

LETTERS TO THE EDITOR EDİTÖRE MEKTUPLAR

Jejunal lipoma causing intussusception and gastrointestinal bleeding

Kanama ve intussepsiyon nedeni olarak jejunal lipom

To the Editor

Gastrointestinal lipomas are rare benign tumors commonly found in the colon near the ileocecal valve (1). They are slowly growing, submucosal tumors, which frequently may cause intussusception but rarely bleeding. Because the small intestine is relatively inaccessible to routine endoscopy, diagnosis of small intestinal neoplasms is often delayed. However, a relatively new imaging technique, double-balloon enteroscopy (DBE), may permit earlier, nonoperative diagnosis. Here, we report a patient who presented with unexplained abdominal colicky pain and previous massive gastrointestinal hemorrhage due to an ulcerated jejunal lipoma, which was first recognized by computerized tomography (CT) and subsequently verified by DBE.

A 61-year-old woman was admitted to our hospital due to abdominal colicky pain for two years. She denied any loss of appetite and had no weight loss. In her medical history, she had a massive gastrointestinal bleeding (hematemesis and hematochezia) six months ago, for which she was transfused repeatedly. However, at that point in time, neither the upper nor lower gastrointestinal endoscopy, performed in another hospital. Despite antacid therapy there was no pain relief. On physical examination, she was pale and had mild abdominal tenderness. Laboratory results were normal except for hypochromia, microcytic anemia (Hct 18.3%), and low ferritin levels [(5 ng/ml (10-350 ng/ml)]. Searching for the etiology of abdominal pain and iron deficiency anemia, a gastroscopy and colonoscopy were performed for the second time, but no specific findings were detected. A subsequent CT showed thickening of the jejunum,

and “target sign” typical for invagination, and homogeneous fat density mass image almost filling the lumen (Figure 1a). A subsequent peroral DBE disclosed an ulcerated, yellowish, sessile submucosal

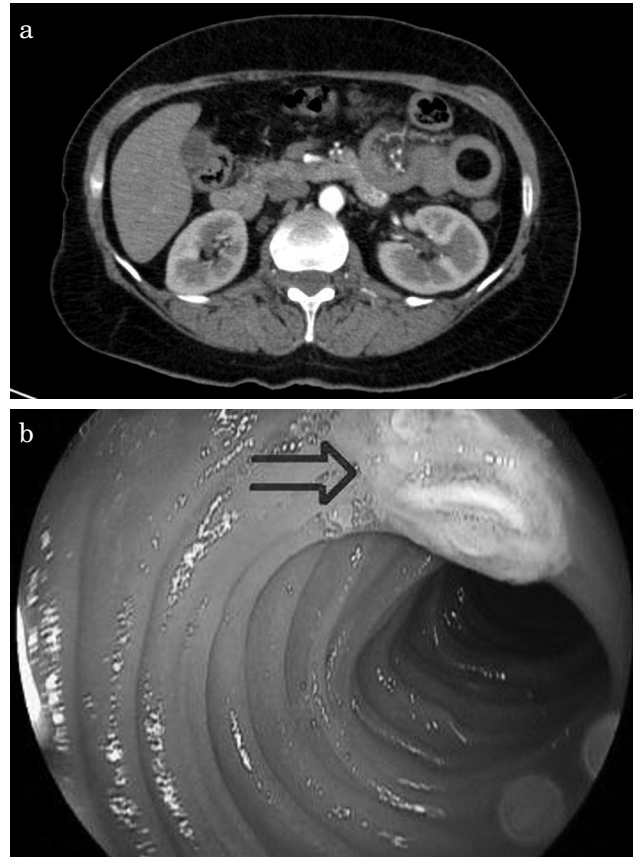


Figure 1a. Axial CT postcontrast image showing fat density round lesion as the lead point of jejunojejunal intussusception in the left upper quadrant. **1b.** Ulcerated jejunal lipoma image taken during DBE.

sal tumor, measuring 15x15 mm in diameter (Figure 1b). The tumor was surgically resected and photomicrographic findings included a tumor composed of mature adipose tissue in the submucosa, which was consistent with a diagnosis of lipoma.

Lipomas constitute about 10% of the gastrointestinal benign tumors (1), and there are limited scattered cases in the literature of intestinal lipomas presented with bleeding (2-3), and bleeding and intussusception (4-6). Symptoms due to invagination in adults tend to be chronic or intermittent and include pain, constipation, weight loss, or a palpable abdominal mass at physical examination. Like in our case, the CT findings in intussusception are usually pathognomonic, including a target-like mass, oral con-

trast material trapped between the opposing walls of the intussusceptum and intussusciens, and a soft-tissue mass secondary to the intussusception, possibly with the accompanying lead point. If blood supply is compromised, bowel-wall thickening, as seen in this case, or intramural air may be seen (7).

Interestingly, our case presented with both abdominal colicky pain due to intermittent intussusception and gastrointestinal bleeding, which prompted us to search for its source. We report this patient to alert physicians to investigate the small intestine in case of unexplained, chronic abdominal pain. Overt or occult bleeding sources can easily be detected and treated by a relatively new, safe technique, namely DBE.

REFERENCES

1. Gill SS, Heuman DM, Mihas AA. Small intestinal neoplasms. *J Clin Gastroenterol* 2001;33:267-82.
2. Zografos G, Tsekouras DK, Lagoudianakis EE, Karantzi-kos G. Small intestinal lipoma as a cause of massive gastrointestinal bleeding identified by intraoperative enteroscopy. A case report and review of the literature. *Dig Dis Sci* 2005;50:2251-4.
3. Baskaran V, Patnaik PK, Seth AK, et al. Intestinal lipoma: a rare cause of lower gastrointestinal haemorrhage. *Trop Gastroenterol* 2003;24:208-10.
4. Ortiz de Solórzapo Aurusa FJ, Yarritu Viilanueva C, Ruiz Adrados E, et al. Gastroduodenal invagination and upper gastrointestinal hemorrhage secondary to gastric lipoma. *Gastroenterol Hepatol* 1997;20:303-5.
5. Baron Y, Priesack W, Sötje G, et al. Hemorrhagic jejunal lipoma with intermittent intussusception. *Eur J Radiol* 1996;22:123-5.
6. Yiğitler C, Ataç K, Yiğit T, et al. A rare cause of bleeding intestinal intussusception in adult: jejunal lipoma. *Ulus Travma Acil Cerrahi Derg* 2007;13:237-40.
7. Slavin JD, Islam QT, Spencer RP, Martinez H. Jejunal lipoma as a source of gastrointestinal bleeding. *Clin Nucl Med* 2000;25:946.

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An atypical initial presentation of Crohn's disease

Crohn hastalığında atipik klinik başlangıcı olan bir olgu

To the Editor

Osteoporosis is prevalent in Crohn's disease (CD), but fracture is an uncommon initial presentation (1-3). A 29-year-old male was admitted with recent onset right ankle pain and hip strain. He had

a history of femur fracture with minor trauma the year before. Physical examination revealed right lower quadrant tenderness. Abnormal laboratory parameters were: hemoglobin: 10.0 g/dl, hematoc-

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rit 30%, mean corpuscular volume: 115 fL, white blood cell count 12,000/mm³, sedimentation rate: 40 mm/hr, total serum calcium: 5.0 mg/dl, ionized calcium: 0.8 mmol/L, phosphorus: 2.2 mg/dl, magnesium: 1.3 mg/dl, alkaline phosphatase: 506 U/L, vitamin B12: 100 ng/L, parathyroid hormone: 651 pg/ml, and vitamin D: 10 nmol/L. The X-ray showed fracture at the right distal fibula (Figure 1). Abdominal ultrasonography showed thickened terminal ileum wall and fistula tract with fluid collection at right lower quadrant. Colonoscopy revealed cobblestone appearance plus strictures at the ileum with patchy, large, deep and linear ulcers throughout the colon and normal rectum. The biopsy from the terminal ileum revealed epithelioid granuloma, ulceration, and lymphocytic and plasmacytic inflammation of the lamina propria. The findings were consistent with CD complicated with osteomalacia. Ileum resection and ileocolectomy were performed, and the patient was discharged in the postoperative period with a treatment of azathioprine, mesalazine, calcium plus vitamin D and vitamin B12. He was well during the follow-up period.

Osteoporosis may be multifactorial in CD. Glucocorticoid therapy, hypogonadism, vitamin D deficiency and low body weight are all contributing factors. Calcium deficiency may occur as a result of inadequate dietary intake or intestinal malabsorption. Secondary hyperparathyroidism due to a lesser degree of vitamin D deficiency results in increased bone turnover, causing predominantly cortical bone loss (4). Inflamed intestinal mucosa cytokines, interleukin-1, interleukin-6 and tumor necrosis factor alpha (5), by increasing the generation and activity of osteoclasts, may be another

possible mechanism. The fracture in our patient may indicate that the disease was present for a long period before the diagnosis. A delay in making a definite diagnosis and thus in management was probably complicated by the osteoporotic fracture.

In conclusion, osteoporosis may be the first presentation, and gastrointestinal symptoms may not dominate even in complicated CD. When the patient is diagnosed as CD, the evaluation for osteoporosis should be started promptly and when the levels are low, treatment with calcium, magnesium and vitamin D should be implemented.



Figure 1. Arrow shows a fracture at the right distal fibula.

REFERENCES

1. Andreassen H, Rungby J, Dahlerup JF, Mosekilde L. Inflammatory bowel disease and osteoporosis. *Scand J Gastroenterol* 1997;32:1247-55.
2. Bernstein CN, Blanchard JF, Leslie W, et al. The incidence of fracture among patients with inflammatory bowel disease. A population-based cohort study. *Ann Int Med* 2000; 133:795-9.
3. Schoon EJ, Blok BM, Geerling BJ, et al. Bone mineral density in patients with recently diagnosed inflammatory bowel disease. *Gastroenterology* 2000;119:1203-8.
4. Parfitt AM, Gallagher JC, Heaney RP. Vitamin D and bone health in the elderly. *Am J Clin Nutr* 1982;36:1014-31.
5. Gross V, Andus T, Caesar I, et al. Evidence for continuous stimulation of interleukin-6 production in Crohn's disease. *Gastroenterology* 1992;102:514-9.

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Rectosigmoid endometriosis

Rektosigmoid endometriozis

To the Editor

Extra-pelvic endometriosis is most commonly seen in the bowels, with a reported incidence of 5-20%. Rectosigmoid endometriosis (RE) accounts for 71% of cases of intestinal endometriosis (1, 2). Bowel endometriosis might show non-specific symptoms, such as abdominal colic-like pain, nausea, vomiting, constipation, diarrhea, and rectal bleeding (1, 3).

We retrospectively reviewed the charts of six patients with histopathologically proven RE between December 2001 and January 2006 (Table 1). Two patients were treated for infertility, and pelvic endometriosis was found on diagnostic laparoscopy previously. The most common findings in ultrasonography (USG) and computerized tomography (CT) were solitary, necrotic mass adhering to the bowel wall and bowel wall thickening. In one patient, a submucosal lesion was found. The results of abdominal USG and CT did not help in diagnosing intestinal endometriosis. Colonoscopy was performed in five of the six patients. Edema, luminal narrowing and increasing thickness of the bowel wall near the rectosigmoid junction were found in four patients. Submucosal tumoral mass was seen with overlying normal mucosa in one patient. Because biopsies almost always include only the lamina propria, histopathologic findings of biopsy materials were in normal range.

The patients were operated electively. In one patient, in the gynecologic operation due to myoma uteri, an incidental mass was detected at the rectosigmoid junction narrowing the lumen and in or-

der to exclude a bowel tumor, anterior resection-Hartmann colostomy was made, in addition to total abdominal hysterectomy + bilateral salpingo-oophorectomy (TAH+BSO). In two patients, only anterior resection was performed. TAH+BSO and anterior or low anterior resection were performed in the other three patients. In one patient, a whitish plaque over the appendix was found incidentally and appendectomy was added. Histopathological examination revealed appendiceal endometriosis in addition to RE. There was no recurrence over a mean follow-up period of 32 months (11 months-5 years).

Due to the presence of risk of malignant transformation and fibrosis development after medical treatment, optimum therapeutic-diagnostic choice in RE is surgery. Surgery is also indicated in case of intestinal obstruction and severe abdominal pain. Limited surgery or cauterization of lesions following frozen-section pathologic examination could be performed (1, 4, 5).

Rectosigmoid endometriosis may mimic various gastrointestinal diseases, including primary bowel carcinoma, diverticulosis, chronic inflammatory bowel disease and gastrointestinal stromal tumors. It is remarkably difficult to diagnose intestinal endometriosis by endoscopic and radiological imaging methods (2, 5). Since there are no pathognomonic radiological or endoscopic findings, definitive diagnosis could only be done by histopathological examination.

Table 1. The characteristics, radiological/endoscopic findings and surgical treatment in patients with rectosigmoid endometriosis

Patient number	Age	Symptoms	Infertility	Previous surgery	Colonoscopy / rectosigmoidoscopy	Abdominal USG	Abdominal CT	Surgery
1	39	Abdominal pain, constipation	+	Diagnostic laparoscopy for pelvic endometriosis 12 years ago	Mucosal edema, luminal narrowing and increasing thickness of the bowel wall (distance 15- 20 cm from anus)	Left adnexal mass (4 cm diameter) compressing rectum wall	Left adnexal mass causing external compression in rectosigmoid junction	Anterior resection
2	48	Cyclic, colic abdominal pain	-	-	External mass effect and increasing thickness of the bowel wall (distance 15 cm from anus)	Left adnexal necrotic mass (2.6 cm diameter) strictly adhered to bowel wall	Bowel wall thickening at sigmoid colon	TAH+BSO, anterior resection
3	49	Vaginal bleeding	-	-	-	Myoma uteri	-	TAH+BSO, Hartmann procedure
4	42	Pelvic pain, cyclic rectal bleeding	+	Diagnostic laparoscopy for infertility 5 years ago	External mass effect and mucosal hyperemia (distance 13 cm from anus)	Thickening of colonic wall	-	TAH+BSO, anterior resection,
5	51	Pelvic pain, cyclic rectal bleeding	-	-	Luminal obstruction due to mass, mucosal hemorrhage (distance 13 cm from anus)	-	Bowel wall thickening at rectum	TAH+BSO, low anterior resection
6	40	Constipation, pelvic pain	-	-	Submucosal tumoral mass (distance 22 cm from anus) with overlying normal mucosa	-	Submucosal mass lesion in rectosigmoid junction and luminal narrowing	Anterior resection

TAH+BSO: Total abdominal hysterectomy + bilateral salpingo-oophorectomy.

REFERENCES

1. Singh K, Lessells A, Adam DJ, et al. Presentation of endometriosis to general surgeon: a 10-year experience. *Br J Surg* 1995;82:1349-51.
2. Houtmeyers P, Ceelen W, Gillardian J, et al. Surgery for gastrointestinal endometriosis: indications and results. *Acta Chir Belg* 2006;106:413-41.
3. Olive DL, Schwartz LB. Endometriosis. *N Engl J Med* 1993;328:1759-69.
4. Mussa FF, Younes Z, Tihan T, Lacy BE. Anasarca and small bowel obstruction secondary to endometriosis. *Clin Gastroenterol* 2001;32:167-71.
5. Tran K, Kuijpers H, Willemsen W, Bulten H. Surgical treatment of symptomatic rectosigmoid endometriosis. *Eur J Surg* 1996;162:139-41.

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Ansa pancreatica: A rare pancreas ductal variation

Ansa pankreatika: Nadir bir pankreas kanal varyasyonu

To the Editor

The accessory dorsal pancreatic duct fuses with the main ventral duct at the pancreas head region in about 90% of the population, and drains through the major papilla, but it may also remain patent, emptying via the minor papilla. When the dorsal pancreatic duct enters the minor papilla by forming an arch like a reverse 'S' character, this type of ductal anatomy is referred to as 'ansa pancreatica'. It has been proposed in the literature that this type of anatomic variation may predispose to acute or chronic pancreatitis (1-3).

Among the patients who had undergone magnetic resonance cholangiopancreatography (MRCP) at our institution between November 2003 and June 2006, 4 patients (2 M, 2 F, age range: 49-71 years) had ansa pancreatica anatomic variation. MRCP was performed with a 1-T system (Signa Horizon; GE Medical Systems, Milwaukee, WI).

One patient had focal pancreatitis in the pancreatic tail; 1 had recently suffered from acute cholecystitis; 1 complained of nonspecific upper right quadrant pain; and 1 was being evaluated for elevated serum alkaline phosphatase (ALP) and gamma

ma glutamyl transferase (GGT) levels. On the MRCP images of these patients, the dorsal pan-

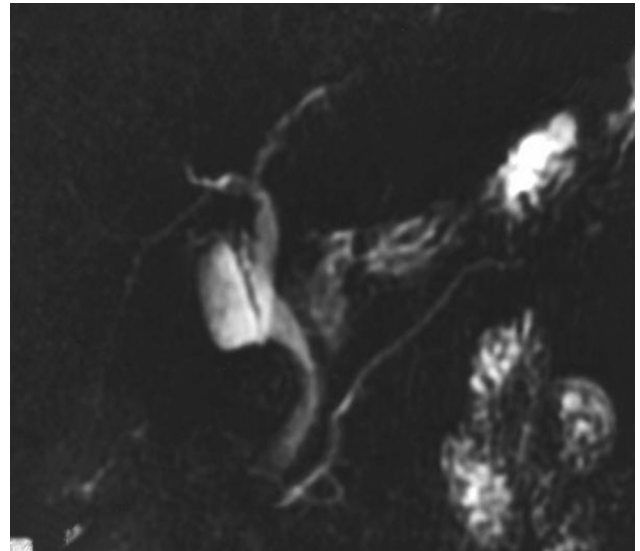


Figure 1. MRCP image in a 57-year-old man with remote acute cholecystitis demonstrating an incidental ansa pancreatica anatomic variation.

creatic duct communicated with an inferior branch of the ventral duct at the uncinat process, draining through the minor papilla (Figures 1, 2). Nevertheless, ansa pancreatica was not considered as a predisposing factor in any of them.

When there is an obliteration in the embryological course of the dorsal pancreatic duct at the confluence level with the ventral duct, the proximal portion of the dorsal duct communicates with the inferior branch of the ventral duct, and a new accessory duct shaped like a reverse 'S' character, terminating in the minor papilla, is formed (1,4). Despite the other side branches of the ventral duct, in the ansa pancreatica type, the accessory duct communicates with the ventral duct at an ob-

lique angle. Although it can be hypothesized that an accessory dorsal duct draining through the minor papilla may prevent pancreatitis by decreasing the pressure in the main duct, in the ansa pancreatica anatomic variation, the drainage is not that matured (1, 3).

'Ansa pancreatica' can be considered as a predisposing factor in patients with idiopathic pancreatitis, can accompany other risk factors such as alcoholism, and can also take part in the etiology of postoperative pancreatitis following pancreaticobiliary surgery (2). As MRCP is being widely used, it is important to recognize and clearly define the pancreas ductal variations, especially in the preoperative period, as they may be clinically significant.

REFERENCES

1. Bhasin DK, Rana SS, Nanda M, et al. Ansa pancreatica type of ductal anatomy in a patient with idiopathic acute pancreatitis. JOP 2006;7:315-20.
2. Koshinaga T, Fukuzawa M. Pancreatic ductal morphological pattern and dilatation in postoperative abdominal pain in patients with congenital choledochal cyst: an analysis of postoperative pancreatograms. Scand J Gastroenterol 2000;35:1324-9.
3. Tanaka T, Ichiba Y, Miura Y, Itoh H, Dohi K. Variations of the pancreatic ducts as a cause of chronic alcoholic pancreatitis; ansa pancreatica. Am J Gastroenterol 1992;87:806.
4. Ishii H, Arai K, Fukushima M, et al. Fusion variations of pancreatic ducts in patients with anomalous arrangement of pancreaticobiliary ductal system. J Hepatobiliary Pancreat Surg 1998;5:327-32.

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Carcinoid tumor of the ampulla of Vater presenting as recurrent acute acalculous cholecystitis attacks

Tekrarlayan akut akalkuloz kolesistit atakları ile görülen ampulla Vateri'nin karsinoid tümörü

To the Editor

Carcinoid tumors are rare neuroendocrine neoplasms that comprise only 1.2% to 1.5% of all gas-

trointestinal tumors. The ampulla of Vater is an extremely rare location for carcinoid tumors (less

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than 0.3% of all GIS carcinoids) (1).

A 63-year-old woman was admitted with nausea, pain in the upper abdominal region and jaundice. She had upper abdominal discomfort over the last two years and was treated for acute cholecystitis three times. Blood biochemistry evaluation revealed a total serum bilirubin of 3.66 mg/dl, direct bilirubin 2.75 mg/dl, aspartate aminotransferase 320 IU/L, alanine aminotransferase 263 IU/L, alkaline phosphatase 154 IU/L, and gamma glutamyl transferase 428 IU/L. Radiologic examination revealed no stones in gall bladder, thickness in the wall, dilated common bile duct (10 mm) and periportal, peripancreatic, and mesenteric lymphadenopathies. An endoscopic retrograde cholangiopancreatogram (ERCP) was performed and a swollen, ulcerated papilla was encountered, and papilla biopsy was performed (Figure 1). The immunohistochemical examination of the specimen showed the mass on the ampulla of Vater to be a carcinoid tumor.

Pancreatoduodenectomy with extended lymph node dissection was performed. The pathological examination confirmed the carcinoid tumor, 1 x 1.7 cm in diameter. Surgical margins were tumor-free and all lymph nodes dissected were negative for tumor infiltration. No chemotherapy was planned and the patient remains disease-free three years after the operation.

The characteristics of carcinoid of the ampulla of Vater (CAV) are unclear. The most common presenting symptoms of CAV are according to its mass effect, which are jaundice (59%) and epigastric pain (37%). The occurrence of carcinoid syndrome is rare (2), and this makes the diagnosis more difficult.

Detection of the tumor is not easy due to its size and mucosal sparing. ERCP and duodenoscopy are more helpful in the diagnosis of CAV and differential diagnosis for other types of periampullary tumors, papillary stenosis, or impacted stones, which may produce similar findings (2). Large and deep biopsies are needed to verify the tumor.

Our patient presented with jaundice and acute acalculous cholecystitis. Her medical history revealed recurrent acalculous cholecystitis attacks before jaundice was observed. ERCP was the ma-

ior helpful test for diagnosis in our patient.

Since the review of patients with ampullary carcinoid revealed that 25% of those patients have von Recklinghausen's disease, the clinical features of the disease such as skin lesions (café au lait spots) should be informative for the clinician regarding the location of the lesion that causes jaundice (3).

Carcinoid of the ampulla of Vater can be accepted as a benign disease considering the fact that five-year survival is over 90%. However, recent studies agree on the fact that achieving a tumor-free area is the key factor for good prognosis. Metastasis, rather than size of the tumor, has a direct relationship with the prognosis (4). In our case, computerized tomography scan suggested local lymph node involvement. We then performed a pancreaticoduodenectomy deciding that resection with larger tumor-free margins and resection of the lymph nodes in the neighborhood of the tumor would be the optimal approach in the patient. The patient is still well with no signs of a recurrence.

In conclusion, all patients with relapsing acalculous cholecystitis attacks should be examined for CAV, since early detection of this kind of tumor may improve the postoperative survival.

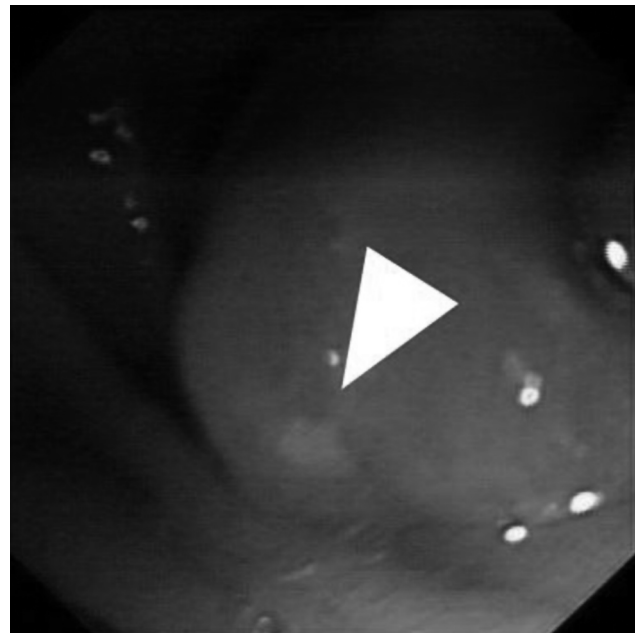


Figure 1. ERCP demonstrates the enlarged ampulla of Vater.

REFERENCES

1. Godwin JD 2nd. Carcinoid tumors. An analysis of 2,837 cases. *Cancer* 1975;36:560-9.
2. Clements WM, Martin SP, Stemmerman G, Lowy AM. Ampullary carcinoid tumors: rationale for an aggressive surgical approach. *J Gastrointest Surg* 2003;7:773-6.
3. Davis GB, Berk RN. Intestinal neurofibromas in von Recklinghausen's disease. *Am J Gastroenterol* 1973;60:410-4.
4. Suzuki H, Kuwano H, Masuda N, et al. Carcinoid of the papilla of Vater; a case report. *Hepatogastroenterology* 2003;50:371-3.

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