A Rare Finding of Primary Aortoduodenal Fistula by Gastrointestinal Bleeding SPECT/CT Scan: A Case Report

Finding PADF by GI bleeding SPECT/CT

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Abstract

BACKGROUND
Primary aortoduodenal fistula (PAFD) is a rare cause of gastrointestinal (GI) bleeding, consisting of the presence of abnormal channels between the aorta and the GI tract without any previous vascular intervention which results in massive intraluminal hemorrhage.

CASE SUMMARY
A 67-year-old man was hospitalized after suffering from coffee ground vomiting, tarry stool passage, and abdominal colic pain. Following this, he was repeatedly admitted due to gastrointestinal (GI) active bleeding and hypovolemic shock. Intermittent and spontaneously stop bleeder was too difficult to find after multiple GI endoscopy, angiography, computed tomography angiography (CTA), capsule endoscopy, and $^{99m}$Tc-red blood cell (RBC) scan. He received supportive treatment every time and was discharged without signs of rebleeding. Therefore, it was suggested that he be admitted longer to find the bleeder. CTA was performed soon after the bleeding, and a small aortic aneurysm was found at the renal level, in contact with the fourth portion of the duodenum. A $^{99m}$Tc-RBC single-photon emission computed tomography/computed
tomography (SPECT/CT) scan was arranged when the patient had bleeding symptoms and showed an active bleeder at the duodenum level. With clinical symptoms (intermittent massive GI bleeding with hypovolemia shock, dizziness, dark red stool passage, and bloody vomitus) and the result of abdominal CTA and \(^{99m}\text{Tc-RBC}\) SPECT/CT scan, the suspicion of duodenal bleeding caused by the small aneurysm arouse, and primary aortoduodenal fistula was highly suspected. Afterwards, he underwent duodenum excision and duodenum-jejunum anastomosis. A 7-mm saccular aneurysm arising from the anterior wall of the abdominal aorta near the left renal artery was found during surgery; thus, percutaneous intravascular stenting of the abdominal aorta was arranged. The symptoms of GI bleeding improved postoperatively.

CONCLUSION

We suggest additional evaluation with \(^{99m}\text{Tc-RBC}\) SPECT/CT scan can help in the diagnosis of rare cause of active GI bleeding.

Key Words: Gastrointestinal active bleeding, Computed tomography angiography, \(^{99m}\text{Tc-red blood cell scan}, \) Single-photon emission computed tomography/Computed tomography, Primary aortoduodenal fistula

Kuo CL, Chen CF, Su WK, Yang RH, Chang YH. A Rare Finding of Primary Aortoduodenal Fistula by Gastrointestinal Bleeding SPECT/CT Scan: A Case Report. World J Clin Cases 2023; In press

Core Tip: We describe a 67-year-old man was recurrently admitted due to gastrointestinal (GI) active bleeding and hypovolemic shock. According to clinical symptoms, abdominal CTA, and \(^{99m}\text{Tc-RBC}\) SPECT/CT scan, duodenal bleeding may be caused by the small aneurysm, and primary aortoduodenal fistula was highly suspected. Then, he underwent duodenum excision and duodenum-jejunum anastomosis. The symptoms of GI bleeding improved postoperatively.
INTRODUCTION
In scientific literature between 1951 and 2010, 253 cases of PADF were reported. PADF conditions are difficult to clinically diagnose, as the clinical characteristic is a 'herald GI bleeding', with hematemesis and melena, while patients are in a severely deteriorated condition. Despite the low incidence of PADF, delay in diagnosis and treatment has been historically associated with extremely high mortality[1-10]. For the treatment of GI bleeding, it is necessary to localize the bleeding site, which is usually done with an endoscopy. Other methods include angiography, computed tomography angiography (CTA), capsule endoscopy, and 99mTc-labeled red blood cell (RBC) scanning; each of these techniques has limitations[11-17]. Herein, we present the first case of a PADF in our hospital and discuss its diagnosis and management. This study was approved by the Mackay Memorial Hospital Institutional Review Board (IRB No. 23MMHIS055e).

CASE PRESENTATION

Chief complaints
A 67-year-old man was hospitalized after suffering from coffee ground vomiting, tarry stool passage, and abdominal colic pain.

History of present illness
The patient had severe anemia due to active GI bleeding.

History of past illness
The patient had long-term GI hemorrhage, hypertension, hyperlipidemia and renal function insufficiency.

Personal and family history
The patient’s personal and family history was unremarkable.
**Physical examination**

He was repeatedly admitted due to active GI bleeding and hypovolemic shock. Intermittent and spontaneously stop bleeder was too difficult to find after multiple GI endoscopies, angiographies, CTAs, capsule endoscopies, and 99mTc-RBC scans. The patient received supportive treatment every time, and was discharged without signs of rebleeding.

**Laboratory examinations**

Blood tests showed low hemoglobin and hematocrit.

**Imaging examinations**

In the beginning, esophagogastroduodenoscopy (EGD) revealed gastric ulcers and initially suspected Dieulafoy lesions (Figure 1a). Furthermore, small bowel endoscopy showed several shallow ulcers with pigmented spots in the middle part of the jejunum and proximal ileum, and hemorrhage of small bowel origin was suspected (Figure 1b). EGD presented duodenal diverticulum on the third portion (Figure 1c). Nevertheless, recurrent episodes of bleeding occurred and 99mTc-RBC scan and colon endoscopy were arranged. 99mTc-RBC scan showed radioactivity in the middle ascending colon to hepatic flexure 17.5 h after radiotracer injection; however, since the interval between images was 5.5 h, the intermittent intestinal bleeding could be from the proximal to ascending colon, cecum, ileum, or jejunum (Figure 1d). The follow-up colonoscopy showed two outpunching with dark brown content and mild oozing in the transverse colon, for which the bleeding condition was highly suspected (Figure 1e). The patient received multiple image examinations during this period, but he still experienced repeated GI bleeding and was admitted to the hospital. Therefore, it was suggested that he be admitted longer to find the bleeder. This time, CTA was performed soon after the bleeding, and a small aortic aneurysm was found at the renal level, in contact with to the fourth portion of the duodenum (Figure 2a). 99mTc-RBC scan was arranged while the patient still had bleeding symptoms and showed an active bleeder at the duodenum.
level in the lateral view at 19 h; other anterior views did not show any significant bleeding points (Figure 2b). Subsequent confirmation through single-photon emission computed tomography/computed tomography (SPECT/CT) found it might be the aortic fistula pumping the blood into duodenum (Figure 3).

**FINAL DIAGNOSIS**
Based on clinical symptoms, GI endoscopy (colonoscopy, EGD, single balloon enteroscopy, capsular enteroscopy), abdominal CTA and $^{99m}$Tc-RBC SPECT/CT scan, the suspicion of duodenal bleeding caused by the small aneurysm arouse, and PADF was highly suspected.

**TREATMENT**
The patient underwent excision of the duodenum and duodenum-jejunal anastomosis. A 7-mm sacular aneurysm arising from the anterior wall of the abdominal aorta near the left renal artery was found during surgery. Percutaneous intravascular stenting of the abdominal aorta was performed. The operative procedure is shown in Figure 4.

**OUTCOME AND FOLLOW-UP**
The symptoms of GI bleeding improved after surgery.

**DISCUSSION**
EGD is the first step in diagnosing active gastrointestinal bleeding. However, it has low detection rate for aortoenteric fistula as the distal parts of the duodenum may not be visualized. CT has the highest detection rate for aortoenteric fistula and is helpful for patients whose bleeder was not found by initial EGD or bleeding continue after treatment. $^{99m}$Tc-RBC scan is often adopted when the amount and frequency of bleeding is lower. It had low resolution and frequently has marked lag between the onset of bleeding but can continuously monitor for hours. In this case we showed $^{99m}$Tc-RBC
SPECT/CT can localize the bleeder if patient has active GI bleeding and can help in the diagnosis. PADF is extremely rare, with an incidence rate at autopsy of 0.04%–0.07%. A fistula most commonly originates between an abdominal aortic aneurysm (AAA) and the duodenum. Because of the close approximation and fixed nature of the duodenum, the expanding nature of the AAA causes irritation and inflammation, resulting in eventual fistulization over the passage of time. Thus, PADF is prone to occur in the 3rd part of the duodenum, accounts for two-thirds of cases. And rest of the one-third occur in the 4th part of the duodenum. The classical presentations of PADFs are upper GI bleeding (64%), abdominal pain (32%), and a pulsatile abdominal mass (25%). GI bleeding is almost invariably present, but the first few episodes are usually mild and self-limiting. In the beginning, these so-called ‘herald bleeds’ may be the only manifestations of fistulization. High proportion of reported cases had a long period - up to months - of sparse and self-limiting episodes. In these cases, this is easily misinterpreted as chronic GI bleeding, which is easily underestimated by patients or confused with peptic ulcer disease by physicians. Consequently, herald bleeds may stay unnoticed until the sudden occurrence of massive hemorrhage precipitates the patient into hemorrhagic shock. These symptoms are very similar to our case, where the patient experienced intermittent GI bleeding for about a year.

A timely and accurate PADF diagnosis may be challenging due to insidious episodes of GI bleeding, which are frequently underdiagnosed until the occurrence of massive hemorrhage. Thence, PADF diagnosis can be challenging not only due to the variation and nonspecificity of symptoms, but also because it is difficult to definitively identify with imaging modalities. Saers et al. analyzed the detection rate of the primary aorto-enteric fistula using diagnostic tools in 81 patients (showed in Table 1). EGD is the first step in diagnosing upper GI bleeding. However, in most cases only proximal portion of the duodenum is visualized. This results in low sensitivity in diagnosing aorto-enteric fistula with a detection rate of 25%. The most valuable diagnostic tool for the diagnosis of PADF is CTA with intravenous contrast because it can reveal all the
associated common findings: AAA, absence of a clear separation between the duodenal and aortic walls, signs of retroperitoneal inflammation, and air in the retroperitoneum or thrombus[4, 5, 7]. Angiography could demonstrate the extravasation of contrast agent into the bowel; however, contrast medium present in the GI tract in only about a quarter of these patients. Therefore, angiography is not reliable in diagnosing PADF[4, 9]. Other modalities, such as ultrasound, colonoscopy, enteroclysis, $^{99m}$Tc-RBC scan, and colonic radiography do not contribute to the diagnosis of PADF and are only of limited value[9].

The above literature indicates that $^{99m}$Tc-RBC scan is not significantly helpful in the detection of PADF. The precise anatomic localization of the bleeding site is not possible due to the limited resolution of scintigraphy, and there is frequently a marked lag between the onset of bleeding and clinical findings. The advantage of using $^{99m}$Tc-RBC scan is that it can continuously monitor the entire GI tract for up to approximately 24 hrs. Additionally, $^{99m}$Tc-RBC scan is a noninvasive method that can detect bleeding with high sensitivity, localize the bleeding area, and could help to approximate bleeding volume. This advantage to perform continuous imaging increases the likelihood of detection of intermittent bleeding over other techniques that are limited to only a single time point or periodic sampling[11-14].

In this case, CTA first found a small aortic aneurysm at the renal level, connected with the fourth portion of the duodenum. Then, $^{99m}$Tc-RBC scan was arranged when the patient still had hemorrhage symptoms. We also performed a SPECT/CT scan because it is better at diagnosing bleeding points and collecting information about the etiology than planar images[16-17]. Moreover, SPECT/CT may contribute by confirming the aortic aneurysm and determining its clinical significance[17]. In conclusion, if a bleeding site is found on $^{99m}$Tc-RBC planar imaging, SPECT/CT study is recommended for confirmation. Furthermore, after reviewing the CT scans performed a year ago, the aneurysm had existed but was ignored due to its small size, no loss of the aneurysmal wall, no air leak in the retroperitoneum, and lack of inflammation. Consequently, the PADF in this patient is easy to ignore and can only be detected by CTA.
CONCLUSION

EGD is the first step in diagnosing active gastrointestinal bleeding. However, it has low detection rate for aortoenteric fistula as the distal parts of the duodenum may not be visualized. CT has the highest detection rate for aortoenteric fistula and is helpful for patients whose bleeder was not found by initial EGD or bleeding continued after treatment. $^{99}\text{mTc}$-RBC scan is often adopted when the amount and frequency of bleeding is low. It had low resolution and frequently has marked lag between the onset of bleeding but can continuously monitor for hours. In this case we showed $^{99}\text{mTc}$-RBC SPECT/CT can localize the bleeder if patient has active GI bleeding and can help in the diagnosis of PADF.

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