Haemosuccus pancreaticus and seven episodes of recurrent unlocalised upper gastrointestinal bleeding

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SUMMARY

Upper gastrointestinal (GI) bleeding is a common medical condition that results in extensive morbidity and mortality, as well as substantial healthcare costs. While there is variation among society and consensus guidelines, the approaches to assessment and evaluation are generally consistent. Our case describes a man in his 40s who presented with seven episodes of recurrent upper GI bleeding over 2 years secondary to haemosuccus pancreaticus. While rare, this case study highlights key principles to the initial diagnostic approach that, in appropriate clinical contexts, should be applied to patients with unlocalised upper GI bleeding. We further perform a complete systematic review of similar cases available in PubMed (36 patients in 24 case reports) to further refine these diagnostic principles.

BACKGROUND

In computer science and machine learning applications, edge cases are an opportunity to better understand and improve existing algorithms. An edge case is a specific scenario that occurs only at an extreme (usually the maximum or minimum) of operating parameter—the computer science equivalent of a 'rare presentation'. By assessing the performance or understanding the output of the method or algorithm to 'rare presentations', developers begin to understand the limitations of the aforementioned methods/algorithms and can begin to improve them.[1] Thus, while upper gastrointestinal (GI) bleeding is common (annual incidence of 1 in 1000),[2] exploring the diagnostic odyssey of a case study of haemosuccus pancreaticus (a rare cause of bleeding, <1% of upper GI bleeds)[3] reveals both the strengths and weakness of current diagnostic approaches to GI bleeding management.[4–6] This is especially critical given the acuity and substantial risk for recurrence of this rare presentation, which often requires aggressive resuscitation and intervention to prevent morbidity and mortality. Aside from case reports, there is no data or systematic investigation into the diagnosis/treatment of haemosuccus pancreaticus, and how that fits into the broader approach to assessment and evaluation of upper GI bleeds. This case report (and systematic review) on our patient with seven episodes of recurrent upper GI bleeding highlights the importance of the identification of particular risk factors and certain diagnostic principles that should be applied when recurrent GI bleeding remains unlocalised.

CASE PRESENTATION

A man in his 40s with chronic pancreatitis complicated by pseudocysts and alcoholic cirrhosis (Model for End-Stage Liver Disease [MELD] score of 10), in the context of chronic and significant alcohol use since he was a teenager, was referred to our pancreatic surgery clinic following seven hospitalisations for upper GI bleeding for the past 2 years, including three admissions in the 3 months leading to definitive diagnosis.

His first hospitalisation in our hospital system occurred 3 months prior to his presentation to us when he presented with haematemesis and abdominal pain. At this point, the patient had a known history of presumed, unlocalised, multifocal small bowel bleeds, with four prior admissions at outside hospitals over the past 2 years. Given radiographic evidence of cirrhosis on the right upper quadrant ultrasound from earlier in the year and radiographic evidence of varices on admission, he was empirically treated for possible variceal bleed with octreotide and intravenous proton pump inhibitors. He underwent oesophagogastroduodenoscopy (OGD) and colonoscopy, which demonstrated a small Mallory-Weiss tear with a clean base and no active bleeding, and otherwise normal EGD and colonoscopy. Video-capsule endoscopy (VCE) was subsequently performed, requiring trans-pyloric placement as the capsule did not leave the stomach on the first attempt. A red blood cell scan was also considered at this time, but the capsule study was likely higher yield given the resolution of bleeding. The OGD/VCE showed grade 1 varices, as well as redemonstrated the previously noted small non-bleeding Mallory-Weiss tear with a clean base. There was also oozing blood at the entrance of the duodenum, which at the time was thought to be related to scope trauma. He required a total of 2 units of packed red blood cells (pRBCs) during this hospital stay to maintain a haemoglobin >7 g/dL. Given the resolution of bleeding and haemodynamic stability, additional diagnostic (eg, cross-sectional imaging) and therapeutic workup was deferred.

Within 24 hours of discharge, the patient re-presented with recurrent haematochezia and an episode of haematemesis. CT angiogram (figure 1) demonstrated new haemorrhagic conversion of an enlarging pancreatic tail pseudocyst, and possible haemorrhage into the pancreatic head pseudocysts, on the basis of interval enlargement. Other notable imaging findings included systemic arteriopathy with extreme visceral and renal tortuosity, beading and ectasia, but no acute complications, as well as bilateral iliac artery ectasia. A subsequent EGD study, on the same day, demonstrated

bleeding at the ampulla (figure 2), which was the first direct visualisation of bleeding from the pancreatic duct (ie, haemosuccus pancreaticus).

An endoscopic retrograde cholangiopancreatography (ERCP was considered at the same time to help differentiate from which ductal system (biliary vs pancreatic) the bleeding was originating from. However, given the CT angiogram findings obtained on admission (figure 1), we were confident that the bleeding was pancreatic in origin. The gastroenterologists also determined that no endoscopic procedure would allow for treatment of his condition given the location of the lesion in the distal pancreas and haemorrhagic pseudocysts that would ultimately require surgical intervention for removal. One of the indications for surgical intervention in haemosuccus pancreaticus includes the presence of haemorrhagic pseudocysts for the removal of the pseudoaneurysm and/or pseudocyst.

Similarly, the interventional radiologists determined that a formal angiogram or empiric embolisation would be low-yield in the absence of active bleeding. Part of the hesitation was that the CT angiogram (figure 1) obtained on admission demonstrated the aforementioned haemorrhagic conversion of an enlarging pancreatic tail pseudocyst as well as possible haemorrhage into the pancreatic head pseudocysts on the basis of interval enlargement. However, without definitive extravasation, the interventional radiologists felt that there were likely too many targets for embolisation (eg, splenic for the tail pseudocyst vs possible embolisation for question of possible arterio-cyst fistualisation with gastroduodenal, coeliac or common hepatic arteries). In addition, haemorrhagic pseudocysts would ultimately require surgical intervention for removal and thus the interventionalists felt a surgical intervention was more appropriate. The patient received a total of 1 unit of pRBC.

Within a month, the patient re-presents as a transfer from an outside hospital for recurrent bleeding secondary to haemosuccus pancreaticus. His presenting symptoms included severe abdominal pain (described as 'twisting of his bowels'), dizziness and hyperhidrosis. An emergent bedside EGD revealed bleeding from the major papilla. At the outside hospital, his presentation was complicated by haemorrhagic shock, requiring admission to the intensive care unit and intubation; there was concern for aspiration with his haematemesis and possible procedural interventions. He was stabilised with 3 units of pRBCs, extubated 2 days later and admitted to the floor. Given the need for complex management, the patient was transferred to our institution 7 days later for possible embolisation. A repeat CT angiogram was negative for active extravasation and interventional radiology deferred treatment. The patient remained stable and he was referred to our clinic.

TREATMENT

Given the increasing frequency, severity and recurrence of his bleeds, as well as the likely source of a pseudocyst in the tail of the pancreas that drains into the pancreatic duct (and subsequently into the ampulla), we felt the patient would benefit from an elective distal pancreatectomy and splenectomy. While there are no consensus or society guidelines, indications for surgical intervention include the following[7 8]:

- Haemodynamic instability and/or uncontrolled bleeding that is unresponsive to resuscitation
- Patients with haemorrhagic pseudocysts to remove the pseudoaneurysm and/or the pseudocyst
- Failure of other therapeutic modalities (eg, embolisation or endoscopic interventions)

Surgical intervention may also be considered in patients with negative angiographic results but with high suspicion for haemosuccus pancreaticus. The patient and his family also felt surgical intervention would be the best option, eliminating the uncertainty around the GI bleeding, especially with all other endoscopic or interventional imaging options exhausted. While it is difficult to obtain accurate estimates given the rarity of the diagnosis, case studies suggest that angiographic embolisation has a success rate of about 67%–100%, with a recurrence rate of 30% of bleeding following the embolisation.[8] Comparatively, surgery has a success rate of 70%–80% with a risk of recurrent bleeding between 0% and 5%.[7 8]

The operation was performed, under combined epidural and general anaesthesia, through an upper midline incision. On entering the abdomen, the liver appeared cirrhotic, with multiple venous collaterals in the greater and lesser omentum, and the mid-body of the pancreas was noted to be very indurated and atrophied. We dissected out the splenic artery, close to its origin, and in the process, we entered the pseudocyst, which had cloudy fluid, but did not appear to be haemorrhagic. We subsequently clamped, transected and suture ligated the splenic artery, before incising the inferior border of the pancreas. After development of the retropancreatic plane and appropriate mobilisation, we delivered the distal pancreas and spleen through the incision. The patient tolerated the procedure well and bleeding was estimated at 1.5 L. The degree of fibrosis in the presence of the pseudocyst in the area of the tail of the pancreas made the dissection of the distal pancreatectomy much more complex.

On examining the specimen, the distal pancreatic specimen had an obvious defect anteroposteriorly (ie, a literal hole) that communicated with the main pancreatic duct (figure 3), which we suspected may have been responsible for the recurrent bleeding presentation. During the operation, we noted that there was marked induration consistent with the walls of the pseudocyst, and we found intermittent relation to the splenic artery (the likely source of bleeding).

OUTCOME AND FOLLOW-UP

Following the surgery, the patient recovered well. As his oral intake increased postprocedure, so did his blood glucose, with the majority of his blood sugars >200. He was discharged home on postoperative day 7, following extensive teaching and with appropriate follow-up. He was also administered the Hib, pneumococcal 20-valent conjugate, first meningococcal (groups A, C, Y and W-135) oligosaccharide diphtheria CRM197 conjugate and first meningococcal group B vaccines prior to discharge and was provided with arrangements for follow-up second Menveo and Bexsero vaccinations with his primary care provider as an outpatient. At 12 months following the operation, he has returned to his baseline health status without any recurrence of bleeding.

DISCUSSION

Haemosuccus pancreaticus describes any GI bleed that originates from the pancreatic duct or major papilla.[8] This broad definition, thus, includes any bleeding source in the pancreas, pancreatic duct or pancreas-adjacent structures (eg, splenic artery) that can bleed into the pancreatic duct.[7 8] While it is difficult to reliably estimate the prevalence, the current case series suggests haemosuccus pancreaticus make up <1% of upper GI bleeds.[9–31]

To help situate our patient's presentation within the broader population of those presenting with haemosuccus pancreaticus and understand how to better improve existing evaluation

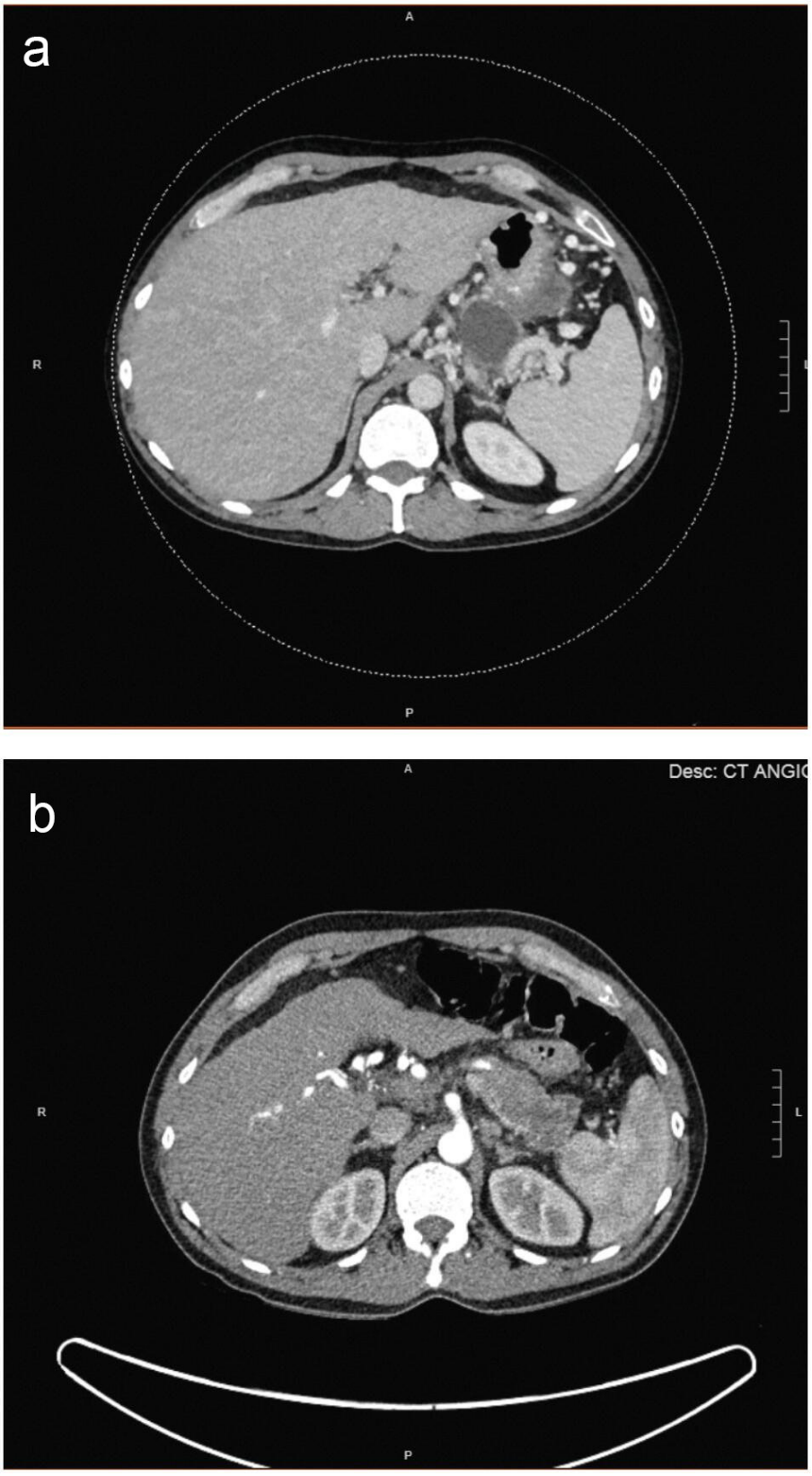

Figure 1 Radiological findings. (A) CT abdomen/pelvis when bleeding from the ampulla was first visualised on endoscopy. The pancreas is notable for parenchymal calcification compatible with chronic pancreatitis and no ductal dilatation. There is 2.1×1.5 cm hypodensity along the pancreatic tail, decreased in size from prior study, likely related to evolving haemorrhage within a pancreatic pseudocyst. (B) First CT abdomen/pelvis when the patient was seen in our hospital system for upper GI bleeding; he was not diagnosed with haemosuccus pancreaticus during that first hospitalisation. Imaging was significant for an unchanged 3.6×3.0 cystic lesion with a high attenuating rim likely arising from the pancreatic tail, abutting the greater curvature of the stomach, consistent with known pancreatic pseudocyst. There was no main duct dilation despite several low-attenuation lesions in the pancreatic head and the uncinate process measured up to 9 mm.

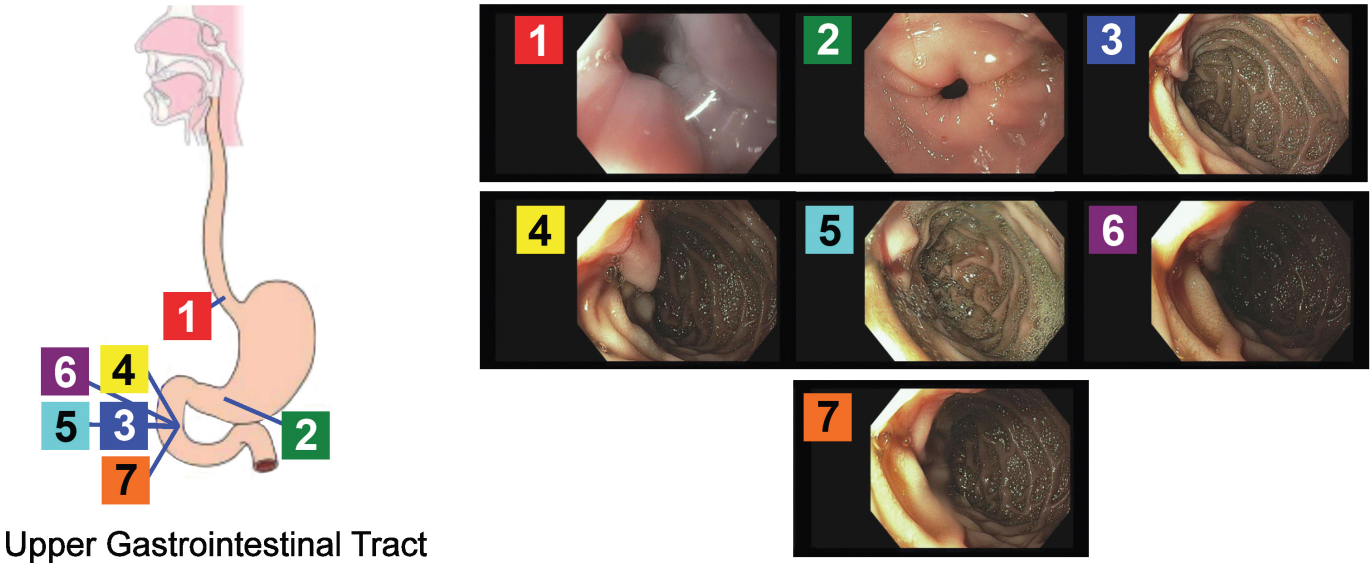

Figure 2 Endoscopic findings. Endoscopic images showing normal mucosa throughout the upper gastrointestinal tract and bleeding in the duodenum from the ampulla of Vater.

algorithms for upper GI bleeds, we performed a comprehensive literature review of all case studies indexed in PubMed that contained the terms 'pseudocyst' and 'haemosuccus pancreaticus' (online supplemental table 1; n=24 studies[9–31] and 36 patients). Two of the original 26 case studies were excluded because the bleeding was unrelated to pancreatic pseudocysts.

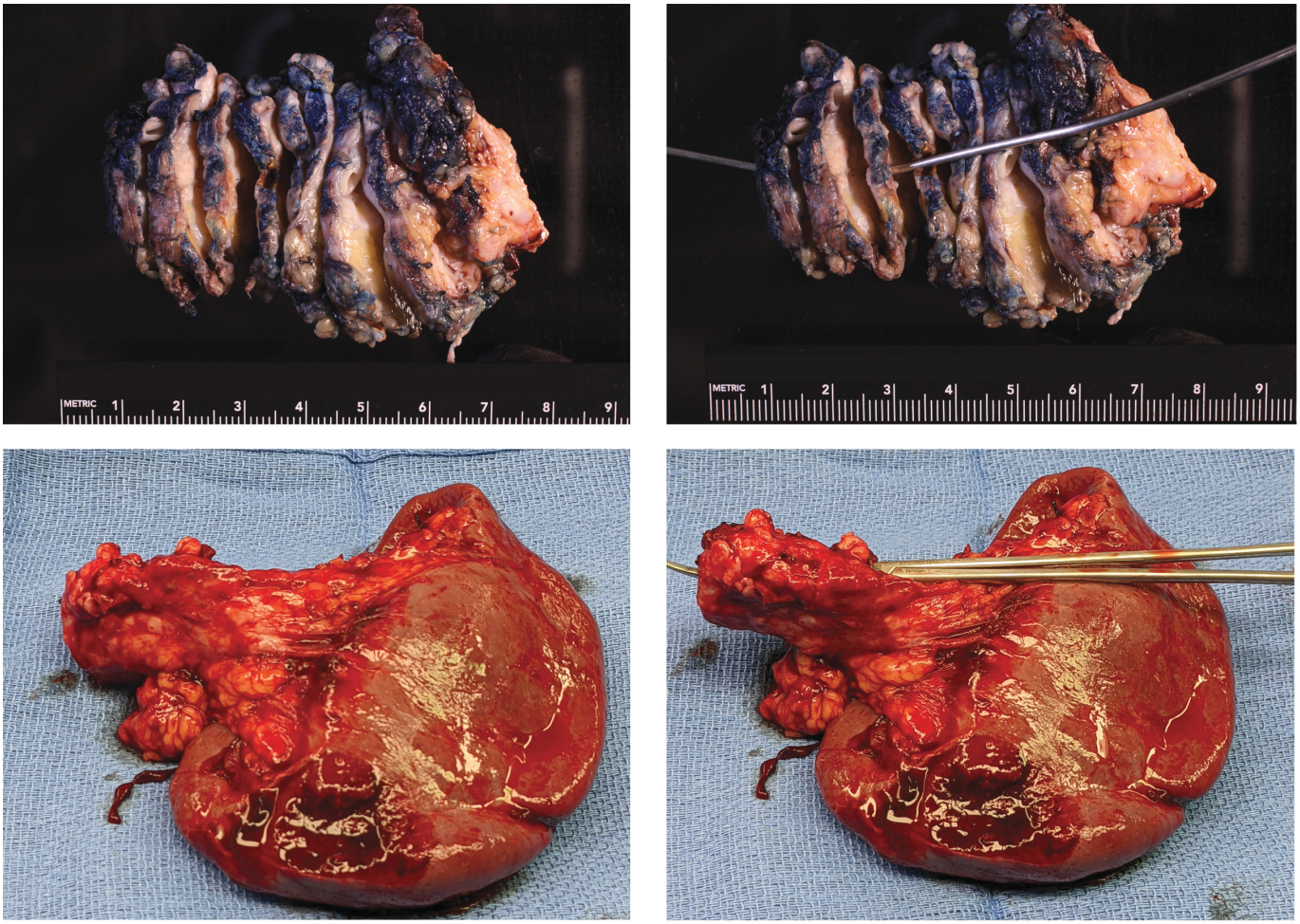

Figure 3 Operative/specimen findings. The distal pancreatic specimen had an obvious defect anteroposteriorly (ie, a literal hole) that communicated with the main pancreatic duct.

Our review reveals that most reported cases of haemosuccus pancreaticus are associated with alcoholic pancreatitis (23 of 33, 70%, online supplemental table 1), and more often involve pseudocysts in the pancreatic head (21 of 34, 62%) when compared with the tail (8 of 34, 24%). Importantly, the size of the pseudocyst was not predictive of bleeding—in many instances, a pseudocyst in the tail was larger than the bleeding pseudocyst in the head of the pancreas. We hypothesise that bleeding from cysts in the head is likely from arterio-cyst fistualisation with an artery other than the splenic (ie, gastroduodenal, common hepatic, right gastric). Two of the 24 case studies did not specify the location. With respect to demographics, 24 of 36 (67%) patients were male with an average age of 46 years old (range 25–67 years)

While our patient is very representative with respect to demographic variables (man in his 40s), he presented with the causal pseudocyst in the tail of the pancreas. Further, unlike 'typical presentations', repeated CT angiograms as well as repeat formal angiography by interventional radiologists in our patient did not reveal active extravasation, even when EGD studies demonstrated active bleeding from the ampulla. Thus, a multimodal approach to diagnosis was critical—a negative angiogram does not rule out a pancreatic aetiology to an upper GI bleed.[8] Similarly, a negative upper GI or EGD is not specific enough to rule out haemosuccus pancreaticus. Interestingly, some case studies report that upper endoscopies initially noted bleeding and/or clots in the duodenum—however, as with our patient, these findings were dismissed as scope injury or non-specific findings.[15 17 27] Thus, high suspicion must be maintained in the appropriate clinical context to diagnose haemosuccus pancreaticus, especially in patients with recurrent unlocalised bleeds.

In addition to the variability of which diagnostic modality provided the final diagnosis, there is also quite a bit of variance with respect to presenting symptoms/signs, as well as physical examination findings. In addition to obvious signs of upper GI bleeding (haematemesis, haematochezia and hypodynamic instability), the presenting findings often correlated well with the location of the pseudocyst and the chronic cause of pancreatitis.[9–31] Additionally, we noticed a pattern of historical and radiological findings that were represented among a minority of case studies (as well as our patient), including a history of vascular malformations and pseudoaneurysm/ectasias. This suggests that vascular ectasia may be an underlying risk factor for developing haemosuccus pancreaticus in patients with chronic pancreatitis. Thus, a thorough history and physical may be informative in cases of unlocalised bleeds.

Learning points

- In the appropriate context (eg, recurrent upper gastrointestinal bleeding with a history of chronic pancreatitis, history of vascular ectasias), bleeding or blood products noted in the duodenum should not be attributed to endoscopic injuries or other causes without ruling out other sources of bleeding (eg, haemosuccus pancreaticus).
- No individual diagnostic modality (endoscopy, angiography or radiological imaging) is specific for the diagnosis—in many cases, many of these diagnostic modalities will be negative when another demonstrates positive findings for bleeding. A multimodal diagnostic approach, including repeated investigations when indicated, is necessary to rule out haemosuccus pancreaticus.
- While the presenting symptoms varied between individuals, they likely reflect the physiological consequences of the location of the pseudocysts and the cause of chronic pancreatitis. However, it is important to note that the largest pseudocyst is not always causally associated with the bleeding. In fact, haemosuccus pancreaticus is most associated with pseudocysts at the pancreatic head, rather than the tail.

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