



Caliber persistent artery of the stomach: An unrecognized cause of massive gastric hemorrhage

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INTRODUCTION

The caliber persistent artery of the stomach, also known as aneurysm, Dieulafoy's exulceration simplex, submucous arterial malformation, gastric arteriosclerosis, and peptic ulcer of peculiar location, was once considered to be a rare cause of massive gastric hemorrhage. Recently, however, the number of reports on caliber persistent artery of the stomach have been increasing, suggesting that this lesion is probably underdiagnosed and not a rare cause of massive gastric hemorrhage^[1-3]. Two patients with caliber persistent artery of the stomach were admitted to our department 2 years ago, and diagnosis was confirmed subsequently by surgical operation and endoscopy and angiography.

CASE REPORT

Case 1

A 64-year-old man with a past medical history of about 1 wk use of self prescribed acetylsalicylic acid (4.5 g daily) for relieving pain from cholecystitis and cholelithiasis presented to our department with

hematemesis consisting of 600 mL of bright red blood accompanied by melena. Emergency endoscopy showed a large amount of blood clots in the gastric cavity, but no active hemorrhage was noted. On admission, physical examination revealed pallor of skin, a heart rate of 100 beats/min, and orthostatic hypotension. The results of laboratory tests showed anemia (hemoglobin 78 g/L). Over the following 7 d, he continued to bleed despite the administration of omeprazole (Losec) and thrombin. Endoscopic examination was repeated, revealing a solitary superficial erosion in normal appearing gastric mucosa near the esophagogastric junction on the posterior wall, and a 0.2 cm × 0.2 cm fresh red tissue was seen at the base, with no active hemorrhage. The patient received altogether about 7000 mL blood transfusion but still had persistent liquid dark stools with recurrent loss of consciousness. Thus, a surgical intervention was given. During the operation, a protruding artery was seen at the posterior wall of the body of the stomach near the cardia. There was a small blood clot on it, and a large amount of blood spurted out as the blood clot was removed. The patient underwent oversewing and ligation of the ruptured vessel and Bancroft operation (Billroth II). Histological examination showed submucous arterial malformation. The patient tolerated the operation well and was discharged 2 wk after the operation.

Case 2

A 49-year-old man experienced his first episode of hematemesis, consisting of 500-1000 mL of bright red blood followed by melena (about 300 g) two times. No history of complaints regarding the gastrointestinal tract was present. He was not taking medications before admission and denied intake of nonsteroidal anti-inflammatory drugs (NSAIDs) and ethanol. On admission, his blood pressure was 12/6.5 kPa, heart rate was 90 beats/min, and hemoglobin was 81 g/L. Emergency endoscopy performed on admission disclosed a large amount of fresh and coagulated blood in the esophagus and gastric and duodenal cavities, but the blood source was not obvious. Conservative management, including blood transfusion, histamine H₂ blockers, and thrombin was instituted. The patient still had recurrent massive hematemesis at times associated with melena. His blood pressure fell to 8/4 kPa. A selective angiography performed on day 2 of hospitalization showed the leakage of contrast from a branch of the gastroduodenal artery. The patient underwent surgical, during which a small, round mucosal defect with a protruding tissue at its base was observed in the small curvature of the body of the stomach. Spurting bleeding from the protruding artery was also be seen. In the gastric cavity, 200-300 mL of fresh blood and about 500 g of blood clots were found. The vessel was ligated and oversewn. Postoperative recovery was excellent, and the patient was discharged from the hospital.

DISCUSSION

Caliber persistent artery of the stomach was first described by

Gallord in 1884 and was later characterized by Dieulafoy in 1896 as the exulceration simplex. This lesion is characterized as follows: a negative past medical history, independence from peptic ulcer disease, sudden onset, increasing bouts of hematemesis, subcardial location, a tiny mucosal lesion, an open submucous artery of seemingly large caliber, failure of conservatism in treatment, and 60.5% overall lethality^[1]. Although it used to be considered a rare cause of massive gastric hemorrhage, more recently this lesion has been identified more frequently. Therefore, it is likely to be an under-recognized and undiagnosed cause than a rare cause of massive gastric hemorrhage^[1-4]. Its incidence as a source of upper gastrointestinal bleeding was shown to range from about 0.3%-6.7%^[5]. Pointner *et al*^[6] found 22 of 1432 patients and Baettig *et al*^[7] found 28 of 480 patients (5.8%) with severe gastrointestinal hemorrhage who underwent emergency endoscopy. Here, we present two cases that we have seen in the recent two years. Thus, physicians' awareness of the existence of this lesion should be heightened to improve the likelihood of uncovering this "unknown cause" of massive gastric hemorrhage.

Caliber persistent artery of the stomach has been reported in a wide age range of patients, from 16 to 93 years^[3,8], with a moderate male predominance^[1,9]. In Pointner's 22 cases, eight cases were female, and 14 were male, ranging in age from 29 to 80 years. Nine of these patients were older than 60 year^[6]. Our two patients were males, 64 and 49 year old.

The bleeding site for this lesion is often within 6 cm of the gastroesophageal junction in the cardia or fundus of the stomach^[10]. In Reilly's review, approximately 98% of the lesions located in the stomach were in its upper portion. Sixty-seven percent were located high in the body and 25% in the gastric fundus^[5]. The recent reports have identified similar lesions in the duodenal bulb^[11], the jejunum^[12,13], and in the right colon^[14]. Endoscopically, the gross appearance of the lesion is that of a solitary lentil sized, round mucosal defect with a protruding artery at the base^[6,15]. Juler *et al*^[4] used multiple tissue staining techniques and showed the following histopathological characteristics: (1) a gastric mucosal defect with fibrinoid necrosis at the base; (2) large thick walled artery loops in the base of the defect; (3) a tortuous dysplastic artery below the muscularis mucosa; (4) large thick walled veins adjacent to the artery; and (5) lymphoid aggregates in the lamina propria.

The typical clinical presentation of this lesion is recurrent and massive hematemesis at times associated with melena, hematochezia, and hypotension. Of the 177 cases described in the literature, 28% presented with hematemesis alone, 51% had hematemesis accompanied by melena, and 18% had melena alone^[5]. There have been inferences in the literature regarding associations with the use of alcohol and NSAIDs. In our case 1, the patient had a past medical history of 1 wk use of acetylsalicylic acid before gastric hemorrhage. It is assumed that the drug injured the mucosa of the stomach, which then triggered a rupture of the arterial branch in the gastric submucosa.

The caliber persistent artery of the stomach is the most dangerous forms of gastrorrhagia. It was observed that 100% of patients who were treated conservatively (*i.e.*, nonsurgically, nonendoscopically) died as a result of hemorrhagic shock^[15]. The mortality rate of the cases published since 1980 ranged from 40.0%-60.5%^[1]. Since this lesion has an extremely high mortality rate, it is necessary for the physicians to consider the existence of the lesion when conservative therapy fails in a patient with a sudden onset massive gastric hemorrhage, and a careful examination should be given as soon as possible. Recently, some authors^[6,16,17] have pointed out that emergency endoscopy is the most effective method of diagnosing the lesion. About 82% of the patients with caliber persistent artery of the stomach were identified with endoscopy. Forty-nine percent of lesions were identified during the initial endoscopic examination, while 33% required a second endoscopy to confidently identify the lesion as the source of bleeding^[5]. However, the source of the bleeding was not identified with endoscopy in 18% of patients evaluated^[5]. An alternative diagnostic method to assist in the diagnosis of this lesion is angiography. According to the literature, this technique was used in 14 patients, and it was helpful in the localization of the bleeding site

in 11 patients^[2,11,12,14,18-20]. Seven of the patients had lesions in the stomach, while the remaining four had one in either the duodenal bulb, jejunum, or ascending colon^[11,12,14]. The source of bleeding in our case 2 was not identified initially by the endoscopy but verified subsequently by angiography, showing that angiography is also an effective diagnostic tool for localizing the lesion.

Regarding treatment, surgical intervention is the treatment of choice^[21]. The most widespread surgical measures used are oversewing and wedge resection^[9,15]. These are safe and often successful procedures^[9]. Endoscopic therapy included epinephrine injection^[7,9,15], polidocanol injection^[6], bipolar electrocoagulation^[6,9], heater probe coagulation^[9], and YAG laser photocoagulation^[9]. According to a review by Reilly *et al*^[5], endoscopic therapy was utilized in 79 patients, and permanent hemostasis was accomplished in 85% of these patients following the first therapeutic endoscopy session. Twelve patients (15%) re-bled. Repeat endoscopic therapy was successful in eight patients (10%), and surgical intervention was needed in four patients (5%). Stark *et al*^[9] reported that endoscopic therapy was successful in 18 of 19 patients (95%), and there were neither deaths due to bleeding nor endoscopic complications. Baettig *et al*^[7] have followed up 21 patients treated by endoscopy for a mean of 28.3 months, and, of these, 20 patients had no recurrence of hemorrhage. Angiography with gel-foam embolization is another therapeutic option available for the treatment of caliber persistent artery of the stomach. With this technique, successful results were achieved in three out of four patients^[18,19].

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