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Review

Hemosuccus pancreaticus: A mini-review

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ABSTRACT

Determining the cause of obscure bleeding in the gastrointestinal tract is the key in treating the disease. Hemosuccus pancreaticus (HP) could be an extremely rare disease. Ordinarily, bleeding in the pancreatic duct is defined as HP. At present, HP is the least frequent cause of upper gastrointestinal bleeding (1/1500), but can lead to massive gastrointestinal bleeding, which is potentially life threatening. Owing to its rarity, HP is difficult to diagnose, and the mortality rate of HP remains high in various studies. The purpose of this study is to expound on the basic symptoms and mechanisms of HP and to describe a potential significant examination method and treatment for usage in clinical practice.

1. Introduction

1.1. Nomenclature

Several terms have been used to describe bleeding from the pancreatic duct, including wirsumgorrhagia proposed by Van Kemmel in 1969, which is commonly used in France, or the equivalent hemowirsungia. The term hemosuccus pancreaticus (HP) was first proposed by Sandblom in 1970, and the term hemoductal pancreatitis was used by Longmire and Rose in 1973. All of these terms attempted to describe the occurrence of bleeding along the pancreatic duct through the papilla. HP is mostly reported in case reports [3,4].

1.2. Definition

HP is defined as bleeding from the ampulla of Vater via the pancreatic duct. It is caused by a bleeding source in the pancreas, pancreatic duct, or structures adjacent to the pancreas, such as the splenic artery and gastric artery. HP is mostly caused by acute or chronic pancreatitis. Aneurysm is also often associated with HP; however, a causal relationship between them has not been established yet [4–7]. However, in some rare cases, HP can be merged with pancreatic tumors and even with pancreatic cystic neoplasms [8,9]. According to some early reports, HP can even arise from trauma iatrogenic causes [10–12].

1.3. Nosazontology

Multiple studies have attempted to explain the reason of bleeding.

1.3.1. Arterial aneurysm

Arterial aneurysm in or out of the pancreas is a main cause of HP. Some studies have verified that the rupture of aneurysm in the splenic artery associated with pancreatitis can cause HP [27]. Pseudoaneurysm of the hepatic, gastroduodenal, or pancreaticoduodenal artery has also been reported. Rupture of a pseudoaneurysm into a pseudocyst is rare; in fact, reports show an incidence rate of 6–17% of intracystic bleeding in patients with chronic pancreatitis. The splenic, common hepatic, gastroduodenal, and pancreaticoduodenal arteries are the areas mostly involved [18]. In some rare cases, aneurysms may not be present; however, pathological or iconographic examinations show findings suggestive of vascular malformations [20].

1.3.2. Inflammatory changes in the pancreas

Acute and chronic pancreatitis may result in pancreatic duct injuries. The corrosion of the pancreatic juice may cause the rupture of the peripheral vascular wall, which lead to HP. Furthermore, some infrequent causes are pancreatolithiasis and pseudocysts of the pancreas, which may lead to inflammation of the pancreatic duct [28].

The unusual propensity of pancreatic pseudocysts associated with false aneurysms suggests that pancreatic enzymes play pathogenic roles [30]. Pseudocysts have been shown to contain activated lytic enzymes. Elastases are observed, in particular. These enzymes exert an elastolytic action on the elastic component of the vessel walls and result in bleeding [31,32]. A pancreatic pseudocyst possesses a significant relationship with the pancreatic duct, and bleeding in the gastrointestinal tract may also occur entirely through this route [31]. According to Zuchelli et al., bleeding is intermittent owing to clot formation in the

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main pancreatic duct [14].

1.3.3. Pancreatic tumors

In many reports, patients with HP have a history of pancreatic tumors (including pancreatic carcinoma, serous cystic neoplasm, mucinous cystic neoplasm, and neuroendocrine tumors) [9,15,21]. In some patients with malignant tumors, Shinzeki et al. reported that the cancer cells in mucinous cystadenoma did not infiltrate the parenchyma or the pancreatic duct [29]. Therefore, they assumed that the presence of malignancy would not be associated with the onset of HP. However, Matsumoto et al. concluded that tumor bleeding via communication between tumor cysts and the pancreatic duct may lead to HP [9]. Further studies, especially verdicts from clinicopathologic data, are needed to elucidate how tumors cause HP.

1.3.4. Other causes

Pancreatic trauma may result in HP. In some case reports, pancreas divisum [22] or iatrogenic operations (HP after endoscopic ultrasound [EUS] needle aspirations or T-tub arrangements) [23–25] may lead to HP.

2. Management

2.1. Manifestation

The age of onset is widely distributed; the mean age range of patients with HP is from 32 to 36 years [13–15]. In terms of sex, men have a higher risk of developing HP than women. The disparity can be as large as five times [13], which still needs to be proven. The clinical symptoms and signs mainly include upper gastrointestinal bleeding, which could be evidenced by hematemesis and melena. The key symptom of HP is melena, which is the most common. Hematemesis is less frequent, and the rupture into the abdominal cavity or the retroperitoneum is rare [15–17]. The characteristic colic pain is a result of the increased intraductal pressure, which is caused by obstruction of the Wirsung duct due to clot formation. The bleeding is usually intermittent, repetitive, and often not severe enough to cause a hemodynamic instability. Thus, most patients show symptoms of chronic anemia or even aggravating anemia. However, some acute cases manifested as severe hematemesis or shock at the early phase and needed prompt blood transfusions [15].

According to various studies, most cases have a history of strictly pancreatic original diseases. Most frequently, the history of pancreatitis has the highest priority. HP is even treated as a complication of chronic pancreatitis [18]. In addition, Sul et al. suggested that even if a patient does not have any evidence of chronic pancreatitis, HP must be included in the differential diagnosis for chronic alcoholics with intermittent upper gastrointestinal bleeding [19]. Significantly, histories of alcohol consumption could be observed in many patients. In some further speculation, asymptomatic chronic alcoholic pancreatitis might be a risk factor of HP.

2.2. Diagnosis

The diagnosis of HP remains challenging. Owing to its rarity, anatomical location, and intermittent symptoms, the diagnosis of HP can be extremely difficult. Blood tests usually do not reveal significant findings, and liver function test results are usually normal apart from an increased serum bilirubin level in the events of pancreaticobiliary reflux. The serum amylase level is also normal, except for episodes of acute pancreatitis [13]. Various diagnostic modalities can aid in the diagnosis of HP. To rule out other causes of gastrointestinal bleeding, endoscopy is essential. In some rare cases, active bleeding can be seen from the duodenal ampulla [26,33,34]. Endoscopy can detect active bleeding via the papilla in only 30% of patients [35]. Pancreatic pseudocysts or aneurysm of the peripancreatic arteries can be found on ultrasonography. Doppler ultrasound or dynamic ultrasound could be used to find vascular abnormalities. Contrast-enhanced computed tomography (CT) scan excellently demonstrates the pancreatic pathology and features of chronic pancreatitis, pseudocysts, and pseudoaneurysms. On pre-contrast CT scan, the characteristic finding of clotted blood in the pancreatic duct, known as the sentinel clot, is seldom seen; however, it attests the bleeding from the pancreas. CT scan may show simultaneous opacification of an aneurysmal artery and pseudocyst or persistence of a contrast within a pseudocyst after the arterial phase [36]. Thus, CT scan remains an important diagnostic modality, which may help allow early therapeutic interventions. Ultimately, angiography remains the gold standard for diagnosis and therapy. It identifies the causative artery and helps delineate arterial anatomy for therapeutic interventions.

2.3. Treatment

2.3.1. Non-surgical treatment

Eradicating the source of bleeding is the only way to cure HP. Considering the influence of pancreatic enzymes, drug therapy seems feeble. Various treatments have been tested. Interventional radiological procedures and surgery are the main potential approaches. Once the patient is hemodynamically stable, interventional procedures are effective as an initial treatment in 67%–100% of cases [37]. If the source of bleeding is found on angiography, interventional radiographic procedures are the first choice for initial management with immediate good results in 79–100% of cases and have an overall success rate of 67% [17,38]. Interventional treatment, i.e., endovascular treatment, includes three methods: mobilization via prosthetic materials, balloon tamponade, and stent grafting. Coil embolization is the most frequently described technique. It stimulates thrombosis in the pseudoaneurysm, which could successfully stop bleeding. Benz et al. described the first successful implantation of an uncoated metal of Palmaz stenting across the aneurysmal segment of the splenic artery [39]. They suggested that implantation of a metal stent may be an effective treatment for HP with a low rate of recurrence and complications. There is a low risk (0.5%) of cellulitis at the femoral access site of embolization [40]; nevertheless, no major septic complications were observed with endovascular treatments [41].

2.3.2. Surgical treatment

For patients with hemodynamic instability, emergency operations are inevitable. In most cases, surgical management of hemorrhagic pseudocysts includes excision of the pseudoaneurysm and pseudocyst. If resection is impossible, the ligation of the artery proximal and distal to the pseudoaneurysm and the abscission drainage of the pseudocyst that enters the gastrointestinal tract are an acceptable alternative; however, these methods are associated with a higher re-bleeding rate [42]. The anatomical position might affect the prognosis. Distal pancreatectomy for bleeding pancreatic pseudoaneurysms in the body or tail of the pancreas is a surgical alternative to angioembolization. When the pseudoaneurysm is located in the head of the pancreas, surgical resection is associated with increased mortality and morbidity rates, and independent angioembolization has been proposed as the recommended treatment modality of choice [13]. Surgical treatment may be appropriate for HP from the pancreatic gland. Nevertheless, the source of bleeding is usually difficult to confirm, which will determine the cutting line of the pancreas. Hence, intraoperative sonography and pancreatoscopy should be performed to confirm the origin of bleeding [27].

2.3.3. New type of treatment

A novel technique of EUS-guided angiotherapy in treating HP was reported by Will et al. for the first time [43]. It can be an important diagnostic and therapeutic tool. Sayilir et al. reported a case of HP in a patient with chronic pancreatitis where embolization for the treatment