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(CASE REPORT)

Gastrointestinal bleeding associated with a ruptured abdominal aortic aneurysm revealing Takayasu's Arteritis

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Abstract

Background: Takayasu arteritis is a rare systemic vasculitis affecting large arteries, leading to stenosis or aneurysmal complications. While stenotic lesions are more common, aneurysms can result in life-threatening events such as rupture and aortoenteric fistulas.

Case Presentation: We report the case of a 41-year-old woman admitted for massive upper gastrointestinal bleeding. Imaging revealed an abdominal aortic aneurysm with a duodenal fistula, ultimately leading to the diagnosis of Takayasu arteritis. The patient underwent emergency aorto-aortic bypass grafting and duodenal fistula closure, followed by immunosuppressive therapy.

Conclusion: This case highlights the diagnostic challenges of Takayasu arteritis, particularly in its rare aneurysmal form. Early imaging and a multidisciplinary approach are crucial for timely intervention and improved outcomes.

Keywords: Takayasu Arteritis; Gastrointestinal Bleeding; Aortoenteric Fistula; Abdominal Aortic Aneurysms

1. Introduction

Takayasu arteritis is a systemic inflammatory disease that primarily affects medium and large arteries, particularly the aorta and its major branches, including the renal, carotid, and subclavian arteries. This condition leads to stenosis, occlusions, or aneurysmal degeneration of these vessels [1].

The disease typically begins with a nonspecific early inflammatory phase marked by constitutional symptoms such as fever, malaise, myalgia, weight loss, and anorexia. Most patients seek medical attention only during the "pulseless" phase, when arterial insufficiencies manifest as hypertension, neurological deficits, or upper limb claudication [1].

The occurrence of upper gastrointestinal bleeding as the initial presentation of Takayasu arteritis is exceedingly rare, particularly when caused by the rupture of an abdominal aortic aneurysm with duodenal fistulization. Aorto-digestive fistulas themselves are an uncommon cause of upper gastrointestinal hemorrhage, accounting for only 0.3% of cases, yet they are often life-threatening [2]. Consequently, the combination of these conditions is exceptionally rare and scarcely documented in clinical practice.

We report the case of a young woman admitted for upper gastrointestinal bleeding due to a ruptured abdominal aortic aneurysm, which ultimately led to the diagnosis of previously unrecognized Takayasu arteritis.

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2. Case report

A 41-year-old woman was admitted to the emergency department after experiencing multiple episodes of profuse hematemesis over the past five hours. She had a history of hospitalization four months earlier for upper gastrointestinal bleeding caused by duodenal bulb ulcers, which had shown good clinical improvement following treatment with proton-pump inhibitors (PPIs) and eradication of Helicobacter pylori.

The patient reported no additional symptoms apart from epigastric pain and gastrointestinal bleeding. Physical examination revealed hypotension with a blood pressure of 80/50 mm Hg, tachycardia at 120 beats per minute, and a respiratory rate of 20 breaths per minute. Abdominal examination showed no abnormalities.

Laboratory tests indicated a low hemoglobin level of 7 g/dL, while other parameters were within normal limits. The patient was admitted to the intensive care unit, where she was stabilized through fluid resuscitation and blood transfusion.

Following stabilization, upper Gastrointestinal Endoscopy was performed, revealing bright red blood originating from the third portion of the duodenum. Further evaluation with an abdominal CT angiography demonstrated inflammatory aortitis complicated by two partially thrombosed pseudoaneurysms of the infrarenal abdominal aorta. The first pseudoaneurysm was located near the origin of the right renal artery, while the second was situated 18 mm distal to it and exhibited a microfistula connecting to the lumen of the third portion of the duodenum.

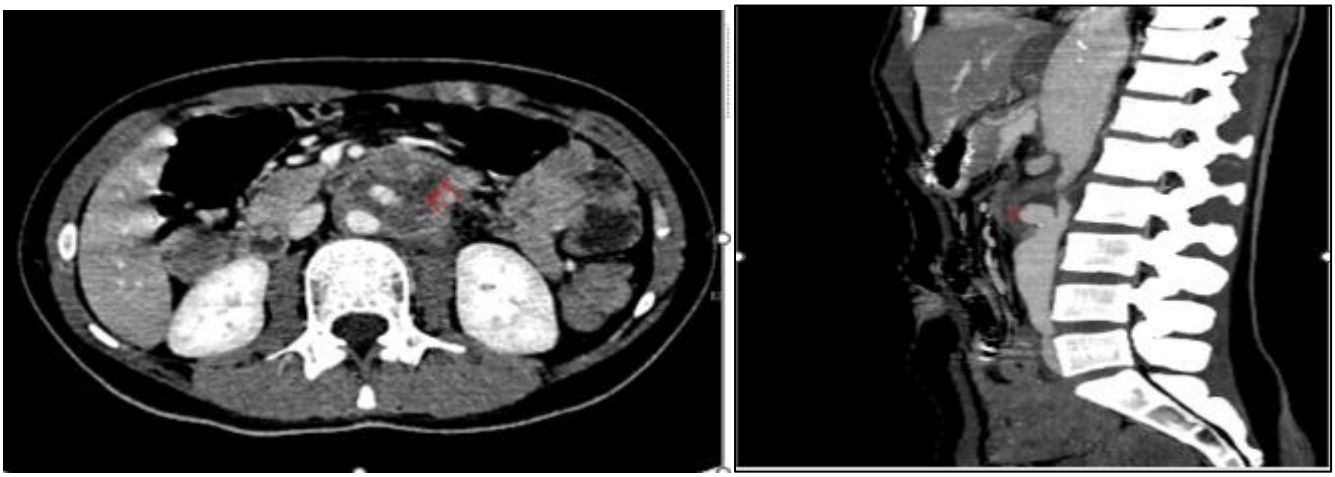

Figure 1 CT images showing inflammatory aortitis complicated by infrarenal pseudoaneurysms, one of which has formed a fistula with the duodenum

Urgent preoperative assessments were carried out, and after ruling out contraindications, the patient underwent aorto-aortic bypass grafting along with closure of the duodenal fistula.

Postoperatively, she was transferred to the internal medicine department for further evaluation.

A chest CT scan revealed aneurysmal dilation of the thoracic aorta, particularly at the aortic arch and descending aorta, with irregular thickened walls and ulcerations, confirming a diagnosis of Takayasu arteritis. She was treated with corticosteroid boluses, followed by three courses of Endoxan and methotrexate from the second course onward.

The clinical course was marked by significant improvement under treatment, with resolution of pain and no recurrence of bleeding during a 12-month follow-up.

3. Discussion

An aortoenteric fistula (AEF) is a direct communication between the aorta and the gastrointestinal tract. It can be classified as primary or secondary. Secondary aortoenteric fistulas occur in patients who have previously undergone aortic prosthetic reconstruction and are caused by erosion of the aortic prosthesis into the gastrointestinal tract. They are ten times more common than primary aortoenteric fistulas [3].

Primary aortoenteric fistulas are typically associated with spontaneous rupture of the expanding aorta into a closely adherent portion of the gastrointestinal tract. This can lead to gastrointestinal bleeding, often of significant volume, threatening the patient's life [3].

The majority of primary aortoenteric fistulas result from the expansion of abdominal aortic aneurysms (AAA), leading to chronic mechanical compression of the gastrointestinal tract. This compression causes fibrotic changes and inflammatory destruction. The third and fourth portions of the duodenum are the most commonly affected, accounting for 80% of AEF cases [3]. The reason for this high incidence of rupture into the third portion of the duodenum is well explained by the combination of anatomicomechanical relations. The third portion of the duodenum is relatively fixed because of its retroperitoneal position, its relation to the superior mesenteric artery, ligament of Treitz, aortic wall, and the vertebral column [4].

The clinical presentation, which includes the classic triad — gastrointestinal bleeding, abdominal pain, and pulsating abdominal mass — occurs in only 25% of primary aortoenteric fistula cases [5].

Upper gastrointestinal endoscopy is the primary examination recommended in cases of gastrointestinal bleeding to obtain valuable diagnostic information in hemodynamically stable patients. However, it rarely provides confirmatory evidence of a primary aorto-duodenal fistula, with a detection rate of 25%, as stable patients often do not present with active bleeding [6, 7]. However, although it does not always allow for the diagnosis of an aorto-duodenal fistula, endoscopy helps guide further investigations. In the case we describe, the presence of active bleeding in the duodenum led us to perform an abdominal CT angiography.

CT angiography of the aorta can determine the size, location, and degree of calcification of an abdominal aortic aneurysm, making it a valuable diagnostic tool for aorto-duodenal fistulas, with a sensitivity ranging from 40% to 90% and a specificity of 33% to 100%. The loss of the aneurysmal wall, the presence of air in the aortic wall, retroperitoneum, or thrombus, as well as focal thickening of the intestinal wall with destruction of the fat plane between the aneurysm and duodenum, or the presence of contrast in the gastrointestinal tract, strongly suggest an aorto-duodenal fistula in the CT findings [6, 8, 9].

However, aortography showing contrast extravasation into the intestine was positive in only 26% of cases [6].

The diagnosis of a primary aorto-duodenal fistula remains difficult. Only 33% to 50% of aorto-enteric fistulas are diagnosed preoperatively [10].

The therapeutic options for primary aorto-duodenal fistula include open surgery and endovascular repair [3]. If left untreated or misdiagnosed, the condition carries a 100% mortality rate. Endovascular aortic repair (EVAR) may serve as a "bridge" therapy in hemodynamically unstable patients, providing temporary stabilization and allowing for subsequent definitive enteric repair. This approach can improve outcomes, particularly when open surgery is challenging or contraindicated [3].

The primary cause of abdominal aortic aneurysms is atherosclerosis, but they can also result from infections, trauma, or inflammatory arteritides like Takayasu arteritis [11]. This large-vessel vasculitis predominantly affects women under 40 years of age, with an incidence of 1.2-2.6 cases per million people annually [3]. While 90% of cases involve stenosis, up to 25% develop aneurysms, primarily in the aorta, subclavian, brachiocephalic, and carotid arteries [3]. The real incidence of rupture of either the abdominal or thoracic aortic aneurysms is not known, but seems to be low [12].

Medical treatment relies on anti-inflammatory and immunosuppressive agents.Corticosteroids remain the cornerstone during active phases, often combined with other immunosuppressive therapies [12].

4. Conclusion

Takayasu arteritis is a rare but serious vasculitis affecting large arteries, leading to stenosis or aneurysmal complications. Although less common, aneurysms can be life-threatening, requiring prompt imaging for diagnosis. Surgical intervention and immunosuppressive therapy are key to management, with endovascular repair as an option in unstable cases. Early diagnosis and a multidisciplinary approach are crucial for better outcomes.

Compliance with ethical standards

Disclosure of conflict of interest

The authors declare that they have no competing interest.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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