INTRODUCTION

Aortoenteric fistula rarely occurs due to the current method of treating abdominal aortic aneurysms by aggressive elective resection (1). It is usually due to erosion of an aortic aneurysm into the intestine, generally at the third and fourth portions of the duodenum (2). In most series, the mortality is extremely high (2-4). The chance of survival in patients presenting with hematemesis or melena is significantly higher when the time interval from the onset of bleeding to operative intervention is short (5). However, in cases with diffuse aortic sepsis or retroperitoneal purulence, morbidity and mortality become higher and an extra-anatomic bypass followed by transabdominal removal of the temporarily placed graft may be required. A positive preoperative blood culture predicts a poor outcome (4).

In our case, the patient presented with chronic primary aortoduodenal fistula extensively involving the duodenum and Gram-negative sepsis. Initially, we performed in situ revascularization with a prosthetic graft and an en bloc resection of the aneurysm with the surrounding structures. After he recovered from sepsis and had been stabilized, a staged extra-anatomic bypass followed by transabdominal removal of the temporarily placed graft was done. This management plan will allow the highest success rate and may be a prudent management protocol for these difficult cases.

CASE REPORT

A 68-yr-old male with a past medical history of cerebral infarction was found to have a large abdominal aortic aneurysm with primary aortoduodenal fistula formation at an outside facility 5 months before. He presented with melena and back pain. Upper gastrointestinal endoscopy and computed tomography were performed at an outside facility, which showed a huge pulsating submucosal mass-like lesion with mucosal hemorrhage on the third duodenal portion and an infrarenal aortic aneurysm suspicious of duodenal erosion (Fig. 1). He received no operative intervention at the request of the family. Five months later, he was admitted to our hospital with melena and a high fever (38.7 ℃). On physical examination, he was noted to be diaphoretic with a diffusely tender abdomen, a pulsating abdominal mass, and gross blood on the rectal examination. His initial hemoglobin level was 6.6 mg/dL and the white cell count was 17,400/µL. Preoperative Gram staining and culture of blood were obtained. Gram staining identified white blood cells and Gram-negative rods. We planned a staged operation: initially, an emergency laparotomy and control of the aorta allowed stabilization of the patient, identification of the fistula, and direct in situ placement of the prosthetic graft followed by an en bloc resection of the aneurysm and the surrounding structures. After he recovered from sepsis and had been stabilized, a staged extra-anatomic bypass followed by transabdominal removal of the temporarily placed graft was done. He received broad-spectrum antibiotics with imipenem cilastatin...
sodium 500 mg and vancomycin HCl 500 mg four times a day, starting preoperatively.

An emergency exploratory laparotomy was performed. During surgical exploration, the duodenum was extensively adhered to the abdominal aortic aneurysm. Mobilization of the duodenum and dissection at a conventional location to the left of the third portion of the duodenum were not feasible. Following isolation of the involved third and fourth portions of the duodenum with GI staplers (Ethicon Endo-Surgery, Inc., Cincinnati, OH, U.S.A.), dissection began under the uncinate process of the pancreas and continued proximally in a plane as close to the aneurysmal wall as possible. After injection of heparin, the aorta was clamped proximal to the aneurysm and to the iliac arteries distal to the aneurysm. The fistula-bearing area of the aneurysm was resected en bloc with the attached duodenum (Fig. 2). Diffuse aortic sepsis and retroperitoneal purulence were noted. Local debridement and in situ revascularization with a PTFE graft (IMPRA Inc., Tempe, AZ, U.S.A.) were done. After an omental pedicle was placed over the graft, pylorus-preserving pancreatoduodenectomy was performed because extensive destruction of the duodenal wall was noted. Preoperative blood culture and cultures from the aneurysm and the perianeurysmal fluid showed a growth of *Enterobacter cloacae*, sensitive to imipenem cilastatin sodium. Imipenem cilastatin sodium and vancomycin HCl were continued intravenously and three serial blood cultures were sterile postoperatively. Seven days after the surgery, follow-up abdomen-pelvis computed tomography showed a large amount of perigraft fluid. A staged axillofemoral extra-anatomic bypass followed by transabdominal removal of the temporarily placed aortic graft with aortic stump closure was performed. Postoperative course was remarkable only for acute renal failure and his blood cell count was normal. However, 20 days after surgery, the patient suddenly died of cardiac arrest due to myocardial infarction. There was no evidence of aortic stump dehiscence or sepsis, and the extra-anatomic bypass was patent.

**DISCUSSION**

Primary or spontaneous aortoenteric fistulae are rare, with their incidence has been reported at 0.04% to 0.07% in autopsy series (6). The incidence of fistulae as a complication of aortic aneurysms is 0.1% to 0.8% (7). The etiology of primary aortoenteric fistulae varies and also includes tuberculosis, syphilis, and other mycotic aortic infections (8-11). In most series, however, the most common etiology is erosion of an atherosclerotic abdominal aortic aneurysm into a segment of intestine with the duodenum being involved in more than 80% of cases (2, 7). The distal portion of the duodenum is fixed, retroperitoneal, and just anterior to the aorta. Because of the close approximation and fixed nature of the duodenum, it is thought that the expanding nature of the aortic aneurysm causes irritation and inflammation, resulting in eventual fistulization over time (11).

Aortoenteric fistula is one of the most challenging problems that confront the vascular surgeons. The mortality rate of untreated aortoenteric fistula with upper gastrointestinal hemorrhage is nearly 100%. With appropriate treatment, survival rates varying between 18% and 93% have been reported (7, 11). Postoperative complications occur in as many as 40% of cases (11). Overall, the postoperative mortality rate is greater than 30% (7). Although surgical repair of the aortic aneurysm and fistula is the standard treatment, there are many options for the operative treatment of the aortoenteric fistula itself. The operative treatment varied depending on the patient’s hemodynamic stability, the extent of infection, and the patient’s vascular anatomy. In cases of primary aortoenteric fistula from abdominal aortic aneurysm and no evidence of gross
infection, anatomic in situ repair with a Dacron or PTFE graft, coupled with repair of the adjacent area of bowel and placement of omentum between the intestinal and aortic repairs, is preferred (11, 12). However, in cases of primary aortoenteric fistula associated with infected abdominal aortic aneurysm, controversy remains over the optimal method because of the continued publication of series with a high mortality, amputation, and aortic disruption rates. The surgeon must choose an operative plan that is appropriate to the urgency and seriousness of the patient’s manifestations of fistula and infection. Diffuse aortic sepsis or retroperitoneal purulence requires an extra-anatomic bypass and aortic stump closure. This has a predictably high likelihood of complications and death. A positive preoperative blood culture is the best predictor of mortality with increased amputation rates due to infection of the extra-anatomic bypass (4). Therefore, in selected cases with sepsis, a prudent management protocol is required.

In our case, the patient presented with chronic primary aortoduodenal fistula extensively involving the duodenum and Gram-negative sepsis. We planned a staged operation. Initially, an emergency laparotomy and control of the aorta allowed stabilization of the patient, identification of the fistula, and direct in situ placement of the prosthetic graft followed by an en bloc resection of the aneurysm and the surrounding structures. The patient recovered from sepsis and was stabilized. After serial blood cultures were sterile, a staged extra-anatomic bypass followed by transabdominal removal of the temporarily placed graft was done. Although the patient died of myocardial infarction and long-term follow-up was not possible, there was no evidence of infection or graft occlusion. In conclusion, the present management plan will allow the highest success rate possible and may be a prudent management protocol for these difficult cases.

REFERENCES

1. Martinez R, Garces D, Podeur L, Abdel Aal K, Laffon M, Castellani L. Ruptured abdominal aortic aneurysm. A ten year experience. J Cardiovasc Surg 1997; 38: 1-6.
2. Buat TJ. Synthetic vascular graft infections. II. Graft-enteric erosions and graft-enteric fistulas. Surgery 1983; 94: 1-9.
3. Moulton S, Adams M, Johansen K. Aortoenteric fistula: a 7 year urban experience. Am J Surg 1986; 151: 607-11.
4. Peck JJ, Eidemiller LR. Aortoenteric fistulas. Arch Surg 1992; 127: 1191-4.
5. Kleinman LH, Towne JB, Bernhard VM. A diagnostic and therapeutic approach to aortoenteric fistulas: clinical experience with twenty patients. Surgery 1979; 86: 868-80.
6. Taheri SA, Kalayat MN, Grippi J, Ricotta JJ, Kale J, Bernhard H. Surgical treatment of primary aortoduodenal fistula. Ann Vasc Surg 1991; 5: 265-70.
7. Sweeney MS, Gadacz TR. Primary aortoduodenal fistula: manifestation, diagnosis, and treatment. Surgery 1984; 96: 492-7.
8. Lee OJ, Kim SH. Aortoesophageal fistula associated with tuberculous mediastinitis mimicking esophageal Dieulafoy’s disease. J Korean Med Sci 2002; 17: 266-9.
9. Reynolds JV, Galloway JM. Primary aortoduodenal fistula: a case report and review of the literature. Ir J Med Sci 1991; 160: 381-4.
10. Pagni S, Denatale RW, Sweeney T, McLaughlin C, Ferneini AM. Primary aorto-duodenal fistula secondary to infected abdominal aortic aneurysms: the role of local debridement and extra-anatomic bypass. J Cardiovasc Surg 1999; 40: 31-5.
11. Lemos DW, Raffetto JD, Moore TC, Menzoian JO. Primary aortoduodenal fistula: a case report and review of the literature. J Vasc Surg 2003; 37: 686-9.
12. Daugherty M, Shearer GR, Ernst CB. Primary aortoduodenal fistula: extra-anatomic vascular reconstruction not required for successful management. Surgery 1979; 86: 399-401.