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Case Report

Diagnostic and Treatment with Propranolol of Diffuse Cavernous Hemangioma of the Rectum in Children: A Case Report

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Abstract

Diffuse cavernous hemangioma of the rectum (DCHR) is a benign vascular malformation, it is a rare entity that occurs mainly in children and young adults. Most patients are initially misdiagnosed for a long time with hemorrhoids, colitis, polyps, and rectal varices secondary to portal hypertension, which often delays treatment. The treatment proposal for this pathology is usually surgical resection of the affected segment, sclerotherapy or ligation of the internal iliac artery.

We report the case of a schoolgirl with recurrent rectal bleeding who was initially diagnosed with hemorrhoids secondary to constipation and who was thereafter treated with propranolol for three years with the subsequent clinical remission.

This is the first case reported in the medical literature of rectal cavernous hemangioma treated with a beta-blocker (propranolol) with a good clinical response, avoiding initial surgical management as previously proposed.

Keywords: *Propranolol; Cavernous Hemangioma; Rectum*

Introduction

DCHR is a benign vascular malformation, which affects young adults and children between 5 and 25 years of age [1], whose pathogenesis has not been fully clarified. This lesion was first described by Phillips B. in 1839 [2] and since then about 350 cases have been described in the medical literature [3]. The most frequently seen clinical manifestation is recurrent painless rectal bleeding of varying intensity, sometimes it can lead to massive bleeding that can endanger the patient's life, so the early diagnosis of this pathology is of vital importance. The lack of knowledge of this entity leads to delay in diagnosis and incorrect treatments with the subsequent morbidity [4].

We report a case of rectal hemangioma that was treated with propranolol for three years with a good clinical response, and that was previously diagnosed with hemorrhoids, avoiding the need for resection.

Case Report

A 6-year-old patient who was evaluated in another medical center for presenting stools with red bloodletting in variable quantity and intermittently since she was 1 year old, for which she was diagnosed with external hemorrhoids and received treatment based on Polyethylene glycol 3350 and topical cinchocaine, without remission of bleeding. Later she attended the external consultation of gastroenterology at the Metropolitan Hospital at the age of 2 years 4 months. It was indicated to maintain treatment with Polyethylene glycol and policresulene ointment with cinchocaine. The patient went back to the hospital at the age of 6 years 4 months because the symptoms

persisted. She had a family medical history of an aunt with a stroke due to a hemangioma. In the physical examination, we obtained the following data: weight: 16.5 kg (p 5%), height: 107 cm (p 5%), BMI: 14.4 kg/m^2. Cardiopulmonary auscultation was normal, visceromegaly was not evident on abdominal examination, joint hyperlaxity was observed. Grade III hemorrhoids were present in the perianal region (See figure 1).

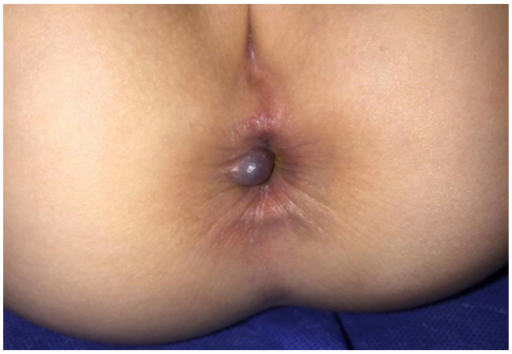

Figure 1: *Grade III hemorrhoids in perianal region.*

A CT- angiography was performed showing a transmural thickening at the rectal level with intramural phleboliths, rectal echoendoscopy showed a hypoechogenic lesion, heterogenous submucosal images and in the rectosigmoidoscopy several lesions in the submucosa with protrusion were observed in the bluish rectal light, these lesions collapsed with the insufflation of air (Figure 2-4). Subsequently, angio CT was performed which showed asymmetric thickening of the rectum with calcifications and adjacent ganglia.

Upon diagnosis of rectal hemangioma, treatment with propranolol was started at 2 mg/kg/day, keeping the heart rate below 80 beats per minute. At the moment the patient is 9 years old and has been with treatment for almost 3 years without any evidence of bleeding or the presence of hemorrhoids.

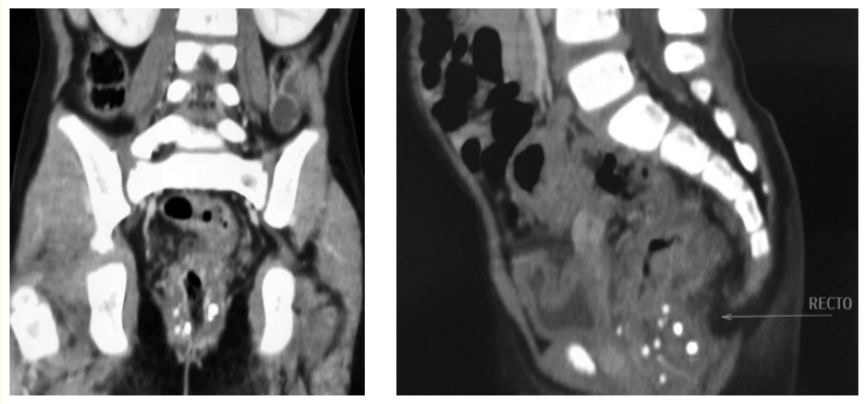

Figure 2 and 3: *CT of the rectum showing intramural phleboliths.*

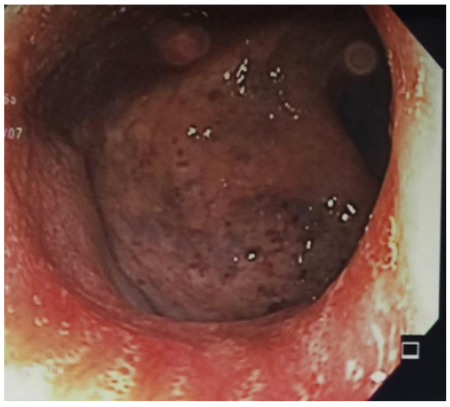

Figure 4: *Colonoscopic findings; lesions in the submucosa with protrusion were observed in the bluish rectal light.*

Discussion and Conclusion

Rectal bleeding is a frequent cause of consultation and DCHR should be taken into consideration as it is commonly misdiagnosed as proctitis, internal hemorrhoids, inflammatory bowel disease or polyps [5,6]. From the first reports of this disease in 1839 until 2012 there were approximately 350 cases reported in the literature [4,6].

The diagnosis even though difficult, is necessary as DCHR has an increased risk of massive bleeding, although around 75% of patients present with anemia and recurrent painless rectal bleeding similar to our patient [5,6]. The diagnosis of DCHR should include a complete clinical history, endoscopic findings, and imaging.

Total colonoscopy is the most important examination for confirming the diagnosis [4]. The main endoscopic findings are multiple bluish polypoid nodules with vascular congestion, our patient had multiple polypoid protrusions with a bluish mucosa that is compatible with the described findings [6]. Additionally, it should be noted that findings in the colonoscopy might be misinterpreted because vessels might become obstructed by thrombi leading to rectal ischemia and inflammatory changes as edema and mucosal ulceration [5]. Endoscopic ultrasound showed a heterogeneous lesion with hypoechoic areas, this is secondary to the sponge-like nature of the cavernous hemangioma [6]. A biopsy was not performed in our patient and it has been contraindicated due to a higher risk of severe hemorrhage.

The patient presented a transmural wall thickening in the rectum with phleboliths in the computed tomography that is compatible with the findings described by Veloso., *et al* [6]. On the other hand, an MRI might also be a satisfactory diagnostic imaging study with less radiation exposure, a higher resolution and much more specific signal intensity of the DCHR [4,5].

The definitive treatment is surgical by pull-through transection and colo-anal anastomosis with a sphincter-saving procedure. Other alternative treatments such as sclerosing injection, endoscopic mucosal resection, and radiotherapy do not appear to be successful. Another treatment for patients that are not eligible for surgery include injections with n–butyl–2–cyanoacrylate, this has only been tested in adults with variable responses [5-7].

Usually, hemangiomas show spontaneous regression, but DCHR are rare and do not behave in this manner, surgical therapy is the treatment of choice [8].

Considering the age of our patient and given the fact that other hemangiomas have been successfully treated with propranolol, we decided to start the patient with 2 mg/kg/day of propranolol with a previous cardiologic assessment. The patient also received PEG 3350

to maintain soft stools and prevent ulcers in the hemangioma. With this therapy, the patient has remained asymptomatic with no further episodes of rectal bleeding. A new CT scan was performed after 3 years of the diagnosis and it showed no significant modifications from the previous study. But since the patient experienced clinical remission no therapy modifications have been indicated.

Propranolol has not been indicated in this specific type of hemangioma but it might be a solution to control the symptoms and to contain the progression of the hemangioma´s growth. Further studies are needed to evaluate this treatment as a possible conservative therapy that could imply avoiding surgery at a young age and the possible complications of surgical treatment.

Bibliography

1. Hakeem AA., *et al*. "CT findings in diffuse rectosigmoid cavernous haemangioma. A case report". *Journal of Gastrointestinal Surgery* 13.5 (2009): 1017-1018.
2. Phillips B. "Surgical cases". Lond Med Gaz 23 (1839): 514-517.
3. Kimura S., *et al*. "Cavernous hemangioma in the ascending colon treated by endoscopic mucosal resection". *Journal of Gastroenterology and Hepatology* 22.2 (2007): 280-281.
4. Wang HT., *et al*. "Diagnosis and treatment of diffuse cavernous hemangioma of the rectum: report of 17 cases". *World Journal of Surgery* 34.10 (2010): 2477-2486.
5. Chatu S., *et al*. "A rare cause of rectal bleeding masquerading as proctitis". *Journal of Crohn's and Colitis* 7.3 (2013): e99-e102.
6. Veloso N., *et al*. "A rare cause of rectal bleeding". *Gastroenterology* 143.5 (2012): e8-e9.
7. J Zurakowski., *et al*. "Diffuse cavernous hemangioma of rectosigmoid colon treated with n−butyl−2−cyanoacrylate injections". *Endoscopy* 40 (2008): 120-121.
8. Rieu PN. "A cavernous haemangioma of the colon as the cause of rectal bleeding in childhood". *Nederlands Tijdschrift voor Geneeskunde* 148.17 (2004): 1409-1410.

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