

Primary Aorto-Digestive Fistula: A Case Report from Madagascar

Maharo Ramifehiarivo^{1*}, Benja Tahina Rafanomezantsoa², Mampiadana Médyno Lovasoa¹, Tsirimalala Rajaobelison³, Toky Mamin'ny Aina Rajaonahary¹, Andriamihaja Jean Claude Rakotoarisoa¹

¹Department of Vascular Surgery, Joseph Ravoahangy Andrianavalona University Hospital Center, Antananarivo, Madagascar

²Department of Medical Imaging, Joseph Ravoahangy Andrianavalona University Hospital Center, Antananarivo, Madagascar

³Department of Vascular Surgery, University Hospital Center Morafeno Toamasina, Toamasina, Madagascar

Email: *mramifehiarivo@gmail.com

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Abstract

Primary aortodigestive fistulas (ADF) are rare abnormal communications between the native aorta and the gastrointestinal tract. We report the case of a 54-year-old hypertensive woman, untreated, with a history of hemorrhagic antral-pyloric ulcer. She had presented with recurrent ulcer pain and a pulsatile abdominal mass for four months. Her treatment consisted of traditional abdominal massage. On admission, she presented with hypovolemic shock secondary to massive digestive hemorrhage, with severe anemia. Abdominal angio-CT revealed a suprarenal aortic aneurysm with gastric fistulization. Despite resuscitation measures, the outcome remained unfavorable due to the lack of surgical management, which is linked to limited resources in Madagascar. Primary ADF are a rare but fatal cause of gastrointestinal bleeding. They should be suspected in patients presenting with the classic triad: gastrointestinal hemorrhage, abdominal pain, and a pulsatile abdominal mass.

Keywords

Aneurysm, Aorta, Fistula, Gastrointestinal Bleeding

1. Introduction

Aortodigestive fistulas (ADF) are defined by the presence of abnormal communication between the aorta and the gastrointestinal tract [1]. Aortodigestive fistulas represent a rare and potentially fatal cause of gastrointestinal haemorrhage [2].

The mortality rate of untreated primary aorto-digestive fistulas is close to 100% due to rapid blood loss in 366 cases collected since 2008 [3].

We report the case of a 54-year-old woman with a history of gastrointestinal

bleeding and hypertension, in whom a primary ADF was discovered secondary to an abdominal aortic aneurysm.

This case highlights the challenges related to the diagnosis and management of primary ADF particularly in resource-limited settings.

2. Case Report

A 54-year-old woman presented to the emergency department with a second episode of massive upper gastrointestinal bleeding. She had a history of untreated grade II hypertension known for 03 years and an antropyloric ulcer diagnosed a year earlier by upper gastro intestinal fibroscopy, having failed to mention the presence of a flailing abdominal mass during the examination. At that time, she was not known to have any abdominal aneurysmal disease.

For four months, she had been experiencing recurrent, moderate epigastric pain, radiating to the back, with no calming factors and no food intake. There were no associated digestive signs. There was no notion of fever. As a response, she consulted a traditional healer and underwent frequent abdominal massages as treatment, but without improvement.

The disease progressed to hematemesis (three episodes of bloody vomiting) and melena (three black stools within 24 hours), prompting her visit to the emergency department.

On admission, she was in a state of hypovolemic shock (tachycardia: 110 bpm, hypotension: 70/40 mmHg), oxygen desaturation 88% on room air, with a palpable pulsatile epigastric mass in a patient with a body mass index of 18.4 kg/m², an unbloated, supple abdomen and rectal examination revealing black stools. Other physical examinations were unremarkable. The patient was very pale and had severe normochromic normocytic anemia (hemoglobin 02 g/dL) with no other blood line disorders, and no biological inflammatory syndrome.

An abdominal CT angiography revealed a suprarenal aortic aneurysm with significant mural thrombosis (**Figure 1**). Active contrast extravasation was detected, confirming the presence of a gastric fistula (**Figure 2**).

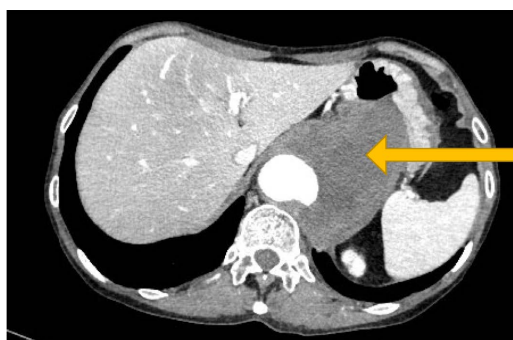


Figure 1. Contrast-enhanced abdominal CT scan in the arterial phase showing a suprarenal aortic aneurysm with significant mural thrombosis (yellow arrow). Source: Department of medical imaging, Joseph Ravoahangy Andrianavalona university hospital center.



Figure 2. Contrast-enhanced abdominal CT scan in the arterial phase showing an aorto-gastric fistula (yellow arrow). Source: Department of Medical Imaging, Joseph Ravoahangy Andrianavalona University Hospital Center.

Resuscitation measures were initiated, including fluid replacement, blood transfusion, and vasopressor support. Treatment of the ulcer pathology was also initiated empirically, including intravenous proton pump inhibitors and antibiotic therapy for eradication of *Helicobacter pylori*. No endoscopic examinations were performed. The patient died within 24 hours of admission following hemorrhagic shock, without any appropriate surgical management.

3. Discussion

Primary aorto-enteric fistulas occur due to the spontaneous development of a connection between the gastrointestinal tract and the native aneurysmal aorta. They are considered rare, with an incidence rate of 0.04% to 0.07% among patients who died from massive gastrointestinal bleeding and 0.69% to 2.36% among patients with abdominal aortic aneurysms [4]-[6]. Secondary ADF occur in the context of a prosthetic aorta and are more common, with a postoperative incidence rate ranging from 0.5% to 2.3% [5].

Primary ADF typically develop on an unrepaired aortic aneurysmal sac that erodes into the gastrointestinal lumen due to predisposing factors [7]. These factors include atherosclerosis (60% - 85%), infection (15%), and rarer causes such as malignant tumors and duodenal ulcers [8]-[12].

Proposed theories for ADF formation include direct mechanical wear and inflammatory destruction triggered by infection, foreign bodies, or erosion. Some studies also suggest a role for matrix metalloproteinases in this process. The characteristic site of fistula formation is the fixed retroperitoneal portion of the distal duodenum, located just anterior to the aorta [5].

According to Gad *et al.*, 73% of primary ADF were associated with an atherosclerotic abdominal aortic aneurysm, while 26% were linked to mycotic or traumatic aneurysms [13].

Although our patient's only cardiovascular risk factor was untreated grade II hypertension, which is also an atheromatous risk factor, chronic pulsatile me-

chanical trauma to the abdomen following repetitive massage promoted erosion of the already fragile digestive wall and fistula formation on an already aneurysmal aorta.

Birket *et al.* demonstrated in 2020, in the context of bilateral vertebral artery dissection, that even the application of low-intensity force to the neck, such as during massage, could cause vascular injury [14]. Although the cervical and abdominal vascular anatomical relationships are different; in a patient classified as underweight according to BMI ($<18.5 \text{ kg/m}^2$) [15], the aorta remains easily accessible to abdominal palpation and subject to trauma during the act of massage. However, traditional massage remains a very frequent and first-resort therapy in Madagascar, which could be an aggravating factor in certain vascular or digestive pathologies if the indication is not well established. The traditional massage received by the patient may have contributed to the enlargement of the aneurysmal sac and served as a risk factor for rupture and fistulization.

Collectively, most (83%) primary and secondary ADF occur in the duodenum, most often in the third portion (57%) [3] [16]. The digestive segment affected in our case was the stomach. The exact incidence of aorto-gastric fistula has not yet been established in the literature. In our case, the aneurysmal sac was large and of suprarenal topography. Its expansion to the gastric level could explain the irritation and inflammation of the gastric wall leading to digestive fistulization.

A hemorrhagic primary ADF may present with a herald bleed. The classic triad of ADF includes a pulsatile abdominal mass, gastrointestinal bleeding, and abdominal pain, but this combination is present in less than 25% of cases [17].

Our patient had a history of minor upper gastrointestinal bleeding one year earlier, followed by a massive hemorrhage that led to her admission. ADF should be suspected in any patient presenting with massive gastrointestinal bleeding [17]. In the literature, the initial minor bleeding preceding massive hemorrhage is referred to as a “sentinel bleed” [2]. Vasospasm, thrombus formation, and reactive intestinal contraction contribute to the temporary closure of the fistulous tract [18].

The time interval between a sentinel bleed and massive hemorrhage varies: in 70% of patients, it is within 6 hours; in 50%, within 24 hours; and in 29%, it exceeds one week [19].

The three most commonly used modalities for evaluating a patient with this presentation are abdominal CT angiography, upper gastrointestinal endoscopy, and aortography/arteriography [17].

Upper gastrointestinal endoscopy is useful for ruling out other causes of upper gastrointestinal bleeding, such as peptic ulcers and esophageal varices [20]. Endoscopy should be the initial diagnostic modality of choice in a hemodynamically stable patient. However, a negative endoscopy does not exclude the possibility of an aorto-enteric fistula. The presence of an ulcer or erosion adjacent to an intraluminal thrombus in the duodenum, with or without an extrinsic pulsatile mass, is highly suggestive of the diagnosis [17]. In our case, endoscopy was not performed

due to the patient's hemodynamic instability and the lack of emergency endoscopic services in our center.

Abdominal CT angiography is a non-invasive diagnostic tool recommended in the literature due to its high specificity (85% - 100%) and variable sensitivity (50% - 94%) [21]. The loss of the aneurysmal wall or the fat plane between the aorta and the duodenum, as well as the presence of retroperitoneal air, are highly suggestive of an aorto-duodenal fistula on CT imaging.

Angiography/aortography can provide a detailed anatomical description, aiding in surgical planning and vascular reconstruction. However, with the advent of high-resolution CT, invasive angiography has become largely obsolete [17]. According to a retrospective study by Peck and Eidemiller, very few primary aorto-enteric fistulas are diagnosed via angiography [12].

Our case was marked by the importance of mural thrombosis and massive exsanguination. This could be explained by the delay in management, given the diagnostic and therapeutic errancy prior to the decision to seek medical advice.

Mortality from acute digestive haemorrhage secondary to ADF is around 100% in the absence of urgent surgical management [20]. Treatment consists in surgically disconnecting the vascular tree from the digestive tract and performing an aortic repair or even an aortic graft and repairing the digestive tract [22]. Endovascular treatment involves excluding the fistula with a covered aortic stent graft, and is currently an attractive treatment option [2].

Surgical planning for a ADF must take into account the underlying etiology, the degree of retroperitoneal contamination and active bleeding. The duodenum should be closed in two layers, with a layer of omentum between the aorta and duodenum. The aorta should preferably be reconstructed with a dacron or polytetrafluoroethylene (PTFE) in situ graft, with the aneurysmal wall closed over the graft. Extra-anatomical reconstruction may be necessary in cases of extensive contamination or mycotic aneurysms [17].

Advances in endovascular techniques over the last few decades have made them an attractive alternative to traditional open surgery for the treatment of primary ADF. Endovascular procedures offer hope to patients who might otherwise face significant risks during conventional surgery [23].

In our case, only drug treatment with resuscitative measures was performed; open and endovascular surgical repair were not performed due to the lack of suitable surgical infrastructure limiting therapeutic options, leading to a fatal outcome.

4. Conclusion

Primary ADF is a rare but potentially fatal cause of gastrointestinal bleeding. They should be suspected of patients presenting with the classic triad of massive gastrointestinal bleeding, abdominal pain and a pulsatile abdominal mass. Abdominal CT angiography remains the diagnostic gold standard, confirming communication between the aorta and the digestive tract and guiding management. In set-

tings where surgical resources are limited, management remains a challenge, leading to high mortality.

Declaration of Consent

The authors certify that they have obtained all consent forms from the family of the deceased patient. In the form, the patient's family gave their consent for their images and other clinical information to be reported in the journal. The family understands that no patient's name or initial will be published and that efforts will be made to conceal identity, but anonymity cannot be guaranteed.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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