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# Acute Pancreatitis Caused by Ampullary Duodenum Adenoma in a Patient with Adenomatous Polyposis Coli with Billroth II Reconstruction After Distal Gastrectomy

Authors' Contribution:  
Study Design A  
Data Collection B  
Statistical Analysis C  
Data Interpretation D  
Manuscript Preparation E  
Literature Search F  
Funds Collection G

AEF **Takao Iemoto**  
AEF **Tsuyoshi Sanuki**  
F **Takayuki Ose**  
F **Tomoo Yoshie**  
F **Katsuhide Tanaka**  
F **Ayaka Sasaki**  
F **Shohei Abe**  
F **Tetsuyuki Abe**  
F **Mika Miki**  
F **Ryoko Futai**  
F **Yuta Inoue**

Department of Gastroenterology, Kita-Harima Medical Center, Ichiba, Ono, Hyogo, Japan

**Corresponding Author:** Tsuyoshi Sanuki, e-mail: [tssanuki@gmail.com](mailto:tssanuki@gmail.com)  
**Conflict of interest:** None declared

**Patient:** **Male, 73**  
**Final Diagnosis:** **Pancreatitis**  
**Symptoms:** **Upper abdominal pain**  
**Medication:** —  
**Clinical Procedure:** —  
**Specialty:** **Gastroenterology and Hepatology**

**Objective:** **Rare disease**

**Background:** Adenomatous polyposis coli is an autosomal dominant hereditary disorder. Duodenal adenocarcinoma and adenoma, which are extracolonic lesions, not only affect the prognosis of patients but also cause acute pancreatitis.  
**Case Report:** We present the case of a 73-year-old male. He had undergone proctocolectomy for familial adenomatous polyposis and distal gastrectomy (Billroth II reconstruction with Braun anastomosis) for gastric ulcer; he presented with acute pancreatitis caused by ampullary duodenum adenoma. Double-balloon endoscopy showed 2 adenomatous polyps in the major papilla and descending limb of the duodenum. Based on the findings of endoscopy and biopsy, the duodenal polyps were diagnosed as adenomas and classified as Spigelman stage II.

**Conclusions:** Our case report suggests that duodenal surveillance is necessary for patients with adenomatous polyposis coli. In addition, surveillance using double-balloon endoscopy is useful for patients with an altered gastrointestinal anatomy.

**MeSH Keywords:** **Adenomatous Polyposis Coli • Gastroenterostomy • Pancreatitis**

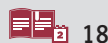
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## Background

Adenomatous polyposis coli (APC) is an autosomal dominant hereditary disorder with a high tendency for cancerous change. Prophylactic total colectomy decreases the risk of colorectal cancer in patients with APC. Duodenal adenoma occurs in up to 90% of patients with APC [1]. The cumulative risk of duodenal cancer is as high as 10% by 60 years of age, which presents a 100- to 300-fold greater risk in affected patients than in the general population [2]. Furthermore, duodenal adenocarcinoma is one of the primary causes of death in patients with APC.

The severity of duodenal polyposis is assessed using Spigelman classification. The risk of developing duodenal cancer with Spigelman stage III and IV adenomatosis is significantly higher than the risk with stage 0–II adenomatosis. Therefore, stage IV disease is an indication for pancreaticoduodenectomy [3]. Conversely, the risk of developing duodenal cancer is very low in patients with stage I and II diseases. The management of stage I and II disease is limited to follow-up [4]. However, duodenal adenocarcinoma and adenoma cause pancreatitis. Although several case reports have described the association between acute pancreatitis and ampullary carcinoma and adenoma in patients with APC [5,6], it is uncertain whether the treatment for ampullary duodenum adenoma causes acute pancreatitis. Furthermore, few articles have described how to screen patients with gastrectomy.

In this article, we present a case of acute pancreatitis caused by ampullary duodenum adenoma (Spigelman stage II), which developed in a patient with APC who underwent distal gastrectomy and Billroth II reconstruction. We detected the ampullary duodenum adenoma using double-balloon endoscopy. Duodenal surveillance is necessary for patients with APC with an altered gastrointestinal anatomy.

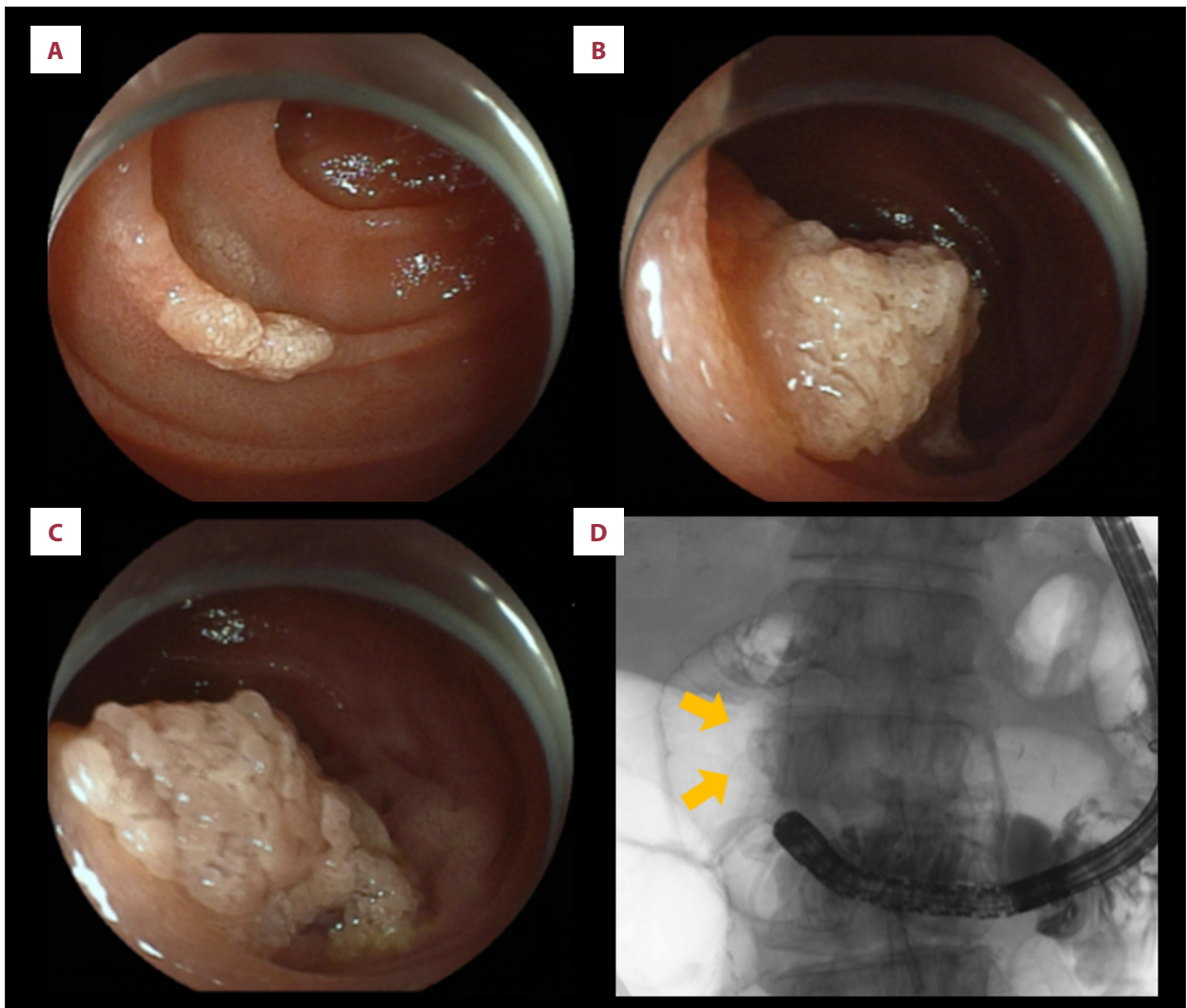
## Case Report

A 73-year-old male was admitted for upper abdominal pain. He had undergone distal gastrectomy and Billroth II reconstruction with Braun anastomosis for gastric ulcer at the age of 23 years, proctocolectomy for APC at the age of 43 years, and laparoscopic omental flap transposition for unexplained gastric perforation at the age of 71 years. In addition, he had been treated with a gonadotropin-releasing hormone antagonist for prostate cancer for 4 years. He had no history of alcohol consumption or medication use that could have caused acute pancreatitis. Physical examination on admission revealed tenderness upon palpation of the epigastrium. Blood analysis showed elevation of the following parameters: the white blood cell count (13480/mm<sup>3</sup>), C-reactive protein (0.56 mg/dL), and serum pancreatic amylase (1220 IU/L). The serum levels of



**Figure 1.** Contrast-enhanced computed tomography showing pancreatic enlargement and an area of poor contrast from the body to the tail of the pancreas.

aspartate, alanine aminotransferase, alkaline phosphatase,  $\gamma$ -glutamyl transferase, triglyceride, carcinoembryonic antigen, carbohydrate antigen 19-9, and serum IgG4 (29.4 mg/dL) were within normal ranges. Abdominal ultrasonography did not reveal gallbladder stones or common bile duct stones. Contrast-enhanced computed tomography revealed pancreatic enlargement and an area of poor contrast between the body and the tail of the pancreas (Figure 1). Under the diagnosis of severe acute pancreatitis (according to the scoring system of the Japanese guideline for acute pancreatitis), conservative therapy (fluid resuscitation, intravenous infusion of gabexate mesilate) was performed. His general status immediately improved with the conservative therapy. Although endoscopic ultrasonography through the remnant stomach showed the pancreatic body and tail, the pancreatic head and ampullary duodenum tumors were not detected because of the altered gastrointestinal anatomy. Therefore, we could not evaluate the depth of the lesion's infiltration and whether it infiltrated the bile or pancreatic ducts. Upper gastrointestinal endoscopy could not reach the duodenal papilla because of the flexion of the Braun anastomosis. Double-balloon endoscopy showed 2 adenomatous polyps in the duodenum: a tumor in the major papilla and another in the descending limb (Figure 2A–2C). Fluoroscopy also showed a tumor in the major papilla (Figure 2D). However, it was difficult to identify the minor papilla. Based on the biopsy findings, these tumors were diagnosed as tubular adenomas with mild dysplasia. It seemed that the severe acute pancreatitis was caused by the occlusion of the main pancreatic duct due to ampullary duodenum adenoma. Based on the findings of endoscopy and biopsy, the duodenal adenomas were classified as Spigelman stage II [4]. The patient did not prefer to undergo pancreaticoduodenectomy owing to the presence of prostate cancer and his history of 3 abdominal surgeries.



**Figure 2.** Double-balloon endoscopy showing 2 adenomatous polyps in the duodenum: (A) tumor in the descending limb, and (B, C) tumor in the major papilla. (D) Fluoroscopy showing a tumor in the major papilla (arrow).

## Discussion

Numerous studies have shown that duodenal and ampullary adenomas can be found in 40–100% of patients with APC [7,8]. Duodenal ampullary tumors associated with APC are considered to be slow-growing tumors, and the overall risk of developing cancer in patients with APC is less than 5% [2]. Using the Spigelman classification, several studies have assessed the severity of duodenal tumors. Spigelman stage IV disease is an indication for pancreaticoduodenectomy; conversely, stage I and II disease are not indications for surgery because of the low risk of developing cancer. The recommended surveillance interval in stage I and II disease is 3 to 5 years [3]. On the other hand, Sourrouille et al. demonstrated progression of the Spigelman score over time in patients with APC. Moreover, they have suggested that the Spigelman score should be modified in consideration of ampullary abnormalities, such as increased size and ulceration [9].

Several reports have described acute pancreatitis caused by ampullary duodenum tumors of Spigelman stage IV [2,10]. The affected patients underwent surgery and experienced no recurrent acute pancreatitis. However, there are few reports on acute pancreatitis caused by ampullary duodenum adenomas. Therefore, the optimal treatment for ampullary duodenum adenoma of Spigelman stages I and II is unclear. A previous article suggested that relapsing acute pancreatitis is an indication for surgery for premalignant ampullary tumors in APC [5]. Patients with APC and acute pancreatitis caused by ampullary duodenum adenoma should be considered for surgical therapy because acute pancreatitis is a lethal disease [11].

In our case, autoimmune pancreatitis and drug-induced pancreatitis were ruled out. Our patient had an ampullary adenoma with mild atypia, which seemed to have caused acute pancreatitis. Upper gastrointestinal endoscopy could not reach

the duodenal papilla because of the altered gastrointestinal anatomy. We were able to diagnose the patient's condition using double-balloon endoscopy. Double-balloon endoscopy has been recently used for pancreatobiliary diseases in patients with an altered gastrointestinal anatomy, and its efficacy and safety have been confirmed [12,13].

Endoscopic ampullectomy is an effective therapeutic procedure for ampullary duodenum adenomas [14]. However, ampullary duodenum adenomas frequently recur after this procedure, at a rate reported to be 58.3% [15]. Moreover, a previous study reported that the tumor recurrence rate with local treatment, such as endoscopic resection of ampullary tumors, is almost 100% [16,17]. Recurrence is dependent on the presence of an intraductal tumor, the resection margin, and the surgical histology [18]. These factors would be difficult to predict before endoscopic ampullectomy.

In our case, we could not determine the whole picture of the tumor because of poor stability during double-balloon endoscopy. Furthermore, the biliary and pancreatic orifices could not be identified. It was difficult to perform a complete en bloc excision of the entire neoplasm; therefore, we did not perform endoscopic ampullectomy. For similar reasons, pancreatoduodenectomy has been recommended for noninvasive ampullary

duodenal tumors in patients with APC [5]. However, pancreatoduodenectomy is one of the most difficult surgeries with a high rate of major complications. We did not perform a pancreatoduodenectomy because of the multiple risk factors identified for our patient (presence of prostate cancer and 3 previous abdominal surgeries).

## Conclusions

We present the case of a patient with APC who developed severe acute pancreatitis due to ampullary adenoma. Our report suggests that duodenal surveillance is necessary for patients with APC. In addition, surveillance using double-balloon endoscopy is useful for patients with an altered gastrointestinal anatomy.

## Department and Institution where work was done

Department of Gastroenterology, Kita-Harima Medical Center, Ichiba, Ono, Hyogo, Japan

## Conflicts of interest

None.

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