CASE REPORT

Primary Aortoduodenal Fistula Caused by Tuberculous Aortitis Presenting as Recurrent Massive Gastrointestinal Bleeding

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Upper gastrointestinal bleeding from primary aortoduodenal fistula (PADF) is unusual and fatal. The etiology of PADF from tuberculous aortitis is rare. We report a 69-year-old male patient who suffered recurrent hematemesis and hematochezia with hypovolemic shock of unknown origin. Initial endoscopy failed to lead to a diagnosis. A bleeder over the third portion of the duodenum was found after the third endoscopy. Exploratory laparotomy showed a ruptured aortic pseudoaneurysm with an aortoduodenal fistula. Dacron graft repair of the aorta and simple closure of the duodenal fistula were carried out. Pathologic examination revealed tuberculous aortitis. The patient survived and was symptom-free following operation and antituberculous therapy. Review of the literature revealed that the clinical presentations in this disorder are insidious. The endoscopic findings are atypical. We conclude that so-called "herald bleeding", a history of tuberculous infection or aortic aneurysm and a high degree of suspicion are critical for successful diagnosis. Early diagnosis and surgical exploration are needed for timely and successful management. [*J Formos Med Assoc* 2008;107(1):77–83]

Key Words: aortitis, aortoduodenal fistula, gastrointestinal bleeding, tuberculosis

Aortoenteric fistula (AEF) is a direct communication between the aorta and intestinal lumen. There are primary and secondary forms. Primary AEF is usually due to erosion of an abdominal aortic aneurysm into the intestine. Secondary AEF is caused by reconstructive procedures on the abdominal aorta. Primary AEF is a severe life-threatening disease because of the difficulty of diagnosis and fatal exsanguinating hemorrhage. The incidence of primary AEF in the general population is about 0.07%. In patients with abdominal aortic aneurysm, the incidence may increase to 0.69–2.36%.[1] The predominant sites of primary AEF are in the third and fourth portions of the duodenum. About 54–78.5% of primary AEF comes from aortoduodenal fistula.[2,3] The predominant causes of primary AEF is aortic aneurysm or atherosclerosis, and about 70% of primary aortoduodenal fistula (PADF) is related to the same etiology.[4] Septic aortitis-induced PADF is unusual.[4–7] In addition, tuberculous aortitis (TBA)-related PADF is rare.[8–11] Early diagnosis of this disease is difficult but crucial. The typical presentation of the clinical triad of fistula (gastrointestinal hemorrhage, abdominal pain, pulsating abdominal mass) is noted in only 11–25% of patients with PADF.[2,12,13] In this report, we describe a case of PADF with the presentation of

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Received: March 6, 2007
Revised: April 24, 2007
Accepted: June 5, 2007

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recurrent massive upper gastrointestinal bleeding with hypovolemic shock. The clinical presentation was insidious and the endoscopic findings were atypical. The final diagnosis relied on surgical laparotomy and pathologic study even though endoscopy had found the bleeder.

Case Report

A 69-year-old male patient had a history of colon cancer, for which he had undergone radical resection 2 years previously, and pulmonary tuberculosis (TB), for which he had received a complete 6-month course of anti-TB treatment. The anti-TB regimen had consisted of a combination of four drugs (rifampin, isoniazid, pyrazinamide, ethambutol) for 2 months, and a combination of three drugs (without pyrazinamide) for an additional 4 months. After 6 months, anti-TB treatment was stopped and he has been disease-free for about 1 year already. He received regular follow-up in a local hospital for the colon cancer and pulmonary TB; no evidence of cancer recurrence was noted. History of other systemic diseases, major operations or smoking, alcohol and drug abuse were denied. However, he had been suffering from early satiety and frequent acid reflux in the recent 2 years.

He experienced episodes of bloody vomitus and tarry bloody stool passage about 2 weeks before he was transferred to our hospital. Initially, he was admitted to a local hospital. The first endoscopy led to suspicion of angiodysplasia or Dieulafoy's lesion of the gastric fundus. Heating probe coagulation was performed but failed to prevent re-bleeding. Repeated massive upper gastrointestinal bleeding associated with hypovolemic shock occurred thrice during hospitalization in the local hospital. An additional two courses of thermal coagulation by endoscopic heater probe were performed to treat the suspicious bleeder at the gastric fundus. Gradually, the active bleeding seemed to subside but tarry stool passage persisted. He was discharged from the local hospital and referred to our clinic for further evaluation. Physical examination findings were not unusual. Upper gastrointestinal endoscopy was repeated immediately and showed grade A reflux esophagitis (by Los Angeles classification) and some erosive patches over the gastric fundus. No active bleeder could be found. He received proton pump inhibitor at home.

Unfortunately, 2 days later, he was sent to our emergency room as a result of a fainting spell which led to a fall that resulted in lacerations of the frontal skin. Tarry stool passage and anemia were noted at the same time. Initial blood pressure was 120/80 mmHg, pulse rate was 96/min and body temperature was 36.5°C. The hemoglobin level was 11.3 g/dL and hematocrit was 33%. No thrombocytopenia, coagulopathy or abnormal liver function could be found. Sudden onset of bloody vomitus followed by hypovolemic shock and collapse (blood pressure dropped to 83/66 mmHg, pulse rate dropped to 51/min) developed 4 hours after wound treatment. Emergency resuscitation was performed, including adequate fluid challenge, blood transfusion (2 units of whole blood and 4 units of packed red blood cells), intubation of endotracheal tube and ventilator support. Hemoglobin level dropped to 8.8 g/dL.

Re-examination revealed a mild distended abdomen at the periumbilical area without tenderness or muscle guarding. Bowel sounds were hypoactive. Digital rectal examination showed bloody stool passage. No palpable mass, shifting dullness, increased splenic dullness or engorgement of superficial vein could be found. Chest X-ray showed old interstitial fibrosis over bilateral upper lung fields (Figure 1). Upper gastrointestinal endoscopy was repeated and showed clear gastric content without any active bleeders. Colonoscopy was not performed due to the patient's relatively unstable condition.

Unfortunately, nasogastric drainage showed fresh blood again 4 hours after admission. A third endoscopy disclosed a slowly oozing bleeder over the third portion of the duodenum with blood clot adhesion (Figure 2). Diverticular bleeding was suspected initially. Two hemoclips were applied,