Turk J Gastroenterol 2004; 15 (4): 253-257

Acute gastrointestinal bleeding due to primary aortoduodenal fistula: Report of two rare cases

Primer aortoduedonal fistül nedeni ile akut gastrointestinal kanama: Nadir görülen iki vaka raporu

Ufuk TÜTÜN, Ayşen AKSÖYEK, Ahmet Tulga ULUS, Ali İhsan PARLAR, M. Kamil GÖL, Murat BAYAZIT

Türkiye Yüksek İhtisas Hospital, Clinic of Cardiovascular Surgery, Ankara

Primary aortoduodenal fistulas are among the rare causes of gastrointestinal hemorrhage and are defined as communications between the native abdominal aorta and the duodenum. The mortality rate is very high if undiagnosed and untreated. Two male patients, 61- and 76-years-old, were admitted to the emergency unit at different times with the chief complaints of abdominal pain, gastrointestinal hemorrhage and pulsatile mass in their abdomen. The first case experienced sudden massive upper gastrointestinal bleeding while being prepared for an emergency operation in the intensive care unit, and cardiac arrest developed within a few minutes. After resuscitation and successful surgical operation, the patient woke up without any neurological defect or sequelae and was extubated at the 9th postoperative hour. The second patient, who had been wounded by gun shot 30 years previously was admitted to the hospital because of simple gastrointestinal hemorrhage. A para-aortic pseudo-aneurysm connected with the duodenum was diagnosed by computed tomography. After successful surgical operation, the patient was discharged. In this report, a case of ruptured primary aortic aneurysm and another case of para-aortic pseudo-aneurysm connected with the duodenum, both of which were treated successfully by surgical operation, are presented.

Key words: Aorta, aneurysm, duodenum, fistula, rupture, pseudo-aneurysm

Native abdominal aort anevrizması ve duodenum arasındaki ilişki olarak tanımlanan primer aorta duodenal fistül çok nadir olarak rastlanılan akut gastrointestinal kanama nedenidir. Eğer teşhis ve tedavi edilmezse yüksek mortaliteye sahiptir. Farklı zamanlarda, biri 61 yaşında diğeri 76 yaşında iki erkek hasta karın ağrısı, karında pulsatil kitle ve akut gastrointestinal kanama şikayetleri ile acil servisimize başvurdu. Bunlardan ilk gelen hasta, acil operasyon için yoğun bakım ünitesinde hazırlanırken ani gelişen hemoptizi ile birkaç dakika içinde kardiak arrest oldu. tik hasta resüstasyon ve başarılı cerrahi sonrası, postoperatif 9'uncu saatte herhangi bir sekel kalmadan uyandı. Basit bir gastrointestinal kanama şikayeti ile başvuran diğer hasta 30 yıl önce ateşli silah yaralanmasına maruz kalmış. Komputerize tomografide paraaortik pseudo anevrizma duodenumlada ilişkili idi. Başarılı cerrahi sonrası bu hastada taburcu edildi. Bu makalede duodenuma rupture olan primer aortik anevrizma ile gelen bir vaka ile paraaortik pseudoanevrizması olan diğer bir başka vakanın başarı ile tedavi edildiği sunulmuştur.

Anahtar kelimeler: Aorta, anevrizma, duedonum, fistül, rüptür, pesudo-anevrizma

INTRODUCTION

Abdominal aortic aneurysm (AAA) is a localized dilatation of the abdominal aorta, which arises below the renal arteries and involves the latter abdominal aorta. Abdominal aortic aneurysms do not usually cause a significant symptom unless rapid expansion, leak or rupture into the retro-peritoneal field or another tissue occurs (1-3). Primary aortoduodenal fistulas are very rare and are defined as communications between the native aorta and the duodenum; gastrointestinal bleeding represents the first symptom (3). Secondary aortoduodenal fistulas arise between a suture line of a vascular graft and the intestine; the complication often occurs months to years after the original surgery (4-5).

In this report, a case of a ruptured primary aortic

Address for correspondence: Ufuk TÜTÜN
Doktorlar 91 Sitesi 688. Sokak No: 11/11 06530 Ümitköy,
Çayyolu-Yenimahalle, Ankara, Turkey
Fax: +90 312 466 56 88
E-mail: ufuktutun@hotmail.com

Manuscript received: 26.07.2004 **Accepted:** 09.11.2004

aneurysm to the last part of the duodenum and another case of a ruptured para-aortic pseudoaneurysm to the 4th part of the duodenum are presented.

CASE REPORT

Case 1

A 61-year-old male patient was admitted to the emergency unit of our hospital with the chief complaints of abdominal pain and a pulsatile mass in his abdomen. He had been admitted to a local community hospital 15 days previously with acute gastrointestinal hemorrhage, which was treated medically. His medical report included the records of his physical and laboratory examinations. A pulsatile mass was palpated in the middle abdominal area and the abdominal ultrasonography performed revealed aneurysmal dilatation of the abdominal aorta. His hematocrit level was 27% at his admittance to our hospital. An AAA was seen on computed tomography (CT) scan performed immediately after he was hospitalized to our cardiovascular intensive care unit and suggested a contained aortic rupture in the retro- peritoneal area (Figure 1). There was no evidence of fistulization of the aorta to any part of the gastrointestinal tract. Abdominal pain and manifest anemia were also consistent with the diagnosis of a ruptured AAA. While being prepared for an emergency operation in the intensive care unit, a sudden massive upper gastrointestinal hemorrhage began, and within a few minutes cardiac arrest developed. There was no time for further investigation, such as an endoscopic examination or angiography.

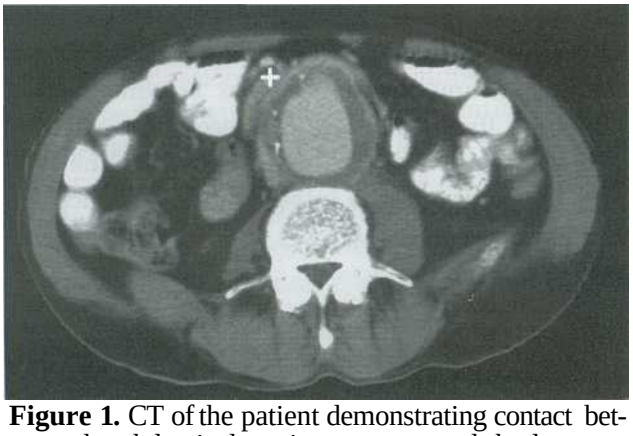

Figure 1. CT of the patient demonstrating contact between the abdominal aortic aneurysm and the last part of the duodenum. +, The contact point between the abdominal aorta and last part of the duodenum

Massive blood and clear crystalloid solutions were infused via an immediately administered subclavian vein catheter. While a cardiovascular surgery resident was performing external cardiac massage, the patient was immediately transferred to the operating room.

An emergency laparotomy with median incision was performed, while external cardiac massage was maintained in the operating room. Aortic bleeding was first controlled by placing an aortic cross clamp into the abdominal aorta at the level of the diaphragmatic hiatus. This maneuver provided hemodynamic stability, and further external cardiac massage was no longer needed. Afterwards, the abdominal aorta and duodenum were carefully and delicately dissected. A small defect in the 4th part of the duodenum, which was in communication with the abdominal aorta, was discovered (Figure 2). A tubular dacron graft, 12 cm in length and 20 mm in diameter, was interposed to the infrarenal and suprailiac part of the aorta. The defect at the duodenum was sutured primarily by 3/0 prolene suture. Partial omentopexy was performed over the aortic graft.

Figure 2. An illustration of the aortoduodenal fistula. A, defect on the anterior wall of abdominal aorta; B, defect on the posterior wall of the last part of the duodenum

Histopathological examination of the resected AAA was unremarkable. Sections of the specimen were stained with hematoxylin and eosin and with stains for collagen elastin. Intimal inflammatory debris, cholesterol crystals, foam cells, patch calcification and medial degeneration were observed. Substantial chronic inflammatory aggregates including lymphocytes and plasma cells with macrophages were present in the adventitia and media of the atheromatous segments of the aorta. Germinal centers were found within the wall.

After a successful operation, a surprisingly uneventful postoperative course was observed. The patient awoke without any neurological defect or sequelae and was extubated at the 9th postoperative hour. Intestinal motility was present on the 3rd postoperative day, but oral intake was allowed on his 7th postoperative day, and decompression was applied during that period with a nasogastric Levine tube. Serologic tests were performed in the early postoperative period from blood samples and were negative for Brucella, Salmonella and syphilis.

The patient was discharged on the 10th postoperative day without any further complications. One month after his discharge from the hospital, he had no complaint other than mild incisional pain.

Case 2

A 76-year-old male patient was admitted to the emergency unit of our hospital with acute gastrointestinal hemorrhage. He had been wounded by a gun shot in the abdominal region 30 years previously, for which no abdominal operation had been performed.

An AAA was seen on CT scan, which was performed immediately before he was admitted to our cardiovascular intensive care unit, suggesting a para-aortic pseudo-aneurysm in the retroperitoneal area (Figure 3). Palpation of the retroperitoneal pulsatile mass during median laparotomy revealed thrill over the mass, which was consistent with the CT scan. When the duodenum was dissected from the pulsatile mass, we observed a small hole on the posterior surface of the duodenum connected with the abdominal aorta.

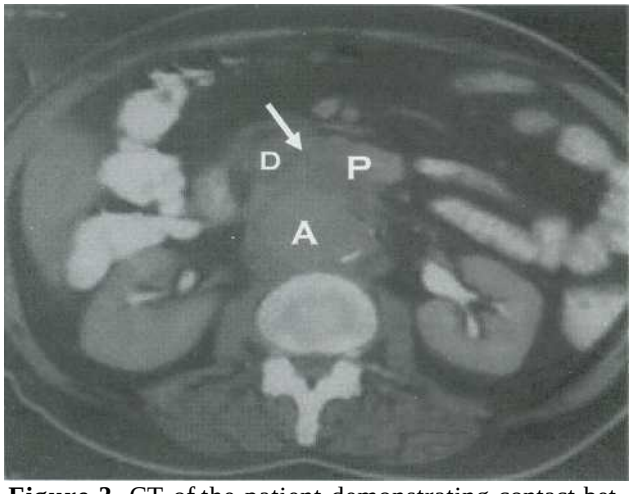

Figure 3. CT of the patient demonstrating contact between the pseudo-abdominal aortic aneurysm and the last part of the duodenum. The contrast medium, which passes to the duodenum from the para-aortic pseudo-aneurysm, is visible. A: aorta, P: pseudo-aneurysm, D: duodenum

During the operation, the abdominal aorta and the duodenum were carefully and delicately dissected. Bleeding from the aorta was controlled by placing an aortic cross clamp to the abdominal aorta at the level of the diaphragmatic hiatus. A small defect, communicating with the para-abdominal aortic pseudo-aneurysm with the 4th part of the duodenum, was discovered. A tubular dacron graft, 8 cm in length and 20 mm in diameter, was interpositioned into the infrarenal and suprailiac part of the aorta. The defect at the duodenum was sutured primarily by 3/0 prolene. Partial omentopexy was performed over the aortic graft.

After a successful surgical operation, the patient was discharged on the 7th postoperative day without any further complications.

DISCUSSION

The most common causes of upper massive gastrointestinal hemorrhage are peptic ulcer disease, hemorrhagic gastritis, bleeding of esophageal varices, vascular ectasia, trauma and Mallory-Weiss tears (1). Rare causes are Dieulafoy's lesion (an actively bleeding visible vessel without an ulceration), bleeding from tumors, ulcerative diseases of the small bowel and aorta-enteric fistula due to AAA(I).

Most AAAs remain asymptomatic until a perforation to the gastrointestinal tract occurs. Primary aortoduodenal fistula is a very rare cause of gastrointestinal hemorrhage, but has a very high mortality if undiagnosed and untreated. Primary aorta enteric fistulas often involve the third and fourth portion of the duodenum (88%) and less often the superior duodenum (4%), pars descendens of the duodenum (8%), the jejunum and ileum (2, 3). Frequently, gastrointestinal hemorrhage (hematemesis, 61% and melena, 50%), back pain, fever and pulsatile mass in the abdomen represent the first symptoms (2, 3, 4). Abdominal aortic aneurysms do not usually cause a significant symptom unless rapid expansion, leak or rupture into the retroperitoneal field or other tissues occurs. Diagnosis is difficult because of the nonspecificity of clinical presentation. Upper endoscopy rarely shows an adherent clot or pulsatile mass into the duodenum and can be performed in a relatively stable patient. Abdominal CT with intravenous

contrast is the preferred test for diagnosis (1). Karam et al. reported a very unusual presentation of ruptured infrarenal AAA to the right hemithorax, presenting with hemothorax (6).

Arteriosclerosis of the aorta is the primary cause of primary aorta enteric fistulas and accounts for more than two-thirds of the cases in the literature (2, 4). Other causes of primary aorta-enteric fistulas are chronic infective diseases such as syphilis, salmonellosis, brucellosis and tuberculosis (2). Histological and biochemical evaluations of patients with aneurysmal aorta revealed that low µ-1-antitrypsin levels, high intercellular adhesion molecules, interleukin-1 and tumor necrosis factor levels may contribute to the development of ruptured aneurysms (7-9). To our knowledge, there is no case report in the literature regarding posttraumatic aortic pseudoaneurysm with bleeding into the duodenum 30 years after the event.

Indeed, the classic clinical presentation of ruptured aortic aneurysm is the triad of sudden onset abdominal or flank pain, shock and the presence of a pulsatile mass in the abdomen. Symptoms of a patient with a primary aortoduodenal fistula are characterized by alternating relapse and remission, which is why surgical treatment is usually delayed. Most of the reported patients with aortoduodenal fistula died (2, 10). As the surgical treatment is usually delayed, such events are unfortunately managed as emergencies with very high preoperative and intraoperative mortality rates, but successful series, such as Van Olffen et al.'s, have been reported as well. They presented a successful series of six cases of primary aorta enteric fistula surgically treated at three different hospitals over a period of 15 years (4).

Primary aortoduodenal fistula as a result of AAA is an uncommon problem, so diagnosis is not easy. Some authors have reported that an aortoduodenal fistula may be diagnosed in some patients with a pulsatile abdominal mass, abdominal and back pain, and intermittent gastrointestinal hemorrhage, if suspected by the clinician (2, 4).

Surgical treatment is different from that of the secondary aortoduodenal fistula. It is recommended that repair of the aortic aneurysm consist of an interposition synthetic graft and primary repair of the gastrointestinal tract (4). If extensive destruction of the intestinal wall has taken place, segmental resection with end-to-end anastomosis is preferred. Aortobifemoral bypass with a synthetic graft is the most practical and preferred method for restoring limb circulation in secondary aortoduodenal fistula after the infected graft is removed.

In selected cases, endovascular technique, which is an alternative to traditional surgical treatment, is recommended. A covered stent may be an alternative in selected cases of AAAs with aorta esophageal fistula, aorta pulmonary fistula or aneurysms of the main branches of the abdominal aorta (11-13). If an infection is suspected in an aorta enteric fistula, a covered stent is not a suitable alternative for treatment (14). However, this technique may not be suitable in a patient with life-threatening bleeding.

In conclusion, in patients presenting with gastrointestinal hemorrhage and pulsatile abdominal mass, primary aorta enteric fistula should always be kept in mind, thereby improving the outcomes of the patients thus affected.

REFERENCES

1. Funaki B. Endovascular intervention for the treatment of acute arterial gastrointestinal hemorrhage. Gastroenterol Clin North Am 2002; 31: 701-13.
2. Seiferth J, Castrup HJ. Rupture of abdominal aortic aneurysm into the duodenum. Dtsch Med Wochenschr 1969; 94: 940-5.
3. Spinelli F, Mirenda F, Mandolfino T, et al. Primary aortoduodenal fistula including the afferent loop of a Billroth II anastomosis. A case report. J Cardiovasc Surg 2002; 43: 711-4.
4. Van Olffen TB, Knippenberg LH, van der Vliet JA, Lastdrager WB. Primary aorta enteric fistula: report of six new cases. Cardiovasc Surg 2002; 10: 551-4.
5. Chang MW, Chan YL, Hsieh HC, Chang SS. Secondary aortoduodenal fistula. Chang Gung Med J 2002; 25: 626-30.
6. Karam PG, Novick RJ, Glickman LT, Blundell PE. Right hemothorax: An unusual presentation of ruptured infrarenal abdominal aortic aneurysm. Can J Surg 1986; 29: 133-4.
7. Cohen JR, Faust G, Tenenbaum N, et al. The calcium messenger system and the kinetics of elastase release from human neutrophils in patients with abdominal aortic aneurysms. Ann Vase Surg 1990; 4: 570-4.
8. White JV, Hass K, Phillips S, Comerota AJ. Adventitial elastolysis is a primary event in aneurysm formation. J Vase Surg 1993; 17: 371-81.
9. Davis CA, Pearce WH, Haines GK, et al. Increased ICAM-I expression in aortic disease. J Vase Surg 1993; 18: 875-80.
10. Dvorac J. Perforation of the jejunum by an aneurysm of the abdominal aorta. Rozhl Chir 1991; 70: 469-71.

11. Shiraishi S, Watarida S, Matsubayashi K, et al. Successful management of an aorta esophageal fistula resulting from an aneurysm of the thoracic aorta with a covered stent. Cardiovasc Surg (Torino) 2002; 43: 95-8. Jpn Circ J

12. Campagna AC, Wehner JH, Kirsch CM, et al. Endovascular stenting of an aorta pulmonary fistula presenting with hemoptysis: a case report. Cardiovasc Surg (Torino) 1996; 37: 643-6.

13. Isaki H, Koizumi J, Tamura T, Miyamoto K. Covered stent implantation by the puncture method for the treatment of small aneurysm of the common iliac artery. 2000; 64: 99-102

14. Tiesenhausen K, Amann W, Koch G, et al. Endovascular stent graft infection - a life-threatening complication. Vasa 2000; 29: 147-5.