

## Hemosuccus pancreaticus following Parathyroidectomy for Primary parathyroid adenoma

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### ABSTRACT

Hemosuccus pancreaticus is defined as intermittent bleeding from the pancreatic duct into the GI tract, usually caused by the rupture of an pseudoaneurysm, which is usually associated with pancreatitis, abdominal surgery, and can result in life-threatening hemorrhage. Hyperparathyroidism is a rare cause of pancreatitis, it should be suspected in all patients with a history of primary hyperparathyroidism. When these diseases coexist, the course of pancreatitis can be rather severe and often complicated. This report detail a case of Hemosuccus pancreaticus due to hyperparathyroidism associated pancreatitis and its successful emergency conservative management by Embolization Techniques.

**Keywords:** Hemosuccus pancreaticus, hyperparathyroidism, transcatheter arterial embolization, endoscopy

### INTRODUCTION

Among the rare causes of gastrointestinal hemorrhage, hemosuccus pancreaticus (HP), also known as Wirsungorrhagia or pseudohemobilia, was first described by Lower and Farrell in 1931 (1). Because of the rarity and consequent unfamiliarity with the condition, the diagnosis is difficult-to-make. Patients with HP present with abdominal pain has a "crescendo-decrescendo" character, and GI hemorrhage manifesting as silent anemia with melena or as intermittent massive bleeding into GI tract. HP is a serious and rare complication of chronic pancreatitis which may result from pseudoaneurysms of peripancreatic arteries (such as the gastroduodenal artery) (2).

Hyperparathyroidism is an extremely uncommon condition implicated as a cause of pancreatitis (3).

Some studies suggest that correction of hyperparathyroidism and normalized serum calcium levels may ameliorate the associated pancreatitis (4,5), however, surgical resection of primary parathyroid adenoma has been questioned. To this point, the surgery may not reverse the pancreatic pathology and long-term therapy for exocrine and endocrine pancreatic insufficiency may be required (6)<sup>P</sup>. Here, we report a patient with pri-

mary hyperparathyroidism who developed associated pancreatic diseases and sequent HP after removal of a parathyroid adenoma. The patient was treated successfully with selective TAE despite of the complication of duodenal hematoma due to technical reasons.

### CASE PRESENTATION

A 42-year-old male presented at the emergency room for evaluation of melena and severe abdominal pain that had occurred in the last 4 month. The patient's medical history revealed he had undergone surgery for a benign parathyroid adenoma in February 2004, Serum calcium and phosphorus levels returned to normal and urinary cAMP and PTH returned to nearly normal after the operation. From January 2008 to January 2010, he experienced a few episodes of epigastric dull pain and melena. There were 8 attacks during this year. One month latter, the patient suddenly fell anorexic, then develop occasional nausea and vomiting of gastric contents, and had persistent anemia with a hemoglobin level dropped to 6.5 g/dL, on 30 march 2010, he experienced one episode of hematemesis of 200 mL in vaule. One weeks later, Abdominal Contrast-enhanced computed tomography (CT) scan in another hospital confirmed chronic pancreatitis with cystic lesion at the pancreatic head (Figure 1a). Bilateral renal calculi. On 3

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June 2010, he had abdominal pain again, and Gastrointestinal endoscopy (GIE) revealed duodenal hemorrhage trickling from the papilla. On 21 June 2010, he was admitted to our emergency room, the concentration of hemoglobin decrease to 4.8 g/dL. The patient was resuscitated with crystalloids and transfusion. The melena occurred only once, after arriving at emergency room, two days later, he was referred to our department for further evaluation.

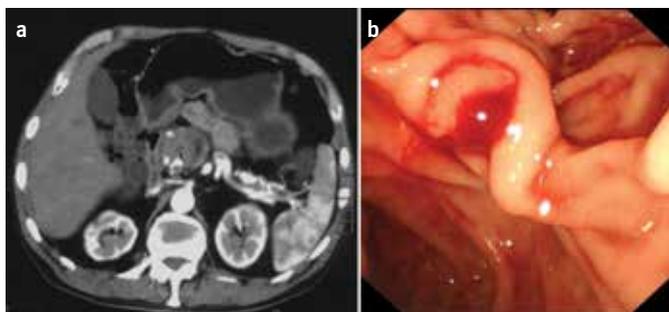
On 26 June, Duodenoscopy demonstrated intermittently active bleeding coming out of papilla again (Figure 1b). She suddenly vomiting, became hemodynamic unstable. Based on the clinical picture and past history, bleeding from a pseudoaneurysm due to the pseudocyst, that is, Hemosuccus pancreaticus was suspected. Emergent Angiography of the celiac artery was performed the next day. It showed a pseudoaneurysm in the gastroduodenal artery (Figure 2a), Through a 3F microcatheter (Terumo corp, Japan) was inserted into the celiac artery and selective embolized gastroduodenal artery (GDA) by TAE (Figure 2b). The subsequent CT scan control performed 6 days after Embolization in this study showed the complete occlusion of the gastroduodenal artery. Additional findings include a retroperitoneally mass located in descending part of duodenum (8 cm × 8 cm) (Figure 3a). In the next day, a repeat endoscopy confirmed no active bleeding in the upper gastrointestinal tract and a large hematoma (5 cm × 3 cm) occupying duodenum near the papilla duodeni (Figure 3b). Laboratory findings revealed an increase value of bilirubin (68.9 μmol/L) and transaminase (AST 64 μmol/L,

ALT 42 μmol/L). 9 days after conservative treatment, an abdominal CT study was obtained before discharge shows the hematoma was decreased in the size to 6 cm. His clinical status was gradually improved. After discharge, the patients were followed and had no further bleeding episode for 2 years.

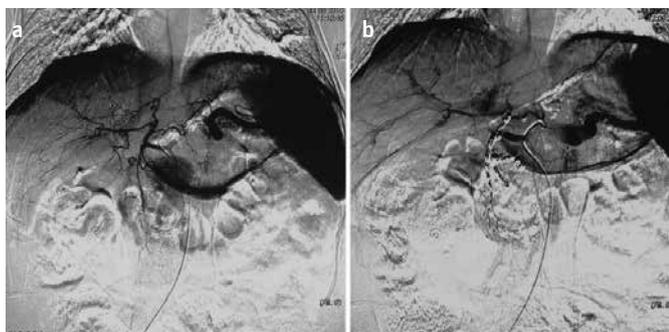
**DISCUSSION**

Primary Hyperparathyroidism (PHPT) is a rare cause of pancreatic disease with a reported prevalence of 1.5% of chronic pancreatitis cases (7). These patients with hyperparathyroidism associated pancreatitis may develop complications (such as pancreatic pseudocysts, calcification), and surgical correction of parathyroid disease can resolve pseudocysts spontaneously. While, the effect of parathyroidectomy on pancreatic pseudocysts complicated with PHPT is less clear due to paucity of information regarding the clinical characteristics of pancreatic disease in hyperparathyroidism (5,8).

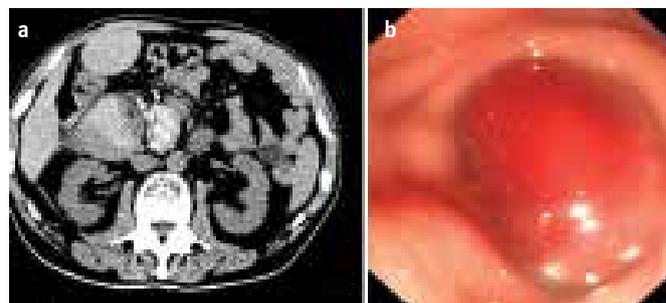
In the present case, we did not know whether the pseudocyst was formed, because the patient was asymptomatic on the abdomen when he underwent parathyroidectomy. The possible pathophysiological mechanism that leads to pancreatitis and related complications seems more related to hypercalcemia induced by PHPT (9), though the higher serum calcium values turn to normal after Parathyroidectomy. Pyrah et al. (10) had attempted to classify the circumstances under which pancreatic disease was found in association with PHPT in 1966, classification was modified to four major classes by Jacob et al. (6), that is to say, PHPT can present as pancreatitis with or without pancreatic calcification according to class 4. PHPT induced hypercalcemia is a risk factor for pancreatitis. It activates the transformation of trypsinogen into trypsin (11), this activation process plays a pivotal role in toxicity to pancreas because it also initiates the pancreatic enzyme cascade (12) as well as, calcium ions cause calculus deposition within the pancreatic ductules and parenchyma (13), with consequent pancreatic calcification and inflammation. The emerging chronic pancreatitis and blockage of a major branch of the pancreatic duct by protein plugs, calculus or localized fibrosis (14) attribute to pseudocysts adjacent to head which shown on our CT examination.



**Figure 1. a,b.** Contrast enhanced multi-slice abdominal CT scan showing the solid mass with a diameter of 5 cm involving the head of the pancreas (small rod) (a). Endoscopy reveals bleeding from papilla of Vater (b).



**Figure 2. a,b.** The 3-Fr catheter is selectively inserted into the gastroduodenal artery, showing an irregular lumen and filling of the distal section of the mass (a). The triangles show the arteries to be closed with stainless steel fibrillar coils (b).



**Figure 3. a,b.** CT scan of abdomen showing artifact (arrow) from coils in the origin of the GDA, and a retroperitoneally mass (triangle) located in descending part of duodenum (8 cm × 8 cm) (a). Endoscopy confirmed no active bleeding in the upper gastrointestinal tract and complete obstruction at the descending level of the duodenum (b).

Although PHPT who developed pancreatic pseudocysts is a quite rare evenience, another rare but potential life-threatening concomitant complication of Hemosuccus pancreaticus result from pseudocyst wall abutting the gastroduodenal artery occurred in the same patients was only by chance (15). In our present case, Angiography showed gastroduodenal artery pseudoaneurysm with no active hemostasis was detected. we hypothesis that the pseudoaneurysm result in HP, whose is characteristic with feature of intermittent bleeding from the papilla, and the appearance of bleeding pseudoaneurysm may be normal when performed occlusion between bleeding episodes. Whether selective angio-embolization provides permanent hemostasis or is just a temporizing process is controversial. Hemosuccus pancreaticus is traditionally treated by surgical method of distal pancreatectomy, but transcatheter selective arterial embolization has become popular to be an effective and organ-preserving alternative for this disorder when it is caused by a primary aneurysm or pseudoaneurysm (16). Based on the CT findings that the pseudoaneurysm was located in the head of the pancreas in this patient, surgical resection is associated with increased mortality and morbidity (17), consider the patient also in a bad general condition, therefore angioembolization alone has been recommended as the initial therapeutic method to surgery. In fact, the pseudoaneurysms are successfully treated with Angioembolization, The subsequent CT scan and repeat endoscopy performed for the patient in this study showed the therapeutic efficacy of TAE in the treatment of upper gastrointestinal bleeding caused by the pseudoaneurysms of GDA. While, Selective embolization of the GDA stump and/or pseudoaneurysm frequently caused some complications because it is technically challenging (18). We hypothesize that in this patient, "duodenal hematoma" was facilitated by ischemia of the duodenum after TAE, and contributed to the development of secondary obstructive cholangitis (19), present as increasing bilirubin and transaminase levels in this case.

In conclusions, our case was first described as Hemosuccus pancreaticus can occur following for Primary parathyroid adenoma after Parathyroidectomy, although Chronic pancreatitis and sequent complations due to hyperparathyroidism often relieves some clinical manifestations on majority of patients underwent parathyroidectomy (4,5,9). Early diagnosis and correct management is essential in preventing the bleeding. The diagnosis of hemosuccus pancreaticus requires a high level of expertise. Upper gastrointestinal endoscopy rarely reveals active bleeding from the ampulla of Vater but rules out other sources of GI bleeding (20), and angiography is the first approach to characterizing the exact bleeding site with subsequent treatment by interventional radiology (21).

**Conflicts of Interest:** No conflict of interest was declared by the authors.

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