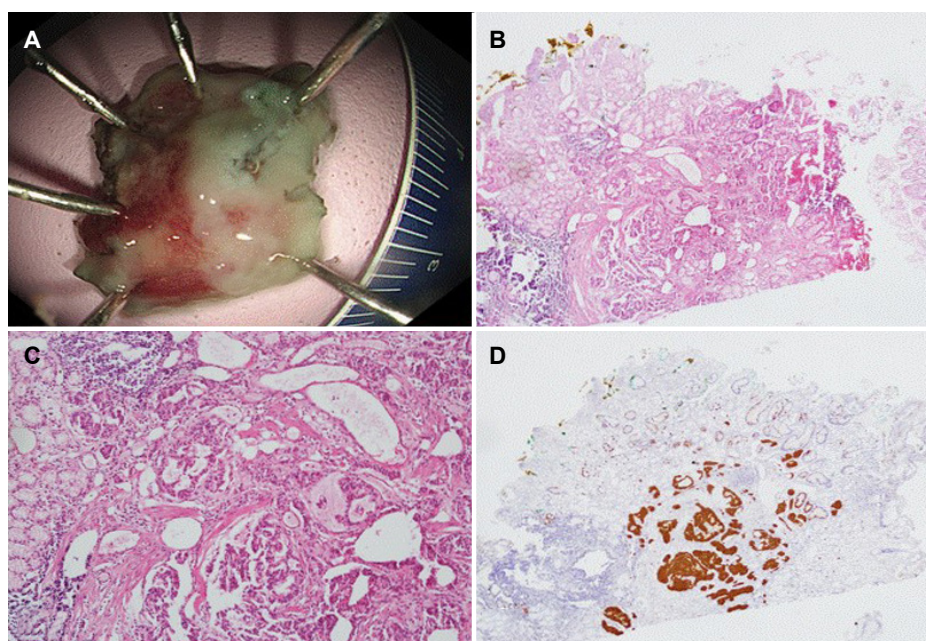


Fig. 2. Resected specimen. (A) En bloc resection was performed. (B) Hematoxylin and Eosin (H&E) staining shows a microscopic view of the neuroendocrine tumor (×100). (C) High power demonstrated nests of bland tumor cells with round to oval nuclei (H&E, ×200). (D) Synaptophysin positivity by immunohistochemistry was noted (Diaminobenzidine, ×40).

DISCUSSION

Duodenal NET are malignant tumors that require early intervention. It is classified into five clinical subtypes, with more than 90% of cases categorized as non-functioning duodenal NETs.^{2,4} These tumors are often asymptomatic and are typically found in the first and second parts of the duodenum.

The endoscopic findings included small polypoid lesions or hemispherical subepithelial tumors, with or without central depression. The tumor surface may exhibit a reddish or yellowish color. The diagnosis was established using an endoscopic biopsy. Endoscopic ultrasonography is recommended for tumors >10 mm to assess deep invasion and metastasis.¹

The absence of standardized treatment modalities stems from the lack of long-term surveillance research.^{5,6} The National Comprehensive Cancer Network (NCCN) recommends an endoscopic resection as the preferred treatment for well-located duodenal tumors lacking evidence of deep or lymphatic invasion.^{2,5}

While EMR is a safe and minimally invasive therapeutic option, its en bloc resection rates are inferior to ESD.^{6,7} In cases of an incomplete resection (R1 resection), the ENETS considered ESD to be feasible. Despite being a valuable method for duodenal mass removal, ESD encounters challenges

with tumors in anatomically difficult regions, such as the pyloric canal. In this case, the lesion was located immediately beyond the pylorus, presenting limited visibility exacerbated by pyloric contractions. Endoscopic handling of a submucosal dissection is a major obstacle in the pylorus-duodenum. This necessitates a new approach for achieving higher complete resection rates and lower complication rates.

POEM, utilized for achalasia and refractory gastroparesis, involves myotomy within the submucosal tunnel, with mucosal entry closure using a metallic clip.⁸⁻¹⁰ Open POEM is a modified technique achieved without a submucosal tunnel and has been reported for achalasia with submucosal fibrosis.¹¹ Recently, the authors' team reported the efficacy of open gastric POEM for preventing stenosis after ESD. This approach involves direct endoscopic visualization of the circular muscle and cutting the pyloric ring during or immediately after ESD.¹² This prevents pyloric contraction, facilitating submucosal dissection. In the present case, improvements in the sight of view and technical processes enabled straightforward ESD using an open gastric POEM, even with some fibrosis caused by prior EMR.

The open gastric POEM technique has not yet been standardized. The inner circular muscle cutting may vary in length according to the lesion site or the absence of pyloric

contraction. Although complications, such as bleeding or perforation, are possible, their risk is minimal, according to a recent report.¹² Therefore, open gastric POEM can be a challenging procedure in endoscopic therapy for early gastric cancer or duodenal masses around the pylorus.

This paper reported a successful endoscopic dissection using open gastric POEM for duodenal NET near the pylorus. Nevertheless, further studies will be needed to determine the safety and efficacy of open gastric POEM.

REFERENCES

1. Panzuto F, Ramage J, Pritchard DM, et al. European Neuroendocrine Tumor Society (ENETS) 2023 guidance paper for gastroduodenal neuroendocrine tumours (NETs) G1-G3. *J Neuroendocrinol* 2023;35:e13306.
2. Hoffmann KM, Furukawa M, Jensen RT. Duodenal neuroendocrine tumors: classification, functional syndromes, diagnosis and medical treatment. *Best Pract Res Clin Gastroenterol* 2005;19:675-697.
3. Fujihara S, Mori H, Kobara H, et al. Management of a large mucosal defect after duodenal endoscopic resection. *World J Gastroenterol* 2016;22:6595-6609.
4. Mandair D, Kamieniarz L, Pizanias M, et al. Diagnostic features and management options for duodenal neuroendocrine neoplasms: a retrospective, multi-centre study. *Sci Rep* 2022;12:15762.
5. Jung SW. Endoscopic treatment of gastric and duodenal neuroendocrine tumors: present and future. *Korean J Helicobacter Up Gastrointest Res* 2021;21:29-34.
6. Kim GH, Kim JI, Jeon SW, et al. Endoscopic resection for duodenal carcinoid tumors: a multicenter, retrospective study. *J Gastroenterol Hepatol* 2014;29:318-324.
7. Mahmud N, Tomizawa Y, Stashek K, Katona BW, Ginsberg GG, Metz DC. Endoscopic resection of duodenal carcinoid tumors: a single-center comparison between simple polypectomy and endoscopic mucosal resection. *Pancreas* 2019;48:60-65.
8. Bechara R, Ikeda H, Inoue H. Peroral endoscopic myotomy: an evolving treatment for achalasia. *Nat Rev Gastroenterol Hepatol* 2015;12:410-426.
9. Inoue H, Minami H, Kobayashi Y, et al. Peroral endoscopic myotomy (POEM) for esophageal achalasia. *Endoscopy* 2010;42:265-271.
10. Khashab MA, Stein E, Clarke JO, et al. Gastric peroral endoscopic myotomy for refractory gastroparesis: first human endoscopic pyloromyotomy (with video). *Gastrointest Endosc* 2013;78:764-768.
11. Liu W, Zeng XH, Yuan XL, et al. Open peroral endoscopic myotomy for the treatment of achalasia: a case series of 82 cases. *Dis Esophagus* 2019;32:1-7.
12. Lee WD, Song JS, Kim BS, et al. Safety and efficacy of prophylactic gastric open peroral endoscopic myotomy for prevention of post-ESD stenosis: a case series (with video). *J Dig Dis* 2022;23:220-227.