



Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Primary aorto-enteric fistula



Andrew C. Gordon*, Mayank Agarwal

Croydon University Hospital, London, United Kingdom

ARTICLE INFO

Article history:

Received 8 October 2015

Received in revised form

12 December 2015

Accepted 13 December 2015

Available online 17 December 2015

Keywords:

Gastrointestinal haemorrhage

Aorto-duodenal fistula

Retroperitoneal metastasis

ABSTRACT

INTRODUCTION: Primary aorto-enteric fistula (PAEF) is a life threatening, spontaneous erosion and communication of the aorta and intestinal tract. Unlike secondary AEF, which occur following aortic surgery, they are extremely rare. The low clinical suspicion and difficulty in obtaining a definitive diagnosis make for a dismal prognosis.

CASE PRESENTATION: A literature review highlighted aetiology which included gallstone erosion, carcinoma of the pancreas and duodenal diverticulum. With written consent, we present the case of a 59 year old female, brought to the hospital following an episode of haematemesis and later found to have an AEF, secondary to metastatic retroperitoneal carcinoma - an extremely rare aetiology.

DISCUSSION: There is far less literature on primary AEF when compared to secondary AEF. Furthermore, there is a variation in aetiology. Identifying the presence of a 'herald' bleed appears to be significant.

CONCLUSION: Aorto-enteric fistulae must always be considered as a potential diagnosis in the setting of an acute upper GI haemorrhage with no apparent cause.

© 2015 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

Primary aorto-enteric fistula (PAEF) is a life threatening complication, defined as a spontaneous erosion and communication of the abdominal aorta into the intestinal tract [1]. They are distinguished from secondary AEF, which occur following aortic surgery; the latter being far more prevalent [1]. Aorto-duodenal fistulae (ADF) are the most common type (~60%) owing to the close relationship of the duodenum (in particular D3/4 which is tethered to the retroperitoneum), with the underlying abdominal aorta. They usually present as a massive gastrointestinal haemorrhage, and a diagnosis is often delayed owing to the rarity of the condition. The low clinical suspicion is compounded by the difficulty in obtaining a definitive diagnosis, making for a dismal prognosis.

The aetiology of PAEF varies, but is commonly a result of pathological changes within the aorta, such as atherosclerotic or mycotic aneurysms [2,3]. Less frequent causes include gallstone erosion [4], carcinoma of the pancreas [5] and duodenal diverticulum [6].

In this report, we present the case of a 59 year old female who was brought to the emergency department following an episode of haematemesis and was found to have an AEF, secondary to metastatic retroperitoneal carcinoma.

Verbal and written, signed consent was gained from the patient's husband (next of kin), to publish an anonymous report on the findings of this case.

1.1. Case report

A 59 year old Afro-Caribbean female, with previous total abdominal hysterectomy (endometrial cancer) and recent partial gastrectomy for secondary malignancy, presented to the emergency department (ED) with one episode of haematemesis and ongoing severe abdominal pain. Although there was no report of previous haematemesis or haematochezia, she was being investigated for ongoing chronic anaemia; for which both previous oral gastroduodenoscopy (OGD) and colonoscopy were inconclusive. There was no history of abdominal aortic aneurysms (AAA).

Shortly after presentation, her haemoglobin level on arterial blood gas was 5.4 g/dL and she proceeded to have another prolonged episode of haematemesis. She received two units of packed red cells, and one unit of platelets and fresh frozen plasma, before being taken for an urgent OGD, as per the consensus of the ED, anaesthetic and surgical teams. At endoscopy, a massive active upper GI bleed was evident, but the source was not found, and Adrenalin injections were futile. She was transferred to the hospital's high dependency unit for full resuscitation and inotropic management.

Despite this, she continued to deteriorate and, 8 h on from admission, she underwent an emergency exploratory laparotomy. Intra-operative haemostatic control proved very challenging, despite receiving over 20 units of packed red cells intraoperatively (with accompanying blood products to avoid clotting disorders). The source of bleeding was not identified until the surgeons moved into the fourth part of the duodenum (D4), where a pulsatile mass and abnormal communication were palpable. At this point, the

* Corresponding author.

E-mail address: andrewg2910@googlemail.com (A.C. Gordon).

clinical picture was indicative of an AEF and the decision was taken to stabilize and limit blood loss as best as possible, considering the patient's current state, with a view to returning for definitive surgery if and when formal imaging could be obtained. Despite aggressive management, the patient deteriorated further post operatively and passed away several hours later, in the intensive care unit.

At post mortem, the cause of death was confirmed as a massive GI haemorrhage secondary to an aorto-duodenal fistula. A retroperitoneal mass, measuring, $6 \times 5 \times 3$ mm, perforated into D4; intraoperative samples sent for histology and immunohistochemical testing were found to be consistent with a poorly differentiated carcinoma, likely small cell in origin. It transpired this lady had been in remission for uterine malignancy, for which she had previously undergone a total hysterectomy and partial gastrectomy (for metastatic disease).

All work reported in line with the CARE criteria guidelines [7].

2. Discussion

Since its first description in 1829 [8], PAEF remains a rare finding; one study reported incidence rates of 0.04–0.07 at autopsy, whilst Yao and Pierce found less than 200 reported PAEF in the literature at time of publication. Moreover, by far the commonest aetiology abdominal aortic aneurysms (atherosclerotic and mycotic), with metastatic disease accounting for less than 1% of reported cases [9]. Classically, upper gastrointestinal haemorrhage (64%), abdominal pain (32%) and a palpable abdominal mass (25%) form the triad of an AEF [10], but as Saers and Scheltinga found, this is only true in ~11% of cases [11]. Less common symptoms include back pain, melena, fever, or syncope. Other subtle points are very useful and if identified early on, can prevent an acute deterioration. One such example is the 'herald' bleed, which temporarily seals off through thrombus formation, but can precede a lethal exsanguination. In the present case, the patient was not actively bleeding on arrival, but did report an episode of haematemesis approximately two hours prior to arrival. However, a review by Steffes and O'Leary found that in 29% of cases, the time between initial bleed and death was greater than one week, highlighting the significance of recognising this sign in the management and prognosis of AEF [12]. Aside from the obscurity of an AEF, the modality of investigations used to confirm the diagnosis is debatable. In the majority of cases, which present with a major GI haemorrhage, OGD will be the first line investigation. Although, as we found in the present case, this is often inconclusive. A recent review of the literature found that computed tomography (CT) was the modality of choice in this scenario, as opposed to OGD or angiography [11]. It concluded that, in patients with AAA, a negative OGD suggests an AEF. However, in the case presented herein, there was no evidence of an AAA, and the diagnosis was clouded by a previous partial gastrectomy. Furthermore, patients are not always haemodynamically suitable, at time of presentation, to undergo a CT.

In untreated cases of AEF, the mortality rates is almost 100%, with the only definitive treatment being surgical. However, several techniques are available, and are broadly categorized into endovascular grafting and the 'classical' extra-anatomical repair, with the former now being the favoured approach. Several studies and literature reviews have identified much higher mortality rates in patients undergoing extra-anatomical bypasses as opposed to in-situ repair [13,14]. Furthermore, Flye and Thomson found that delaying surgical intervention until after a massive GI haemorrhage increases mortality to ~74% [15].

Although the literature on the precise aetiology of this case report is scarce, it is clear that AEF must always be considered, and actively ruled out in the setting of GI haemorrhage. This particu-

lar case reinforces why it is especially so in patients with anaemia of unknown cause, and previous malignancy, presenting with an upper GI haemorrhage. This is a potentially lethal clinical phenomenon, which albeit, is relatively rare, but one where early diagnosis and prudent management can have huge effects on prognosis.

Conflicts of interest

No conflicts of interest.

Funding

None.

Ethical approval

None.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Andrew Gordon

- Dealt directly with case.
- Writing bulk of report.

Mayank Agarwal

- Oversaw case.
- Edited report.

Guarantor

Andrew Gordon.

References

- [1] F. Puccio, G. Pandolfo, S. Chiodini, F. Benzi, M. Solazzo, Primary aorto-duodenal fistula as a late complication of radiotherapy: report of a case and review of the literature, *Case Rep. Gastroenterol.* 2 (September (3)) (2008) 415–423, <http://dx.doi.org/10.1159/000164621>, Epub 2008 Nov 14. PubMed [citation] PMID: 21897792, PMCID: PMC3166804.
- [2] J.P. Reckless, I. McColl, G.W. Taylor, Aorto-enteric fistulae: an uncommon complication of abdominal aortic aneurysms, *Br. J. Surg.* 59 (June (6)) (1972) 458–460, PubMed [citation] PMID: 4537432.
- [3] S. Pagni, R.W. Denatale, T. Sweeney, C. McLaughlin, A.M. Ferneini, Primary aorto-duodenal fistula secondary to infected abdominal aortic aneurysms: the role of local debridement and extra-anatomic bypass, *J. Cardiovasc. Surg. (Torino)* 40 (February (1)) (1999) 31–35, PubMed [citation] PMID: 10221382.
- [4] N.R. Broadbent, D.E. Taylor, Gallstone erosion of the aorta, *Aust. N. Z. J. Surg.* 45 (May (2)) (1975) 207–208, PubMed [citation] PMID: 1059408.
- [5] A.D. Ghaphery, R. Gupta, R.A. Currie, Carcinoma of the head of the pancreas with aortoduodenal fistula, *Am. J. Surg.* 111 (April (4)) (1966) 580–583, PubMed [citation] PMID: 5910668.
- [6] M.F. Adinolfi, W. Hardin, M.D. Kerstein, Aortic erosion by duodenal diverticulum: an unusual aortoenteric fistula, *South Med. J.* 76 (August (8)) (1983) 1069–1070, PubMed [citation] PMID: 6879282.
- [7] CARE Guidelines for case reports (2013). Retrieved from <http://www.care-statement.org/>.
- [8] J.S. Gelister, J.A. Fox, Primary aortoenteric fistula, *J. R. Soc. Med.* 80 (July (7)) (1987) 459–460, PubMed [citation].
- [9] Lemos, DW, et al., Primary aortoduodenal fistula: a case report and review of the literature, *J. Vasc. Surg.* 37 (3) (2003) 686–689, journal.

- [10] M.S. Sweeney, T.R. Gadacz, Primary aortoduodenal fistula: manifestation, diagnosis, and treatment, *Surgery* 96 (September (3)) (1984) 492–497, PubMed [citation] PMID: 6236571.
- [11] S.J. Saers, M.R. Scheltinga, Primary aortoenteric fistula, *Br. J. Surg.* 92 (February (2)) (2005) 143–152, Review. PubMed [citation] PMID: 15685700.
- [12] B.C. Steffes, J.P. O’Leary, Primary aortoduodenal fistula: a case report and review of the literature, *Am. Surg.* 46 (March (3)) (1980) 121–129, PubMed [citation] PMID: 6966478.
- [13] P. Goshtasby, D. Henriksen, C. Lynd, L.P. Fielding, Recurrent aortoenteric fistula: case report and review, *Curr. Surg.* 62 (November–December (6)) (2005) 638–643, Review. PubMed [citation] PMID: 16293501.
- [14] F.S. Lozano, et al., Primary aortoduodenal fistula: new case reports and a review of the literature, *J. Gastrointest. Surg.* (February) (2008), journal.
- [15] M.W. Flye, W.M. Thompson, Aortic graft-enteric and paraprostatic-enteric fistulas, *Am. J. Surg.* 146 (August (2)) (1983) 183–187, PubMed [citation] PMID: 6224432.

Open Access

This article is published Open Access at sciedirect.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.