

RELATO DE CASO

DOENÇA HEMANGIOMATOSA MÚLTIPLA ENVOLVENDO TRATO GASTROINTESTINAL: DESCRIÇÃO DE UM CASO CLÍNICO
MULTIPLE HEMANGIOMATOSE DISEASE INVOLVING GASTROINTESTINAL TRACT: A CASE REPORT **ACESSO LIVRE**

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RESUMO

Hemangioma é um tipo de tumor vascular benigno, originado a partir de distúrbios na angiogênese e está presente em cerca de 4% a 5% da população. Dentre as complicações mais comuns destacam-se: ulcerações e sangramentos, nos casos de hemangiomas localizados no trato gastrointestinal, comprometimento da via aérea e da visão, complicações viscerais, obstrução do canal auditivo e até déficit na alimentação ou na emissão de sons devido a sua presença na região da cavidade oral. O hemangioma de cólon e reto é uma causa rara de hemorragia digestiva baixa. O objetivo deste estudo é demonstrar um caso de doença hemangiomatosa múltipla em uma paciente internada e submetida a tratamento no Hospital Geral Público de Palmas (HGPP).

Palavras-chave: Hemangioma. Ulcerações. Gastrointestinal.

ABSTRACT

Hemangiomas are a type of benign vascular tumor, originated from angiogenesis disorders and prevalent in approximately 4% to 5% of the population. Among the most common complications are: ulcerations and bleeding, in cases of those located in the gastrointestinal tract; impairment of airways and vision; visceral complications; obstruction of the ear canal causing deficit in hearing; or problems in eating or in the emission of sounds due to its presence in the oral cavity. Colon and rectum hemangiomas are a rare cause of lower digestive hemorrhage. The aim of this study is to demonstrate the case of multiple hemangiomatose disease in an inpatient undergoing treatment at the Palmas General Public Hospital (HGPP).

Keywords: Hemangiomas. Ulcerations. Gastrointestinal.

INTRODUCTION

Hemangioma is a type of benign vascular tumor, originated from angiogenesis disorders, that is, it arises from pre-existing blood vessels^{1,2}. These tumors represent 6% of benign vascular tumors and are most prevalent in females³. Hemangiomas of the colon and rectum are rare and scarcely described in literature.

Hemangiomas of the colon and rectum are a rare cause of lower gastrointestinal bleeding and, undoubtedly, rectal bleeding is the most frequent clinical element of this vascular pathology. In approximately half of patients, hemangiomas cause ulceration of the mucosa which bleed to within the gastrointestinal lumen⁴.

Depending on the intensity of the intestinal bleeding, the individual may present alterations to physical examination such as sweating, dizziness, asthenia, fatigue, pallor and laboratory signs of severe anemia and hypovolemic shock⁵.

Although low digestive bleeding is the typical clinical manifestation of a patient with hemangiomas in the intestinal tract and the symptoms begin as early as still in childhood, diagnosis is usually made only in adulthood, especially after the third decade of life. As this is a condition of dragged elucidation and diagnosis, often, the patient is submitted to erroneous treatments due to mistakes in the exclusion of differential diagnoses such as inflammatory bowel disease and hemorrhoids⁶.

This case report aims to discuss the case of an outpatient at the hematology service of the Hospital Geral Público de Palmas (HGPP) after being diagnosed with multiple hemangiomatose disease.

CASE REPORT

KRBO, female, student, resident to Nativity-TO, born in 28/04/1984. The patient sought medical care in 2000 presenting multiple hemangiomas throughout the body associated with asthenia. The patient was found to have severe anemia. In the same year, she underwent upper digestive endoscopy (UDE) to investigate if hemangiomas were present in the digestive tract, which showed no alterations. Later, still in 2000, the patient presented hemangiomas in the oral cavity, so a colonoscopy was performed and showed multiple hemangiomas and some polyps, but without evidence of active bleeding. In 2001 she was hospitalized for severe anemia and the following year she started using Noripurum (Iron(III)-hydroxide polymaltose complex) and Folifer (ferrous bisglycinate and folic acid). In an outpatient consultation in June 2002, she reported having an episode of paresthesia in the left leg and arm lasting about six hours, in addition to headache and nausea. A new colonoscopy was performed, which showed multiple hemangiomas in the transverse, descending, sigmoid and rectal colons, and a cranial computed tomography (CT) without abnormalities. During 2002, the doses of Noripurum and Folifer were reduced until their suspension. UDE was performed on 01/08/2004, which showed violate nodulation, similar to those of the palate, in the gastric body. According to the pathological report (AP), the lesion "could not be ruled out as a gastric polyp".

In 2004, the patient reported on consultation that she had blackened stools and epigastric pain and, while her laboratory tests of June, 2004 were Hb:9 and Ht:27, following the melena episode in 2004, her exams changed to Hb:7.3 and Ht:23.1. For this reason, the patient required blood transfusion. One month later, the patient returned referring melena of onset one week earlier and asthenia, with Hb:6.0, needing to be transfused again.

After several outpatient consultations with no complaints or physical changes, in 2005, the patient returned to the outpatient clinic complaining, once again, of darkened stools that began about 24 hours earlier. At the time, the exams were: Hb: 8.5 and Ht: 27.8. The conduct was expectant, and after 15 days, the patient returned with tests of Hb : 5.5 and Ht: 19.7.

In 2006, the laboratory tests and her clinic improved, without melena and with tests reaching Hb: 12 and Ht: 31.6 in March. Then, the patient presented progressive worsening of laboratory parameters, with Hb reaching Hb: 6.8 and Ht: 19 in August. Her exams continued with low Hb and Ht values until there was an improvement in late 2006, reaching Hb: 9.8 and Ht: 30 in November. In 2007 her exams were consistently below Hb: 8 and Ht: 25. In August, 2008, patient presented Hb:4.8 and Ht:18.6, needing once again to be transfused. Laboratory parameters improved with yet another relapse in November 2008, when values fell to Hb:4,5 and Ht:17.

In early 2010, the patient required a new transfusion due to tests showing Hb: 6.8 and Ht: 24.6. In June 2010, a fecal occult blood test was done, coming back as positive. It was then that omeprazole exchanged for lansoprazole, always keeping the Noripurum. In late 2010, the patient underwent a colonoscopy, which demonstrated two polyps in the transverse colon, with characteristics compatible with that of hemangiomas. In 2012, she complained of asthenia and presented Hb: 6 and Ht: 19,9, once again needing blood transfusion. A few months later, another transfusion was required after Hb and Ht parameters worsened. In July 2012, a hemicolectomy was scheduled for the removal of the bleeding polyps evidenced in colonoscopy in 2010. However, the surgery was not performed and the patient required a new transfusion in August of that year. A new colonoscopy was performed in December 2012, once again showing 1.5 cm hemangiomas in the sigmoid colon and 1-2.5 cm in the transverse colon liver angle. In January 2013, a new transfusion was performed after exams presented Hb: 5.5 Ht: 10.9. The rest of the year was uneventful and without further transfusion until November, when she presented Hb: 6.3 and Ht: 19.3, and again in December when Hb: 7.3. Colonoscopy was performed in 2013 for resection of the polyps, but this was not performed due to the lack of specific material.

The next transfusion performed on the patient was in 2014, when she presented Hb: 5.4. In August of 2015, a head CT scan was performed (without abnormalities) due to complaints of headache. In October she underwent a new transfusion after complaints of asthenia and tachycardia on exertion, and her exams were Hb: 5.2 and Ht: 21.2. Results of an UDE in 2016: normal esophagus, several sessile and pediculated polyps in the stomach (one removed for biopsy), including one in the anterior wall suggestive of recent

bleeding, and normal duodenum. In addition, a sample of a bleeding pedunculated lesion on the patient's right thigh was sent to biopsy. Anatomopathological results showed the following: fundus gland polyp, negative for *H. pylori*, and capillary hemangioma. In September 2016, the patient was transfused again after complaints of darkened stools and Hb: 6.1, and again in November after Hb: 6.2 and Ht: 18.1. On May 25, 2017, the patient underwent a chest CT, which showed an increase in the cardiac area, a sheet of pericardial fluid and a subcutaneous device in the right pectoral region with a catheter projected over the right atrium, without other changes. An echocardiogram was performed in 2017, which confirmed pericardial effusion, but without signs of diastolic restriction, and an ejection fraction of 74%. Transfusions were performed in July, August, October, November and December of the same year. Magnetic resonance imaging performed in late 2017 was normal. The patient was transfused again in February, May, June, July, August, November, and twice in December 2018. New UDE was requested in April 2017, returning with normal results. In September she started coughing with greenish discharge. CTs of the upper abdomen, chest, and pelvis were requested. The upper abdominal CT returned with no alterations. The thoracic CT had the following results: chest with opacities in left upper and lower right pulmonary lobes, and small pleural effusion on the right side. The pelvic CT had no alterations as well. In October 2018, she presented pulmonary thromboembolism, evidenced by images of right pulmonary artery filling failure, mosaic pulmonary attenuation (suggestive of perfusion alterations) and images suggestive of interstitial thickening or linear atelectasis. Rivaroxaban was exchanged for Warfarin in late 2018. In 2019, the patient had two transfusions done: one in January and another February.

She is currently in use of *Valeriana officinalis*, Ultrafer (polymaltose iron), Propranolol, Atenolol, Omeprazole, Noripurum (Iron(III)-hydroxide polymaltose complex) (no report of dosage and interval by the patient), Xarelto (rivaroxaban), beginning 09/10/2018, Warfarin, started 26/12/2018, Aspirin, 12/03/2019, and finally continuous use of folic acid (could not specify dosage).

DISCUSSION

Hemangiomas are present in approximately 4% to 5% of the population. The frequency amongst women is 2 to 3 times more than men, and the complications also tend to affect more females⁷.

The main risk factors for the complications in individuals with hemangioma, are the location of the tumor and its size. Among the complications, the most commonly found are the formation of ulcerations, bleeding, in cases of hemangiomas located in the gastrointestinal tract, airway and vision compromise, visceral complications and others, depending on the anatomical location, such as auditory canal obstruction and deficit in feeding or sound emission due to location in the oral cavity region⁸.

Large hemangiomas, regardless of their location, deserve special attention due to the high risk of culminating in high-output heart failure⁹.

With regards to methods of diagnostic imaging, endoscopic tests such as fiberoptic bronchoscopy, endoscopy and colonoscopy can view hemangiomas located on mucosal surfaces of the upper airway and gastrointestinal tract and are recommended in these cases. Magnetic resonance imaging is the most accurate noninvasive exam for the diagnosis of hemangiomas, with sensitivity ranging from 50 to 70% and specificity from 90 to 95%¹⁰. The diagnosis of colorectal hemangioma is suspected on colonoscopy, with the presence of vascular ectasia. A biopsy is less used in these cases due to the greater risk of bleeding, being reserved only for patients with atypical lesions¹¹.

Cervical and rectal hemangiomas are classified into two categories: polypoids and non-polypoids. The non-polypoid form is the most common and has a pervasive pattern of involvement. In these cases, the therapy employed is the resection of the affected segment. When they are located in the sigmoid and rectum, the technique of low colorectal or colon-anal anastomosis is employed to preserve the anal sphincters. In small polypoid lesions, endoscopic resection may be employed¹².

Regardless of the treatment of choice, it is essential to emphasize the importance of knowledge of this pathology in order to highlight the possible differential diagnoses, recognize the main complications and thus establish the most appropriate treatment plan and therapeutic approach for each case of hemangioma disease.

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