

Double Primary Aortoenteric Fistulae: A Case Report of Two Simultaneous Primary Aortoenteric Fistulae in One Patient

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Aortoenteric fistula is a rare but potentially fatal condition causing massive gastrointestinal bleeding. In particular, double primary aortoenteric fistulae are vanishingly rare. We encountered a 75-year-old male patient suffering from abdominal pain, hematochezia, hematemesis, and hypotension. His computed tomography images showed abdominal aortic aneurysm and suspected aortoenteric fistulae. During surgery, we found two primary aortoenteric fistulae. The one fistula was detected between the abdominal aorta and the third portion of the duodenum, and the other fistula was detected between the abdominal aorta and the sigmoid colon. We conducted the closure of the fistulae, the exclusion of the aneurysm, and axillo-bifemoral bypass with a polytetrafluoroethylene graft. The patient was discharged with no complications on the 21st postoperative day.

Key words: 1. Aneurysm
2. Aorta
3. Fistula

CASE REPORT

A 75-year-old man visited a local hospital complaining of abdominal pain and melena. His computed tomography showed a 7 cm-sized abdominal aortic aneurysm and suspected an aortoenteric fistula (Fig. 1). He needed surgery, and was transferred to Pusan National University Hospital. When he arrived at the emergency room, his blood pressure was 70/40 mmHg, his heart rate was 120/min, and the body temperature was 36.4°C. He complained of abdominal pain, hematochezia, and hematemesis. Owing to persistent hypotension and bleeding, we had to perform an emergency operation.

A median laparotomy incision was made and the abdominal aorta was exposed. We detected two aortoenteric fistulae.

One fistula was located between the abdominal aorta and the third portion of the duodenum, and the other fistula was detected between the abdominal aorta and the sigmoid colon (Fig. 2). After the disconnection of each fistula, primary repair of the duodenum and the sigmoid colon was carried out. We worried about graft infection, and thus we carried out the right axillo-bifemoral bypass with a ring enforced 8 mm polytetrafluoroethylene graft. Next, we exclude the aneurysm with an oversewn infra-renal aorta and both common iliac arteries. We checked for adequate circulation to both of the patient's feet. He was transferred to the intensive care unit in critical condition. He stayed at the intensive care unit for 7 days and was transferred to the general ward in a tolerable condition. Ileus occurred on the 9th postoperative day, so we

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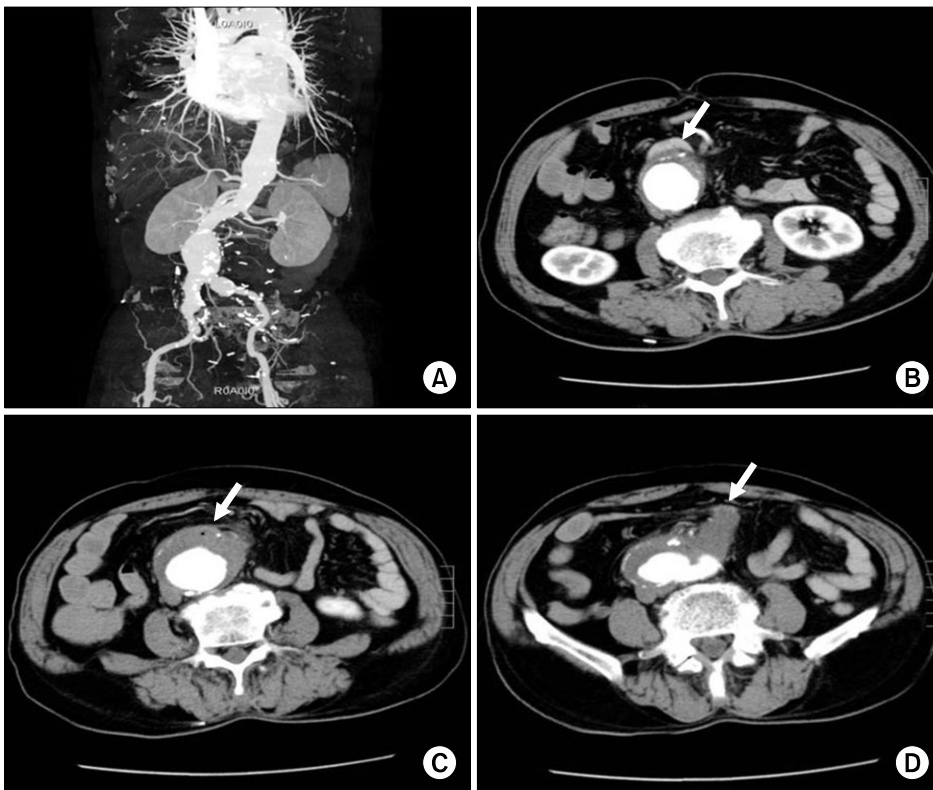


Fig. 1. Preoperative computed tomography (CT). (A) 3-Dimensional reconstructive CT showed abdominal aortic aneurysm. (B) Anterior wall of the aneurysm abutted on posterior wall of the duodenum (arrow). (C) Ectopic air bubble around the aorta (arrow). (D) The aneurysm showed consealed rupture to the left inferolateral aspect abutting with the sigmoid colon (arrow).

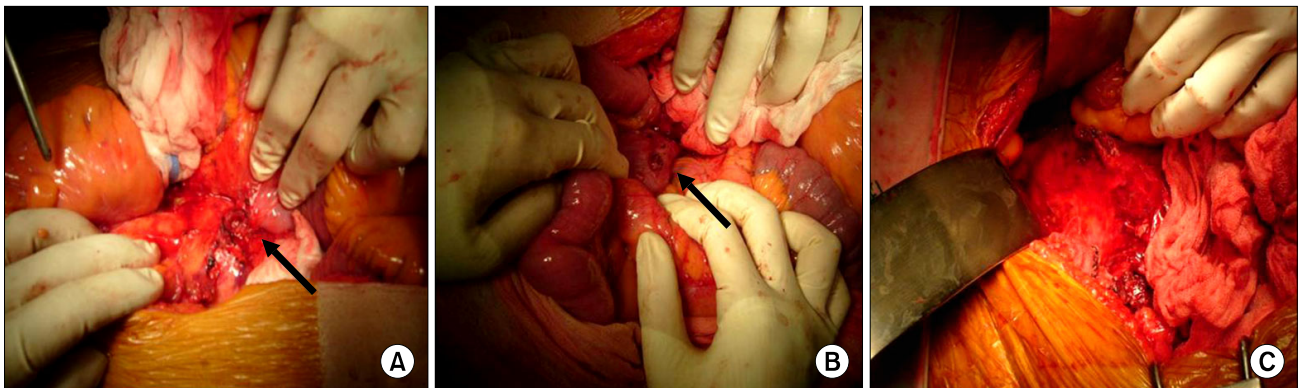


Fig. 2. Operative findings. (A) The aorta communicated with the duodenum (arrow). (B) The surface of disconnected fistula of the sigmoid colon (arrow). (C) The abdominal aortic aneurysm.

were stopping oral intake and total parenteral nutrition began. Then ileus recovered on the 15th postoperative day. He was discharged on the 21st postoperative day with no complications. Over the four-month follow-up period, no infection signs were noted, and the graft patency was well-maintained (Fig. 3).

DISCUSSION

Primary aortoenteric fistula is a communication between the native aorta and the gastrointestinal tract with an incidence rate of 0.1% to 0.8% [1,2]. The first report of successful treatment of two simultaneous primary aortoenteric fistulae in one patient by Varekamp and Spruijt [3] in 1998,

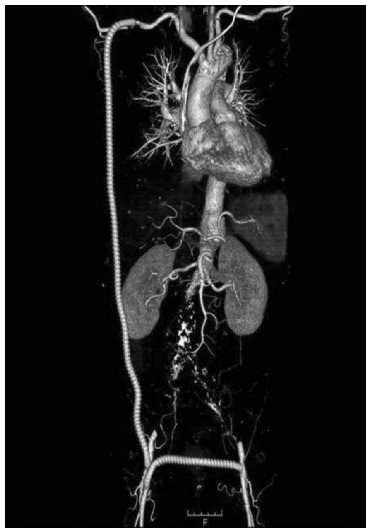


Fig. 3. Follow-up computed tomography. The graft patency was well maintained.

and we could not find other report on the double primary aortoenteric fistulae.

The most common cause of primary aortoenteric fistula is atherosclerotic aortic aneurysm. However, tuberculosis, syphilis, infection, cancer, and foreign bodies are known to cause primary aortoenteric fistula [4]. Most aortoenteric fistulae communicate between the aorta and the third part of the duodenum [5], because the duodenum overlies the infrarenal aorta. Some cases have reported fistulae occurring among the esophagus, small and large bowel, and stomach [5].

Gastrointestinal bleeding, abdominal pain, and pulsating abdominal mass constitute the classical clinical symptom triad of primary aortoenteric fistula. However, only 11% of patients exhibit all of these findings [5]. Despite the communication between the aorta and gastrointestinal lumen, septicemia and fever are rarely observed [1]. “Herald bleed” is a minor bleed, which is usually self-limited. However, in most cases, massive hemorrhage occurred within hours to a month after the herald bleed, and one-third of the cases occurred within 6 hours [5]. It is theoretically desirable that diagnosis and surgical treatment should be carried out within this short time period.

Currently, intravenous contrast-enhanced computed tomography (CT) aortograms are recommended as an initial tool for the diagnosis of primary aortoenteric fistula [6]. The CT find-

ings of the aortoenteric fistula are as follows: air bubbles around the aorta, loss of the fat plane between the aorta and gastrointestinal tract, extravasations of aortic contrast material into the enteric lumen, and bowel wall edema around the aorta.

The mortality rate of the surgery for the primary aortoenteric fistula is as high as 30% to 40% [5]. For successful outcome, the following four principles of surgery should be considered: control the bleeding, repair the bowel defect, selectively restore distal circulation, and eradicate any associated infection [3]. For distal circulation, *in situ* aortic reconstruction or extra-anatomic bypass is generally selected. In the previous reports [7], when *in situ* aortic reconstruction was performed, the incidence of graft infection was not particularly high. However, in our case, because there were two repair sites in the bowel, we thought that the possibility of graft infection or secondary aortoenteric fistula formation might be somewhat increased. Therefore, we carried out the axillo-bifemoral bypass, and ensured that the decision was appropriate. In recent years, endovascular aortic aneurysm repair (EVAR) has been regarded as a treatment for primary aortoenteric fistulae in elderly or hemodynamically unstable patients. Although the success rate of short-term treatment is relatively high, the risk of reinfection remains. Therefore, EVAR can be employed as a bridging procedure for definitive surgical repair [8]. In order to control the infection, Saers and Scheltinga [5] recommended at least one week of antibiotic therapy in negative cultures and 4 to 6 weeks of antibiotic therapy in positive cultures. In our case, we did not prepare intraoperative cultures. Thus, we used antibiotics for the four-week follow-up period.

In conclusion, the incidence of double primary aortoenteric fistulae is a vanishingly rare condition with a high mortality rate, but rapid and accurate diagnosis coupled with appropriate surgical treatment can achieve good results. In our case, we successfully treated a patient suffering from this condition via an extra-anatomic bypass and primary closure of duodenum and sigmoid colon. Of course, long-term follow-up will still be necessary, but the short-term success of the procedure is clear.

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