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Digestive Hemorrhage of Rare Causes: A Case Report of Primary Aorto-duodenal Fistula in the Hepato-Gastroenterology Department of the Souro Sanou University Hospital of Bobo-Dioulasso (Burkina Faso)

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ABSTRACT

Primary aortoduodenal fistula (PADF) is a rare cause of life-threatening digestive hemorrhage. The clinical picture is one of massive upper gastrointestinal hemorrhage with hemodynamic instability. We report the case of a 41-year-old female patient who was admitted to our department after a stay in the medical emergency department for hematemesis, hematochezia and hemodynamic instability. An abdominal CT scan was used to make the diagnosis, but due to the inadequate technical facilities for cardiovascular surgery in our hospital, she was evacuated to another hospital in Ouagadougou. Her subsequent progress was marked by her death a few days after evacuation. This case shows that in addition to the availability of diagnostic tools in our healthcare facilities, the need for qualified staff, particularly in vascular surgery, is imperative.

Keywords

Vascular surgery, Primary aortoduodenal fistula (PADF), CT scan, Digestive hemorrhage.

Introduction

Gastrointestinal bleeding is a frequent, life-threatening medical and surgical emergency in gastroenterology, with a variety of causes, the most common of which are peptic ulcers and esophageal varices [1]. However, certain etiologies are rare and pose significant diagnostic and therapeutic challenges. These include primary aortoduodenal fistula (PADF), an extremely rare but potentially life-threatening condition characterized by abnormal communication between the aorta and duodenum [2-4]. The fistula is said to be primary when it occurs on a native aorta, unaffected by any previous surgical treatment, otherwise it is said to be secondary.

We report here a case of primary aorto-duodenal fistula (PADF) observed in the hepato-gastroenterology department of the Souro Sanou University Hospital in Bobo-Dioulasso (Burkina Faso). This case observation aims to enrich the scientific literature on this rare pathology, highlighting the clinical, diagnostic and therapeutic challenges encountered in a context of limited resources.

Clinical Observation

A 41-year-old housewife was admitted to the medical emergency department of Souro Sanou University Hospital on January 28, 2024 with hematemesis and hematochezia, which had been present for 24 hours. This symptomatology was accompanied by dizziness and asthenia. There was no notion of loss of consciousness, pain or sensation of abdominal mass. No history of aggressive gastrointestinal drugs, anticoagulants, epigastralgia, peptic ulcer disease or vascular surgery, particularly aortic surgery. However, the patient reported blackish, foul-smelling, liquid stools that had been passing intermittently for around four months.

Clinical examination on admission to the emergency department revealed anemia, hemorrhagic syndrome and hemodynamic instability with tachycardia at 123 beats/minute and polypnea at 24 cycles per minute. The blood count showed hemoglobin at 5.5 g/dL normocytic normochromic, white blood cells at 4890/µL and platelets at 399000/µL. The immediate course of action in the medical emergency department was to transfuse 02 packed red blood cells, after which the patient was transferred to the hepato-gastroenterology department on January 29, 2024.

Examination on admission to the hepato-gastroenterology department revealed:

- World Health Organization (WHO) stage 2 general condition, hemodynamically stable with blood pressure 120/60 mmHg, pulse 94 beats/minute, oxygen saturation 98% on room air, respiratory rate 18 cycles/min and temperature 36.7ºC;
- mucocutaneous pallor;
- persistent melena;
- abdomen not enlarged, without collateral venous circulation or stellate angioma, supple and depressible, with no intra-abdominal mass or signs of peritoneal irritation.

The patient was put on continuous omeprazole injection 40mg.
An upper gastrointestinal endoscopy (UGIE) performed on January 30, 2024 was non-contributory, as there was no bleeding lesion or bleeding stigma.

The evolution was marked on January 31, 2024 by the occurrence of abundant hematochezia associated with a drop in the hemoglobin level to 4.8 g/dL, which led to the transfusion of 02 packed red blood cells.

As bleeding persisted, with alternating hematochezia and melena, a colonoscopy was performed after stabilization of the hemodynamic state, which required the administration of eight bags of red blood cells. Colonoscopy was non-contributory, as it was severely hampered by the presence of blackish blood, despite adequate preparation with Macrogol.

Recurrent bleeding in hospital required a second oesogastroduodenal endoscopy on February 16, 2024, under optimal conditions. This revealed active jet bleeding in the second duodenum, limiting visibility of its exact origin.

The UGIE was then completed by an abdominal angioscanner which revealed:

- a saccular aneurysm of the sub-renal abdominal aorta measuring 54x52x36 mm with parietal thrombosis and significant edematous peri-aneurysmal infiltration, complicated by duodenal (2nd and 3rd duodenums) and jejunal fissuring with signs of active bleeding marked by the presence of blushes (Picture 1A);
- another saccular aneurysm, approximately 35 mm in diameter and measuring 13x10 mm, with parietal thrombosis and no signs of fissuring (Picture 1B);
- a bifocal fusiform aneurysm at the initial portion of the left common iliac artery, measuring 17 mm in diameter and approximately 52 mm in height, with no signs of fissuring (Picture 2C and D);
- moderate intraperitoneal fluid effusion in the iliac fossae and cul-de-sac of Douglas (Picture 2C);
- no signs of hypoperfusion of the abdominopelvic viscera, with correct opacification of the celiac trunk, superior mesenteric artery and renal arteries.

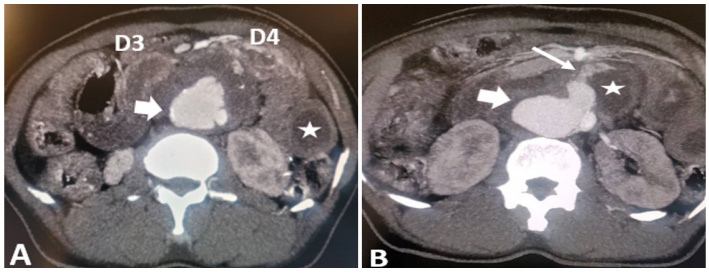

Figure 1: Abdominal CT scan after contrast injection in parenchymal window, axial section (A; B) depicting an atheromatous saccular aneurysmal dilatation of the sub-renal abdominal aorta with parietal thrombosis, asymmetric contours of the circulating channel (arrowhead) and perfect visualization of an anterior fistulous tract between the aorta and the third duodenal (arrow) with vascular blush in the digestive lumen at D3 and D4.

Note dilatation with fluid stasis and diffuse parietal thickening of the jejunum.

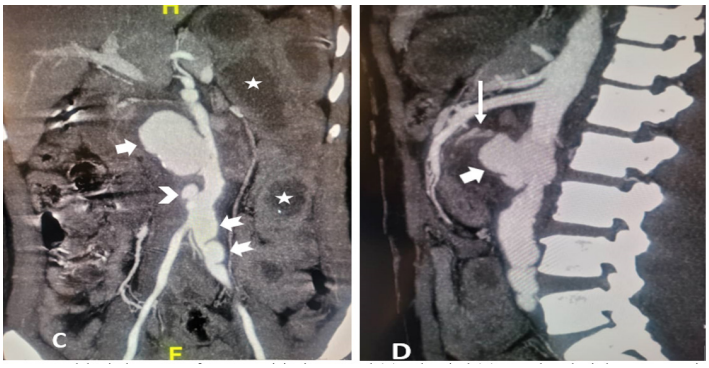

Figure 2: Abdominal CT scan after contrast injection, coronal (C) and sagittal (D) parenchymal window reconstruction depicting right anterolateral saccular atheromatous aneurysm of the sub-renal abdominal aorta with parietal thrombosis, asymmetric contours of the circulating channel (arrowhead) and perfect visualization of an anterior fistulous path between the aorta and the third duodenal (arrow).

Presence of other atheromatous aneurysms, respectively saccular of the abdominal aorta (chevron), fusiform and saccular of the initial portion of the left common iliac artery (notched arrow).
Note dilatation with fluid stasis and diffuse parietal thickening of the jejunum (star).

In the absence of a vascular surgeon in our hospital, the patient was evacuated on February 19, 2024 to the Tengadogo University Hospital in Ouagadougou, which has a vascular surgery technical platform. At the time of evacuation, the patient had received 14

bags of red blood cells, and the latest hemoglobin level on the hemogram was 7.3 g/dL.

The subsequent evolution at the Tengandogo University Hospital was marked by the patient's death eight days (February 27, 2024) after admission to the hospital, before any hemostasis surgery.

Discussion

The case we describe highlights important elements in the positive diagnosis of PADF. It can be difficult because of its rarity, nonspecific symptoms and the difficulty of confirming it with available imaging modalities.

Indeed, the first case of PADF was described by Sir Astley Cooper in 1824, but the incidence of PADF is very low, at 0.007 per million inhabitants, with fewer than 400 cases reported in the literature, and its diagnosis is most often delayed [2,5-7].

The classic clinical presentation of PADF is the triad of gastrointestinal bleeding, abdominal pain and pulsatile abdominal mass. It is complete in only 11% of cases, hence the non-specificity of the symptoms. In our patient's case, the clinical form was essentially a digestive hemorrhage [8]. Digestive bleeding occurs in 94% of cases of PADF [8]. It is a digestive hemorrhage of high origin, which may be externalized by an upper (hematemesis) or lower route (melena or even hematochezia in cases of high flow). It often takes the form of a “sentinel” hemorrhage, as illustrated in the case described here: a hemorrhagic episode of variable abundance. This bleeding ceases spontaneously thanks to the formation of a clot, favored by arterial hypotension secondary to blood loss. This sentinel hemorrhage may evolve recurrently, followed by massive hemorrhage due to clot mobilization, which is lethal in the absence of rapid treatment. The interval between the first hemorrhagic episode and massive hemorrhage varies from a few hours to several months [8]. In our case, the first episode occurred four months earlier, characterized by the onset of melena.

Oesogastroduodenal endoscopy, the key examination for any upper gastrointestinal bleeding, has two objectives in the event of an PADF. Firstly, it highlights the fistula, such as arterial bleeding linked to ulceration in the 3rd or 4th duodenum (D3 and D4), and secondly, it excludes other possible causes of upper gastrointestinal bleeding [9]. However, in 50% of cases of PADF, it does not reveal any pathology, as our case shows [10]. The first UGIE was non-contributory, as the fistula most often develops between the infra-renal aorta and the third or fourth retroperitoneal portion of the duodenum, due to the anatomical proximity of the two structures, whereas D3 and D4 are rarely explored during conventional UGIE [11]. UGIE plays an important role and is only cost-effective when examining the distal duodenum, and should therefore be performed with particular attention to the distal portion of the duodenum, for the diagnosis of this pathology.

The most common cause of PADF is atheromatous aneurysms, which account for 83% of cases [8]. Inflammatory (vasculitis) and infectious causes are classic: syphilis, tuberculosis. Other etiologies are more rarely described, such as tumors, radiation lesions, diverticulitis, ulcers and foreign-body ingestions [8]. In our case, no cause could be identified that could explain the occurrence of this PADF. Despite the patient's lack of aneurysm risk factors, atheromatous workup was not performed. Indeed, once the diagnosis of PADF had been made, the emergency after stabilization of the patient was surgical hemostasis of the hemorrhage, hence the urgent evacuation of the patient, thus preventing exploration of this frequent atheromatous origin and of the other causes described in the literature.

Emergency surgery is the only curative treatment for this serious condition, without which mortality can reach 100% due to massive digestive hemorrhage or sepsis secondary to digestive contamination [7,12]. Thus, early diagnosis and treatment are imperative to reduce mortality due to PADF [13]. Emphasis in management is placed on control of bleeding, rehabilitation of the gastrointestinal tract and vascular restoration with maintenance of distal perfusion [11]. Given the complexity of this pathology, management must be carried out by specialized centers with multidisciplinary experience. Management is well codified once the diagnosis has been made, but made difficult in our context due to insufficient technical resources and a lack of qualified vascular surgery skills. In our case, this explains the evacuation of the patient, the delay in her treatment and the fatal outcome.

Conclusion

PADF is a rare but potentially fatal condition, requiring rapid and appropriate diagnosis and management; hence the need to strengthen the technical platforms and human resources in surgical vascular pathology in our hospital.

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