

## Case Report

# Intestinal cavernous hemangioma as a cause of anemic syndrome and gastrointestinal bleeding

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## ABSTRACT

There are multiple causes of non-variceal gastrointestinal bleeding, some more commonly diagnosed than others. Therefore, the finding of a cavernous hemangioma in the jejunum should make us consider patients who present with anemic syndrome due to occult gastrointestinal bleeding, once the more common pathologies have been ruled out, this after extensive studies, including upper endoscopy, colonoscopy, imaging studies and some cases endoscopic capsule. It is described that cavernous hemangiomas account for 0.05% of gastrointestinal neoplasms and up to 7-10% of benign tumors of the small intestine. They can also be found in up to 7.7% of patients with gastrointestinal bleeding.

**Keywords:** Intestinal hemangioma, Anemic syndrome, Surgery

## INTRODUCTION

Intestinal hemangiomas (IH) are uncommon benign vascular tumors responsible for only 0.05% of all cases of gastrointestinal neoplasms that may occur anywhere in the gastrointestinal tract as single or multiple lesions.<sup>1,2</sup> They originate from fast-growing embryonic mesodermal tissue and are characterized by a proliferation of endothelial cells and are also referred to as vascular malformations since their pathology consists of a collection of malformed blood vessels.<sup>2,3</sup> Risk factors for the development of it including female sex, white non-hispanic race, prematurity, low birth weight, and multiple gestation have been well described.<sup>4</sup> Prematurity rates and associated complications have increased over the past decades, but how these changes relate to incidence of it is unknown.<sup>5</sup>

They are classified as cavernous and capillary, the first one is the most frequent.<sup>6</sup> Capillary hemangiomas are a proliferation of small capillaries composed of thin-walled,

blood-filled spaces lined by endothelial cells. cavernous hemangiomas consist of large blood-filled spaces or sinuses lined by single or multiple layers of endothelial cells. On the other hand, cavernous hemangiomas may infiltrate large segments of the intestine and mesentery.<sup>1</sup>

This pathology might be considerate with clinically evident, such pain and gastrointestinal bleeding with frank melena, severe anemia, and orthostatic symptoms.<sup>6</sup> Less commonly, intestinal hemangiomas may cause bowel obstruction, intussusception, and even perforation.<sup>8</sup>

IH are associated as part of several genetic syndromes, including Von-Hippel-Lindau, Maffucci syndrome, Osler Weber-Rendu disease, Klippel-Trénaunay syndrome, or the congenital blue rubber bleb nevus syndrome.<sup>1,9</sup> The diagnosis of intestinal hemangioma can be challenging due to their location, they are often not detected during upper endoscopy or colonoscopy.<sup>9</sup> Magnetic resonance imaging and colonoscopy were unable to detect the lesion.

furthermore, computed tomography (CT) imaging can detect focal calcification in gastrointestinal hemangioma as a degenerative change.<sup>10</sup> Gastrointestinal endoscopy, including esophagogastroduodenoscopy, colonoscopy, and capsule endoscopy, can also be useful diagnostic tools for gastrointestinal hemangioma.<sup>3</sup> The surgery is the treatment of choice, while in those unresectable or diffuse IH radiation, cryotherapy or arterial embolization have been used with limited success.<sup>6</sup> Moreover, intestinal hemangioma should be considered in patients who present pain and gastrointestinal bleeding despite the few bibliographical stock on the subject secondary to the rarity of the disease; but potentially lethal in working-age patients, therefore of this article report.

## CASE REPORT

A 40-year-old male with no chronic degenerative diseases and a positive history of alcoholism and smoking. He reports receiving multiple blood transfusions on several occasions due to anemic syndrome.

He reports starting with the current condition in 2017, experiencing fatigue, weakness, and paresthesia in the pelvic limbs, for which he was treated at a general hospital with blood transfusions and oral iron treatment due to an haemoglobin (Hb) level of 7 g/dl.

Diagnostic investigations were performed, with a result of esophagitis and follicular gastritis associated with *H. pylori* were detected. Treatment for *H. pylori* eradication was administered, but as there was no improvement and the anemic syndrome persisted, the patient was transferred to the hematology and gastroenterology departments for further diagnostic workup.

He was admitted to UMAE 25 on 09 February 2022, denying the presence of nausea, vomiting, melena, hematochezia, abdominal pain, or symptoms consistent with anemic syndrome.

Upon admission, the following vital signs were recorded: blood pressure: 140/89 mmHg, heart rate: 90 bpm, respiratory rate: 19 rpm, temperature: 36.5 °C, O<sub>2</sub> sat: 98%, weight: 98 kg, height: 1.72 m, and body mass index (BMI): 33.1.

Physical examination revealed no abnormalities.

### Laboratory results

On 28 September 2021, negative fecal occult blood test, white blood cells (WBC) 4.08, Hb 12, haematocrit (Hct) 39.2, mean corpuscular volume (MCV) 80.1, mean corpuscular haemoglobin (MCH) 24.4, platelet (PLT) 288, fibrinogen 323, prothrombin time (PT) 11.4, partial thromboplastin time (PTT) 30.4, international normalised ratio (INR) 0.9, glucose (GLU) 114, Urea 32.1, blood urea nitrogen (BUN) 15, and creatinine (CR) 1.1.

On 04 January 2022, glycated haemoglobin (HbA1c) 5.7, Hb 13.6, Hct 43.2, WBC 6.2, PLT 248.9, fibrinogen 392, PT 11.8, INR 1.0, PTT 31.9, GLU 131, Urea 34.2, BUN 16, and Cr 1.1.

### Imaging studies

On 08 March 2018 endoscopy revealed grade A esophagitis according to the Los Angeles classification and acute follicular gastritis.

On 14 January 2019 colonoscopy showed normal parameters.

On 19 July 2019 contrast-enhanced abdominal CT scan showed non-obstructive renal lithiasis and Bosniak I cortical renal cysts.



**Figure 1: Abdominal tomography: presence of wall thickening at the level of the jejunum.**

On 20 May 2021 antegrade enteroscopy showed normal findings.

On 17 June 2020 esophagus and stomach were normal, with cholesterol deposits in the second portion of the duodenum.

On 09 August 2021 retrograde enteroscopy showed normal findings.

On 28 September 2021 intestinal transit study revealed normal characteristics.

On December 2021 video capsule endoscopy showed vascularized tumor in the jejunum with active bleeding. Surgical resection of the tumor with support from transhepatic enteroscopy is recommended.

### Surgical procedure

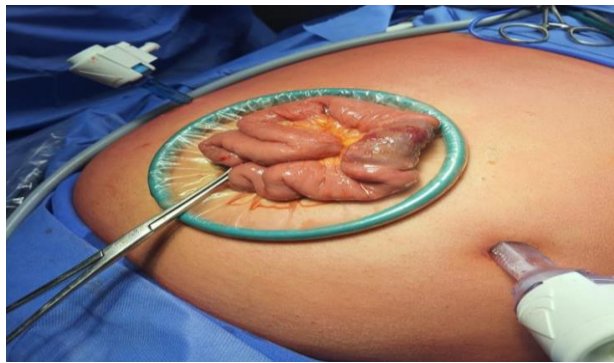
Diagnostic laparoscopy plus intestinal resection plus mechanical side to side anastomosis.

### Surgical findings

Vascularized tumor involving half of the circumference of the distal jejunum, 200 cm from the angle of treitz, without observing any lesion. a 24 cm resection was performed, and an anastomosis was made 200 cm from the angle of treitz and 180 cm from the ileocecal valve.



**Figure 2: Laparoscopic image of the tumor.**



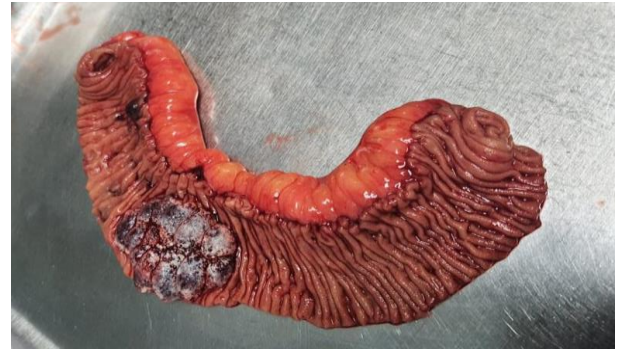
**Figure 3: Jejunum with presence of lesion.**



**Figure 4: Resected intestinal specimen.**

The histopathological result obtained was positive for cavernous hemangioma of the jejunal segment.

The patient has shown good postoperative progress and has been discharged from the digestive and endocrine surgery service with follow-up by the gastroenterology department.



**Figure 5: Resected intestinal specimen.**

### DISCUSSION

The small intestine is the most common site of gastrointestinal hemangiomas, which represent approximately 7–10% of all benign tumors in the small intestine therefore our initial hypothesis was despite of the rarity to the disease its lethality potential for working-age patients that is why justifies the need to increase published knowledge about this disease.<sup>1</sup> Differential diagnosis includes those who show up as a gastrointestinal (GI) bleeding. It should be considered as a discard pathology versus the most common causes of it, but not completely ignored. GI bleeding as a complication of peptic ulcers occurring in 15% of ulcer patients and is the main cause of bleeding, responsible for 28–59% of it.<sup>12</sup> Differs from IH of the background of NSAID use, *Helicobacter pylori* infection and the acute upper gastrointestinal bleeding as an emergency situation.<sup>13</sup> Another differentials diagnosis are oesophagitis who is a form of peptic ulcer disease, but usually only causes minor acute bleeding, variceal bleeding most frequent decompensating event in patients with cirrhosis and carcinoma or lymphoma of the stomach, when at an advanced ulcerated stage, commonly bleed but this usually results in occult blood loss.<sup>12,14</sup> Occasionally, hemoptysis may be confused for hematemesis. Ingestion of bismuth-containing products or iron supplements may cause stools to appear melanic and certain foods/dyes may turn emesis or stool red.<sup>15</sup> The main characteristics of IH are low digestive bleeding, abdominal pain, and chronic symptoms of anemia associated to risk factors. IH is rarely mentioned in reviews as a cause and differential diagnosis of intestinal bleeding.<sup>12,13,15</sup> However complications such as intussusception, obstruction, acute lethal bleeding or intestinal perforation demonstrate the importance of both diagnosis and early treatment. In this report, the case of a male patient of reproductive and worker age was described who presented a history of persistent anemia with evolution of years without timely detection, conditioning the requirement of blood transfusions and absences from work on some occasions. The aforementioned secondary to the late diagnosis and treatment of the disease, thus increasing its morbidity and mortality.



## CONCLUSION

Due to the rarity of this condition, it is common for the diagnosis of these patients to be delayed, as there are multiple other causes of gastrointestinal bleeding. Suspicion of this condition should be raised or the more common pathologies causing gastrointestinal bleeding should be ruled out in order for this pathology to be considered.

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