We present the case of an 18-year-old male patient that was referred to our Gastroenterology Department with a history of intermittent painless hematochezia since childhood. During such instances, he was diagnosed with bowel intussusception, eosinophilic gastroenteritis, and inflammatory bowel disease at 4, 6, and 8 years old, respectively. He underwent treatment with 5-aminosalicylic acid for two years, without improvement of symptoms. He was then lost to follow-up until our observation.

His physical examination was unremarkable except for digital rectal examination, which found a nodular compressible mass by the palpating finger. Blood tests revealed a mild iron deficiency anemia (hemoglobin 120 g/L, MCV 84 fL, ferritin 3 ng/mL). The colonoscopy showed an extended reddish and bluish multinodular submucosal mass in the rectum (Fig. 1), suggesting diffuse cavernous hemangioma of the rectum (DHCR). The magnetic resonance imaging showed diffuse thickening of the entire rectum extending into the distal sigmoid with the mesorectum revealing multiple serpiginous structures, corresponding to abnormal blood vessels (axial T2 SPAIR weighted) (Fig. 2).

Fig. 1. Colonoscopic findings: bluish nodular submucosal mass (A); bluish and reddish serpentine varicosities (B and C).

Fig. 2. Axial T1 weighted MRI (A): concentric hypointense thickening of the rectal wall (asterisk) and a heterogeneous mesorectal fat (white arrow). Sagittal T2 weighted (B) and axial T2 SPAIR weighted (C) MRI: diffuse thickening of the entire rectum (asterisk) extending into the distal sigmoid and the mesorectum reveals multiple serpiginous structures with moderate to high intensity corresponding to abnormal blood vessels (white arrows).
After discussion, we considered to perform a sphincter-sparing procedure, namely pull through transection and coloanal anastomosis. However, intervention was ruled out by the patient because of his fear of anal incontinence and permanent colostomy. We adopted a conservative strategy with clinical surveillance and iron supplementation. At present, the patient remains with intermittent rectal bleeding, referring poor quality of life due to his ongoing symptoms.

DISCUSSION

This is a rare case of DHCR. Despite being a benign disease, the management of DHCR requires a sphincter mucosectomy and pull-through coloanal sleeve anastomosis which has become the first-line procedure (1-3). Surgical outcomes are non-expectable in 32% of cases, with permanent sphincter lesion or with incomplete DHCR removal (1). As in this case, surgeons or patients refusal to perform the intervention is common, representing a challenge to the clinical follow-up (4).

REFERENCES