Secondary Aortoduodenal Fistula

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Secondary aortoenteric fistula (SAF) is now recognized as an uncommon but exceedingly important complication of abdominal aortic reconstruction. The complication often occurs months to years after the original surgery. The main clinical manifestation of the disease is always upper gastrointestinal bleeding. Treatment of the disease is early surgical intervention. The mortality is high if no prompt operation. We present a case of secondary aortoduodenal fistula (SADF) found 20 days after aortic reconstructive surgery, with the clinical presentation of upper gastrointestinal bleeding. Even immediate exploratory laparotomy was performed, the patient died 48 hrs after the surgical management. Because of the increasing number of elective aortic aneurysm repairs in the aging population, it is likely that more patients with SAF will present to the clinical physicians in the future. So, a high index of suspicion is necessary for prompt diagnosis and treatment of this actually life-threatening event. (Chang Gung Med 2002;25:626-30)

Key words: secondary aortoenteric fistula, abdominal aortic reconstruction, upper gastrointestinal bleeding, aortic aneurysm, secondary aortoduodenal fistula.

Secondary aortoenteric fistulas are an uncommon but lethal complication of aortic reconstructive surgery. Because of the advancement of vascular surgery and aggressive surgical treatment, the frequency of secondary aortoenteric fistulas will likely increase. The complication often occurs months to years after the original surgery, with an incidence of 0.4%-4%. Bastounis et al. reported that the mean interval from the initial operation to the onset of upper gastrointestinal bleeding was 32 months. The 20-year experience with secondary aortoenteric fistulas at the Johns Hopkins Medical institutions showed the average interval to be 2.8 years. The longest postoperative interval for an aortoenteric fistula reported in the literature was 27 years, in which an aortocolic fistula developed after aortofemoral bypass surgery; the shortest postoperative interval was 2 days, recorded in 1974, in which a paraprothetic-enteric fistula developed after resection of a ruptured abdominal aortic aneurysm with graft interposition.

Dubost et al. first described homograft replacement of an abdominal aortic aneurysm in 1952. One year later, the first reported secondary aortoenteric fistula was reported by Brock in a case involving an aortic homograft and the duodenum. In 1956, Claytor et al. presented the first aortoenteric fistula caused by a prosthetic graft of the aorta. In 1958, Mackenzie et al. demonstrated the first successful repair of a secondary aortoenteric fistula between a synthetic graft and the intestine. Due to the anatomical proximity, the majority of cases involve the duodenum with the proximal suture line of an aortic prosthesis. Prompt diagnosis and surgical intervention is the only possible treatment that preserves the patient's life. Although most patients
have so-called "herald bleeding" before fatal exsanguination, diagnosis has rarely been made before laparotomy or autopsy. In our case, the final diagnosis was made during exploratory laparotomy. Nonetheless, our case exemplifies several of the important clinical features that may lead to a diagnosis of an aortoenteric fistula preoperatively.

**CASE REPORT**

An 81-year-old man presented to a community hospital with a history of severe lower abdominal pain for 4 days. The severe pain was sudden in onset, and was not associated with radiation. Associated symptoms included cold sweating and dizziness. The patient had no relevant history. Because hypovolemic shock occurred at the community hospital, he was transferred to Chang Gung Medical Hospital under the impression of a ruptured abdominal aortic aneurysm (AAA). On arrival at the Chang Gung Medical Center Emergency Department, the patient was awake and alert. His blood pressure (BP) was 140/76 mmHg, with a pulse of 90 beats/minute, and a temperature of 36 °C. On physical examination, his skin was pale and he was mildly diaphoretic. Abdominal examination showed hypoactive bowel sounds, no palpable mass and no audible bruit. Laboratory investigation showed hemoglobin of 6.9 mg/dl, hematocrit of 20.5%, leukocytes of 11,600/mm³ and impaired renal function, with blood urea nitrogen of 42 mg/dl and creatinine of 2.8 mg/dl.

The patient was taken for computed tomography (CT) to evaluate whether an aneurysm existed in the abdomen. Emergency CT revealed an AAA 3 cm in diameter, with an adjacent pseudoaneurysm 6 cm in diameter. Massive hematoma was noted in the right anterior pararenal space. The patient was awake and alert. His blood pressure (BP) was 140/76 mmHg, with a pulse of 90 beats/minute, and a temperature of 36 °C. On physical examination, his skin was pale and he was mildly diaphoretic. Abdominal examination showed hypoactive bowel sounds, no palpable mass and no audible bruit. Laboratory investigation showed hemoglobin of 6.9 mg/dl, hematocrit of 20.5%, leukocytes of 11,600/mm³ and impaired renal function, with blood urea nitrogen of 42 mg/dl and creatinine of 2.8 mg/dl.

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four cases with less than a 3-week interval, with 2 days being the shortest time to fistula development.\(^3,^5\) This confirms the necessity of being highly suspicious of an aortoenteric fistula after aortic reconstructive surgery when a patient presents with abdominal pain, gastrointestinal hemorrhage and sepsis.

Two types of secondary aortoenteric fistulas are generally described. Type 1, termed a true aortoenteric fistula or graft-enteric fistula, with or without a pseudoaneurysm, develops between the proximal aortic suture line and the bowel. This type of fistula is the most common, and often initiates "herald bleeding" following massive gastrointestinal bleeding potentially leading to fatal exsanguinations.\(^3,^10-12\) The main clinical manifestation of this type is always upper gastrointestinal bleeding (76\%),\(^13\) which might be either hematemesis or melena with equal frequency. Sepsis and abdominal pain are relatively rare with this type of fistula. Type 2, or a paraprosthethic-enteric fistula, develops no communication between the bowel and the graft. It accounts for 15\%-20\% of secondary aortoenteric fistulas.\(^13\) In this type of fistula, bleeding occurs from the edges of the eroded bowel by mechanical pulsations of the aortic graft. Sepsis is more frequently associated with this type of fistula (57\%).\(^13\) In addition to sepsis, gastrointestinal hemorrhage (30\%),\(^13\) abdominal pain (20\%),\(^13\) septic emboli in the lower extremities, septic arthritis, multicentric osteomyelitis and hypertrophic osteoarthropathy have been described.\(^12,14-16\)

The exact pathogenesis of secondary aortoenteric fistulas is unknown, but two mechanisms have been proposed. One is the constant pulsating motion of the graft on the bowel wall, and the other is adhesion of an already infected, inflamed graft site to the wall of the gastrointestinal tract.\(^5,^17-18\) In addition to the above mechanisms, infection or partial rupture of the suture line of the anastomosis and inadequate coverage of both the graft and proximal anastomosis with retroperitoneal tissue may contribute to SAF.\(^5\)

In our case, Klebsiella pneumoniae was cultured from the resected AAA. The relatively high risk of either graft or suture line infection was considered. Busuttil et al., based on animal and clinical studies, proposed that a sufficient concentration of bacteria is necessary for SAF to form.\(^19\) Therefore, we considered that the concentration was sufficient to develop a fistula in our case because of the positive AAA culture. The most common organisms cultured are enteric pathogens and Klebsiella species.\(^5,^20\) Generally, broad-spectrum antibiotic coverage following an operative procedure for SAF is necessary and ranges from 2-6 weeks according to the degree of graft infection. Patients with positive arterial wall cultures are at higher risk for subsequent disruption of the vascular anastomosis and hemorrhage and should receive 6 weeks intravenous antibiotics followed 6 months of oral administration.\(^2,^5\) If appropriate antibiotics are administered as soon as possible to patients presenting with sepsis after mycotic aneurysmectomy with graft replacement, the infectious illness may be controlled to prevent the complication of a SAF.

Because of the nonspecific nature of the clinical history and physical findings, diagnosis of aortoenteric fistula is difficult to make preoperatively. There is no single diagnostic investigation which has a very high specificity and sensitivity, including upper gastrointestinal endoscopy, computed tomography, angiography or gallium 67 CT. Nevertheless, exploratory laparotomy is the only method that can definitely establish a diagnosis. Once the SAF is identified, the surgical procedures most commonly employed are graft excision, oversewing of the aortic stump, repair of the gut defect and placing a new graft in situ or extra-anatomic bypass.

The mortality rate during surgery and in the postoperative period is relatively high, averaging about 50\%-60\%.\(^5,^7,^11\) The major complications postoperatively include recurrence of the fistula, aortic stump disruption and infection of the new graft. Blowout of the aortic stump is the most common complication usually caused by residual infection. To prevent complications, an atraumatic operative technique, aseptic operative field, appropriate antibiotics and proper covering of the graft with periaortic tissue are important.

In summary, a high index of suspicion of secondary aortoenteric fistula is required in any patient who presents with sepsis, abdominal pain and gastrointestinal hemorrhage after aortic reconstructive surgery. Because of the increasing frequency of aortic reconstructive surgery, more cases of aortoenteric fistulas will be encountered in the ED. Early diagnosis and prompt surgical management are mandatory for saving the life of a patient with an aortoenteric fistula.
REFERENCES

次發性主動脈十二指腸瘻管

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次發性主動脈十二指腸瘻管是腹腔主動脈重建手術後相當少見但嚴重的併發症，通常在術後幾個月到幾年之內發生。上消化道出血是它最常出現的症狀，外科手術是唯一的治療方式。若未早期發現與治療，死亡率相當的高。我們報告一例主動脈瘤患者，在術後第20天出現上消化道出血併發貧血性休克，經緊急剝腹探查手術，發現在主動脈與十二指腸間有一瘻管產生，雖經手術切除，病患還是在48小時後死亡。隨著老年化人口與主動脈重建手術的增加，在未來，次發性主動脈十二指腸瘻管發生率會有增加的趨勢。臨床醫師必須有高度的警覺性與認知，才能診斷此種高危險性急症。 (長庚醫誌 2002;25:626-30)

關鍵字：次發性主動脈十二指腸瘻管，腹腔主動脈重建手術，上消化道出血，主動脈瘤，次發性
主動脈十二指腸瘻管。