

Primary Aortoduodenal Fistula in Patients with Rheumatoid Arthritis

Romatoid Artritli Hastada Primer Aortoduodenal Fistül

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Abstract

Primary aortoduodenal fistula (PADF) develops between the aorta and duodenum in the presence of an aortic aneurysm. Although rare, it causes life-threatening conditions. A common clinical presentation is gastrointestinal bleeding. Palpable abdominal mass and abdominal pain are other clinical symptoms. Recently, computed tomography scan with intravenous contrast and endoscopy have been commonly used for diagnosis. Despite physical examination, endoscopy, and radiological evaluation, diagnosis can be challenging due to uncommon location of gastrointestinal bleeding. Here we present a case of PADF in combination with morgagni hernia and acute mesenteric ischemia in a patient with rheumatoid arthritis.

Keywords: Aortoduodenal fistula, rheumatoid arthritis, gastrointestinal bleeding

Öz

Primer aortoduodenal fistül (PADF), aort ve duodenum arasında anevrizma varlığında gelişir. Nadir bir durumdur, fakat yaşamı tehdit edici durumlara neden olur. Yaygın klinik sunumu gastrointestinal kanamadır. Ele gelen abdominal kitle ve abdominal ağrı diğer klinik semptomlardır. Günümüzde tanıda yaygın olarak intravenöz kontrastlı bilgisayarlı tomografi ve endoskopi kullanılmaktadır. Gastrointestinal kanamanın yaygın olmayan lokalizasyonu nedeniyle tanı koymak zor olabilir. Bu çalışmada, romatoid artritli hastada, akut mesenterik iskemide ve morgagni hernisi kombinasyonunda PADF olgusu sunulmuştur.

Anahtar Kelimeler: Aortoduodenal fistül, romatoid artrit, gastrointestinal kanama

INTRODUCTION

Primary aortoduodenal fistula (PADF) develops between the aorta and duodenum in the presence of an aortic aneurysm. It is a rare entity but causes life-threatening conditions. Till date, only 267 PADF cases have been reported in the literature (1). Secondary aortoduodenal fistula, which occurs after aortic surgical interventions (implantation of aortic prosthesis), has a higher incidence than PADF. The most common etiological factor for the development of PADF is abdominal aortic aneurysm. The common clinical presentation is gastrointestinal bleeding (1). This is the first report of PADF in a patient with rheumatoid arthritis.

Here we present a case of PADF in combination with morgagni hernia and acute mesenteric ischemia in a patient with rheumatoid arthritis.

CASE PRESENTATION

A 79-year-old female presented to our emergency ward with symptoms of hematemesis and abdominal pain. She was diagnosed with rheumatoid arthritis 40 years ago and she was under treatment with steroid. Physical examination showed tachycardia and hypotension. Laboratory tests were as follows: hemoglobin, 4.4 g/dL; creatinine, 3.3 mg/dL; K, 6.9 mEq/L; WBC, 6700 cells/mcl. After a three-unit erythrocyte replacement, endoscopy was performed. There was no active sign of bleeding on endoscopy. Computed tomography (CT) scan revealed occlusion of the inferior mesenteric artery and strangulated intestinal segments in the pouch of morgagni hernia (Figures 1, 2). Emergency surgery was performed with a prediagnosis of acute mesenteric ischemia. During the surgery, ischemia and necrosis were observed in the left and sigmoid colon (Figure 3). Anterior resection was performed and end stoma was created. Transverse colon redacted to the abdomen from the thorax, (hernia pouch). Resection was not required due to intact transverse colon. During the surgery, only 300 ml of dark colored blood was aspirated using a nasogastric (NG) drainage kit. The patient was transported to the intensive care unit. After 30 min, approximately 1500 mL of fresh blood was observed in the NG drainage kit. Emergency exploration was performed and PADF was diagnosed with gastrotomy and duodenotomy (Figure 4). Unfortunately, the patient died on the operating table due to irreversible hemorrhagic shock and cardiac arrest. Therefore, approval for publication could not be obtained from the patient.

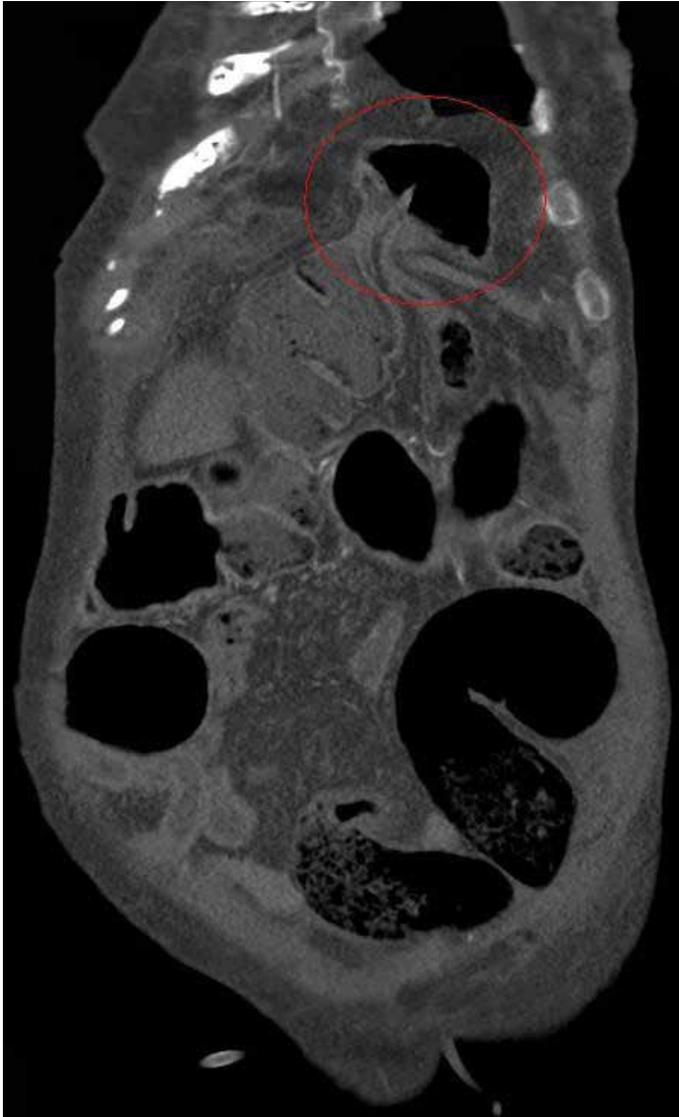


Figure 1. Strangulated intestinal segments in the pouch of morgagni hernia on computed tomography scan



Figure 3. Ischemia and necrosis in the left and sigmoid colon

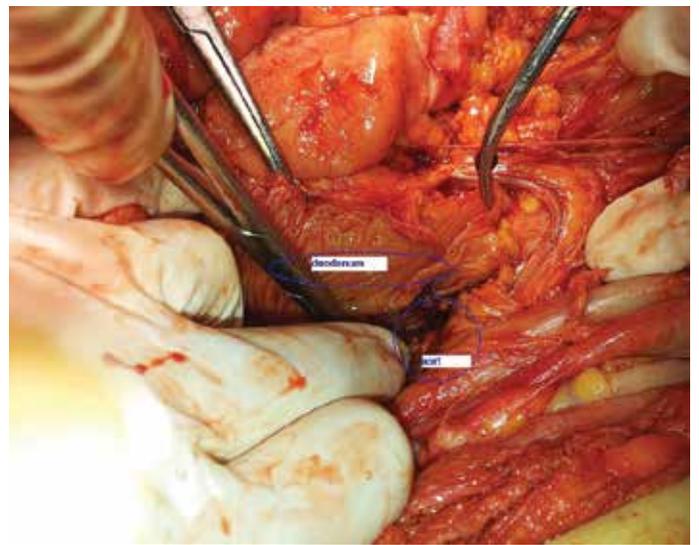


Figure 4. Primary aortoduodenal fistula between the third part of duodenum and abdominal aorta

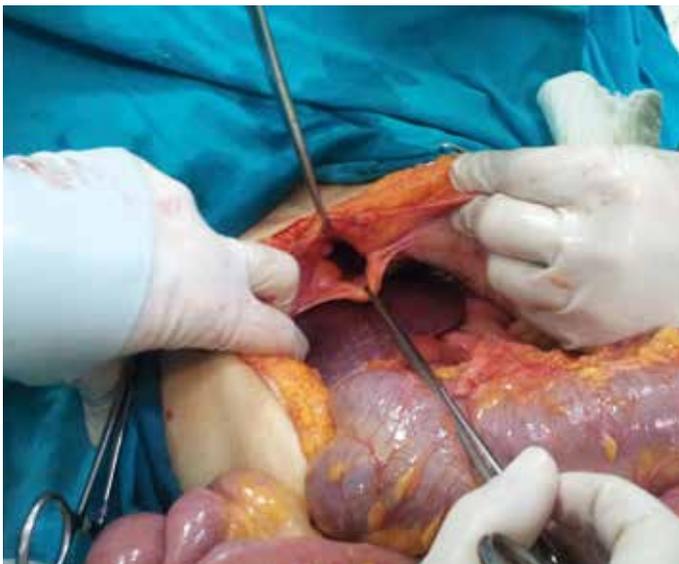


Figure 2. Strangulated intestinal segments in the pouch of morgagni hernia



Figure 5. The disappearance of the fat plane between the abdominal aorta and duodenum and aortic aneurysm on computed tomography

DISCUSSION

Primary aortoduodenal fistula is a rare clinical entity that causes massive gastrointestinal bleeding. The incidence of PADF is 0.04%–0.07% (2). The most common etiological factor for PADF is aortic aneurysm. Other possible factors are primary or metastatic tumors, foreign bodies, radiotherapy, and infections as (diverticulitis, appendicitis, etc) (1). Aortic aneurysm is more common in patients with rheumatoid arthritis than in the general population (3). Furthermore, long-term steroid use may induce aneurysm by inhibiting the synthesis of chondroitin sulfate or granulation and making the connective tissue fragile (4). This is the first report of PADF in a patient with rheumatoid arthritis. We hypothesized that the process of chronic inflammation in rheumatoid arthritis and steroid use may have contributed to the formation of PADF in this case.

Primary aortoduodenal fistula generally occurs between the third part of duodenum and abdominal aorta. It may occur in other regions of the gastrointestinal (GI) tract such as the esophagus, jejunum, ileum, and colon (1). Gastrointestinal bleeding is the most common clinical symptom (64%) in patients with PADF. Other symptoms are abdominal pain (32%) and palpable mass in the abdomen (25%) (5). The first bleedings are known as “herald bleeds” and they are usually mild and self-limited. After hours, days, or weeks, massive bleeding occurs and results in hemorrhagic shock (6). In this case, presentation and location of PADF was typical, but concomitant clinical entities such as acute mesenteric ischemia and strangulated morgagni hernia might hinder the preoperative diagnosis of PADF.

In the diagnosis of PADF, CT scan, endoscopy, and angiography can be used. Recently, CT scan with intravenous contrast and endoscopy have been commonly used (7). CT findings of air in the retroperitoneum, loss of aneurysmal wall, and disappearance of the fat plane between the aneurysm and duodenum indicate PADF. Pathognomonic finding of PADF is contrast leakage into the gastrointestinal tract (8). Retrospective evaluation of CT scan revealed the disappearance of the fat plane between the abdominal aorta and duodenum and aortic aneurysm (Figure 5). PADF is fatal without treatment; after surgical treatment, it has 18%–93% mortality rate (9).

Endovascular interventions may be helpful in hemodynamically stabilizing the patient (10). Current treatment approaches of PADF are en bloc resection and reconstruction with aortic graft, extra-anatomic bypass, and endovascular repair. For duodenal lesion a primary suture and jejunal patch is recommended (11).

CONCLUSION

The diagnosis of Primary aortoduodenal fistula is difficult and PADF has high mortality rate. It should be considered in the differential diagnosis of patients with massive GI bleeding and aortic aneurysm. In case of undetectable focus of GI massive bleeding in patients with rheumatoid arthritis, keeping PADF in mind may save lives.

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