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Life-threating upper gastrointestinal bleeding due to a primary aorto-jejunal fistula

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ABSTRACT

INTRODUCTION: Primary aorto-enteric fistula (AEF) is an uncommon life-threating condition. Only 4% of them involve the jejunum or ileum and its mortality ranges from 33 to 85%.
PRESENTATION OF CASE: A 54-year-old female was admitted to the Emergency Department with syncope and hematemesis. The esophagogastroduodenoscopy found a pulsatile vessel in the second portion of the duodenum. A computed tomography scan showed an AEF with an infrarenal aortic aneurysm and iliac artery thrombosis. During surgery, an infrarenal aortic aneurysm complicated with an aorto-jejunal fistula was found. An axilo-bifemoral bypass, open repair of the aneurysm and segmental small bowel resection with primary suture of the jejunal defect were performed.
DISCUSSION: Depending on previous aortic grafting, AEF can be classified as primary or secondary. Primary AEF is usually caused by an untreated abdominal aortic aneurysm, commonly presenting an infectious etiology. The main clinical sign is a "herald" hemorrhage. The EGD is considered as the first step in diagnosing AEF. The treatment of choice for AEF is emergent surgery. Use of broad-spectrum antibiotics is mandatory in the postoperative period to avoid fistula recurrence.
CONCLUSION: AEF is a rare entity with a high mortality. High clinical suspicion is essential to make a correct diagnosis, which is crucial for the prognosis of these patients, such is the case of our patient. If hemodynamic stability is achieved, it allows to employ surgical strategies in which extra-abdominal bypass is performed before fistula is treated.

1. Introduction

Primary aorto-enteric fistula (AEF) is an extremely unusual disease, with an incidence less than 1%, according to autopsy studies in general population [1,2]. More than 80% of them involve the duodenum, mainly in the third and fourth portions [1]. The jejunum and ileum are only affected in 4% of cases [3]. Mortality ranges from 33 to 85%, being an early diagnosis the most important prognostic factor [1,4,5,6].

Our aim is to report a case of a female affected by an aorto-jejunal fistula, presenting with an upper gastrointestinal bleeding.

2. Presentation of case

A 54-year-old female patient was admitted to the Emergency Department with syncope and an episode of hematemesis. Previous medical reports were unremarkable, the only outstanding features were a smoking habit of 34 packs/year and 16 g/day of alcohol consumption.

Upon arrival, the patient was hemodynamically stable and asymptomatic. Physical examination was anodyne. Full blood examination showed an hemoglobin of 10.2 mg/dl (two months before was 14 mg/dl). Coagulation parameters were normal.

A cranial computed tomography (CT) scan was performed with no pathological findings. Initial esophagogastroduodenoscopy (EGD) revealed plenty of traces of blood in the stomach with uncomplicated hiatal hernia, no active bleeding was found.

In-hospital observation for 72 h revealed no hemodynamic instability regardless of several stools compatible with melenae. The hemoglobin level was stable. An EGD that was performed after 48 h of admission did not show any traces of blood nor

Abbreviations: AEF, aorto-enteric fistula; EGD, esophagogastroduodenoscopy; CT, computed tomography; PTFE, polytetrafluoroethylene.

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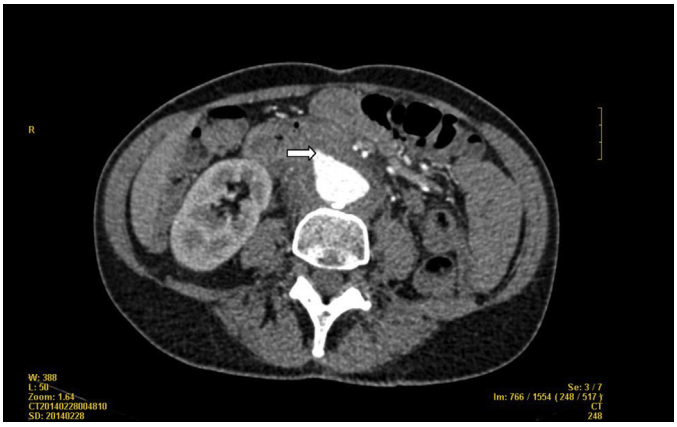

Fig. 1. Axial CT of the abdomen with intravenous contrast in arterial phase demonstrating an AEF associated with an infrarenal calcified fusiform aneurysm.

other pathological features. Being asymptomatic, the patient was discharged.

The patient was readmitted 2 days later due to syncope. Upon arrival she presented hypotension (blood pressure 90/40 mmHg) and a heart rate of 92 bpm. Fluid replacement was started, gaining hemodynamic stability. Hemoglobin was 8.4 mg/dl, requiring transfusion of 2 units of packed red blood cells. The patient presented a new episode of melenae.

A new EGD was performed, finding a pulsatile vessel in the second portion of the duodenum, with no active bleeding. An irregular fusiform aortic aneurysm with maximum diameters of 63 × 41 × 54 mm was found on CT scan imaging. The aneurysm contacted with the third duodenal portion and extended itself toward the iliac bifurcation, containing a 1 cm mural thrombus in the right iliac artery. A duodenal AEF originating from the aneurysm could be seen within a distance of 4.3 cm from the origin of both renal arteries (Fig. 1).

The patient underwent an urgent laparotomy. An inflammatory infrarenal aortic aneurysm was found. Ligation of both common iliac arteries and infrarenal aorta was performed. Vascular perfusion was assured with an axilo-bifemoral bypass using a polytetrafluoroethylene (PTFE) prosthesis. Finally, resection of the aortic-jejunal fistula was performed with subsequent primary suture repair of the defect (Figs. 2–4, ,).

During the procedure, the patient required low dose vasoactive drugs and transfusion of 5 additional units of packed red blood cells. The patient was admitted to the Intensive Care Unit postoperatively, presenting a favorable evolution.

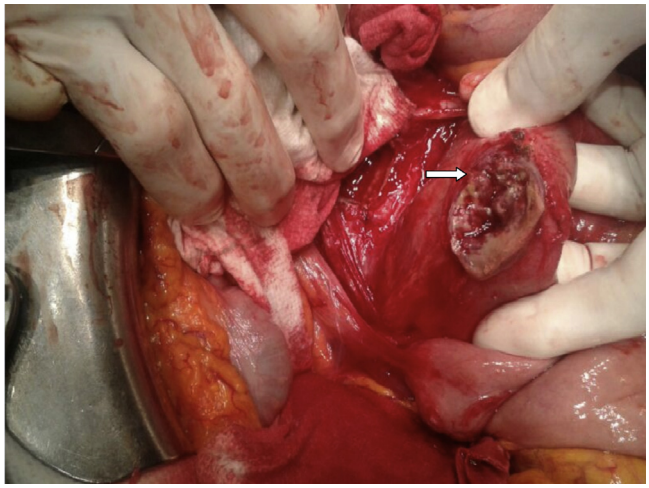

Fig. 2. External view of the affected jejunum loop after en bloq resection of the aneurysmatic aortic defect was carried out.

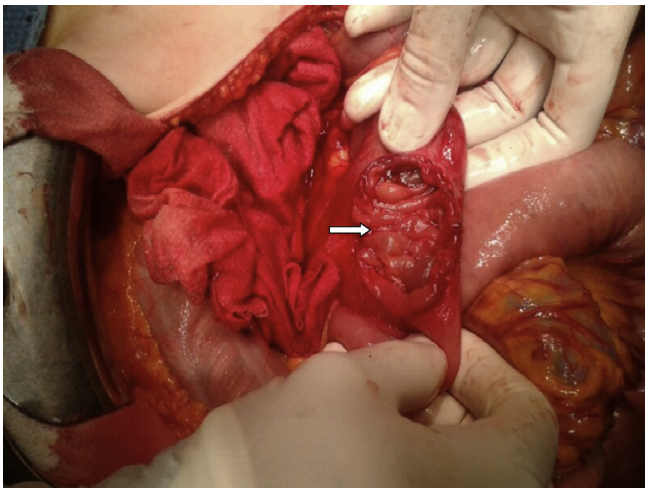

Fig. 3. Open jejunum after segmental wedge resection of the affected segment was performed.

Being hemodynamically stable and asymptomatic, the patient was transferred to the ward on postoperative day 6.

She was discharged 11 days after the procedure without signs of active bleeding and with a stable hemoglobin analysis. The pathology report of the surgical specimen identified an AEF with no presence of pathogens. The culture was positive for gram positive aerobic flora, *Streptococcus viridians* and coagulase-negative *Staphyloccocus*, being treated with Piperacillin/Tazobactam for 4 weeks.

Follow-up CT scan 1.5 months later revealed no evidence of recurrent AEF. The patient remains in good condition with no further gastrointestinal bleeding.

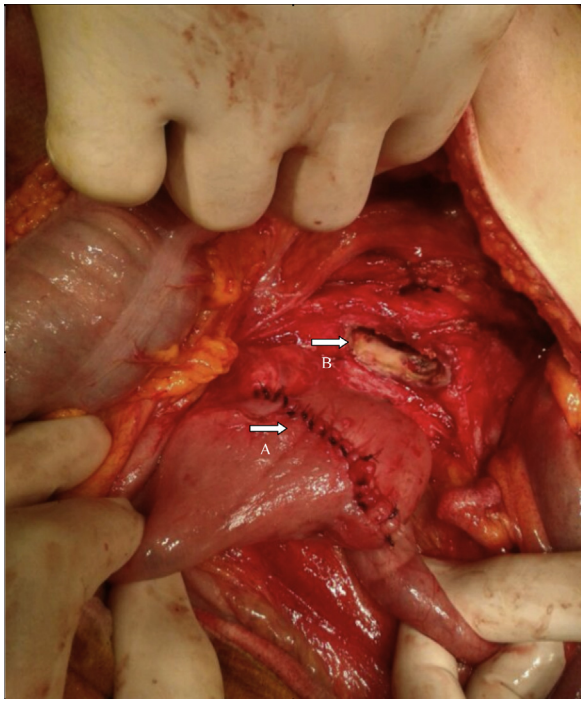

Fig. 4. Primary suture of the jejunal defect (A). The aneurysmatic aortic defect still remains opened in the picture, showing the intern fistulous orifice (B).

3. Discussion

Fistula formation between the aorta and the intestinal tract was first described in 1829 by English surgeon Sir Astley Cooper [7].

AEF can be classified as primary, regarding a spontaneous communication between the lumen of an aortic aneurysm and an intestinal loop without previous grafting, or secondary, in patients with aneurysms who have undergone reconstruction procedures [8,9]. Before 1960, primary AEF were the most commonly encountered. Over the past three decades, the prevalence of secondary AEF has risen, with an incidence of 4% [2]. The prevalence of primary AEF is 0.1–0.8% over all aortic abdominal aneurysms [8].

Primary AEF is usually caused by an untreated abdominal aortic aneurysm [2]. This ensues atherosclerosis as the most frequent etiology of primary AEF. Other causes are carcinoma, ulcers, gallstones, diverticulitis, appendicitis and foreign bodies [1].

Although the physiopathology remains uncertain, primary AEF is thought to be caused by the erosion of the digestive tract by the continuous beating of the aneurysmatic wall, which causes an ischemic effect, creating a communication between both structures [10].

The typical clinical picture presents with the triad of gastrointestinal bleeding, abdominal pain and pulsatile mass. This is not very common and it appears in less than 40% of cases [11,12]. This makes the diagnosis extremely challenging. As seen in our patient, the main clinical sign is a "herald" hemorrhage, resulting from of a small fistula tamponaded by thrombus formation [13]. Other signs that can be present are sepsis or hypovolemic shock.

Acute clinical suspicion is essential for making a prompt diagnosis, required for successful outcome [5,6]. The initial diagnostic procedure of choice is controversial, however EGD is considered by most authors as the first step in diagnosing AEF [2,9]. Although its sensitivity and specificity are very low, 38% and 18–50%, respectively, it is a useful tool to exclude other causes of bleeding [7,14,15]. In our case, the first two EGD were not useful, but the third one suggested the diagnosis. CT scan is a non invasive test that can confirm the clinical or endoscopic suspicion. Some authors favor it as the first diagnostic test for a bleeding with uncertain origin [16]. In our patient, the CT scan confirmed the diagnosis, although no diagnostic test was able to predict the level of the fistula. Percutaneous angiography may be considered but is rarely of value [2]. In many cases, the definitive diagnosis is done in the operating theater [16,17].

The treatment of choice for AEF is urgent surgical repair [7,9,18]. Resection of the aneurysm and vascular perfusion with an axilobifemoral bypass was traditionally the technique of choice [9]. If the hemodynamic situation allows it, nowadays, trends stand up for in situ reperfusion. Several authors defend that it has less morbidity and mortality than the classical technique. Regarding digestive tract repair, usually a primary suture of the defect is sufficient [7,14].

Another important point in the management of AEF is antibiotic therapy. Postoperative broad-spectrum antibiotic treatment is mandatory for one week, if the culture is negative, and for 4–6 weeks, if the culture is positive. Despite correct antibiotic treatment, postoperative infectious complications and fistula recurrence are not unusual [7,9,14,16,19]. Our patient was treated with Piperacillin/Tazobactam for 4 weeks and she has not shown fistula recurrence until the present time.

4. Conclusion

In conclusion, AEF is a rare entity whose correct diagnosis is usually not easy, but it will be crucial for the prognosis of these patients. If hemodynamic stability is achieved, extra-abdominal bypass performed before fistula treatment will be a good and safe surgical option, which makes possible a good outcome, such as in the present case.

Conflict of interest

The authors declare that there is no conflict of interest in undertaking this article.

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Ethical appoval

Nothing to declare.

Aurhor contribution

Elena Fernández de Sevilla – writing the paper, design, data collection.

Juan Andrés Echeverri – writing the paper.

Miriam Boqué – drafting the article and revising critically.

Silvia Valverde – data collection.

Nuria Ortega – data colection.

Ana Gené – data collection.

Nivardo Rodríguez – drafting the article ad revising it critically. Attending surgeon.

José María Balibrea – drafting the article and revising it critically. Attending surgeon.

Manel Armengol – drafting the article and revising it critically. Final approval.

Guarantor

Elena Fernández de Sevilla.

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.

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