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Case Report

Aortojejunal Fistula on Healthy Aorta Due to Jejunal Diverticulum

Della Schiava N[1*], Cazauran JB[2,3], Lermusiaux P[1,3] and Millon A[1,3]

[1]Department of Vascular and Endovascular, University Hospital of LYON, France

[2]Department of Hepato-biliary and Pancreatic Surgery, University Hospital of Lyon, France

[3]Claude Bernard Lyon 1 University, Lyon, France

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***Corresponding author:**

Della Schiava Nellie, Department of Vascular and Endovascular, University Hospital of LYON, Hôpital Edouard Herriot, France, Tel: +33-4-72-11-78-30 ; +33-4-72-11-76-76, Fax: +33-4-72-11-75-57, E-mail: dellaschiava.nellie@neuf.fr; nellie.della-schiava@chu-lyon.fr

1. Abstract

Aortoenteric Fistula (AEF) is a rare cause of massive gastrointestinal bleeding. Primary AEF are rare and generally the consequence of an aortic aneurysm. The duodenum and esophagus are the main locations of AEF. Diagnosis is difficult and patients are often hemodynamically unstable at the time of management because of a long delay between hospital admission and diagnosis. Yet, surgery remains possible. Endovascular techniques yield lower morbi-mortality rates. We present the first case of primary aortojejunal fistula on non-aneurysmal aorta treated efficaciouslyby stent graft placement. Gastroenterologists must be aware of this etiology to reduce the delay to diagnosis and improve survival rates.

2. Keywords: Primary aortojejunal fistula; Jejunal diverticulum; Endovascular repair; CT scan; Gastrointestinal endoscopy

3. Introduction

Aorto-Enteric Fistula (AEF) is a rare cause of massive intestinal bleeding. It generally occurs as a complication of previous aortic surgery [1].

Primary Aorto-Enteric Fistula (PAEF) occurs on native aorta with a reported incidence of 0.04-0.07% in large autopsy series [2, 3]. It predominantly occurs as a complication of an Abdominal Aortic Aneurysm (AAA) (incidence varies between 61% and 98%) with compression and erosion of the fixed parts of the adjacent bowel tract, such as the esophagus and duodenum [4, 5]. Fistula can also occur with other parts of the gastrointestinal tract but incidence is lower [5]. Other causes of PAEF with healthy aorta are rare [5-7].Whatever the cause, PAEF leads to massive gastrointestinal tract bleeding and is often lethal. The diagnosis is difficult because endoscopy often shows bleeding, but not the cause of the bleeding, even when endoscopy is repeated. Because PAEF is such a rare cause of gastrointestinal bleeding, gastroenterologists do not systematically consider this cause and do not perform other exams, such as Computed Tomography (CT). Moreover, CT can fail to depict active bleeding in a number of cases [8]. Thus, the diagnosis may be delayed, and patients are often hemodynamically unstable at the time of management, with the result that mortality rates remain high. Yet, surgical repair is possible and with new endovascular techniques, peri-operative morbidity and mortality rates have decreased significantly [9, 10].

4. Case Report

We present the case of a Primary Aortojejunal Fistula (PAJF) on healthy aorta without any

Abdominal Aortic Aneurysm (AAA) that was successfully treated by endovascular stent graft, despite a long delay between the patient's hospital admission and diagnosis.

A 68year old patient with a history of hypertension and no surgical antecedents was brought to our emergency department with a twelve-hour history of melena and dizziness. A first episode of isolated melena had occurred 15 days previously, and the patient had received antibiotics. First physical examination showed a hemodynamically stable patient with soft abdomen and no evidence of infection. Biology results reported a serum hemoglobin level of 6.5 g/dL without any other abnormality. Transfusion of 2 packs of Red Blood Cells (RBC) was initiated.

Major melena recurrence occurred 3 hours after admission and was followed by hemorrhagic collapse and brief, reversible circulatory arrest motivating transfer of the patient to the intensive care unit, with infusion of noradrenaline, and transfusion of 6 packs of RBC and 5 units of fresh frozen plasma.

A first emergency gastro duodenal endoscopy revealed blood in the duodenum but no cause of bleeding.

First CT and one repeat gastro duodenal endoscopy did not reveal any active bleeding and no sign of intra-abdominal sepsis. CT diagnosed sinuous aorta but no aortic aneurysm.

After a new hemorrhagic collapse the next day requiring another massive transfusion, another CT examination finally revealed active bleeding at the left infra-renal aortic wall with extravasation of iodine into the first jejunum (Figure 1).

The patient was immediately transferred to the operating theatre and effectively treated by aortic infra-renal stent graft placement (C3 Excluder®, GORE Medical, Flag staff, AZ, USA). Peri-operative arteriography confirmed the aortojejunal fistula (Figure 2).

Postoperative CT showed no sign of remaining bleeding and efficient stent placement without leakage (Figure 3).

The patient received intravenous antibiotics (glycol peptide and ticarcillin) to prevent potential peritonitis and stent graft infection.

The third endoscopy, using a duodenoscope, showed a first jejunum diverticulum just after the duodeno-jejunal angle. No remaining hemorrhagic or inflammatory signs were found by a baby-endoscope used for internal exploration (Figure 4).

Follow up reported several complications (spontaneously resolving hemiplegia following collapse and low flow, deep vein thrombosis and pneumonia) but none related to the surgical procedure. The pa-

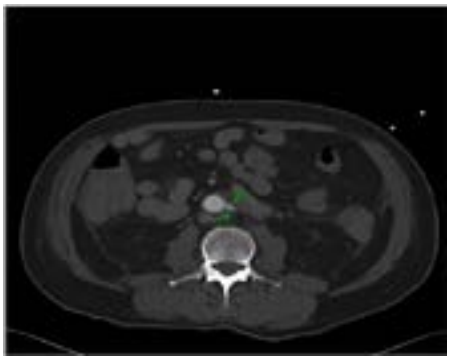

Figure 1: Computed tomography scan showing extravasation of iodine on the left aortic wall.

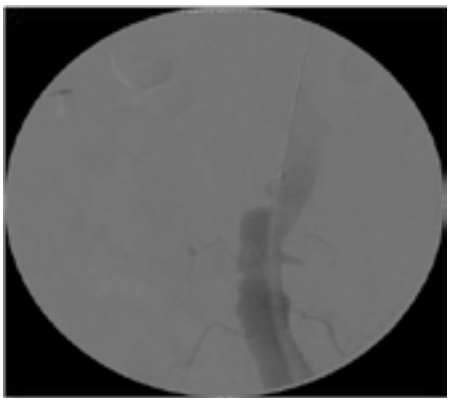

Figure 2: Extravasation of iodine on the left aortic wall during peri-operative angiography

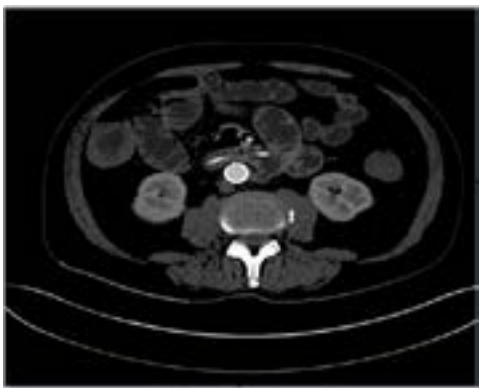

Figure 3: Post-operative CT shows the efficacy of the endoprosthesis

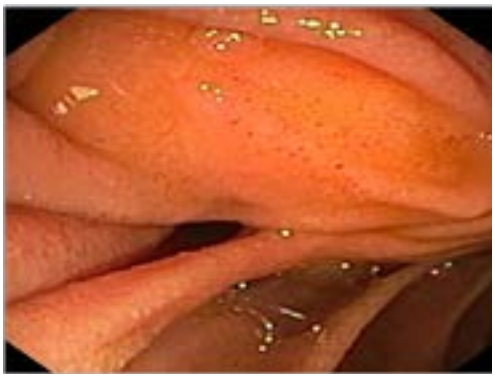

Figure 4: Post-operative endoscopy: jejunal diverticulum without any remaining sign of

tient was discharged from intensive care unit 10 days after surgery and from hospital two months after surgery.

Three month follow-up found no evidence of clinical sepsis or recurrent bleeding and the PET-CT found no sign of endovascular stent graft infection. The antibiotics were stopped. No surgical or endoscopic procedure was performed on the diverticulum.

4. Discussion

To the best of our knowledge, this is the first reported case of a successfully treated PAJF occurring on non-aneurysmal aorta. A previous PAJF was reported on an autopsy case report [11], and several studies reported secondary aorto-jejunal fistula or PAJF with aortic aneurysm [8, 12].

As reported in the literature, AEF is a difficult diagnosis, even when an aortic aneurysm is known or discovered on first examination. AEF generally occurs on the fixed part of the bowel, most commonly on the transverse or third portion of the duodenum, followed by the other parts of the duodenum and esophagus. The mobile parts of the digestive tract such as the jejunum, ileum and sigmoid colon are involved in less than 5% of cases each, and the stomach in 2% [5].

Cases of PAEF without an underlying AAA are extremely rare. Causes include primary aortic infections (tuberculosis, syphilis and mycotic agents), trauma, penetrating duodenal ulcer, sigmoid diverticulitis, digestive tract tumors, para-aortic radiation, duodenal or esophageal diverticulum with or without infection, and swallowed foreign bodies [6, 7, 13].

In this case, aortojejunal fistula seemed to be caused exclusively by mechanical erosion by the jejunal diverticulum. The first part of the jejunum is mobile but remains in close anatomical contact with the abdominal aortic wall. This proximity could explain the progressive aortic wall erosion. As generally reported, AEF requires early recognition following the initial bleeding to prevent the stress of massive hemorrhage which, when associated with sepsis, comorbidity and major surgery, leads to poor survival rates. Specific CT and endoscopic signs are quite rare and diagnosis depends mostly on repeated procedures when an AEF is suspected. The reported rates of sensitivity for CT vary between 40% and 90%, greater than that of other diagnostic methods [8].

However, PAEF has a very low reported incidence and in patients without an underlying AAA, is rarely suspected on first approach as illustrated in our report, and this precludes rapid diagnosis.

Nonetheless, several studies have reported improved survival rates with endovascular stent graft placement in case of primary or secondary AEF. A recent review by Antoniou et al reported 5 cases of endovascular treatment of abdominal PAEF from 1990 to 2008, with only one post-operative complication represented by persistent sepsis with a follow-up between 6 and 21 months [9].

Another study reported no 30-day mortality in 15 patients treated by endovascular stent graft [14]. Risk of septic recurrence was 60% but all cases were secondary AEF with an increased risk of sepsis due to prior prosthetic material at the time of the AEF [14]. Antoniou et al found 5 secondary stent-graft infections on 23 patients who did not have evidence of infection pre-operatively [9]. Nevertheless, the endovascular approach provides rapid control of bleeding and avoids open surgery in a hemodynamically unstable patient. It yields excellent survival rates.

When associated with endovascular surgery, the necessity of surgical resection of the diverticulum could be discussed. In our case, we managed the diverticulum with antibiotics only, with satisfactory results, as also reported [15].

Aorto-jejunal fistula is a possible diagnosis of massive gastrointestinal bleeding even in the absence of AAA. Endovascular stent graft repair is possible and seems to be a safe alternative providing fast hemostasis and low morbidity-mortality rates.

It is important for gastroenterologists to be aware of this etiology of gastrointestinal bleeding, in order to improve survival rates.

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