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CLINICAL REPORT

Unexpected double-primary aortoenteric fistula resulting in massive bleeding after induction of anesthesia

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Abstract We report a case of a patient with a double-primary aortoenteric fistula with an abdominal aortic aneurysm. A 75-year-old man was taken to the operating room for the repair of an abdominal aortic aneurysm and a suspected aortoenteric fistula between the aorta and sigmoid colon. Sudden onset of massive bleeding through the nasogastric tube occurred after the induction of anesthesia. Surgical exploration confirmed an unexpected aortoduodenal fistula. Primary aortoenteric fistula is extremely rare and difficult to diagnose, and may cause fatal bleeding. The possibility of the presence of aortoenteric fistula, including multiple types, should be considered in the anesthetic management of abdominal aortic aneurysm.

Keywords Primary aortoenteric fistula · Abdominal aortic aneurysm · Gastrointestinal bleeding

Introduction

A primary aortoenteric fistula (AEF) is an open communication caused by the spontaneous erosion of the aorta into the gastrointestinal tract, described first by Sir Astley Cooper. This condition is very often fatal owing to unpredicted massive bleeding. Without surgical repair, the mortality rate is nearly 100 % [1]. The incidence of primary AEF is very low; fewer than 300 cases have been reported in the published data. Although computed tomography (CT) scans and esophagogastroduodenoscopy (OGD) are available, the definite diagnosis of primary AEF is still difficult. These diagnostic challenges that have a low incidence are dangerous for the patient. We report our experience of anesthesia for a patient with unexpected double-primary AEF (which is a very rare entity) with an abdominal aortic aneurysm.

Case report

A 75-year-old man with no specific past history was admitted to a local hospital for the evaluation of abdominal pain and a small amount of melena. He underwent CT scanning. A 7-cm-sized abdominal aortic aneurysm and aortoenteric fistula between the aorta and sigmoid colon was suspected on the CT scan, with no definite evidence of the aortoenteric fistula (Fig. 1). He was transferred to our hospital for operation. On arrival at the emergency room, his vital signs were blood pressure of 70/40 mmHg and tachycardia of 120 bpm. He complained of abdominal pain and hematochezia. Irrigation of nasogastric tube showed no active bleeding. In spite of continued transfusion of fluid and blood, the hypotension continued and the patient was taken to the operating room with suspected rupture of an abdominal aortic aneurysm (AAA) or aortoenteric fistula between the abdominal aorta and sigmoid colon. The patient's BP was 130/80 mmHg in the operating room before the induction of anesthesia. Induction of anesthesia was done with midazolam 5 mg, rocuronium 50 mg, and sevoflurane 2 vol %. During induction, systolic BP was

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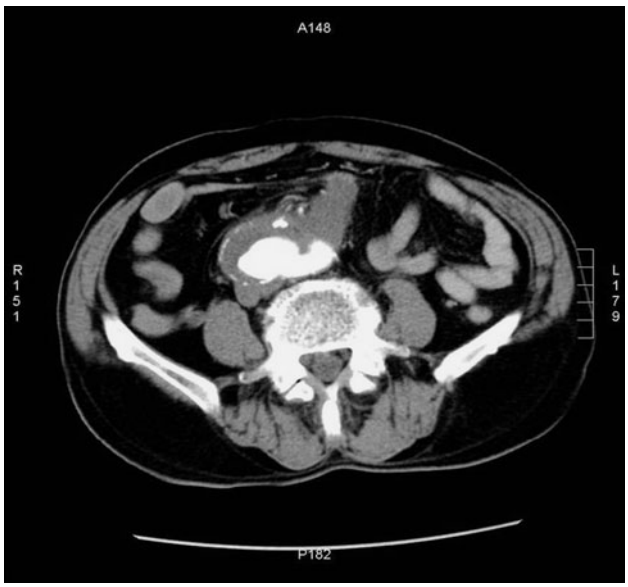

Fig. 1 Computed tomographic scan shows a large infrarenal aortic aneurysm with concealed rupture, abutting the sigmoid colon

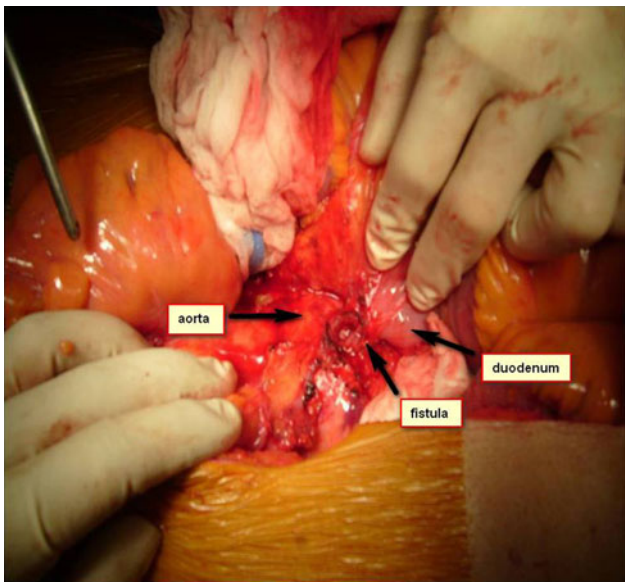

Fig. 2 Intraoperative finding of aortoenteric fistula (AEF) between the third portion of the duodenum and the abdominal aorta is seen

maintained. After induction, massive bleeding suddenly occurred through the nasogastric tube and the systolic BP subsequently dropped to 80 mmHg. To find the origin of the bleeding, exploratory laparotomy and intraoperative OGD were performed simultaneously. OGD showed a blood clot in the stomach but no active bleeding. On laparotomy, an unpredicted aortoduodenal fistula was found between the third part of the duodenum and the aortic aneurysm; in addition, impending rupture of an infrarenal-type abdominal aortic aneurysm was found (Fig. 2), and a suspected aortoenteric fistula between the aorta and sigmoid colon was also found. It took some time to figure out the origin of the bleeding. Fortunately, we had prepared for massive bleeding because of the possibility of rupture of the AAA. Therefore, we were able to prevent severe and possibly fatal hypotension, using a rapid infusor (FMS 2000; Belmont Instrument Corp., Billerica, MA). Both fistulae were disconnected and repair of the duodenum and sigmoid colon was performed. An axillo-bifemoral bypass and resection of the aneurysm were done. The patient was transferred to the intensive care unit (ICU) and received mechanical ventilation for 7 days because of pneumonia. Apart from his lung condition, his recovery was uneventful and he was discharged without any complications.

Discussion

Aortoenteric fistula (AEF) is a direct communication between the abdominal aorta and the gastrointestinal tract. AEF is classified as primary or secondary. Secondary AEF can arise mainly after AAA repair, and a common cause is graft infection [2]. Primary AEF, more rare than secondary, occurs spontaneously, with an incidence rate of 0.04–0.07 % at autopsy, and is commonly associated with AAA (83 % of primary AEF) [1, 3, 4]. In one report, 1 % of all acute aneurysms had an associated primary AEF [4]. The most common site of primary AEF is the duodenum (54 %), especially the third portion. Other sites are the esophagus, jejunum, sigmoid colon, stomach, and ileum [1, 3]. The presence of two simultaneous primary AEFs has been reported in only two patients previously [5, 6].

Because of massive and life-threatening bleeding, without surgery, primary AEF leads to 100 % mortality. Therefore, early detection and intervention should be done. However, primary AEF poses diagnostic challenges owing to the subtleties of its clinical symptoms and evolution [7]. Commonly used diagnostic methods include CT scanning, OGD, and arteriography. However, the detection rates for each of these modalities are 61, 25, and 26 %, respectively [3]. Thus, the exact diagnosis is still difficult. In one report, despite the use of repeated CT scanning, the diagnosis of primary AEF was not made [8]. CT findings suggestive of primary AEF are as follows: air within the aortic wall, focal bowel-wall thickening, disruption of aortic fat cover, and contrast penetrating into the bowel [3]. In the present patient, we used enhanced CT scanning, and found a concealed rupture of an aneurysm abutting the sigmoid colon. Therefore, we had a suspicion that there was an aortoenteric fistula between the aortic aneurysm and sigmoid colon. However, a definite diagnosis was not reached, and the presence of an aortoduodenal fistula was not even

considered until it was found during laparotomy. After the patient's massive bleeding, we performed OGD intraoperatively. OGD is regarded as the most useful diagnostic method for searching upper gastrointestinal bleeds, but it has limitations in diagnosis too. Identification of a fistula below the third portion of the duodenum is especially difficult, because the angulation between the third and fourth portions of the duodenum is very acute [9]. In our patient, blood clots were found, but no active bleeding focus was found. Eventually, we found the origin of the bleeding during laparotomy. In our patient, the primary AEF had existed in two areas simultaneously. We did not expect this rare entity, and therefore it took a long time to find the cause of the bleeding. In a patient with primary AEF, suspicion is the most important diagnostic tool and a laparotomy seems to be the definitive method of diagnosis.

Our patient bled three times. First, he bled a small amount of melena at the local hospital. Second, there was massive hematochezia in the emergency room of our hospital. Finally, there was upper gastrointestinal bleeding in the operating room after the induction of anesthesia. The first minor bleeding was considered to be herald bleeding, which is common for AEF and is later followed by a life-threatening bleed [1]. The second bleed was considered to be a typical catastrophic and life-threatening bleed which caused unstable vital signs. This second bleed seemed to have been due to the aortosigmoidal fistula, because the hematochezia was fresh-colored and irrigation through the nasogastric tube was negative for blood. Until this point the bleeding sequence in our patient was similar to that described in the literature. However, there was an unexpected massive bleed through the nasogastric tube after the induction of anesthesia.

This bleed was unusual in view of the fact that it occurred just after anesthesia induction.

Intermittent bleeding in AEF is thought to be due to vasospasm, thrombus formation, and contraction of the bowel around the fistula tract. The thrombus that plugs the fistula may be pushed out owing to the fluid used in resuscitation and owing to elevated blood pressure.

In our case, the patient had received a considerable volume of fluid and blood in the emergency room, to restore the hypotension. At the time of anesthesia induction the patient's BP was 130/80 mmHg and central venous pressure (CVP) was 7 mmHg; these values were higher than those in the emergency room. After the unexpected massive bleed, systolic pressure dropped to 80 mmHg and thereafter was maintained below 100 mmHg.

Consequently, we think that the causes of the bleed in our patient were elevated BP and instillation of the large volume of fluid during anesthesia induction and loss of contraction of the bowel around the fistula owing to the use of muscle relaxants. Massive bleeding during anesthesia induction can interfere with intubation and induce the aspiration of blood owing to regurgitation of blood within the upper gastrointestinal tract. Fortunately, the patient's nasogastric tube was kept in place. Therefore, we were able to find out about the bleed early and prevent aspiration during intubation. The duodenum, especially the third portion, is the most common site of primary AEF, but it is difficult to view this portion using OGD. Thus, keeping a nasogastric tube in place and monitoring the bleed through this tube seems to be useful when an aortoenteric fistula is suspected.

In the literature, controlled hypotension and avoidance of overaggressive fluid therapy are accepted lifesaving methods in the management of ruptured aortic aneurysm [10]. We think these suggestions should be applied to the induction and maintenance of anesthesia in patients with primary AEF as well. Saers and Scheltinga [3] have also suggested it is helpful to maintain systolic tension at 60–100 mmHg.

In conclusion, primary AEF is a very rare and often fatal disease. Therefore, early diagnosis and treatment is essential. However, this entity is difficult to diagnose. Consequently suspicion is the most important diagnostic tool. Primary AEF should be strongly suspected in cases of gastrointestinal bleeding associated with AAA, and we should also keep in mind that multiple primary AEF is possible as well. In anesthesia for these patients, caution and preparation for the occurrence of massive bleeding are needed, including the use of a nasogastric tube, large-bore intravenous route, rapid infuser, and vasoactive drugs. In addition, elevated blood pressure and excessive fluid therapy should be avoided to prevent additional bleeding during the induction as well as maintenance of anesthesia.

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