

## Ligation-assisted endoscopic submucosal resection with circumferential mucosal incision for duodenal carcinoid tumor

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### Abstract

Here we present the case of a 64-year-old female with a duodenal carcinoid tumor treated by ligation-assisted endoscopic submucosal resection (ESMR-L) with circumferential mucosal incision (CMI). Band ligation was effective in resecting the duodenal carcinoid tumor after CMI, with an uneventful post-procedural course. Histopathological examination showed clear tumor margins at deeper tissue levels. Thus, in the present case, ESMR-L with CMI was useful for the treatment of duodenal carcinoid tumor.

**Key words:** Carcinoid tumor; Submucosal tumor; Duodenum; Ligation-assisted endoscopic submucosal resection; Endoscopic mucosal resection; Endoscopic submucosal dissection; Ligation device

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**Core tip:** This report presents the case of a 64-year-old female with duodenal carcinoid tumor treated by ligation-assisted endoscopic submucosal resection (ESMR-L) with circumferential mucosal incision (CMI). Band ligation was effective in resecting the duodenal carcinoid tumor after CMI. The post-procedural course was uneventful without bleeding and perforation. Histopathological examination showed clear tumor margins at deeper tissue levels. Thus, ESMR-L with CMI can be effective for the management of duodenal

carcinoid tumors.

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## INTRODUCTION

Carcinoid tumors are relatively rare, slow-growing tumors that show low-grade atypia. Typically, gastrointestinal carcinoid tumors are found incidentally during screening esophagogastroduodenoscopy (EGD) and colonoscopy<sup>[1]</sup>, with the most common location being the rectum, followed by the stomach, duodenum, and appendix. Carcinoid tumors also have excellent 5-year survival rates of 98.9%-100% after curative resection, provided there is no vessel infiltration, they are within the submucosa, and they measure < 10 mm in diameter<sup>[2-5]</sup>. Therefore, conventional endoscopic mucosal resection (EMR) is usually sufficient for the resection of small submucosal carcinoid tumors diagnosed by endoscopic ultrasound (EUS). However, those at deeper locations within the submucosa are not always amenable to complete resection by conventional EMR.

Some studies have recently reported that ligation-assisted endoscopic submucosal resection (ESMR-L) and endoscopic submucosal dissection (ESD) can facilitate complete resection of carcinoid tumors<sup>[6-8]</sup>. Furthermore, the efficacy of ligation-assisted endoscopic enucleation (EE-L) has been reported for the treatment of submucosal tumors (*e.g.*, leiomyomas and gastrointestinal stromal tumors)<sup>[9]</sup>. However, in cases where the submucosa has significant fibrosis, it may be impossible to dissect the fibrosis by ESD alone or to suck the tumor through a ligation device. In this setting, ESMR-L with circumferential mucosal incision (CMI) may be effective. In this report, we therefore describe our positive experience using this technique to treat a duodenal carcinoid tumor identified by mass screening EGD.

## CASE REPORT

A 64-year-old female underwent a screening EGD that revealed a 7-mm submucosal tumor in the anterior wall of the duodenal bulb (Figure 1). EUS revealed the lesion to be a 6 mm × 5 mm hypoechoic, uniform tumor with a regular margin within the third layer. Therefore, we performed ligation-assisted ESMR with CMI after obtaining written informed consent from the patient.

The procedure was performed as follows. Firstly,



Figure 1 A 7-mm carcinoid tumor presents in the anterior wall of the duodenal bulb.

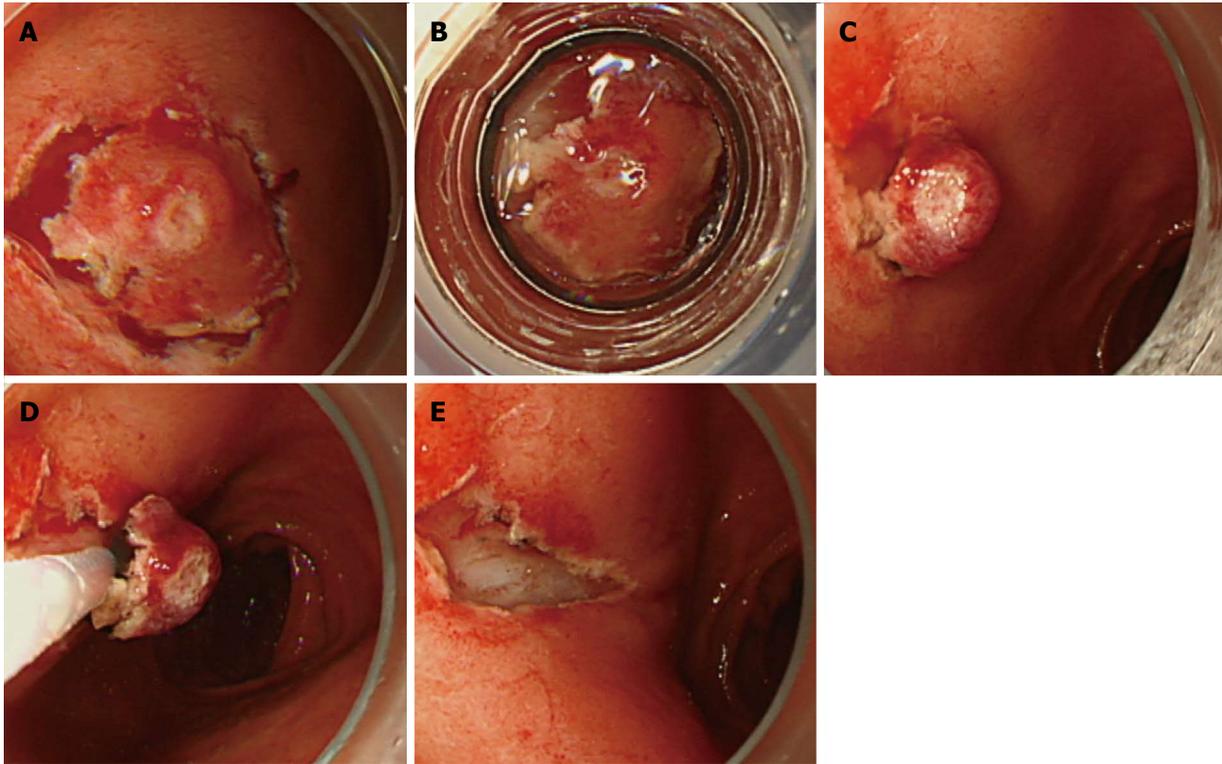
to elevate the tumor, we injected sodium hyaluronate in the underlying submucosa. After lifting the mucosa, we used a FlushKnife BT (DK2618JB; Fujifilm, Tokyo, Japan) to make a hemircumferential mucosal incision around the tumor. Although we attempted ESD, it was not possible because of fibrosis. Therefore, in addition to the hemircumferential mucosal incision (Figure 2A), we suctioned the tumor with a ligation device (pneumo-activate EVL device; Sumitomo Bakelite, Tokyo, Japan) (Figure 2B), which changed the shape of the tumor to a pseudopolyp that was suitable for band ligation (Figure 2C). Next, we performed polypectomy beneath the rubber band using a snare in ENDOCUT mode (Figures 2D and E). Histopathological examination revealed a G1 neuroendocrine tumor measuring 5.5 mm × 4.6 mm with clear tumor margins at deep tissue levels (the tumor cells were positive for chromogranin-A, synaptophysin, and CD56) (Figure 3). The post-procedural course was uneventful without bleeding and perforation.

## DISCUSSION

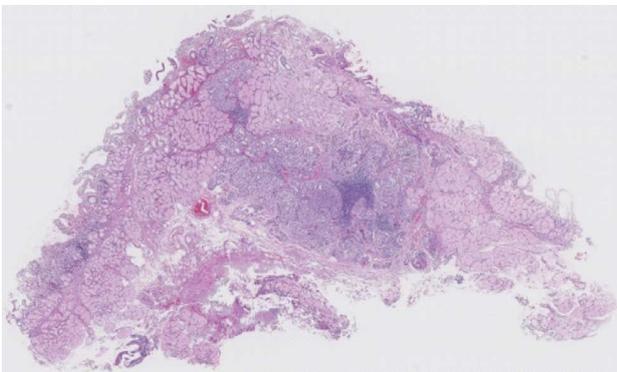
Recently, the efficacy of ESD has been reported for carcinoid tumors that have not spread beyond the submucosa. However, as observed in the present case, submucosal dissection may be impossible when complicated by severe fibrosis. Although histopathological examination of the resected tumor failed to show severe fibrosis, we believe this resulted from the influence of hyperplasia of Brunner's glands. It is believed that submucosal injection is particularly difficult in the duodenal bulb in comparison with application in other parts of the duodenum because of the presence of many Brunner's glands.

Another important consideration is the report by Shida *et al.*<sup>[10]</sup> who performed ESD with a ligation device (ESD-L). In our opinion, naming the procedure as ESD-L is not appropriate because it is not performed to dissect the submucosa. Consequently, we would argue that ESMR-L with CMI is more appropriate.

Band ligation was effective for resecting the



**Figure 2** Ligation-assisted endoscopic submucosal resection with circumferential mucosal incision for a carcinoid tumor in the anterior wall of the duodenal bulb. A: After circumferential mucosal incision with a FlushKnife BT; B: The duodenal carcinoid tumor was sucked into a ligation device; C: A band ligation created a pseudopolyp; D: Snaring was performed beneath the rubber band; E: An artificial ulcer observed after removal of the tumor without bleeding and perforation.



**Figure 3** Histopathological examination shows *en bloc* resection of carcinoid tumor.

duodenal carcinoid tumor after CMI because it was relatively easy to center and suction the duodenal carcinoid tumor with band ligation. However, band ligation can cause mucosal bleeding, making it difficult to ensure appropriate suction at the center of the tumor without CMI. Therefore, it is possible that band ligation after CMI facilitates easy suction of the submucosal tumor.

In ESMR-L with CMI, the tumor is cut by snaring beneath the rubber band after band ligation. Therefore, perforation is possible if the muscularis is involved, making it necessary to identify such involvement. This is particularly likely when band

ligation involves the muscularis of the thin duodenal wall. Thus, if the ligation band involves the muscularis, the lesion should be cut over the rubber band to avoid perforation, although this may affect histopathological examination.

The advantage of ESD is that tumors can be resected endoscopically, *en bloc*, and regardless of their size or the presence of fibrosis. However, considerable skill is needed to maintain an appropriate line of dissection when there is extensive fibrosis of the submucosa. In such a setting, there is an increased risk of exposure of the lower tumor margin at the time of dissection, which can influence the histopathological evaluation. In contrast, ESMR-L with CMI ensures that tumor involvement in the deeper tissue level is included.

In this case, ESMR-L with CMI had minimal burning effects on the cutting margin because the tumor was cut by snare beneath the rubber band. However, during ESD, the dissection line is precisely determined under direct vision and may prove to be superior to ESMR-L with CMI for curative resection and histopathological examination. Therefore, further investigation is warranted to compare these procedures.

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advice.

## COMMENTS

### Case characteristics

A 64-year-old female with a submucosal tumor in the duodenal bulb on the anterior wall.

### Clinical diagnosis

A submucosal tumor was incidentally found by screening esophagogastroduodenoscopy.

### Differential diagnosis

Gastrointestinal stromal tumor, liposarcoma, angiosarcoma, granular cell tumor, and Brunner's glands.

### Laboratory diagnosis

WBC 6.8 k/ $\mu$ L; HGB 13.50 g/dL; Gastrin 115 pg/mL; no remarkable findings for the other laboratory tests.

### Imaging diagnosis

Esophagogastroduodenoscopy revealed a submucosal tumor in the duodenal bulb on the anterior wall. Endoscopic ultrasound revealed a hypoechoic and uniform tumor, 6 mm  $\times$  5 mm diameter, with a regular margin within the third layer.

### Pathological diagnosis

Histopathological examination showed carcinoid of duodenum, positive for chromogranin-A, synaptophysin, and CD56.

### Treatment

The patient was treated by ligation-assisted endoscopic submucosal resection with circumferential mucosal incision.

### Related reports

Few cases of ligation-assisted endoscopic submucosal resection with circumferential mucosal incision for carcinoid tumor have been reported in the literature.

### Term explanation

Carcinoid tumor, also called neuroendocrine tumor, is a relatively rare tumor which is occasionally found during screening endoscopy.

### Experiences and lessons

This case report described ligation-assisted endoscopic submucosal resection with circumferential mucosal incision for duodenal carcinoid. Band ligation was effective in resecting the duodenal carcinoid tumor after circumferential mucosal incision.

## Peer-review

The authors have described a case of endoscopic resection for duodenal carcinoid tumor. The article highlights that band ligation facilitated complete resection with sucking the tumor after circumferential mucosal incision.

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