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CASE REPORT

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Gastrointestinal infantile hemangioma: A rare cause of digestive tract bleeding in children to consider

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Abstract

Intestinal hemangiomas are an infrequent cause of gastrointestinal bleeding and chronic anemia in infants, which diagnosis depends on high suspicion. Propranolol has been described as an effective treatment.

KEYWORDS

gastrointestinal bleeding, hemangioma, infantil

1 | INTRODUCTION

Infantile hemangiomas are benign vascular tumors that can affect 5%–10% of infants.[1] They present as isolated skin lesions and visceral localization. When they are located in gastrointestinal tract symptoms include melena, hematochezia, or cause chronic anemia and require blood transfusion. The diagnosis depends on a high suspicion.[2]

Most infantile hemangiomas present as isolated skin lesions and less frequently they have visceral localization, the liver being the most commonly affected organ, although they can also be located in the central nervous system and gastrointestinal tract. The most common anatomical sites of infantile hemangiomas that affect the gastrointestinal tract are intestine colon and rectum; although they can occur in any segment of the digestive tube.[3]

Typically, infantile hemangiomas are not present at birth, but can appear in the first weeks of life. They have a period of accelerated growth in the first months followed by a period of stabilization with slow growth and subsequently regress in the course of following months or years. The diagnosis depends on the age of the patient, anatomical location, and presenting symptoms.

The most frequent manifestation of gastrointestinal infantile hemangioma is digestive tract bleeding, and its severity depends on the anatomical site where it is found. It can present as melena, hematochezia, or cause chronic anemia and require blood transfusion. In children, GI hemangiomas are an infrequent cause of GI bleeding and due to its low frequency, its diagnosis depends on a high suspicion.

We describe the case of a visceral infantile hemangioma that affected the gastrointestinal tract in a 10-month-old infant who developed hematochezia recurrent and chronic anemia.

2 | CASE REPORT

A 10-month-old female infant was evaluated by her pediatrician for hematochezia. The first episode was at 7 months of age being of little quantity. Intestinal infectious process

was suspected indicating antibiotic treatment. In the following 2 months, the girl had 2 episodes of rectal bleeding being evaluated by performing abdominal ultrasound discarding intussusception. Next suspicion was possible food allergy, indicating diet, and change from dairy formula to elemental diet; however, the appearance of evacuating blood persisted, being referred to a pediatric gastroenterologist for its approach and diagnosis at the age of 10 months.

In her evaluation, the patient had vital signs within normal parameters and physical examination adequate coloring of integuments, soft and depressible abdomen, not painful on palpation, without visceromegaly or palpable masses. Initial laboratory tests reported hemoglobin of 8.9 g/L, hematocrit of 26.8%, leukocytes 9.260/mm^3, and platelets of 243,000 /mm3. Clotting times were normal with a TP 15.8 sec, TTP 23.4 s, and an INR 1. We decided to admit the patient to the Pediatric Intensive Care Unit due to profuse bleeding and low hemoglobin to perform blood transfusions. A colonoscopy is performed (Figure 1) showing the presence of small angiodysplasia lesions in the colon at the splenic flexure and transverse colon extending approximately 5 cm of mucosa, no evidence of active bleeding or ulceration of the surrounding mucosa. The patient persists with bloody bowel movements so erythrocyte scan is done labeled with 99mTc-pyrophosphates (Figure 2) which shows zones of colon splenic angulation radiotracer hyper-uptakes that extend toward the descending colon. With this finding, it is decided to perform angio-CT aorto-iliac (Figure 3) evidencing the presence of irregular hyper-uptake areas of neovascularization at the splenic flexure level and in most of the mesentery, and that project in the path of the splenic and superior mesenteric artery.

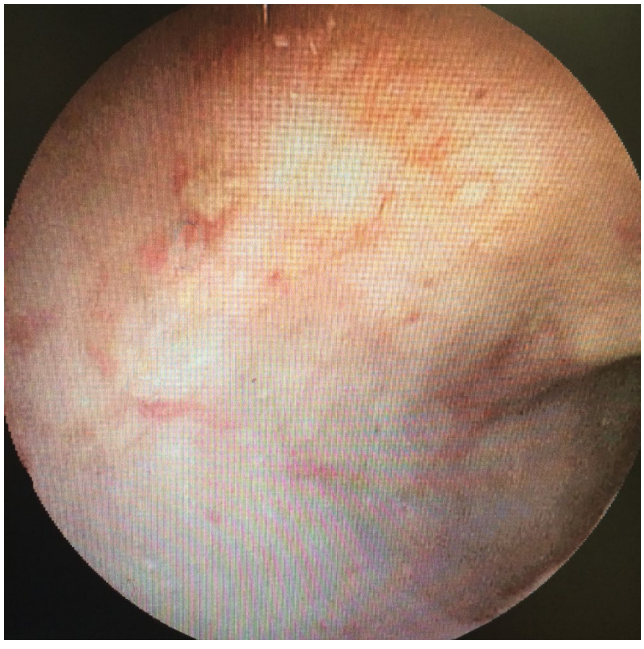

FIGURE 1 Colonoscopy showing the presence of small angiodysplasia lesions in the colon at the splenic flexure and transverse colon located 40 cm from the anal margin

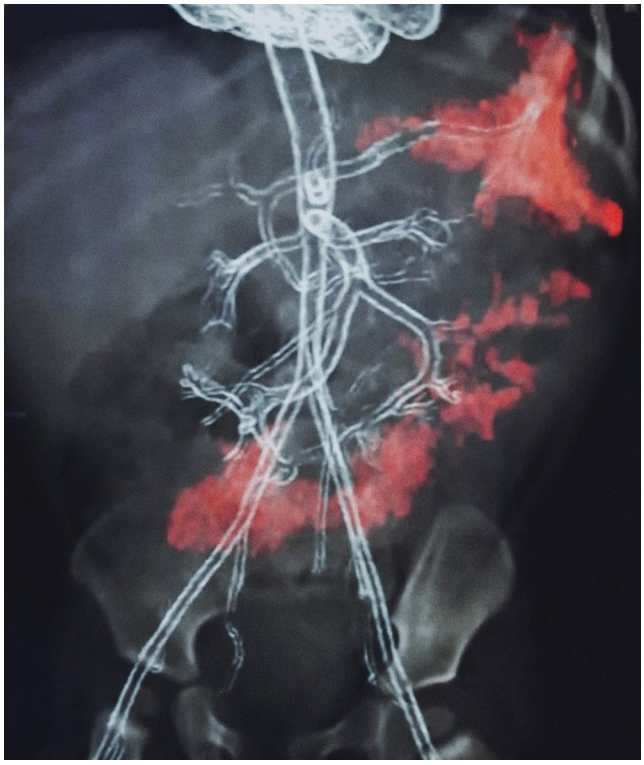

FIGURE 2 Aorto-iliac angioTac showing the presence of irregular hypercaptant areas of neovascularization at the splenic flexure and in most of the mesentery

Peritoneal and mesenteric vascular malformation is diagnosed including splenic flexure of the colon and ascending colon. Finally, a mesenteric angiography (Figure 4) for diagnosis and possible treatment with embolization which confirms the presence of hypervascularity in the splenic flexure of the colon and transverse colon, for diagnosis and possible treatment with embolization which confirms the presence of hypervascularity in the splenic flexure of the colon and transverse colon, in the territory of the mesenteric artery higher, including the mesentery the behavior suggests the presence of an intestinal hemangioma therefore no embolization was performed.

The patient was diagnosed with intestinal hemangioma with involvement of mesentery, propranolol treatment was started at a dose of 1 mg/kg per day and increased to 3 mg/kg the following days. The rectal bleeding disappears completely within the next 14 days after starting the treatment with propranolol which lasted for 6 months. No side effects were observed (hypoglycemia, bradycardia, or respiratory distress). At the end of treatment with propranolol,

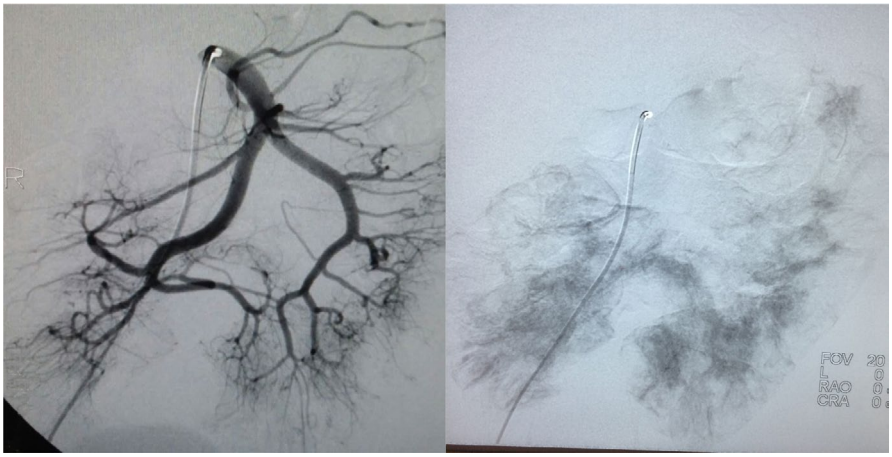

FIGURE 3 Abdominal angiography where capillary staining of the spleen is reported, in the superior mesenteric arterial territory with early venous drainage, suggesting the presence of intestinal hemangioma

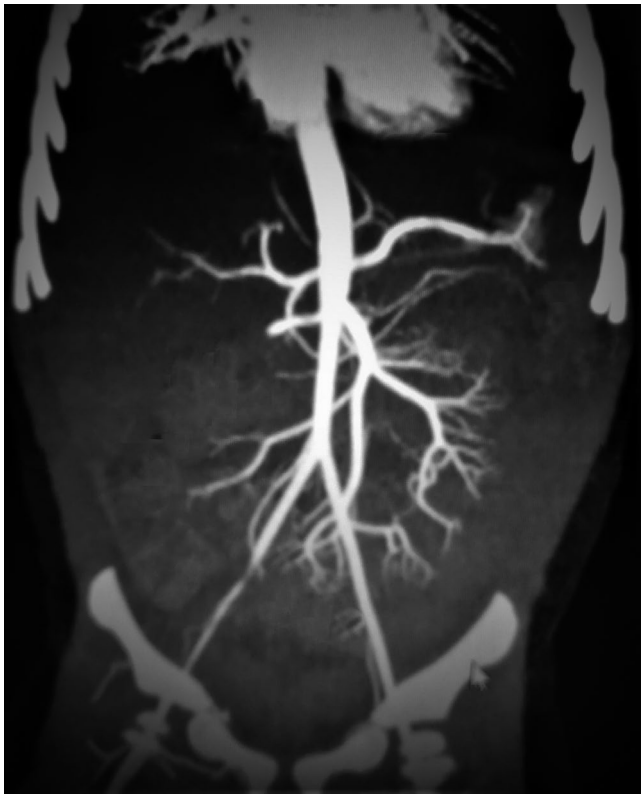

FIGURE 4 Abdominal angiography after the propranolol treatment

a new AngioTac study was performed demonstrating the disappearance of the hemangioma.

3 | DISCUSSION

Infantile hemangiomas are the most common benign tumors in childhood. They are alterations characterized by abnormal proliferation of endothelial cells and abnormal architecture of blood vessels, in contrast with vascular malformations which are congenital structural abnormalities of vascular morphogenesis.[4]

Hemangiomas can appear in any organ, as single or multiple lesions and are characterized by rapid growth followed by involution in the first years of life. However, visceral hemangiomas occur most frequently in the liver. Hemangiomas that affect the gastrointestinal tract commonly involve the small intestine and present with upper and/or lower gastrointestinal bleeding. It has been reported that up to 80% of patients with intestinal hemangiomas begin their clinical picture with bleeding from the digestive tract or obstruction. Similar to what has already been reported in other studies, the patient opened her clinical picture with bleeding from the lower digestive tract.[5]

Gastrointestinal (GI) hemangiomas can occur in association with syndromes such as Maffucci syndrome, Klippel Trénaunay syndrome, and benign neonatal hemangioma.[6]

In a study published by Soukoulis et al. in 2015, 16 patients with intestinal hemangiomas were analyzed, the frequent form of presentation was gastrointestinal bleeding (melena and hematochezia) and the most frequent location was small intestine and mesentery.[4]

As in infantile hemangiomas that occur in other areas of the body, intestinal hemangiomas require systemic therapy for their treatment.[7,8] In the beginning, corticosteroids were the treatment of choice,[8] however, after the discovery of the effects of propranolol on hemangiomas, since 2014, it has been the recommended treatment, reserving the use of corticosteroids for patients in whom beta-blockers are contraindicated or for those with no response to treatment.[9]

On occasions, surgical exploration should be considered, especially in those patients in whom non-invasive treatment to induce remission or resolution of bleeding is insufficient or fails.[10] In our case, the final diagnosis was made by abdominal angiography and the response to treatment was appropriate and effective.

Intestinal hemangiomas are not found on physical examination, despite being large masses. Given the few discoveries on physical examination, the diagnostic

suspicion of intestinal hemangiomas is confirmed by imaging studies.[10]

In the last 20 years, multiple case reports of gastrointestinal bleeding secondary to hemangiomas have been published in the pediatric population. In most of these cases, laparotomy was required for diagnosis.[5] In this case, medical treatment with propranolol at a dose of 3 mg/kg/day was carried out for a period of 6 months.

Propranolol treatment is generally well tolerated and has no adverse effects; however, the most frequent adverse effects include bronchial hyperresponsiveness, hypoglycemia, symptomatic hypotension, and bradycardia.[9]

Our patient did not present any contraindication for the use of beta-blockers, intrahospital therapy is started with propranolol at 1mg/kg with a gradual increase until reaching 3 mg/kg and after three days of hospitalization in Pediatric Intensive Care Unit, she is discharged to continue management out-of-hospital.

During medical treatment with propranolol, the patient shows improvement, without any episode of gastrointestinal bleeding, as well as adverse effects associated with the medication.

Unlike skin lesions, intestinal hemangiomas do not have an objective parameter which is visible to evaluate the efficacy of the treatment; therefore, the duration of therapy is reserved for the clinical course of the patient.[11] In our patient 6 months after stopping treatment with propranolol, a new CT angiography was performed which showed the reduction of the hemangioma until it almost disappeared. This case demonstrates that such injuries must be considered in the differential diagnosis of unexplained gastrointestinal bleeding.

The incidence of intestinal hemangiomas is low and manifests itself clinically as bleeding from the digestive tract. It is important to consider intestinal hemangioma within the differential diagnosis of gastrointestinal bleeding in all patients in whom the etiology is not clear and after excluding the prevalent pathologies in children. Although infantile cutaneous hemangiomas are common, infantile visceral hemangiomas are fewer and can present a significant diagnostic challenge.

This case demonstrates the need to consider a broad list of differential diagnosis.

4 | CONCLUSIONS

In conclusion, gastrointestinal hemangiomas are a rare cause of gastrointestinal bleeding that should be considered in the differential diagnoses of patients with gastrointestinal bleeding. Imaging with ultrasonography, CT scanning, and MRI might be helpful in achieving a primary diagnosis without the need for a biopsy, which represent risk of vascular and easily bleeding lesions. Propranolol therapy was clinically effective in our patient. More review articles are needed to define a diagnostic and management algorithm for these patients.

5 | KEY POINTS

1. Intestinal hemangiomas are infrequent cause of GI bleeding and represent a significant diagnostic challenge. It is important to consider within the differential diagnosis of gastrointestinal bleeding in all patients in whom the etiology is not clear
2. Most infantile hemangiomas present as isolated skin lesions and less frequently they have visceral localization and are characterized by rapid growth followed by involution in the first years of life.
3. The efficacy of propranolol in the treatment of hemangiomas has been described since 2008. Its mechanism of action remains unclear.

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CONFLICT OF INTEREST

None.

AUTHOR CONTRIBUTIONS

All authors contributed equally to this work.

ETHICAL STATEMENT

All information related to the identity of the patient will remain confident.

DATA AVAILABILITY STATEMENT

Research data are not shared.

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