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Case report

Case report: two cases of spontaneous intramural duodenal hematoma associated with pancreatitis

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Abstract. Intramural duodenal hematoma (IDH) is a rare entity which is generally associated with trauma. Spontaneous (nontraumatic) intramural duodenal hematoma is associated with bleeding disorders, anticoagulation therapy, alcoholism, pancreatitis, tumors and duodenal ulcers. We report two cases of spontaneous intramural duodenal hematoma in middle-aged men who subsequently developed pancreatitis. The underlying pathophysiology is still unclear. In the cases described, it is not clear whether the intramural duodenal hematoma contributed to the development of pancreatitis or chronic pancreatitis has contributed to the development of IDH.

Key words: pancreatitis, duodenal hematoma, computed tomography

Introduction

We herein report two cases of intramural duodenal hematoma (IDH) in middle-aged men with no risk factors to suggest hematoma. Intramural duodenal hematoma is an uncommon condition that has been usually reported after blunt abdominal trauma, especially in children. Retroperitoneal fixation and disruption of rich submucosal and subserosal vascular plexus contribute to its development (1). Nontraumatic IDH is generally associated with bleeding disorders, anticoagulation therapy, alcoholism, pancreatitis, tumours and duodenal ulcers. It can also occur after endoscopic procedures (2). However, according to available literature, to date, the association between intramural duodenal hematoma and pancreatitis is still unclear. This study describes two cases of IDH with subsequently developed pancreatitis, one of which was Groove's pancreatitis.

Case Reports

Case 1

A 45-year-old male was admitted to the emergency department with a complaint of periumbilical abdominal pain for the last two weeks. The day before admission, the pain intensified and was associated with vomiting. He denied fever, hematemesis, hematochezia or abdominal trauma. He had a surgical history of appendectomy and left inguinal hernioplasty. He reported no other comorbidities and did not use any drugs regularly, but consumed alcohol occasionally. Examination revealed an afebrile, hemodynamically stable male with upper abdominal pain and voluntary guarding but without a rebound. Initial laboratory investigations revealed normal haemoglobin value and white cell count. Serum amylase was 136 U/L (reference range 23 – 91 U/L), lipase 481 U/L (ref.range

13 – 60 U/L) and gamma-glutamyl transferase 118 U/L (reference range 11 – 55 U/L). Markers of renal function and C-reactive protein (CRP) were normal. The remainder of the laboratory data was within normal limits. Contrast-enhanced abdominal computed tomography (CT) revealed a markedly dilated horizontal part of the duodenum filled with the content of mixed absorption coefficients with thickened walls in places (Figure 1).

The wall and folds of the descending section of the duodenum and proximal jejunum were also thickened. Upper gastrointestinal endoscopy showed a stenosed segment in the descending portion of the duodenum which could not be traversed by the endoscope. Due to high suspicion of duodenoduodenal intussusception, the patient underwent an urgent exploratory laparotomy which revealed the following: dilated duodenum and hematoma of the II and III segments of the duodenum. Following the adhesiolysis, the duodenum was mobilized, which triggered spontaneous perforation of the duodenal hematoma. No opening was found in the duodenal wall, nor was palpable resistance in the lumen detected. An interventional gastroenterologist did the intraoperative esophagogastroduodenoscopy till the proximal jejunum and no duodenal stenosis or intraluminal tumor process was found. The further postoperative course was uneventful, and the patient was discharged on the eighth postoperative day in a stable condition.

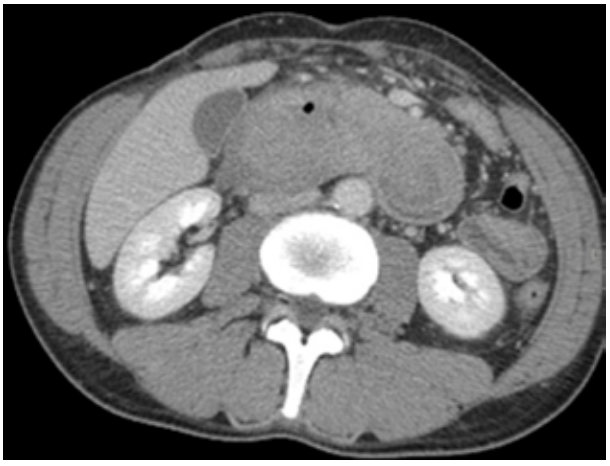

Figure 1. Contrast-enhanced abdominal computed tomography shows a markedly dilated horizontal part of the duodenum filled with the content of mixed absorption coefficients with thickened walls in places.

At the regular follow-up, three months after the initial surgery, the patient complained of pain in the epigastrium and anorexia, but he denied nausea and vomiting. In the area of the uncinatus process towards the head of the pancreas, CT showed a cystic-solid expansive lesion that was inseparable from the postbulbar segment of the duodenum. In addition, moderate amount of free intraperitoneal fluid and enlarged lymph nodes with a shorter diameter of up to 0.9 cm were noted. Radiological findings indicated a suspected neoplasm of the pancreatic head. The body and tail of the pancreas had a normal CT appearance with no visible focal lesions. Another surgical procedure was indicated. The duodenum and the head of the pancreas were firmly adhered to the right liver lobe and hepatoduodenal ligament. Cephalic duodenopancreatectomy (CDP) was performed with a standard reconstruction including a two-layer pancreaticojejunostomy, a single-layer hepaticojejunostomy and a double-layer gastrojejunostomy. Histopathological examination demonstrated the diagnosis of Groove pancreatitis (GP) (Figure 2).

Case 2

A-48-year old male patient presented with hematemesis and upper abdominal pain. The pain was non-radiating and located in the epigastrium with a severity of 7/10. He had a past history of diabetes and paranoid schizophrenia. His appetite was decreased,

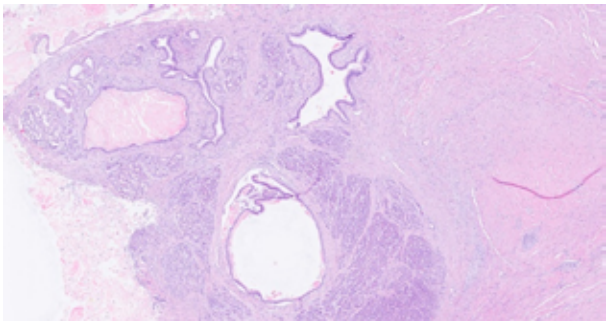

Figure 2. Ectopic pancreatic tissue within the "pancreatoduodenal groove" with dilated ducts containing homogeneous eosinophilic secretions.

but his bowel habits were usual and no fresh blood in the stool was visible. He smoked cigarettes for several years, but denied alcohol consumption. Blood pressure was 135/85 mmHg with a pulse rate of 96 beats/min, and physical examination demonstrated no palpable abdominal mass or organ. A digital rectal examination revealed the stool with a small amount of blood. On initial laboratory evaluation, haemoglobin was 159 g/L (normal 138 – 175 g/L), white blood cell count 15.5 H x109/L (normal 3.4 - 9.7 x109/L), C-reactive protein 34.6 mg/L (normal < 5 mg/L), bilirubin 40, alkaline phosphatase 156 U/L (normal 60 - 142 U/L), gamma-glutamyl transferase 259 U/L (normal 11-55 U/L), alanine aminotransferase 64 U/L (normal 12-48 U/L). Initial serum amylase was not evaluated.

Esophagogastroduodenoscopy showed hematinized contents with luminal narrowing. No bleeding lesion was found. A biopsy was performed and pathohistological analysis showed signs of inflammation. Control blood tests revealed a drop in hemoglobin from 159 g/L to 133 g/L, serum amylase level of 243 U/L (normal 23 – 91 U/L), serum lipase level of 133 U/L (normal 13 – 60 U/L) and an increase in C- reactive protein from 34.6 mg/L to 184.3 mg/L. Contrast-enhanced abdominal computed tomography was requested for further evaluation and found a large intramural hematoma (axial dimensions up to 10x7 cm) in the wall of the descending (Figure 2) and horizontal segment of the duodenum without clearly visible tumor process and without signs of active bleeding. Gross distension of the stomach was also noted. A nasogastric tube was placed for gastric decompression. We decided to manage the patient conservatively because there were no signs of active bleeding. After three days, a control CT was done which showed progression of hematoma size but still with no noticeable active bleeding. A narrow zone of inhomogeneous adipose tissue and lamellar fluid content along the pancreas was also observed. Given the elevation of inflammatory parameters and signs of biliary obstruction, surgery was indicated. Evacuation of IDH via duodenotomy was done with a drain left in the paraduodenal space. After 17 days of hospitalization, the patient remained asymptomatic and was safely discharged.

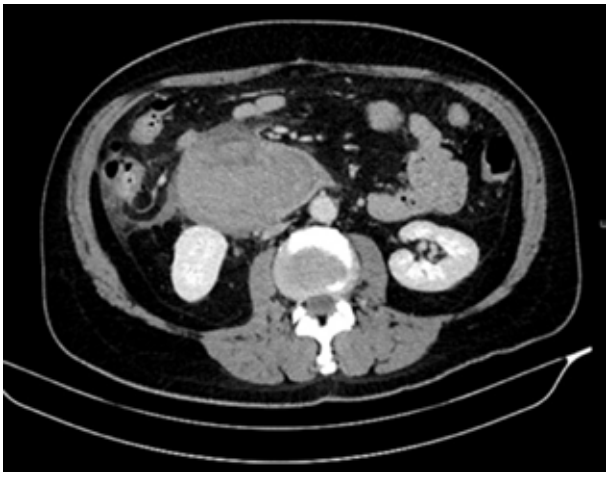

Figure 3. Axial contrast enhanced CT demonstrating intramural duodenal hematoma in the 2nd part of duodenum.

One month postoperatively, the patient was admitted to the emergency department due to upper abdominal pain. Laboratory exams showed high serum amylase levels (2066 H U/L) with slightly elevated inflammatory parameters. The patient was diagnosed with acute pancreatitis. He was managed conservatively with intravenous antibiotics, total parenteral nutrition, pain control and electrolyte correction. A few days before discharge, he was able to consume a low residual diet orally without abdominal discomfort.

Discussion

Spontaneous intramural duodenal hematomas are rare and generally related to anticoagulant treatment. The first description of this condition was made by McLauchlan in 1838 (3). Other causes include bleeding disorders, Henoch-Schönlein purpura, alcoholism, tumours, duodenal ulcers and endoscopic procedures (2). Reports of IDH associated with pancreatitis are infrequent, and it is difficult to clarify the true pathophysiology (4).

None of our patients had risk factors for intramural duodenal hematoma development. In both cases, additional laboratory diagnostic assessment was performed in order to exclude hereditary and acquired bleeding disorders. The first patient described had slightly elevated serum amylase and lipase values, however, no clear signs of acute pancreatitis were described on CT scan. Three months after the initial surgery, a diagnosis of GP was established.

Groove pancreatitis (also known as paraduodenal pancreatitis) is an uncommon form of chronic pancreatitis that involves the anatomical area between the pancreatic head, the duodenum and the common bile duct. Most patients are young men, and there is a strong association between Groove pancreatitis and alcohol abuse. The exact incidence is unknown, but it has been reported to be present in only 2% of pancreatic resections for chronic pancreatitis (5).

To date, according to available literature, the proposed mechanism of acute pancreatitis development implies duodenal obstruction by hematoma (6). In our case it is not clear whether the intramural duodenal hematoma contributed to the development of GP or pancreatitis was just a result of excessive alcohol consumption.

The pathogenesis is considered to be anatomical or functional obstruction of the minor papilla (7).

Our patient had symptoms for three months. It is known that symptoms can range from a few weeks to several years (8). He had upper abdominal pain and did not gain weight since the previous operation. He did not vomit because at the time of diagnosis, duodenal stenosis was not significant. Although biopsy did not provide malignant cells, we found surgery as a treatment of choice due to the intensity and persistence of the patient's symptoms and inability to rule out pancreatic cancer. It was also difficult to distinguish it from pancreatic cancer.

Differentiating Groove pancreatitis and pancreatic carcinoma is difficult based on radiologic features only. Patients with GP are younger than the average patient with pancreatic adenocarcinoma, and carcinoma manifests as a round, irregular mass with infiltrated arteries in the pancreatic head which is rarely seen in Groove pancreatitis (9). Surgical resection and histopathological analysis are required to determine the exact diagnosis.

Histopathological examination found the pancreas firmly fused to the thickened wall of the duodenum. The resected specimen revealed severe fibrosis and inflammatory cell infiltration at the groove area. No evidence of malignancy was present. Pancreatic tissue was predominantly of regular appearance, except in the area adjacent to the duodenum itself which suggest the pure form of GP. Unlike the pure form, the segmental form involves the entire head of the pancreas, with a stenotic and dilated main pancreatic duct (7).

Symptoms of IDH include upper abdominal pain, weight loss, vomiting and nausea due to duodenal stenosis. Jaundice may be seen in patients with involvement of the common duct.

Diagnosis relies on clinical suspicion and gastrointestinal tract radiography. Contrast-enhanced computed tomography is more recommended over magnetic resonance because of its availability and cost-effectiveness (10). This modality helps to exclude the possibility of intestinal obstruction and hollow organ perforation. Laboratory tests may indicate anemia with elevated inflammatory parameters which are nonspecific findings.

Management of patients with IDH in the majority of cases is non-operative (11). Treatment includes fluid resuscitation, blood replacement therapy and analgesia (12). In the first case presented, the surgery was necessary due to high suspicion of duodenoduodenal intussusception.

As noted earlier, we decided to manage our second patient conservatively because there were no signs of active bleeding, but after three days, a control CT showed progression of hematoma size. Due to an estimated risk of bleeding, surgery was indicated.

Lee et al. described successful endoscopic decompression for IDH with gastric outlet obstruction caused by acute pancreatitis (13). Due to minimally invasive nature, it has benefits. Endovascular techniques like arterial embolization are effective for the management of bleeding. It is important to highlight that in case of bleeding in hemodynamically unstable patients, the emergency laparotomy is mandatory.

Conclusion

Intramural duodenal hematoma is an uncommon complication of pancreatitis, but as shown in these two cases, it is difficult to distinguish if pancreatitis leads to IDH or IDH leads to pancreatitis. Regardless of currently unexplained pathophysiological mechanism of this condition, it is important to emphasize the importance of an early suspicion of IDH because of its rarity and fatal consequences in cases of delayed recognition.

Consent for publication: We obtained written informed consent for publication of this report from both of our patients.

Conflict of Interest: Each author declares that he or she has no commercial associations (e.g. consultancies, stock ownership, equity interest, patent/licensing arrangement etc.) that might pose a conflict of interest in connection with the submitted article

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