

[CASE REPORT]

Two Cases of Hemorrhagic Ampullary Lesions Successfully Treated by Endoscopic Papillectomy

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Abstract:

We herein report two cases of hemorrhagic ampullary lesions in which endoscopic papillotomy was performed to control bleeding and resulted in successful treatment. Both patients were pathologically diagnosed with an underlying pathology characterized by inflammatory cell infiltration and capillary proliferation. They also had disposing factors for bleeding, such as antithrombotic therapy and idiopathic thrombocytopenic purpura. Endoscopic treatment was selected because the risk of surgical resection was high due to the patients' hemorrhagic condition. Both patients were successfully treated without any serious adverse events and had an uneventful postoperative course with no relapse of bleeding.

Key words: endoscopic papillectomy, ampulla of Vater, ampullary lesion, hemorrhagic, bleeding

(Intern Med 61: 1843-1848, 2022)

(DOI: 10.2169/internalmedicine.8294-21)

Introduction

Endoscopic papillectomy (EP) is an established and useful endoscopic therapy that is currently considered a reliable alternative therapy to surgery in patients with ampullary tumors. This technique is generally indicated for adenomas but has also been shown to be effective for treating certain early-stage ampullary carcinomas and ampullary neuroendocrine tumors (1-3).

We herein report two cases of hemorrhagic ampullary lesions in which EP was performed to control bleeding and resulted in an uneventful clinical course without recurrent anemia.

Endoscopic papillectomy procedure

We perform EP using a standard duodenoscope in a similar way to snare polypectomy for mucosal lesions. A stan-

dard polypectomy snare is widely opened, the duodenoscope is advanced in the craniocaudal direction, and the ampullary lesion is grasped at the base for en bloc resection. Constant tension is applied to the snare loop during electrosurgery until resection is complete. The lesion is grasped using net forceps and immediately removed. Thereafter, to prevent postprocedural pancreatitis, a prophylactic pancreatic stent is inserted if possible, and prophylactic sphincterotomy or biliary stenting is performed to prevent cholangitis after EP. The procedure is completed by clipping closure of the resected area to prevent postprocedural bleeding and perforation.

Case Reports

Case 1

A 78-year-old man on warfarin for atrial fibrillation un-

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Received: July 12, 2021; Accepted: September 16, 2021; Advance Publication by J-STAGE: December 4, 2021

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derwent upper gastrointestinal endoscopy for the close examination of anemia, which revealed an erythematous and swollen duodenal papilla. Insufflation caused the lesion to bleed easily. Because the bleeding was considered the cause of anemia, the patient was admitted to our hospital on referral for treatment of the bleeding.

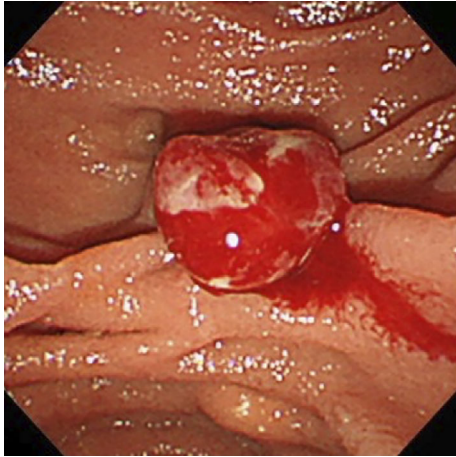


Figure 1. A highly erythematous raised lesion with white coating was found in the duodenal papilla. When washed, the lesion bled easily.

Laboratory tests showed mild anemia. Upper gastrointestinal endoscopy showed no lesions in the esophagus or stomach that could have caused bleeding but did show a highly erythematous raised lesion with white coating in the duodenal papilla. The lesion was hemorrhagic, as oozing was easily caused by washing the lesion (Fig. 1). A biopsy showed moderate lymphocytic/plasmocytic infiltration and neutrophilic infiltration in a granulated duodenal mucosa. Endoscopic ultrasound (EUS) showed no intraductal involvement of the pancreatic duct or bile duct and no tumor invasion of the muscularis propria. Magnetic resonance cholangiopancreatography (MRCP) showed no evidence of malignant tumor causing biliary bleeding, pseudoaneurysm/pancreatic cyst causing hemosuccus pancreaticus, or pancreas divisum. Based on these findings, bleeding from the ampullary lesion was considered the cause of anemia.

Because the patient had the underlying condition of atrial fibrillation, it was difficult to suspend antithrombotic medication. Although we considered surgical resection, the patient strongly preferred endoscopic treatment. Therefore, after receiving informed consent, EP was performed following the withdrawal of warfarin and with a normal coagulation ability [prothrombin time-international normalized ratio (PT-INR) 1.25] (Fig. 2).

Second-look endoscopy on day 7 after treatment showed

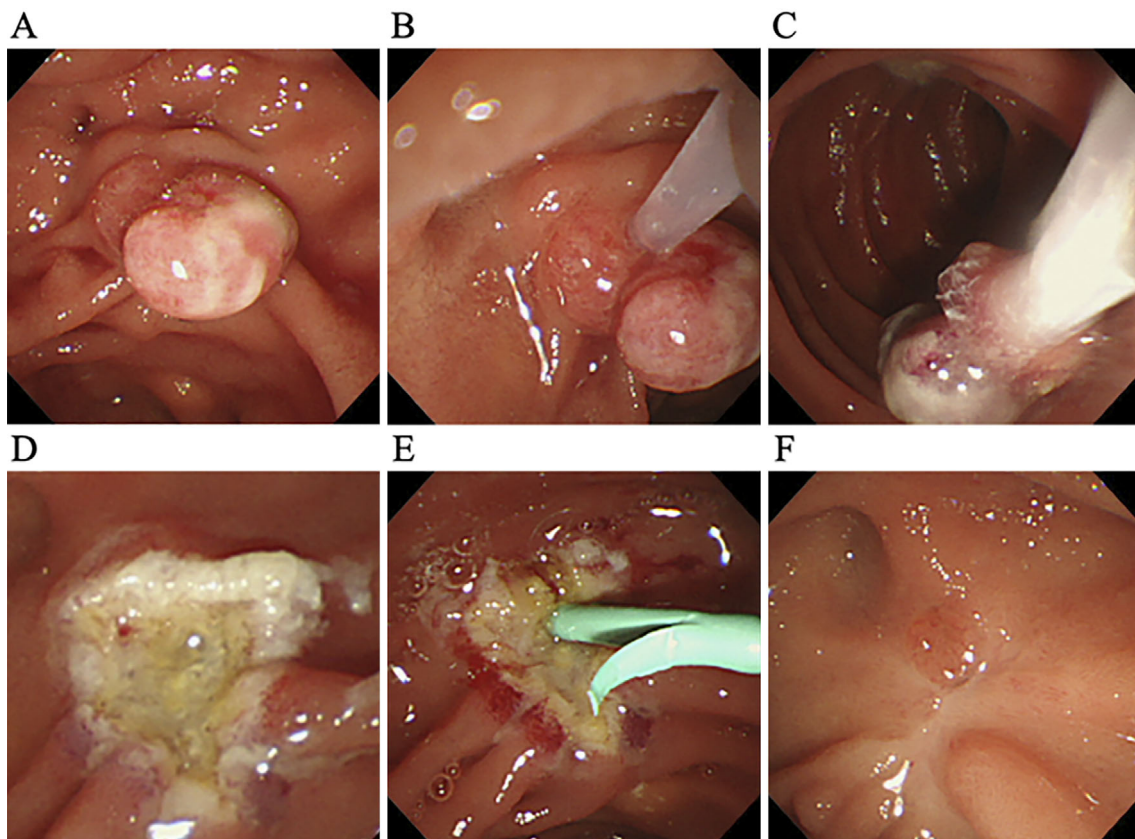


Figure 2. Endoscopic papillectomy (EP). A: The papilla was 8 mm in size. B: The lesion was resected by energizing the snare loop. C: The resected specimen was collected with net forceps. D: The papilla after resection. E: A pancreatic duct stent was placed after EP. F: Appearance of the resection surface one month after EP. The surface is scarred without bleeding.

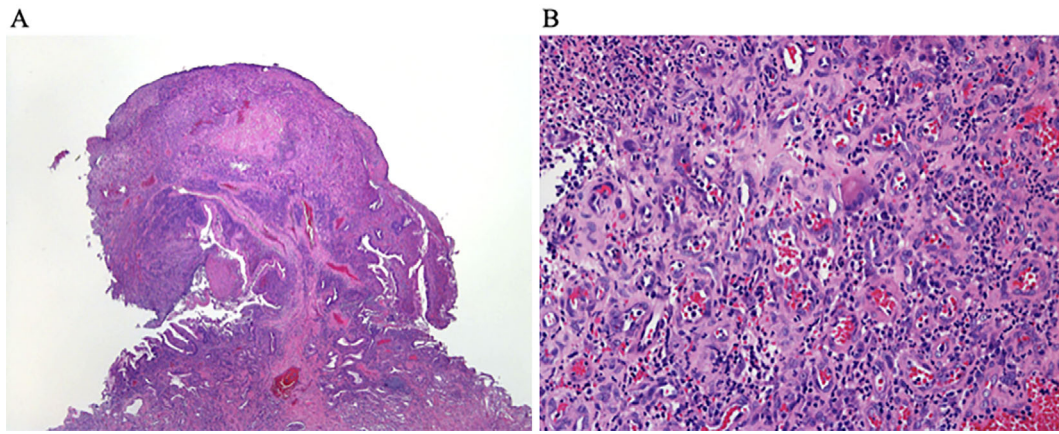


Figure 3. Histological view of the resected specimen. A: Low-power view: The lesion was pedunculated and measured 8 mm in size. B: High-power view: Infiltration of inflammatory cells such as neutrophils and macrophages, proliferation of capillaries and fibroblasts, and proliferation of inflammatory granulation tissue consisting of aggregated foreign body-type giant cells were observed.

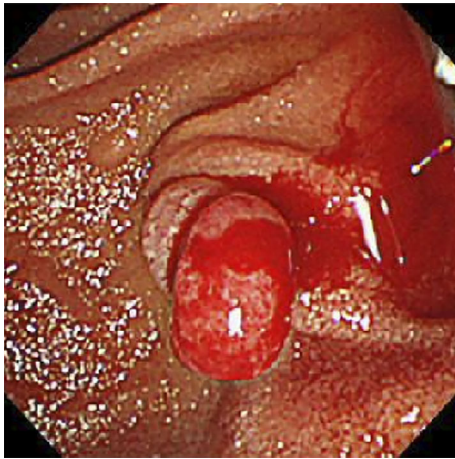


Figure 4. An erythematous, swollen, hemorrhagic lesion was found in the duodenal papilla.

no bleeding from the resection surface or residual lesion. A pancreatic duct stent placed after EP was then removed. The patient started eating on day 8 and was discharged from the hospital on day 12. A histopathological examination showed infiltration of inflammatory cells, such as neutrophils and macrophages, proliferation of capillaries and fibroblasts, and proliferation of inflammatory granulation tissue consisting of aggregated foreign body-type giant cells, leading to a diagnosis of benign polypoid lesion with foreign body granuloma (Fig. 3). No relapse of symptoms has been observed as of three years and six months after treatment.

Case 2

The patient was a 79-year-old man who was being treated for idiopathic thrombocytopenic purpura with *Helicobacter pylori* eradication, steroid therapy, splenectomy, and oral hematopoietic stimulants. He had a history of tarry stool and severe anemia of unknown cause resulting in hemorrhagic shock and temporary cardiopulmonary arrest. He presented

to another hospital with chief complaints of tarry stools and anemia. Upper gastrointestinal endoscopy revealed bleeding from the duodenal papilla (Fig. 4). After unsuccessful hemostasis by argon plasma ablation, the patient was admitted to our hospital on referral for treatment of the bleeding.

Upper gastrointestinal endoscopy showed no lesions in the esophagus or stomach that could have caused bleeding but did show an erythematous, hemorrhagic raised lesion in the duodenal papilla. A biopsy specimen from the lesion was histologically diagnosed as erosive duodenal mucosa with granulomatous changes in the interstitium. EUS showed no intraductal involvement of the pancreatic duct or bile duct and no tumor invasion of the muscularis propria. MRCP showed no evidence of pancreas divisum or a disease causing biliary bleeding or hemosuccus pancreaticus. Based on these findings, bleeding from the ampullary lesion was considered the cause of anemia.

Surgical resection was considered to carry a high risk for this patient because of the idiopathic thrombocytopenic purpura, and he strongly preferred endoscopic treatment. However, his condition was well controlled with hematopoietic stimulants, with a platelet count of 119,000/ μ L and hemoglobin (Hb) level of 12.1 g/dL. Therefore, after receiving his informed consent, EP was performed without the need for preoperative high-dose immunoglobulin therapy or platelet transfusion (Fig. 5).

On day 6 after treatment, calculous cholecystitis occurred, which was treated by EUS guided-gallbladder drainage. Second-look endoscopy was performed on day 14 after treatment, showing no bleeding from the resection surface or residual lesion. The bile duct and pancreatic duct stents placed after EP were then removed. Although conservative treatment of biliary peritonitis after EUS-gallbladder drainage took a long time, resulting in a prolonged hospital stay, the patient had a favorable clinical course thereafter and was discharged on day 24.

A histopathological examination revealed prominent pro-

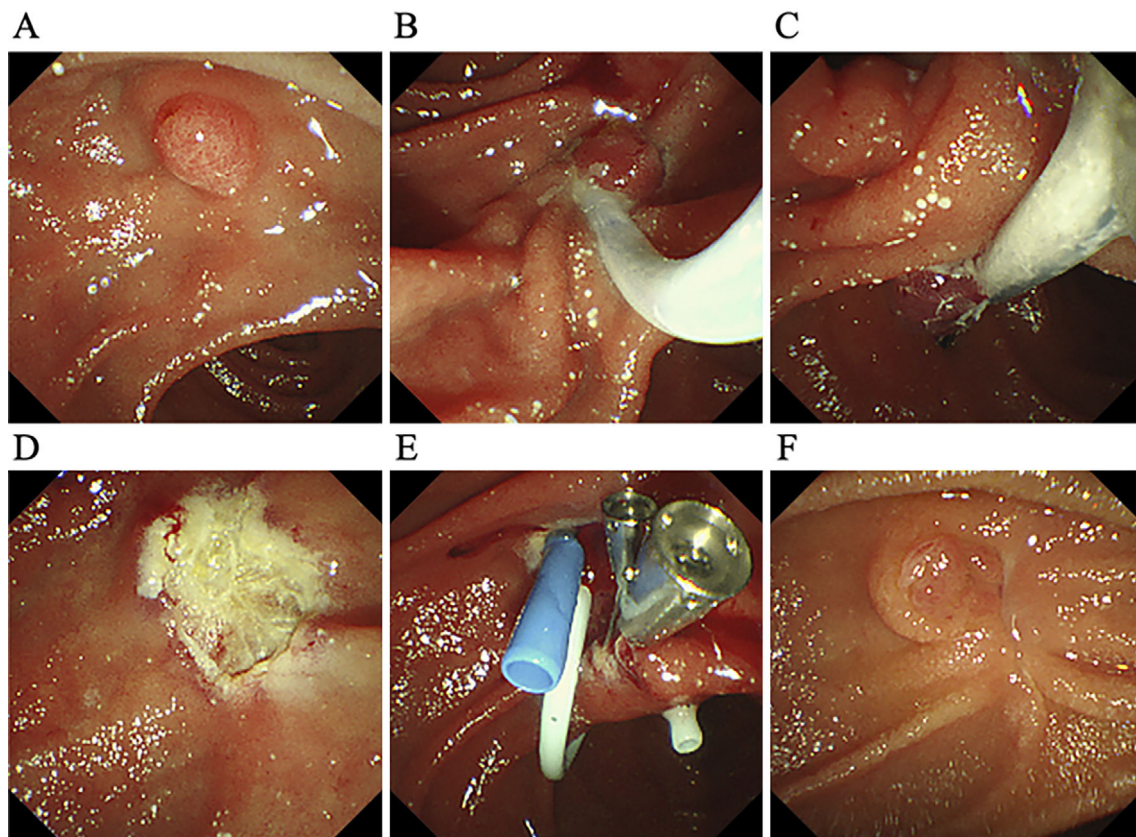


Figure 5. Endoscopic papillectomy (EP). A: The papilla measured 9 mm in size. B: The lesion was resected by energizing the snare loop. C: The resected specimen was collected with net forceps. D: The papilla after resection. E: The anal side of the papilla was plicated with a clip, and bile duct and pancreatic duct stents were placed. F: Appearance of the resection surface one month after EP. The surface is scarred without bleeding.

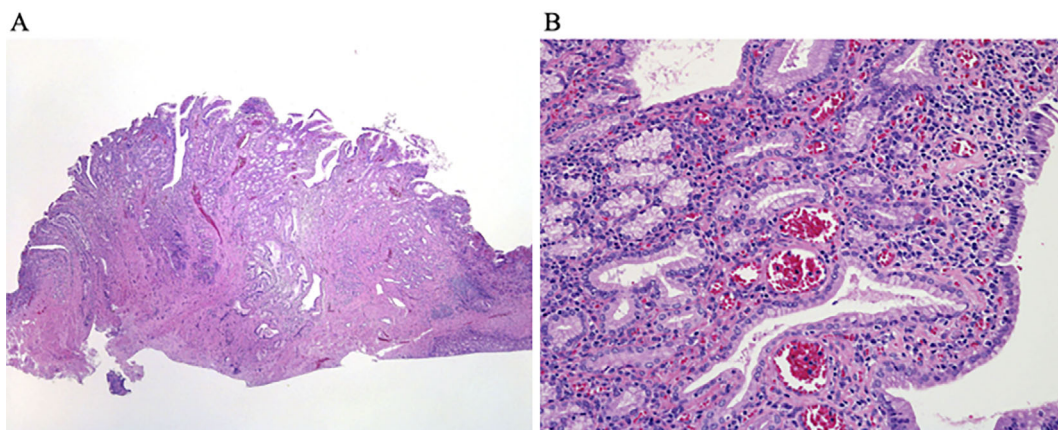


Figure 6. Histological view of the resected specimen. A: Low-power view: The lesion was sub-pedunculated and measured 9 mm in size. B: High-power view: Prominent proliferation of small vessels, mainly capillaries, and mild to moderate infiltration of inflammatory cells, mainly lymphocytes and plasma cells, were observed.

liferation of small vessels, mainly capillaries, and mild to moderate infiltration of inflammatory cells, mainly lymphocytes and plasma cells, leading to a diagnosis of small vessel proliferation with mild epithelial atypia (Fig. 6). No relapse of symptoms has been observed as of four months

since the treatment.

Discussion

Tumors occurring in the duodenal papilla include carcino-

Table. Reported Cases of Endoscopic Resection of Hemorrhagic Duodenal Lesions.

Case	Reference	Age (years)/ Sex	Symptoms	Size (mm)	Location	Treatment	Diagnosis
1	Our case 1	78/Man	Anemia	8	Duodenal papilla	Endoscopic papillectomy	Benign polypoid lesion with foreign body granuloma
2	Our case 2	79/Man	Anemia Melena	9	Duodenal papilla	Endoscopic papillectomy	Small vessel proliferation with mild epithelial atypia
3	(9)	77/Woman	Anemia	10	Duodenal papilla	Endoscopic papillectomy	Pyogenic granuloma
4	(10)	64/Man	Anemia	7	Duodenal mucosa	Endoscopic mucosal resection	Pyogenic granuloma
5	(11)	60/Man	Anemia	8	Duodenal mucosa	Endoscopic polypectomy	Pyogenic granuloma

mas, adenomas, neuroendocrine tumors, and paragangliomas (1-4). The standard treatment for these tumors is pancreaticoduodenectomy. EP, which was first reported by Suzuki et al. (5) in 1983, has demonstrated effectiveness as a less invasive and curative treatment for not only adenomatous lesions but also certain early-stage cancers and neuroendocrine tumors (1-3). Although EP is associated with adverse events, such as hemorrhage, pancreatitis, and perforation, which can be severe and fatal in some cases (6), we took several measures to prevent these adverse events, such as performing clipping closure of the resected area to prevent postprocedural bleeding and perforation and placing a pancreatic stent to prevent pancreatitis. As such, EP is still safer than surgical treatment and also superior in terms of the quality of life. The technique is also useful for performing a total biopsy of ampullary tumors, which tend to be underdiagnosed preoperatively.

In the present cases, EP was performed to control bleeding from a hemorrhagic ampullary lesion. Both patients were pathologically diagnosed with underlying pathologies characterized by inflammatory cell infiltration and capillary proliferation and had disposing factors for bleeding, such as antithrombotic therapy and idiopathic thrombocytopenic purpura. In addition to these factors, exposure to food and digestive fluids might have caused bleeding from the lesions. These features are similar to those observed in pyogenic granuloma (PG).

PG is a painless, hemorrhagic, polypoid granulosomatous lesion that is a type of hemangioma characterized histologically by abundant vascularization and inflammatory cell infiltration. This granulation tissue-like tumor is formed as a result of secondary changes caused by trauma, chronic irritation, or infection and most commonly occurs in the oral cavity and skin and less commonly in the gastrointestinal tract (7). Ikeoka et al. (8) reported 13 cases of PG in the small intestine. All patients underwent radical resection (surgery in 10 and endoscopic treatment in 3) to control bleeding. A total of three cases of duodenal PG have been previously reported and endoscopic treatment was performed to control bleeding in all cases (Table) (9-11). Therefore, in addition to surgery, endoscopic resection is considered a viable treatment option for PG in the gastrointestinal tract.

The lesions in the present cases resembled PG - although this was not confirmed pathologically - in terms of the hemorrhagic nature and histological findings of capillary proliferation and inflammatory cell infiltration. We speculate that originally normal ampullae might have been transformed into lesions pathologically characterized by capillary proliferation and inflammatory cell infiltration in response to certain chronic stimuli.

To treat the present cases, we first attempted to improve the hemorrhagic condition. However, in Case 1, the patient was on long-term antithrombotic therapy for chronic atrial fibrillation, and it was difficult to suspend the therapy. In Case 2, the patient was being treated for idiopathic thrombocytopenic purpura, with failure to control bleeding. The patient also had a history of hemorrhagic shock. Therefore, in both cases, resection of the ampullary lesion itself was deemed necessary to achieve hemostasis. We selected EP because the risk of surgical resection was high due to the patients' hemorrhagic condition, and hemostasis with angiography would make it challenging to identify the responsible vessel due to intermittent and oozing bleeding. Of course, EP itself carries a high risk of procedure-related bleeding, so we performed EP after having taken measures to control bleeding: in Case 1, we withdrew warfarin, and the PT-INR was 1.25 at the time of EP; in Case 2, we administered hematopoietic stimulants, and the platelet count was 119,000/ μ L. Thus, both patients were successfully treated without any serious adverse events and had an uneventful postoperative course with no relapse of bleeding. EP is still considered a high-risk procedure owing to its associated adverse events and might be challenging in cases with hemorrhagic lesions. Therefore, for lesions with a high bleeding risk, EP should be performed by expert endoscopists trained in the management of pancreatobiliary disease at institutions where angiographic embolectomy and surgical hemostasis can be performed immediately if required.

Conclusion

EP can be effective for not only endoscopic treatment of ampullary tumors, including cancers and adenomas, but also controlling symptoms of hemorrhagic ampullary lesions.

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

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