Journal of
Babol University of Medical Sciences
JBUMS
e-ISSN: 2251-7170 p-ISSN: 1561-4107

A Rare Case of Acute Gastrointestinal Bleeding Following an Aortoenteric Fistula

S. M. Mousavi (MD)[1], M. Arabi (MD)[1], A. Ashrafi (MD)[1], H. Ghaedamini (MD)[*1], F. Salmanpour (MD)[2], A. M. Ghaedamini (MD)[3]

1.Department of General Surgery, School of Medicine, Ahvaz Jundishapur University of Medical Sciences, Ahvaz, I.R.Iran.

2.Student Research Committee, Ahvaz University of Medical Science, Ahvaz, I.R.Iran.

3.Student Research Committee, Tehran University of Medical Sciences, Tehran, I.R.Iran.

•**Corresponding Author: H. Ghaedamini (MD)**

Address: Department of General Surgery, Golestan Hospital, Mofid Street, Ahvaz, I.R.Iran.

Tel: +98 (61) 33204539. **E-mail:** Ghaedaminih@gmail.com

Article Type

Case Report

Received:
Sep 3rd 2023
Revised:
Nov 25th 2023
Accepted:
Dec 11st 2023

ABSTRACT

Background and Objective: Aortoenteric fistula (AEF) is a rare but life-threatening condition. Secondary AEF in patients with a history of aortic surgery with symptoms of sudden gastrointestinal bleeding may occur even without evidence in endoscopy and colonoscopy, and delay in diagnosis and lack of timely surgery may lead to the death of the patient, and it is important to consider it. This study introduces a 60-year-old man with a history of aortic surgery who referred to the Golestan Hospital in Ahvaz with a complaint of acute gastrointestinal bleeding and underwent surgery with the diagnosis of secondary AEF.

Case Report: The patient is a 60-year-old man who had a history of Aortobifemoral Bypass surgery three years ago, and was hospitalized in the emergency room due to gastrointestinal bleeding (melena). The patient's heart rate was 110 beats per minute, the patient's systolic blood pressure was 90 mm Hg and diastolic blood pressure was 60 mm Hg (positive tilt test) and the patient's hemoglobin (HB) was 6 g/dl. The patient underwent fluid resuscitation by the gastroenterology service, received pantoprazole, blood and antibiotics, endoscopy and colonoscopy, but no pathological findings were found. A surgical consultation was requested for him, and the patient underwent emergency CT angiography of the abdominal vessels. Due to the prolonged bleeding, unstable hemodynamics and drop in hemoglobin, it was decided to operate the patient with the suspicion of AEF. During the operation, the aorta (in the renal part) was fistulated to the intestines (jejunum, 30 cm from Treitz ligament) through a 5 mm long tract, which was repaired. The day after the operation, the patient was conscious and had stable vital signs, and was discharged the following week with a good general condition. The patient was followed up monthly for one week, three weeks, and then up to six months, which showed normal conditions.

Conclusion: Based on the results of this study, AEF should be considered as a rare but fatal diagnosis in a patient with a history of aortic surgery and lower gastrointestinal bleeding and should undergo emergency surgery.

Keywords: *Vascular Fistula, Intestinal Fistula, Gastrointestinal Bleeding, Iran*.

Cite this article: Mousavi SM, Arabi M, Ashrafi A, Ghaedamini H, Salmanpour F, Ghaedamini AM. A Rare Case of Acute Gastrointestinal Bleeding Following an Aortoenteric Fistula. *Journal of Babol University of Medical Sciences*. 2024; 26: e64.

[DOI: 10.22088/jbums.26.1.64]

Copyright © 2024 Babol University of Medical Sciences. Published by Babol University of Medical Sciences. This work is licensed under a Creative Commons Attribution-NonCommercial 4.0 International license (https://creativecommons.org/licenses/by-nc/4.0/). Noncommercial uses of the work are permitted, provided the original work is properly cited.

Introduction

A fistula from the aorta to the intestines (Aortoenteric Fistula= AEF) is a rare but life-threatening disease. The annual incidence of this disease is about 0.008 per million. This complication occurs in both primary and secondary forms (1). Primary Aortoenteric Fistula (PAEF) is created directly, while Secondary Aortoenteric Fistula (SAEF) occurs after repair of Abdominal Aortic Aneurysm (AAA) or reconstruction of aorta and iliac vessels (2-4). The incidence rate of SAEF after aortic surgical reconstruction varies from 0.36% to 1.6% (5). The incidence rate of AEF is higher in men compared to women due to the fact that the rate of AAA and aortic surgery is higher in men. The male-to-female ratio for PAEF is three to one and for SAEF it is eight to one (6).

Most primary and secondary cases of AEF occur in the third and fourth segments of the duodenum, where the duodenum is closest to the aorta (1). The results showed that other places such as the colon can also be rarely involved (about 5%). On the other hand, early and timely diagnosis of AEF is very difficult and depends on many factors (3). The clinical consequences of this complication are determined according to the timely surgical repair, the revascularization approach used, the type of surgery (emergency versus non-emergency) and the complications (4).

In a study on 22 patients with AEF, Sieber et al. showed that the most common manifestations include gastrointestinal bleeding, abdominal pain, and fever. Ten patients underwent emergency surgery, who were older than others and had more risk factors for respiratory diseases. The death rate of these people was 27%. The results show that despite the rapid diagnosis and treatment of this complication, AEF is still a life-threatening condition (1).

Jheasha et al. reported a 64-year-old man with AEF who had previously undergone aortobifemoral bypass surgery. This patient was referred with symptoms of fever, gastrointestinal bleeding, fatigue and shortness of breath and underwent emergency surgery and responded well to the treatment. Research results show that since the most common manifestations of AEF are caused by a previous surgery, gastrointestinal bleeding and sepsis, delay in surgical treatment can lead to the death of the patient (2).

Rütmann et al. reported a 63-year-old man with a history of abdominal aortic surgery 5 years ago who referred with complaints of abdominal pain and hematemesis and was diagnosed with AEF and underwent emergency surgery. The results of the research show that this case is very rare and misleading and a multifaceted approach, including sufficient expertise of the surgeon, appropriate special care, antibacterial treatment with long-term monitoring is necessary for a favorable short-term and long-term outcome for the patient (5).

The results showed that AEF with sudden lower gastrointestinal bleeding with possible gastric or intestinal origin can occur even without any evidence in endoscopy and colonoscopy. Although this complication is rare, delay in its diagnosis can be associated with increased morbidity and mortality. Therefore, it is always important to pay attention to this issue in people with a history of aortic surgeries with severe gastrointestinal bleeding, and in many cases, they should undergo emergency surgery. This study was conducted with the aim of introducing a 60-year-old man with a history of previous femoral aorta surgery, who referred to the emergency department of Golestan Hospital in Ahvaz with a complaint of acute and severe gastrointestinal bleeding and underwent surgery with the diagnosis of secondary AEF.

Case Report

This research was approved by the ethics committee of Ahvaz Jundishapur University of Medical Sciences with code IR.AJUMS.REC.1402.465. The patient is a 60-year-old man who referred to the internal

[DOI: 10.22088/jbums.26.1.64]

emergency department of Golestan Hospital with a complaint of blood in stool (melena, four times widely since the day before admission). The patient has had a history of bilateral iliac artery stenosis since 4 years ago, coronary artery stenosis since 7 years ago, and asthma since 5 years ago. He underwent Coronary Artery Bypass Grafting (CABG) six years ago. In addition, he had undergone Aortobifemoral Bypass three years ago due to bilateral iliac artery stenosis. He used aspirin 80 mg daily and salbutamol spray (2 puffs every 12 hours) regularly. The patient had no history of smoking or drug use. He did not mention any history of previous gastrointestinal bleeding. When entering the emergency room, the patient's heart rate was 110 beats per minute, the patient's systolic blood pressure was 90 mmHg while lying down and 70 mmHg while sitting, and the patient's diastolic blood pressure was 60 mmHg while lying down and 50 mmHg (positive tilt test). In the examinations, the patient was in a lethargic state (confused and drowsy with impaired attention to time and place). In the abdominal examination, only scars from previous surgeries were observed on the patient's chest and abdomen. Melena was observed in the rectal examination. The patient's tests at admission included hemoglobin= 6 g/dl and hematocrit= 29% (Table 1).

First, the patient was hospitalized by the gastroenterology service and underwent fluid resuscitation, pantoprazole infusion, blood transfusion and antibiotic treatment, and emergency endoscopy was performed. Multiple erosions were seen in the first part of the duodenum (D1) and there was no evidence in favor of bleeding. Also, the patient underwent colonoscopy, the only finding of which was internal hemorrhoid grade I. Due to the prolonged bleeding and lack of recovery, surgical consultation was requested for the patient. In addition to the management of gastrointestinal bleeding by this service, the patient underwent emergency CT angiography of the abdominal vessels based on the opinion of vascular surgery professors; the image of the previous graft site of the femoral aorta surgery was seen in a Y shape. In the anterior wall of the distal aorta, there was a focal outpouching with dimensions of 9×8 mm, which was above the anastomosis site, and indicated the presence of a penetrating wound of the aorta (Figure 1).

Table 1. Laboratory findings of the patient

Variable	Number
White Blood Cell count (mcl)	7900
Hemoglobin (g/dl)	6
Hematocrit	29%
Platelet	143
International Normalized Ratio (INR)	1

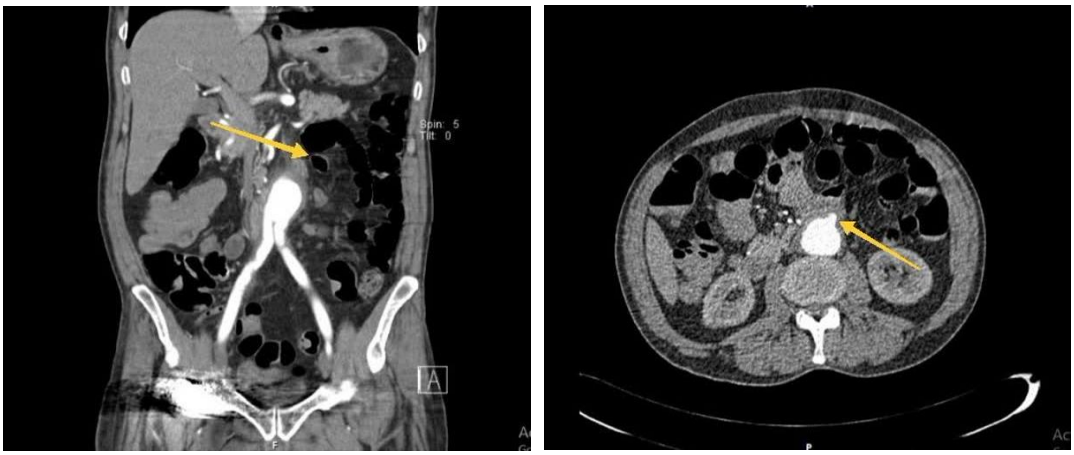


Figure 1. CT angiography of the patient in two transverse and coronal views and suspected aortoenteric fistula

[DOI: 10.22088/jbums.26.1.64]

In the third part of the duodenum, which passed in front of the aorta and adjacent to the wound, no aortic contrast extravasation or definitive evidence of an aortoenteric fistula (aorta to intestine) was detected, but it was still suspected. Due to the prolonged bleeding, unstable hemodynamics and drop in hemoglobin, the decision was made to operate the patient with the suspicion of fistula from the previous site. During the operation, there was a very strong adhesion of the intestines to each other and to the abdominal aorta at the site of the previous graft. After enterolysis, vascular control was taken from the place where the renal artery was separated from the abdominal aorta. The intestines were gently released over the aortic graft, and the aortic graft (in the renal area) was fistulated to the intestines (jejunum, 30 cm from Treitz ligament) through a 5-mm tract (Figures 2, 3).

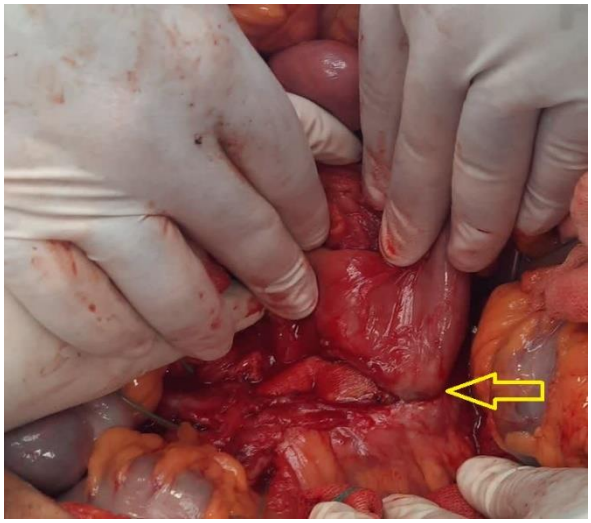


Figure 2. Intraoperative findings of the patient and presentation of the location of the aortoenteric fistula

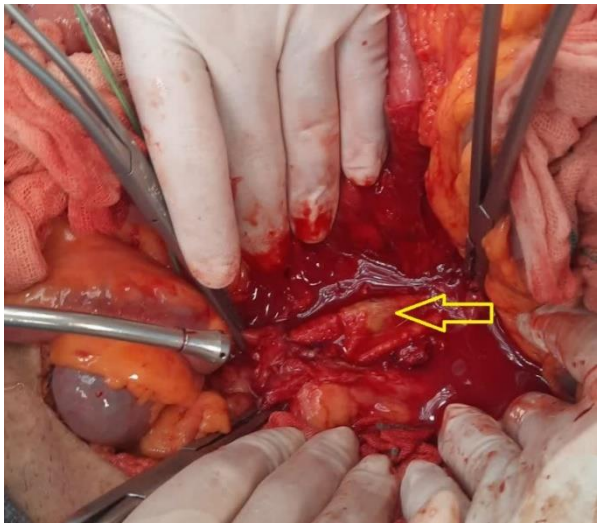


Figure 3. Intraoperative findings of the patient and presentation of the previous graft caused by aortobifemoral bypass

[DOI: 10.22088/jbums.26.1.64]

The intestinal perforation site was repaired and the aortic perforation site was repaired and patched with omentum (the omentum was placed over it as a protector) (Figure 4) and the surgery was successfully completed. A few hours after the operation, the patient was transferred to the intensive care unit of Golestan Hospital. The day after the operation, the patient was conscious and had stable vital signs, and there was no evidence of bleeding or hemoglobin drop. He was discharged a week later with a good general condition and necessary recommendations. One week later, he went to the vascular surgery clinic of Golestan Hospital on an outpatient basis, where he was examined and the examination results were normal. Three weeks later, he came again and was evaluated for infection, bleeding and the health of the digestive tract. The next month, the patient was followed up with a CT angiography of the abdominal vessels, which was normal, and after that, abdominal and pelvic ultrasounds were requested for him monthly for six months, the results of which were also normal.

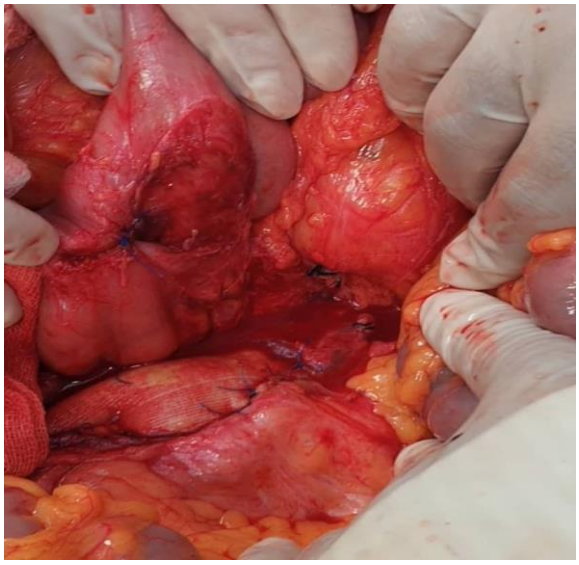


Figure 4. The image after fistula repair (repair of the fistula and intestine and placing a patch between them)

Discussion

In the present study, the age of the introduced participant was 60 years (elderly age group based on the classification of the World Health Organization), which is in line with the results of the studies of Sieber et al. (1), Jheasha et al. (2) and Rüütman et al. (5). Moreover, in reference books, the common age of this complication is 61 years and above (6-8). It is possible that the consistency of vascular and intestinal tissues decreases with age and the chance of fistula formation increases. Therefore, it is necessary to pay attention to the elderly group in terms of the possibility of AEF.

AEF should always be considered as one of the most important differential diagnoses of a rare but fatal condition in patients who refer with a history of aortic surgery and uncontrollable lower gastrointestinal bleeding. Failure to perform surgery on time will endanger the patient's life, which was mentioned in this research. The results showed that the gender of the present participant was male, which was consistent with the results of the studies of Jheasha et al. (2) and Rüütman et al. (5). In reference books, it is reported that the prevalence of this disease is higher in men than in women, the cause of which has not yet been determined (6-8). Therefore, it is suggested to carry out additional research on this topic.

[DOI: 10.22088/jbums.26.1.64]

The clinical symptoms of the presented patient were extensive lower gastrointestinal bleeding (melena) and hemorrhagic shock. According to the results by Sieber et al. (1), the most common manifestations include gastrointestinal bleeding, abdominal pain, and fever. In a study by Jheasha et al., the patient was presented with symptoms of fever, gastrointestinal bleeding, fatigue and shortness of breath (2). Rüütman et al. reported abdominal pain and hematemesis as common symptoms (5). Moreover, in reference books (6-8) and other studies (9-11), the most common clinical manifestations are extensive lower gastrointestinal bleeding, abdominal pain, and hemorrhagic shock, which is caused by rapid outflow of blood from the aorta to the interior of the intestinal lumen. If this is not considered, it causes hemodynamic instability and death of the patient. Other rare symptoms of AEF include fever and sepsis, pulsatile abdominal mass, groin mass, retroperitoneal abscess, limb ischemia, abdominal pain, back pain, and weight loss, but the presented patient did not have these symptoms (10).

The results of the present study showed that the patient had a history of aortofemoral surgery, which is consistent with the results of Sieber et al. (1), Jheasha et al. (2), Rüütman et al. (5), reference books (6-8) and other articles (9-11). A history of abdominal vascular surgery raises the suspicion of AEF because it is difficult to diagnose considering its rarity. The most common risk factors for AEF include aortic or iliac artery aneurysms and previous aortic intervention or surgery. Moreover, other intra-abdominal processes that cause inflammation, infection, or mechanical erosion of the aorta and adjacent structures can also lead to AEF (5). Considering this case, it is necessary for surgeons to cover the prosthesis with an aneurysm sac or retroperitoneum to prevent fistula during the operation (12). In this case, three years had passed since the aortobifemoral bypass operation. Fistula formation can happen at any time after the operation, and no specific time frame has been defined for it, and its cause is often related to the techniques used by the surgeon, the type of graft, and adhesions after the operation.

CT angiography, endoscopy, colonoscopy, and full abdominal and pelvic ultrasound are often used to diagnose AEF, which is consistent with the results of Sieber et al. (1), Jheasha et al. (2), and Rüütman et al. (5). In the present study, CT angiography of abdominal vessels, endoscopy and colonoscopy were used for diagnosis, but no evidence was found in favor of AEF. This is a challenging issue per se. Therefore, young surgeons and internal specialists should always pay attention to the fact that if the clinical conditions of a patient with extensive gastrointestinal bleeding do not improve, despite the normality of other paraclinical procedures (such as endoscopy, colonoscopy, and CT angiography), the probability of fistula is high in a patient with a history of vascular surgery. On the other hand, the incidence of fistula from the aorta to the small intestine is higher than in other areas, such as the stomach, colon, and pancreas, and there is limited information about its survival rate, which is necessary to be considered (6).

If AEF is not treated correctly and on time, the patient's life will be in danger. The patient introduced in the present study underwent emergency surgery, which was consistent with the results of Jheasha et al. (2) and Rüütman et al. (5). According to reference books, the treatment of AEF includes proper resuscitation of the patient, treatment with antibiotic therapy, and emergency repair of the aortic fistula site (6-8), and in the present study, all of the above were done for the patient. According to reference books, the surgical method (open versus endovascular) for AEF repair is considered based on clinical discretion, type of AEF, time of occurrence, comorbidities of the patient, and individual aortic anatomy (10-12). In this study, due to the significant bleeding of the patient, poor clinical conditions, unstable vital signs, medical co-morbidities, poor functional status, as well as the history of CABG and aortobefemoral, and the suspicion of the possible existence of SAEF, open surgery was agreed upon.

[DOI: 10.22088/jbums.26.1.64]

In general, AEF in the present patient is a rare but life-threatening complication caused by the previous aortofemoral bypass, which is diagnosed based on clinical, radiological and paraclinical manifestations. Early diagnosis of this complication is difficult and depends on the surgeon's clinical suspicion. Surgical management for optimal outcomes is dependent on various factors. The death rate of this disease is very high. It is recommended that surgeons pay attention to the possibility of AEF in elderly patients presented with gastrointestinal bleeding and a history of previous vascular surgery, if gastrointestinal bleeding is not controlled despite the normality of paraclinical procedures including endoscopy, colonoscopy, and CT angiography of abdominal vessels.

Acknowledgment

Hereby, the Deputy of Research and Technology of Jundishapur University of Medical Sciences, Ahvaz, for supporting the research, as well as the patient who consented to publish his condition in the form of a research, and the respected personnel of the Radiology and Medical Records Department of Golestan Hospital, Ahvaz, are gratefully acknowledged.

[DOI: 10.22088/jbums.26.1.64]

References

1.Sieber S, Busch A, Sargut M, Knappich C, Bohmann B, Karlas A, et al. A Modern Series of Secondary Aortoenteric Fistula - A 19-Year Experience. Vasc Endovascular Surg. 2024;58(2):185-92.

2.Abu Jheasha AA, Ashhab M, Dukmak ON, Maraqa M, Emar M, Jubran F, et al. Digestive hemorrhage and fever as a result of a double secondary aortoenteric fistula following the repair of a juxtarenal abdominal aortic aneurysm and an infection of the aortobifemoral bypass graft: a case report. Ann Med Surg (Lond). 2023;85(8):4053-9.

3.Freitas BD, Dionisio A, Ferreira B, Azevedo S, Araújo I. Aortoenteric Fistula: A Differential Diagnosis of Anemia. Cureus. 2023;15(8):e42952.

4.Saito H, Nishikawa Y, Akahira JI, Yamaoka H, Okuzono T, Sawano T, et al. Secondary aortoenteric fistula possibly associated with continuous physical stimulation: a case report and review of the literature. J Med Case Rep. 2019;13(1):61.

5.Rüütmann AM, Kals J. Primary and secondary aortoenteric fistulas in a patient with abdominal aortic aneurysm. Int J Surg Case Rep. 2023;107:108344.

6.Rimoldi P, Lista A, Occhiuto MT, Cuccì A, D'Alessio I, Tolva VS. Major Complications of Vascular Surgery. In: Aseni P, Grande AM, Leppäniemi A, Chiara O, editors. The High-risk Surgical Patient, 2nd ed. New York: Springer; 2023. p. 499-510.

7.Ochoa C, Manzur MF. Complications of Vascular Surgery. In: Murga A, Teruya TH, Abou-Zamzam Jr AM, Bianchi C, editors. The Vascular Surgery In-Training Examination Review (VSITE), 3rd ed. New York: Springer; 2023. p. 465-81.

8.Levine JL. Upper Gastrointestinal Bleeding. In: Petrone P, Brathwaite CEM, editors. Acute Care Surgery in Geriatric Patients, 3rd ed. New York: Springer; 2023. p. 423-30.

9.Marques I, Vieira M, Ferreira A, Abreu R, Rodrigues R, Vilaça L, et al. Emergent Endovascular Repair For Primary Aortoenteric Fistula. Port J Card Thorac Vasc Surg. 2023;30(2):63-6.

10.Jenab Y, Hosseini K, Rezaee M. Aortoenteric fistula in a man with history of endovascular aneurysm repair that presented with abdominal pain and anaemia. Eur Heart J Case Rep. 2023;7(4):ytad136.

11.Narayanan A, Hanna J, Okamura-Kho A, Tesar J, Lim E, Peden S, et al. Management of secondary aorto-enteric fistulae: a multi-centre study. ANZ J Surg. 2023;93(10):2363-9.

12.Jiang C, Chen X, Li J, Li H. A case report of successful treatment of secondary aortoenteric fistula complicated with gastrointestinal bleeding and retroperitoneal abscess in an elderly patient. Medicine (Baltimore). 2018;97(24):e11055.

[DOI: 10.22088/jbums.26.1.64]