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Case Report

Laparoscopic Swenson Procedure for Diffuse Cavernous Haemangioma of Rectum in Children: A Case Report

S *Chen*, Q *Chen*

Abstract Diffuse cavernous haemangioma of the rectum (DCHR) is a rare benign lesion of vascular malformation. According to previous studies, the most reported major symptoms of DCHR are recurrent painless rectal bleeding or iron deficiency anaemia. We present a case of an 8 years and 11 months boy who was diagnosed with DCHR by magnetic resonance imaging. The patient underwent Laparoscopic-assisted transanal distal sigmoidectomy and sigmoido-anal anastomosis (Laparoscopic Swenson Procedure), and recovered well. After the operation, the child had short-term faecal incontinence and loose stools, which returned to normal after half a year. Regardless of age and symptoms, early and correct diagnosis and treatment of DCHR and postoperative restoration of normal anal function are necessary. Laparoscopic Swenson procedure may be a good option for the treatment of DCHR.

Key words *Children; Diffuse cavernous haemangioma; Laparoscopic Swenson; Rectum*

Introduction

Diffuse cavernous haemangioma of the rectum (DCHR) is relatively rare,the most common symptoms are recurrent painless rectal bleeding or iron-deficiency anaemia,[1-6] some patients were misdiagnosed as internal haemorrhoids or treated as ulcerative colitis or Crohn's disease.[7,8] Current diagnostic modalities are abdominal X-ray, computed tomography (CT), magnetic resonance imaging (MRI), endoscopic ultrasonography, and colonoscopy, all of which have their own advantages. Compared with the high recurrence rate of palliative care, surgical treatment is the first choice.[9,10] Laparoscopic Swenson Procedure has been used to treat Hirschsprung's disease and its related diseases. However, this approach has not been evaluated in the treatment of patients with DCHR.

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Case Presentation

This case reports a boy aged 8 years and 11 months who first presented with recurrent painless rectal bleeding 7 years ago. In early childhood, bleeding symptoms were thought to be due to anal fissure, and three colonoscopies performed 2 years earlier in the gastroenterology department showed no significant abnormal lesions in the rectum. The patient was admitted to our hospital with recurrent haematochezia. He had no significant medical or family history.

His laboratory tests revealed a haemoglobin level of 10.2 g/dL. Three colonoscopies showed no apparent abnormal lesions. Abdominal MRI revealed the possibility of vascular malformations in the rectum wall and around the rectum (Figure 1).

The patient was diagnosed with DCHR and underwent laparoscopic-assisted transanal distal rectosigmoid resection and sigmoidal-anal anastomosis (Laparoscopic Swenson Procedure).

Specific surgical procedures: A Trocar with a diameter of 10 mm was placed at the umbilical margin, pneumoperitoneum was established by pneumoperitoneum needle puncture (12 mmHg), the main vision lens was inserted, and a Trocar with a diameter of 5 mm was placed in the right lower abdomen and the right upper abdomen. Exploration revealed a diffuse distribution of intestinal wall haemangioma in the distal rectum and sigmoid colon, with prominent anterior wall, and a distribution of vascular malformations in the pelvic soft tissues (Figure 2). The distal mesentery of sigmoid colon and mesorectum were removed and separated to the pelvic floor under the premise of retaining the corresponding vascular arch with forceps and ultrasonic scalpel respectively. After changing to lithotomy position. The colon was pulled out of the anus with a tissue clamp over the peritoneal reflex. The bowel was cut open in a circular way, and about 15 cm of the bowel was pulled out. The edge of the rectum to the dentate line was resected, 1.5 cm of the anterior wall and 0.5 cm of the posterior wall of the rectum were retained. Reverse oblique resection of the sigmoid colon was performed, and the diseased sigmoid colon was completely removed without obvious haemangioma-like tissue at the edge of the resection (resection range: diseased sigmoid colon to low rectum). An end-to-end heart-shaped anastomosis of the proximal sigmoid colon to the rectum was performed

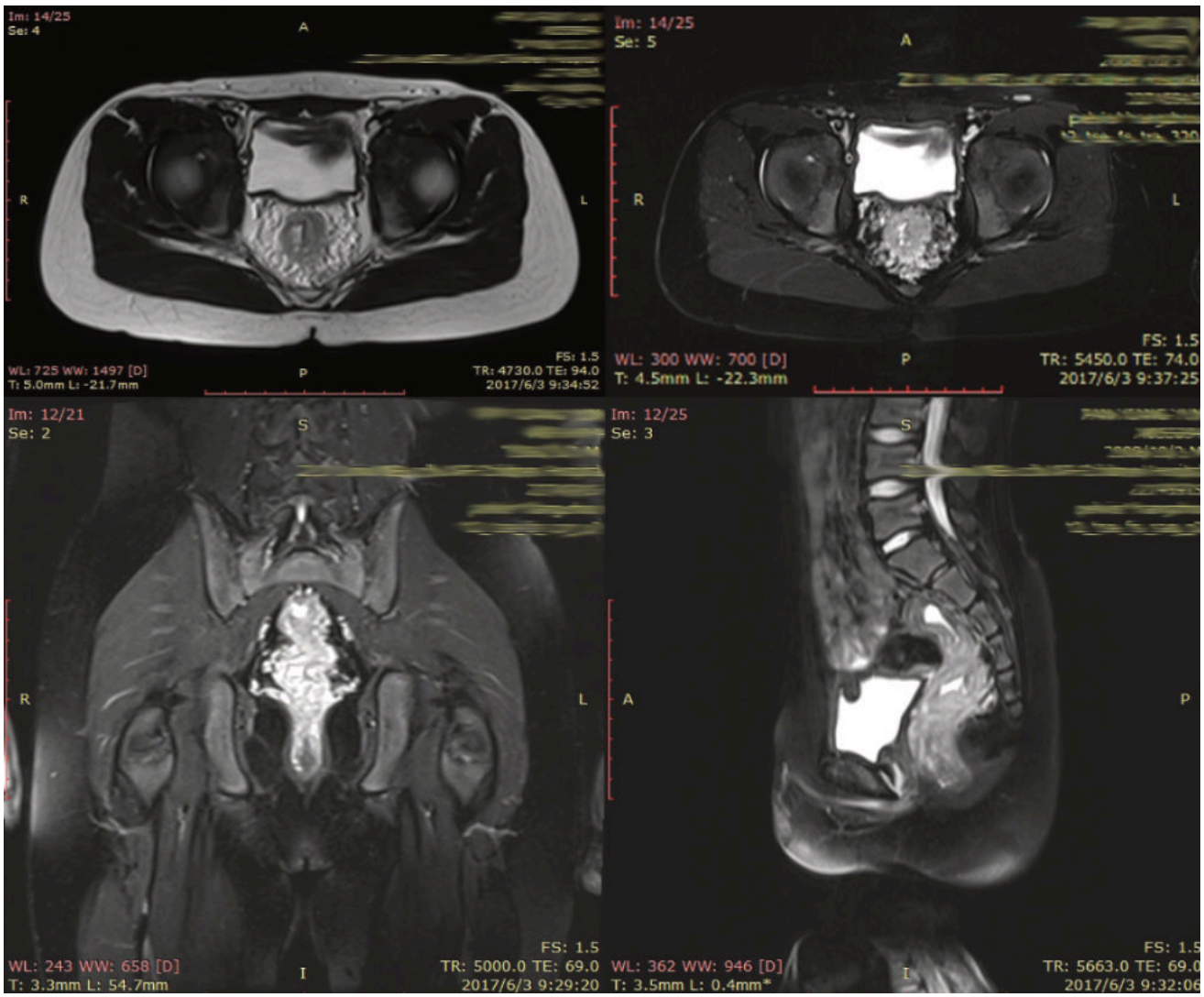

Figure 1 Abdominal MRI showed that the rectal wall was significantly thickened with obvious T1 and T2 signal changes, the inner wall of the rectum was irregular, and the outer side of the rectum was coarse. A thick reticular mass of long T1 and long T2 signals was seen around the rectum. The possibility of rectal wall and perirectal vascular malformations was considered.

(Figure 3). After examination for no active bleeding, anastomotic stenosis and leakage, an anal decompression tube was placed.

The operation time was 170 minutes, blood loss was 20 ml, intraoperative blood transfusion was not required, and the patient recovered well after the operation. The gastric tube was removed on the 3rd postoperative day, and oral administration was started on the 5th postoperative day. The anal decompression tube was removed on the 8th postoperative day and the patient was discharged on the 14th postoperative day. The patient was followed up for 1 week / 1 month / 3 months / 6 months / 13 months / 24 months and no obvious signs of recurrence were found by ultrasound. After the operation, the child had short-term faecal incontinence and loose stools, which returned to normal after half a year.

Discussion

DCHR is a rare benign vascular lesion. It originates from the submucosal blood vessels and is caused by the abnormal development of embryonic mesodermal tissue, accounting for 80% of rectal haemangiomas.[11] Since the first case of rectal haemangioma was reported by Phillips in 1839, about 350 cases have been reported worldwide.[5] It is more common in children and young adults, and the male to female ratio is generally 2:1.[12] According to previous studies, the most commonly reported main symptoms of DCHR are recurrent painless rectal bleeding or iron deficiency anaemia.[1-6] Some patients are misdiagnosed with internal haemorrhoids or treated for ulcerative colitis and Crohn's disease.[7,8] Of course, there are other symptoms including lower abdominal pain, low bowel obstruction and constipation.

The clinical manifestations of DCHR are non-specific symptoms. However, auxiliary examinations have their own characteristics. In the abdominal X-ray findings, closely packed venules were seen in some cases.[13] With the development of colonoscopy, CT, rectal ultrasound, and MRI, abdominal radiography has been replaced by them. Colonoscopy is an important examination to visually

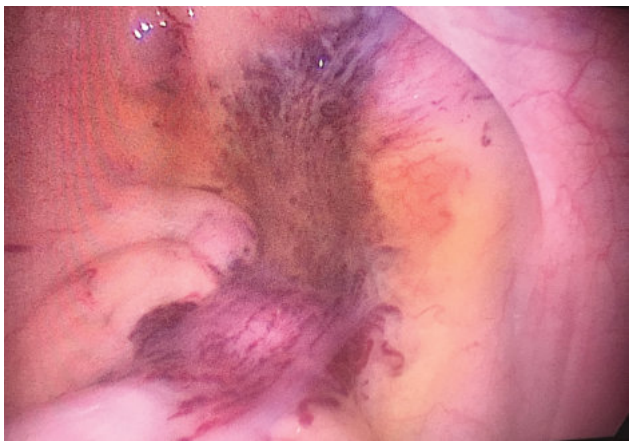

Figure 2 Laparoscopy showed a diffuse distribution of haemangiomas in the distal rectal sigmoid wall.

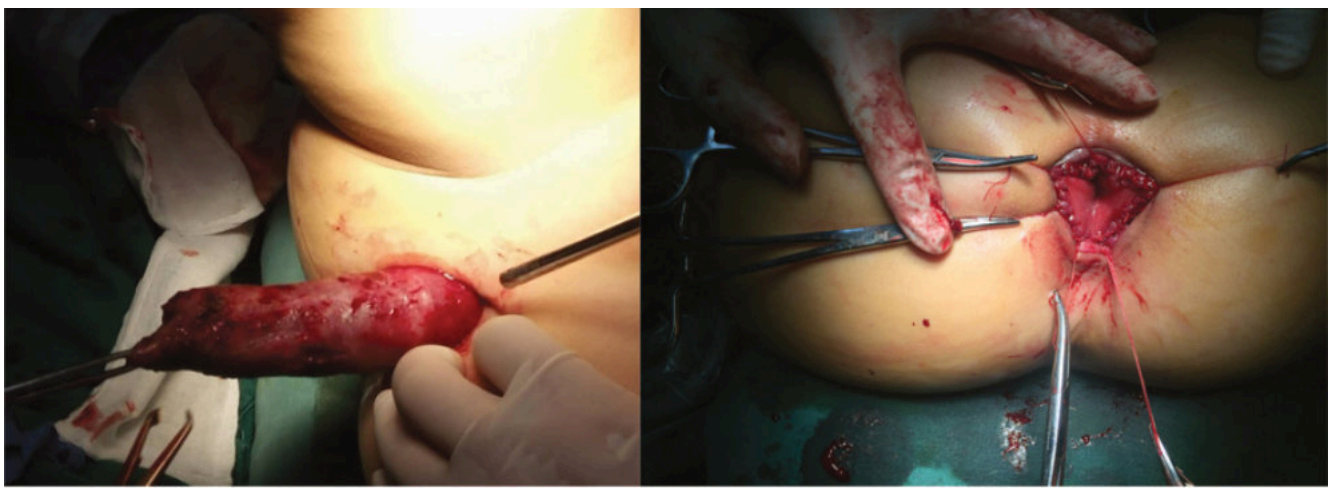

Figure 3 The colon was pulled out of the anus above the peritoneal reflex, the bowel was cut in a circular way, and about 15 cm of the bowel was pulled out. The margin of the rectum to the dentate line was resected, and 1.5 cm of the anterior wall and 0.5 cm of the posterior wall of the rectum were preserved. After the sigmoid colon was cut in the reverse beveling plane, an end-to-end heart-shaped anastomosis was made with the rectum.

observe rectal haemangioma and its lesion length. The typical findings on colonoscopy are congestion and swelling of the rectal mucosa, with soft, distorted, enlarged blue-red or dark-red submucosal vascular lesions.[14,15] CT is another important examination for the diagnosis of rectal haemangioma. Typical CT findings are thickening of the rectal wall, serpentine enlargement of blood vessels around the rectum, and sometimes multiple calcifications in the pelvic region.[16] MRI is superior to CT because of its ability to produce high-resolution and multidimensional soft tissue images.[16] Therefore, it can provide accurate information on the exact size of the rectal haemangioma, the involvement of the sphincter, and the involvement of adjacent structures.

At present, the treatment of DCHR is generally divided into palliative treatment and surgical treatment. The common palliative treatment is low-dose radiation therapy, cryotherapy, vascular ligation therapy, sclerotherapy, and interventional angiography occlusion.[5,6,16,17] Palliative treatment can reduce the amount and frequency of rectal bleeding, but it is prone to recurrence. Therefore, non-surgical treatment of rectal haemangioma is not recommended.[9,10]

Surgical treatment is the first option for DCHR. Among them, endoscopic surgery is widely recommended because of its short operation time and small surgical injury. The selection of endoscopic surgery is also controversial. It is thought that the main advantage of EFTR (endoscopic full-thickness resection), as compared with endoscopic mucosal resection and ESD (endoscopic submucosal dissection), is the lower risk of residual or recurrent haemangioma because haemangiomas sometimes invade into or completely exceed the muscular layer. EFTR (endoscopic full-thickness resection) may be a better treatment option for colorectal cavernous haemangioma involving the muscularis propria layer.[4,18,19] However, the scope of endoscopic surgery is small, and it may not be able to do anything for DCHR cases that spread to the pelvis.

Therefore, the disadvantages of traditional surgery are undoubtedly obvious. Among the traditional surgical options, compared with TME (total mesorectal excision), it is difficult to completely remove the distal rectal lesions, TaTME (transanal total mesorectal excision) can directly remove the lesions deep to the dentate line, which has more advantages.[1,20]

So far, there are few reports about laparoscopic surgery as a further improvement of traditional surgery. Leal et al reported 2 cases of laparoscopy-assisted bowel resection for DCHR, after which the children returned to normal without abnormal defecation and bloody stools.[2] Fujii et al introduced robot-assisted surgery and suggested that robotic surgery has certain advantages over laparoscopic surgery, including three-dimensional imaging, no vibration, dexterity in motion scaling, and minimal bleeding. But robotic surgery requires a longer operation time.[21]

In this case, due to the child's young age and small operable space at pelvic floor and anus, laparoscopic surgery was innovatively combined with TaTME, and Laparoscopic Swenson's operation in Hirschsprung's disease surgery was used for reference. This is the first report on the treatment of DCHR by laparoscopic-assisted transanal distal sigmoidorectal resection and sigmoidoanal canal anastomosis.

The amount of bleeding during the operation was less and the patient recovered satisfactorily. Although the defecation function was abnormal in a short time after operation, the long-term defecation function was good. Based on the susceptibility age of DCHR, this result is acceptable. This experience suggests that the Laparoscopic Swenson procedure may be a new option for the treatment of DCHR, especially in children.

Disclosure

The authors declare no conflict of interest.

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