

Spontaneous aortoenteric fistula involving the sigmoid: A case report and review of literature



Eleonora G. Karthaus, Ivo C.J.H. Post, George J.M. Akkersdijk*

Department of Surgery, Spaarne Gasthuis, Hoofddorp, The Netherlands

ARTICLE INFO

Article history:

Received 6 August 2015
Received in revised form
12 December 2015
Accepted 12 December 2015
Available online 17 December 2015

Keywords:

Primary enteral fistula
Hartmann
Aortic aneurysm
Ruptured aneurysm

ABSTRACT

INTRODUCTION: Primary aortoenteric fistula (PAEF) is a pathological communication between the aorta and any portion of the gastrointestinal tract. The pathology is very rare and easily overlooked during the diagnostic process.

PRESENTATION OF CASE: We report the exceptional case of an 86-year-old man with episodes of abdominal pain and rectal bleeding of unknown cause over a period of 1,5 months due to a PAEF to the sigmoid. A sigmoidectomy was performed and a rifampicin-soaked aortic graft was placed. The patient had an uneventful post-operative recovery. The duration of symptoms, the anatomic location of the fistula and the outcome after surgery makes this case unique.

DISCUSSION: With an incidence of 0.04–0.07% in all patients with aortic aneurysms a PAEF is very rare. Only 2% of PAEF's involves the sigmoid. The most common cause is an atherosclerotic aortic aneurysm. Patients with PAEF can present with a triad of symptoms including gastrointestinal bleeding, abdominal pain and a pulsating mass. A contrast-enhanced computer-tomography scan (CTa) is the most accurate tool to demonstrate a PAEF. Without a strong clinical suspicion, diagnosing a PAEF is hard and frequently delayed. The overall PAEF-related mortality is high (61–100%) and decreases after surgery (30–40%).

CONCLUSION: A primary aortoenteric fistula involving the sigmoid is very rare. Clinical presentation can vary, diagnosis can be difficult and surgical options may differ. Even with low suspicion of PAEF, we recommend performing a CTa. With a high overall mortality of more than 60% due to exsanguinating, surgical treatment is always indicated.

© 2015 The Authors. Published by Elsevier Ltd. on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

1. Introduction

In 1829 Sir Astley Cooper was the first to describe a primary aortoenteric fistula (PAEF), a potential complication of abdominal aortic aneurysms [1]. PAEF is a pathological communication between the aorta and any portion of the bowel. While secondary fistulas after aortic repair are relatively common (0.4–1.6%), PAEF is a very rare complication of abdominal aneurysms with incidences ranging from 0.04 to 0.07% [2–5]. While most cases of PAEF concern involvement of the third or fourth part of the duodenum, in unique cases other portions of the bowel may also be affected [4,6]. In this report we present an unlikely case of a primary fistula from an aortic aneurysm to the sigmoid Fig. 1.

2. Case presentation

An 86-year old male presented to the emergency room with acute rectal bleeding with hemodynamic instability, an estimated

blood loss of 500cc and cramping abdominal pains. The only medical antecedent was a 5.2 cm infrarenal abdominal aortic aneurysm, which had been stable for the past two years. The aneurysm was not painful on palpation. The bleeding had stopped spontaneously while the patient was at the emergency room. During hospital admission a colonoscopy was performed, which revealed no abnormal findings. Although not seen at colonoscopy, the bleeding was attributed to a diverticular haemorrhage or a haemorrhoid. After a 4-day observation and a blood transfusion (2 packed cells) the patient was discharged to follow-up in good health. Three weeks later, the patient was readmitted with rectal bleeding, this time without hemodynamic instability or other symptoms. A surgeon was consulted and an ultrasound of the abdomen was obtained to rule out acute aortic pathology. After a brief observation the patient could be discharged without intervention. However, after five weeks the patient was readmitted with rectal bleeding and severe hemodynamic instability. A CTa revealed peri-sigmoidal fat infiltration adjacent to the abdominal aortic aneurysm. At laparotomy the thickened sigmoid adherent to the aneurysm, just above the level of the bifurcation, was confirmed.

Controlling the infrarenal aorta and iliac vessels, the thickened sigmoid was dissected from the aneurysm. A small quantity

* Corresponding author at: Spaarnepoort 1, 2134TM, Hoofddorp, The Netherlands.
E-mail address: gakkersdijk@spaarnegasthuis.nl (G.J.M. Akkersdijk).

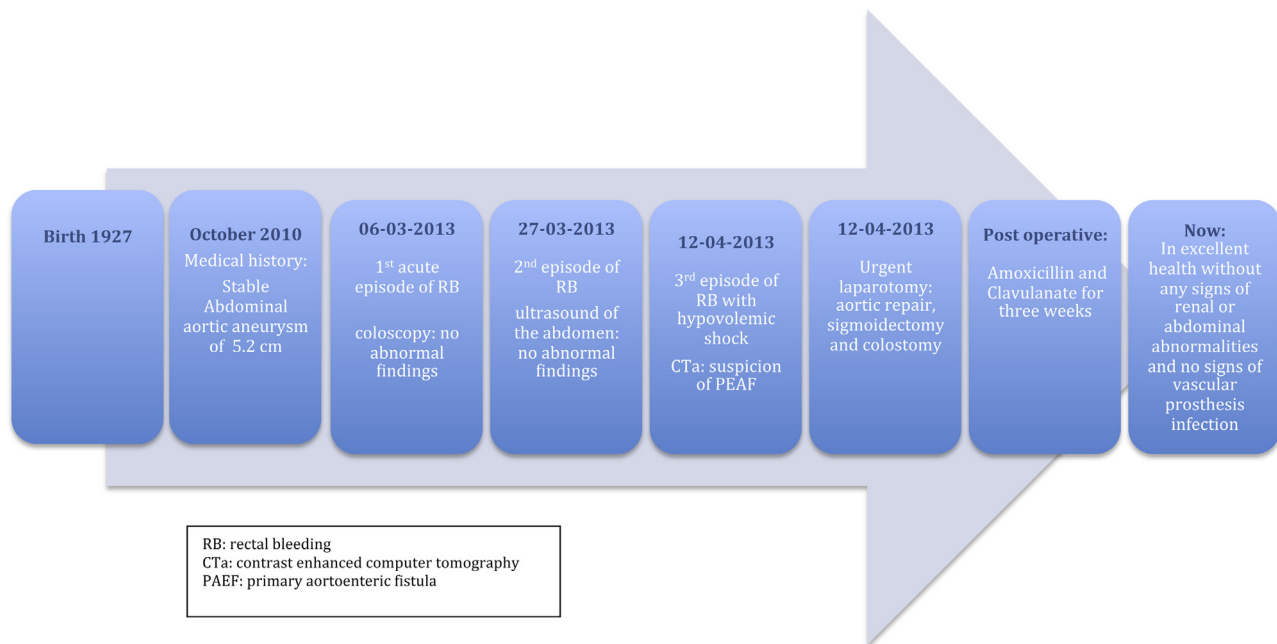


Fig.1. Timeline.

of pus was discharged followed by major bleeding from a 2 cm primary aorta-sigmoid fistula. Following clamping of the aorta, a sigmoidectomy was performed. After extensive peritoneal irrigation, a rifampicin soaked 18–9 mm Dacron infra-renal aorto-bi-iliac bifurcation graft was sutured. The retroperitoneum was closed after rifampicin irrigation with a Gentamicin mesh covering the prosthesis. Finally a colostomy was administered.

The patient had an uneventful post-operative recovery and was treated with amoxicillin and clavulanate for three weeks according to culture results (*Bacteroides fragilis*, sensitive for amoxicillin and clavulanate). Histologic examination of the sigmoid revealed a local erosive perforation, without signs of diverticular disease. Two years after the event, the patient was confirmed to be in excellent health without any signs of renal or abdominal abnormalities and no signs of vascular prosthesis infection.

3. Discussion

Primary aortoenteric fistulas are described to have an incidence of approximately 0.04–0.07% in patients with an aortic aneurysm. More than half of the PEAf's affect the third and fourth duodenal section, and involvement of the sigmoid is extremely rare, which can be explained by its fixed retroperitoneal position and close relation to the aorta and superior mesenteric artery. Approximately one-third of the aortic-enteric fistulas involve the esophagus and in 2% the stomach. Of the reported cases, 15% involves the small bowel and colon, of which only a handful reported (2% of all PEAf's) involvement of the sigmoid [6,7].

Before 1970 infectious diseases like tuberculosis and syphilis caused the majority of PEAf's. With effective treatments now available, infectious etiologies have become rare [3]. Currently most aortoenteric fistulas occur secondary after open abdominal aortic aneurysm repair, mainly aorto-duodenal fistulas. These are called secondary aortoenteric fistulas. However, PEAf's that are more seldom seen than secondary aortoenteric fistulas, are mostly caused by an atherosclerotic aortic aneurysm [8]. In a previously published report 83% of PEAf occurred in the presence of an aortic aneurysm [3]. Other causes of PEAf's include tumors, radiotherapy, diverticulitis, ulcers and foreign body ingestion [7,9–14]. Regarding our

patient, the fistula was most likely the result of spontaneous erosion of the aortic aneurysm causing chronic inflammation enabling aorto-enteric fixation.

Patients with PEAf can present with a classical triad of symptoms, known as Cooper's triad, which includes gastrointestinal bleeding, abdominal pain and a pulsating abdominal mass [3]. However, only 11% of patients shows this classical triad of symptoms [2,3]. Other clinical manifestations can be intermittent back pain, syncope, shock and weight loss [3]. Fever and sepsis are also possible but rarely seen [15,16]. In 94%, a gastrointestinal bleeding is seen at presentation. The time-interval between a first gastrointestinal bleeding and massive bleedings varies from hours to months. In 30% of the patients this interval is less than 6 h, in 50% less than 24 h and in 71% less than a week [3]. For our patient this interval was about 1,5 months, notably longer.

Patients with gastrointestinal bleeding are usually evaluated with a gastroduodenoscopy. However, the detection rate for PEAf with gastroduodenoscopy is 25% [3]. Arteriography has a comparable low detection rate of 26%, whereas colonoscopy and ultrasound have even lower rates, respectively 10% and 0% [4,16]. With a detection rate of 61%, a contrast-enhanced CTa is the most reliable diagnostic tool for the evaluation of a PEAf. Signs that are suggestive for PEAf are the presence of contrast within the gastrointestinal tract, air within the aneurysm wall, bowel wall thickening adjacent to the aneurysm and destruction of the fat cover surrounding the aneurysm [4,8,16]. Besides to the bowel wall thickening proximately to the aneurysm, these other typical findings were not seen in our patient. In retrospect, the use of a CTa earlier in the diagnostic process of this patient with rectal bleeding and a history of abdominal aortic aneurysm could have been indicated. Also an explorative laparotomy would have been justified in this case with long standing symptoms.

Sears et al. looked for abnormalities in laboratory tests and found an elevated ESR and C-reactive protein in the majority of patients, when compared to patients scheduled to undergo elective AAA repair [6]. In our patient C-reactive protein was normal at presentation (6 mg/L).

For the patients that can undergo surgery before they exsanguinate, different surgical treatments have been suggested. These

treatments may vary from simple closure of the defect to extra anatomical bypasses. Most reports give preference to graft insertion and subsequent disconnection and closure of the fistula [2,3,6]. However, in the case of mass contamination several studies have suggested an extra-anatomical bypass [4,5,7,8]. Before 1991 only 32 successful operations for PEAf had been described [17]. In 2005 Sears and Scheltinga described a surgical mortality rate of 30–40% over the last decades [3]. However the overall mortality from PEAf is between 61 and 100% [4]. With these statistics in mind one may advocate for an explorative laparotomy when there is a high suspicion of a PEAf but unable to be demonstrated with diagnostics tools.

4. Conclusion

Primary aortoenteric fistulas are a rare phenomena, occurring in 0.04–0.07% of patients with an aortic aneurysm. In only 2% of these cases the sigmoid is involved. Though rare, the classical presentation includes a triad of gastrointestinal bleeding, abdominal pain and a pulsating abdominal mass. In patients with an abdominal aortic aneurysm, presenting with rectal bleeding without abnormalities on endoscopy, suspicion of a PEAf should be raised. In such cases, the performance of a CTA is essential and should not be omitted, since it is the most accurate diagnostic tool to demonstrate the presence of an aortoenteric fistula. Although the operative mortality of this condition is high, (emergency) laparotomy, (anatomic or extra-anatomic) aortic aneurysm repair and resection of the affected intestines are indicated.

Conflict of interest

All authors have no conflict of interest to declare. In addition, there was no outside financial support for this research.

Sources of funding

All authors have no sources of funding to declare.

Ethical approval

Not applicable, considering it is a case report For this case report on a patient with a primary aortoenteric fistula's we obtained a written informed consent from the concerning patient.

Consent

For this case report on a patient with a primary aortoenteric fistula's we obtained a written informed consent from the concerning patient. All specific patient information is de-identified.

Author contribution

Eleonora G. Karthaus: study concept/design, data collection, data analysis/interpretation, writing the paper Ivo C.J.H. Post: study

concept/design, data collection, data analysis/interpretation, writing the paper George J.M. Akkersdijk: study concept/design, data collection, data analysis/interpretation, writing the paper.

Guarantor

George JM Akkersdijk MD, PhD, Spaarnepoort 1, 2134TM, Hoofddorp, The Netherlands, +31 23 890 7432, gakkersdijk@spaanegasthuis.nl.

References

- [1] A. Cooper, The Lectures of Sir Astley Cooper on the Principles and Practice of Surgery with Additional Notes and Cases by F Tyrell, 5th edn, Haswell, Barrington and Haswell, Philadelphia, 1939.
- [2] R. Voorhoeve, F.L. Moll, J.A. de Letter, T.J. Bast, J.P. Wester, P.H. Slee, Primary aortoenteric fistula: report of eight new cases and review of the literature, *Ann. Vasc. Surg.* 10 (January (1)) (1996) 40–48, PubMed PMID: 8688296.
- [3] S.J. Saers, M.R. Scheltinga, Primary aortoenteric fistula, *Br. J. Surg.* 92 (February (2)) (2005) 143–152, PubMed PMID: 15685700.
- [4] W. Lee, C.M. Jung, E.H. Cho, D.R. Ryu, D. Choi, J. Kim, Primary aortoenteric fistula to the sigmoid colon in association with intra-abdominal abscess, *Korean J. Gastroenterol.* 63 (April (4)) (2014) 239–243, PubMed PMID: 24755749.
- [5] D.O. Kavanagh, J.F. Dowdall, F. Younis, S. Sheehan, D. Mehigan, M.C. Barry, Aorto-enteric fistula: changing management strategies, *Irish J. Med. Sci.* 175 (January–March (1)) (2006) 40–44, PubMed PMID: 16615228.
- [6] S.J. Saers, M.R. Scheltinga, J.M. Salemans, P.J. Kitslaar, Gastroenterological aspects of primary aorto-enteric fistulae (PAEF), *Acta Chir. Belg.* 109 (November–December (6)) (2009) 741–745, PubMed PMID: 20184059.
- [7] S.E. Wilson, M.L. Owens, Aortocolic fistula, a lethal cause of lower gastrointestinal bleeding: report of a case, *Dis. Colon Rectum.* 19 (October (7)) (1976) 614–617, PubMed PMID: 1086196.
- [8] C.W. Lee, S.W. Chung, S. Song, M.J. Bae, U. Huh, J.H. Kim, Double primary aortoenteric fistulae: a case report of two simultaneous primary aortoenteric fistulae in one patient, *Korean J. Thoracic Cardiovasc. Surg.* 45 (October (5)) (2012) 330–333, PubMed PMID: 23130309, PubMed Central.
- [9] N.C. Armitage, K.C. Ballantyne, Primary aortoenteric fistula due to recurrent colorectal cancer. report of a case, *Dis. Colon Rectum.* 33 (February (2)) (1990) 148–149, PubMed PMID: 2298102.
- [10] O. Drogitz, J. Pfeifferberger, W. Schareck, U. Adam, H. Nizze, U.T. Hopt, Primary aortoduodenal fistula as a late complication of para-aortic radiation therapy: a case report, *Der Chirurg: Zeitschrift für alle Gebiete der operativen Medizen* 73 (June (6)) (2002) 633–637, PubMed PMID: 12149951. Primäre aortoduodenale Fistel als Spatkomplikation nach paraaortaler Radiatio. Ein Fallbericht.
- [11] J.L. Grosfeld, K. Eng, Right iliac artery-duodenal fistula in infancy: massive hemorrhage due to whisk-broom bristle perforation, *Ann. Surg.* 176 (December (6)) (1972) 761–764, PubMed PMID: 4540101. PubMed Central.
- [12] J.H. Huang, J.S. Chen, S.H. Chu, K.M. Chiu, Recurrent cervical carcinoma presenting as a primary aortoduodenal fistula, *Ann. Vasc. Surg.* 24 (November (8)) (2010) e5–e7, PubMed PMID: 20800434.
- [13] R.D. Odze, L.R. Begin, Peptic-ulcer-induced aortoenteric fistula. Report of a case and review of the literature, *J. Clin. Gastroenterol.* 13 (December (6)) (1991) 682–686, PubMed PMID: 1761841.
- [14] N. Sevastos, P. Rafailidis, K. Kolokotronis, K. Papadimitriou, G.V. Papatheodoridis, Primary aortojunal fistula due to foreign body: a rare cause of gastrointestinal bleeding, *Eur. J. Gastroenterol. Hepatol.* 14 (July (7)) (2002) 797–800, PubMed PMID: 12169993.
- [15] S. Ramanujam, A. Shiels, G. Zuckerman, C. Prakash, Unusual presentations of aorto-enteric fistula, *Gastrointest. Endosc.* 59 (February (2)) (2004) 300–304, PubMed PMID: 14745412.
- [16] W. Ranasinghe, J. Loa, N. Allaf, K. Lewis, M.G. Sebastian, Primary aortoenteric fistulae: the challenges in diagnosis and review of treatment, *Ann. Vasc. Surg.* 25 (April (3)) (2011) e1–e5, PubMed PMID: 21269801.
- [17] A. Ihaya, Y. Chiba, T. Kimura, K. Morioka, T. Uesaka, R. Muraoka, Primary aortosigmoid fistula treated by descending thoracic aortofemoral bypass, *J. Cardiovasc. Surg* 41 (April (2)) (2000) 325–327, PubMed PMID: 10901546.

Open Access

This article is published Open Access at sciendo.com. It is distributed under the [IJSCR Supplemental terms and conditions](#), which permits unrestricted non commercial use, distribution, and reproduction in any medium, provided the original authors and source are credited.