Primary Aortic Aneurysm Characterized by Herald Gastrointestinal Bleeding

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Case report

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Abstract

Objective: We describe a case series of patients diagnosed with Primary aortic aneurysm characterized by herald gastrointestinal bleeding, reviewed the literature, aimed to identify and treat such patients without delay, reduce fatality rate.

Materials and methods: We reviewed a case series of Primary aortic aneurysm in the Department of Gastroenterology, Beijing Shijitan Hospital Affliated to the Capital Medical University. Each patient was admitted to hospital with gastrointestinal bleeding as main complaint, and was diagnosed by CT or endoscopy. Then characteristics of endoscopy and CT were analyzed.

Results: 3 patients were enrolled. In case 1, hematoma was formed after a ruptured abdominal aortic aneurysm, and compression of the hematoma led to ischemic colitis, abdominal pain and hematochezia were the main symptoms. In case 2 and case 3, aortic gastrointestinal fistula were found in the duodenum and esophagus, respectively, abdominal pain, melena and hematochezia were the main symptoms.

Conclusion: A timely and accurate diagnosis of Primary aortic aneurysm may be challenging due to insidious episodes of GI bleeding, which are frequently under-diagnosed until the occurrence of massive hemorrhage. Clinical physicians should keep a high index of awareness for Primary aortic aneurysm.

Introduction

Primary aortic aneurysms (PAA) are disease with high mortality. Most of the patients have no obvious clinical symptoms in the early stage, so they miss the best time for treatment. Primary aortic gastrointestinal fistula (PAGF) is a complication of an abdominal aortic aneurysma, a rare but life-threatening cause of massive gastrointestinal bleeding. Despite its rarity, it is an important complication, as it is usually fatal unless detected. PAGF causes spontaneous rupture of an aortic aneurysm into the lumen of the adjacent gastrointestinal loop. The formation of a fistula between the aorta and the gastrointestinal tract is extremely rare. The annual incidence of PAGF is estimated to 15/million. Hence, diagnosis and management of aortic aneurysm characterized by herald gastrointestinal bleeding is very challenging. Without surgical treatment, its mortality rate reaches 100%. Even if patients undergo timely surgical treatment, they may still die of complications such as infection. Herein, we describe a case series patients with aortic aneurysm characterized by herald gastrointestinal bleeding and aimed to improve the rate of early diagnosis and early treatment.

Materials And Methods

This was a single-center, retrospective, observational case series. In the following cases, patients were admitted to Department of Gastroenterology, Beijing Shijitan Hospital Affliated to the Capital Medical University with different complaints (hematochezia in three cases and melana in one). In each case, Emergency consultation, physical examination and Laboratory examination failed to accurately diagnose

the disease. Computed tomography (CT) or endoscopy was chosen as the next diagnostic test of choice, and AGIF was detected by CT or endoscopy. Then, CT characteristics and follow-up were analyzed.

Case Presentation

Case 1

In April 2013, a 50-year-old man was admitted to the ward by emergency department with a complaint of hematochezia for 1 day. He had a history of hypertension and coronary heart disease. He had several episodes of abdominal pain or discomfort in the months before this presentation. On physical examination, his heart rate was 106 beats/min with blood pressure of 180/90 mm Hg. Laboratory investigations revealed anemia (hemoglobin, 10.6 g/dL), white blood cell count, 12,950/mm3. Abdominal ultrasound and abdominal vascular ultrasound revealed no obvious abnormality. Computed tomography (CT) showed abdominal aortic sclerosis, aneurysm at the abdominal aorta, along the psoas major muscle and iliopsoas muscle on the left side of the aneurysm, the hematoma was formed, the origin of the inferior mesenteric artery was compressed, which caused intestinal ischemia (Fig. 1). The patient was immediately transferred to vascular surgery for abdominal aortic aneurysm repair. During the interventional procedure, there was a tear on the left side of the middle part of the abdominal aorta, which was about 1.5 cm. Two covered stents (Medtronic, 23*50 mm) were released into the middle part of the abdominal aorta. Re-angiography showed that the abdominal aorta and double iliac arteries were good flow, the blood flow in the stent was good, and no contrast medium extravasation was found. After 10 days of antibiotics therapy, the patient had normal body temperature and negative fecal occult blood, and was discharged from hospital.

Case 2

A 49-year-old man was admitted to our ward with a complaint of hematochezia for 4 days, abdominal pain for 1 day in September 2018. Gastroscopy in local hospital indicates duodenal ulcer and chronic gastritis. He had a history of hypertension 2 years. On physical examination, his heart rate was 90 beats/min with blood pressure of 168/104 mm Hg, with left abdominal tenderness. Laboratory investigations revealed anemia (hemoglobin, 6.9 g/dL). Emergency Computed tomography (CT) showed rupture of abdominal aortic aneurysm and intestinal fistula of abdominal aortic aneurysm (Fig. 2). The patient was immediately transferred to vascular surgery for abdominal aortic aneurysm repair. Unfortunately, the patient suddenly lost consciousness during preparation of surgery, the patient appeared ventricular fibrillation, and eventually died.

Case 3

A 75-year-old man was admitted to our ward with a complaint of intermittent melena for 11 hours in December 2019, without chest pain or hematemesis. He had a history of hypertension and coronary heart disease. On physical examination, his heart rate was 90 beats/min with blood pressure of 130/80 mm

Hg. Laboratory investigations revealed anemia (hemoglobin, 6.5 g/dL), white blood cell count, 16,950/mm3. Endoscopy showed a protruding lesion in the lower segment of the esophagus, with blood clots attached on the lesion surface, which was suspected of AGIF (Fig. 4). Emergency Computed tomography (CT) showed aortic atherosclerosis, thoracic aortic aneurysm and AEF (Fig. 3). The patient was immediately transferred to vascular surgery for thoracic aortic aneurysm repair. One covered stents (Gore, 31-31-150 mm) were deployed into the thoracic aorta. Re-angiography showed that the thoracic aorta was good flow, the blood flow in the stent was good, and no contrast medium extravasation was found. After 30 days of antibiotics therapy, the patient had normal body temperature and negative fecal occult blood, and was discharged from hospital.

Discussion

PAGF is an abnormal communication between aorta and gastrointestinal tract, which is a rare but life-threatening cause of gastrointestinal bleeding, characterized by significant mortality, even with early diagnosis and intervention. The mortality rate is high, the mortality rate of untreated patients is close to 100%, and that of surgical patients is 30%, 40%[7, 8]. The pathophysiological mechanism is not clear, it is believed that due to the continuous beating of aneurysms, constantly squeezing and eroding the digestive tract, resulting in ischemia and necrosis, the formation of fistula between the two is a rare cause of gastrointestinal bleeding, accounting for less than 0.2%[9]. PAGF are most commonly caused by abdominal aortic aneurysms and 85% are caused by atherosclerosis. Other rare causes include infection (aortic arteritis caused by bacteria, syphilis and tuberculosis), tumors, radiation injuries, peptic ulcers, diverticulitis and foreign bodies (such as needle, fishbone or chicken bone). Fistulas can form between the aorta and anywhere from the esophagus to the sigmoid[8]. In our patients, case 1 had a primary aortic aneurysm, which formed hematoma after bleeding, the beginning of the inferior mesenteric artery was compressed by hematoma, which caused intestinal ischemia. Fistula was formed between the aorta and intestine in Case 2, and esophagus in case 3. Clinical manifestations of PAGF are variable, including low-grade fever associated with obscure infected lesions and classical triad of abdominal or chest pain, GI bleeding, and abdominal pulsating mass[10]. In our patients, all had GI bleeding, only one patient had abdominal pain. The sentinel hemorrhage is the initial bleeding prior to catastrophic exsanguinations, and is usually minor and self-limiting owing to formation of thrombus plugging the fistula as a result of hypotension. However, the plug may be dislodged of its canal, leading to further bleeding after the patients become normotensive. Sentinel hemorrhage occurs in a repetitive fashion, the time interval between the initial hemorrhage and final exsanguination ranges from hours to months[8–10]. In our study, this time interval ranges from 11 hours to 4 days. Hypovolemic shock occurred in case 2 during the first sentinel hemorrhage, resulting in death. A high index of suspicion is essential for prompt diagnosis of PAGF. In the absence of clinical suspicion, no single examination can reliably make the diagnosis. Endoscopy is the most useful investigation for origins of GI bleeding, but it can be misleading in finding out coexisting pathological entities. The lack of awareness of PAGF, together with the inaccessibility to distal duodenum and underlying overt GI lesions, are probably responsible for misdiagnosis and delayed appropriate management. If endoscopy failed to reveal informative findings, then CT scan is usually

performed which may give a reported detection rate of 30–61%[11]. A definitive diagnosis at the time of the initial bleeding is essential for timely, life-saving surgery. Absence of an identifiable source of massive GI bleeding strongly prompts a surgical exploration, in order to establish diagnosis and salvage the patients. All cases in our study were discovered during an exploration, and 2 patients survived the operation. Therapeutic approaches in patients with PAGF may be either open surgery or endovascular repair. Menezes et al. showed that there are no differences in overall mortality when comparing endovascular with open aortic aneurysm repair (7.69% versus 11.89%, $P = 0.263$)[12]. However, patients with a ruptured or inflammatory aortic aneurysm were excluded from this cohort. In our study, two cases were treated with endovascular aortic repair (EVAR) by placing aortic stent. While study of Leonhardt showed that the immediate success rate at stopping bleeding was 80% using stent grafts. Despite the inital success, 80% rebled after 2 weeks or longer. 30-day mortality was 40%, which doubled at 6 months[13]. This supports the conclusion that EVAR should be considered only as bridging measure prior to definitive surgical repair. As the risk of infection is high, patients require antibiotic cover following EVAR. Bacteria in the digestive tract may translocate or enter the blood directly, causing septicemia, if blood culture is negative, antibiotics are recommended for 7–10 days, if blood culture is positive, antibiotics should be selected according to the results of drug sensitivity test, and antibiotics should be used for about 6 weeks after operation. In spite of this, postoperative infection complications and fistula recurrence are still possible. In our study, case 1 and case 3 were treated with antibiotics for a month after placing endovascular stent, and so far, the prognosis is good.

Conclusion

In conclusion,, PAGF is a rare cause of gastrointestinal bleeding. Once this disease occurs fatal massive hemorrhage mortality is extremely high, timely and accurate diagnosis in the stage of premonitory hemorrhage is very important.

Declarations

Informed consent

As a retrospective study there is no need to obtain informed consent from patients. The study was approved by the institutional ethical review board in Beijing Shijitan Hospital, Capital Medical University.

Consent for publication

Written informed consent for publication was obtained from all participants.

Availability of data and material

All data generated or analysed during this study are included in this article and its supplementary information files.

Competing interests

The authors declare no competing financial interests.

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Authors' contributions

Yadan Wang contributed to the data analysis and interpretation and drafted the manuscript. Wu Jing is in charge of the overall design of the article. Canghai Wang, Jing Wang, Pengpeng Ding, Yun Wang, Hong Liu and Chunmei Guo contributed to the data interpretation and edited the manuscript. All the authors approved the final version. All authors had access to the data and a role in writing the manuscript.

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Tables

Table 1
Clinical findings in patients with PAGF.

Case No.	Sex/age (yr)	Symptom	Past history	Aortic aneurysm localization	CT findings
1	M/50	hematochezia	Hypertension, Coronary heart disease	Lower segment of abdominal aorta	An aneurysm was formed, on the left side of the aneurysm, hematoma was formed, inferior mesenteric artery was compressed, which caused intestinal ischemia
2	M/49	hematochezia, abdominal pain	Hypertension	Lower segment of abdominal aorta	rupture of abdominal aortic aneurysm and intestinal fistula of abdominal aortic aneurysm was formed
3	M/79	intermittent melena	Hypertension, Coronary heart disease	Middle part of thoracic aorta	rupture of thoracic aortic aneurysm and esophageal fistula of thoracic aortic aneurysm was formed

Figures

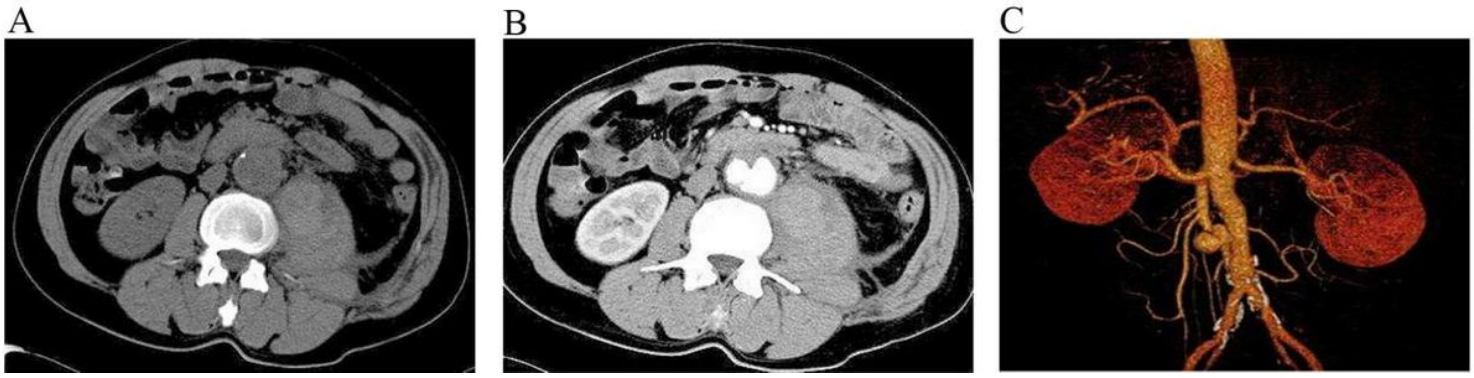

Figure 1

(A) abdominal non-contrast CT scan; (B) abdominal enhanced CT showed abdominal aortic sclerosis, aneurysm at the abdominal aorta, along the psoas major muscle and iliopsoas muscle on the left side of the aneurysm, the hematoma was formed, the beginning of the inferior mesenteric artery was compressed, which caused intestinal ischemia; (C)Three-dimensional reconstruction of abdominal vessels

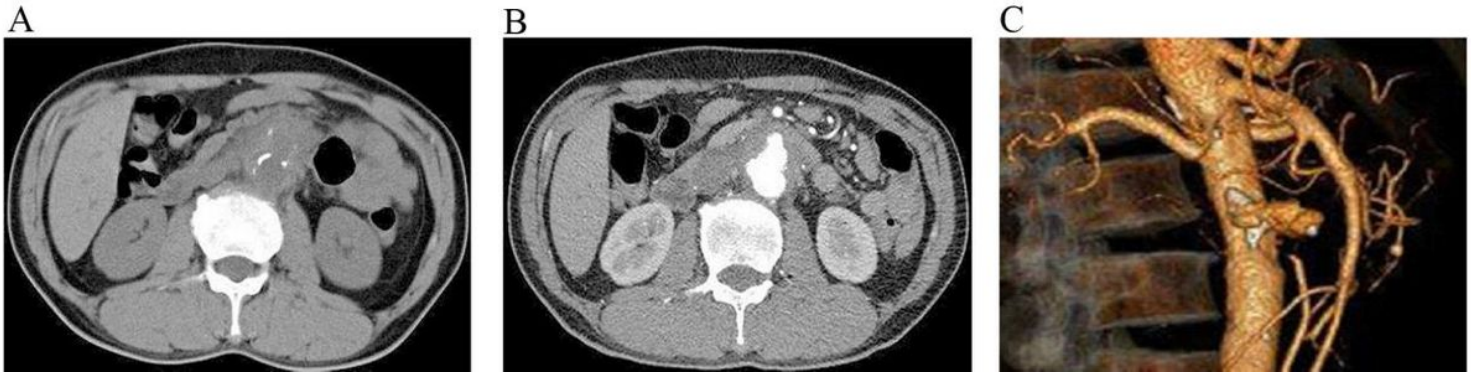

Figure 2

(A) abdominal non-contrast CT scan showed vascular calcification and the boundary of abdominal aorta was unclear. (B) abdominal enhanced CT showed rupture of abdominal aortic aneurysm and intestinal fistula of abdominal aortic aneurysm was formed; (C) Three-dimensional reconstruction of abdominal vessels.

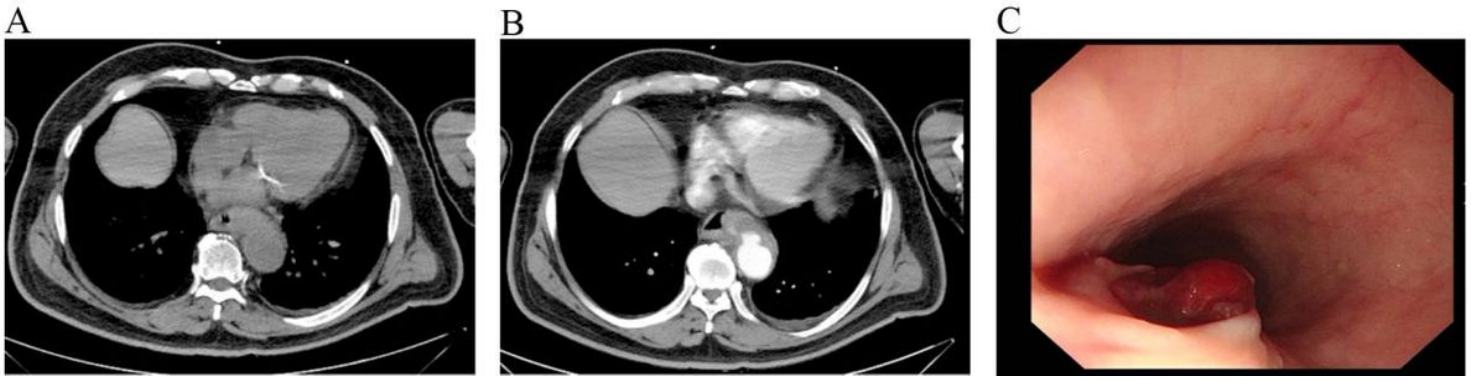

Figure 3

(A) chest non-contrast CT scan showed vascular calcification, and the boundary of abdominal aorta was unclear; (B) chest enhanced CT showed rupture of thoracic aortic aneurysm and esophageal fistula of thoracic aortic aneurysm was formed;(C) Endoscopy showed a protruding lesion at the lower end of the esophagus, and fresh blood clots could be seen on the surface.