

Gastrointestinal hemorrhage caused by adult intussusception secondary to small intestinal tumors

Two case reports

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Abstract

Rationale: Adult intussusception is rarely observed, and the clinical manifestations are very atypical. The most common symptom is abdominal pain, while the incidence of hematochezia is relatively low. We report two cases of adult intussusception secondary to small intestinal tumors with gastrointestinal hemorrhage as the main symptom.

Patient concerns: Two men aged 19 and 54 years were successively referred to our department due to intermittent hematochezia. The hemoglobin levels of the two patients declined progressively, and conservative treatment was ineffective.

Diagnoses: The first patient underwent an abdominal computed tomography angiography examination, which showed that the intestine and its mesentery were tortuous, suggesting an intra-abdominal hernia or intussusception. The second patient underwent an abdominal computed tomography examination, which suggested a high possibility of an intussusception. The two patients were diagnosed as adult intussusception caused by small intestinal tumors.

Interventions: Emergency laparoscopic explorations were performed. Enterointeric intussusceptions caused by ileal tumors were found during surgery. Reduction of the intussusceptions and resection of the ileal tumors were performed.

Outcomes: The patients recovered well after surgery, and postoperative pathology showed that the tumors were a vascular hamartoma polyp and a lipoma.

Lessons: Adult intussusception is very rare, particularly with gastrointestinal hemorrhage as the main symptom. Isolated hamartoma polyp is a rare cause of intussusception in adults. The clinical manifestations of adult intussusception are very atypical, and thus, making a preoperative diagnosis is difficult. Abdominal CT or CTA is an effective diagnostic method for adult intussusception. For adult patients with gastrointestinal hemorrhage caused by intussusceptions, active surgery should be performed when conservative treatment is not effective. Laparoscopic surgery is a safe and effective treatment for adult intussusceptions caused by benign diseases.

Abbreviations: APTT = activated partial thrombin time, CT = computed tomography, CTA = computed tomography angiography, INR = international normalized ratio, MRI = magnetic resonance imaging, PT = prothrombin time.

Keywords: adult intussusception, gastrointestinal hemorrhage, laparoscopic surgery, small intestinal tumor

1. Introduction

Intussusception refers to the telescoping of a part of the intestine and its mesentery into the adjacent intestine, causing symptoms of acute intestinal obstruction in the clinic. Intussusception may lead to intestinal ischemia, intestinal necrosis, or intestinal perforation if

not treated in time. Intussusception occurs most frequently in children aged 5 to 9 months.^[1] Conversely, the incidence of adult intussusception is relatively low, accounting for approximately 5% of all intussusception cases and 1% to 5% of all adult bowel obstruction cases.^[2–4] Most instances of adult intussusception are caused by lesions in the intestinal cavity, including polyps, malignant tumors, and Michael diverticulum, etc.^[2,5,6] The symptoms of adult intussusception are not obvious, and the typical characteristics of abdominal pain, hematochezia, or an abdominal mass that are present in intussusceptions in children are rare in adults, only 12% to 13% of whom have hematochezia.^[5,7] For patients with intussusception caused by small intestinal tumors, the presenting complaints include abdominal pain, abdominal distension, nausea, vomiting, hematochezia, and melena.^[8–12] Recently, 2 cases of adult intussusception with gastrointestinal hemorrhage as the main symptom were treated in our hospital, both of which were caused by secondary intussusception of small intestinal tumors. The cases are reported below, and the literature is reviewed.

2. Case reports

2.1. Case 1

A 19-year-old man was initially referred to our department on November 29, 2017, due to intermittent hematochezia for 2 days.

Editor: N/A.

Statement: Informed consent has been obtained in writing from the patients and is available for review by the editor.

The authors have no conflicts of interest to disclose.

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Medicine (2018) 97:34(e12053)

Received: 27 April 2018 / Accepted: 2 August 2018

<http://dx.doi.org/10.1097/MD.00000000000012053>

During the previous 2 days, the patient had intermittent hematochezia 3 times, with a blood volume of approximately 200 mL each time. The patient was in good health previously and had no surgical history and no chronic diseases, such as hypertension or diabetes. No positive signs were found on an abdominal examination. The abdomen was soft, and no tenderness, rebound tenderness or muscle guarding was observed. Blood tests showed that the patient's white blood cell count was $8.62 \times 10^9/L$ (4.00–10.00), the red blood cell count was $3.01 \times 10^{12}/L$ (4.00–5.50), the hemoglobin level was 60 g/L (120.0–160.0), the prothrombin time (PT) was 14.4 seconds (9.4–12.5), the activated partial thrombin time (APTT) was 25.1 seconds (25.4–38.4), and the international normalized ratio (INR) was 1.38 (0.80–1.20). The rest of the blood test results were generally normal. Gastroscopy showed chronic non-atrophic gastritis accompanied by erosions. After admission, the patient was treated with antacids, hemostatic drugs, blood transfusions, and other symptomatic treatments. The patient was given a blood transfusion of 4 units of red blood cells and 400 mL of plasma, and the next morning the patient's blood tests showed that the hemoglobin level was 53 g/L. An abdominal computed tomography angiography (CTA) examination was performed. The CTA showed that the intestine and its mesentery were tortuous in the right upper abdomen and that the end of the mesentery was ring-shaped, which suggested an intra-abdominal hernia or intussusception (Fig. 1). The CTA also showed a secondary intestinal obstruction. Because the patient's hemoglobin continued to decrease and conservative treatment was ineffective, an emergency laparoscopic exploration was performed. During surgery, an ileal tumor was found 40 cm from the

ileocecal region in the right upper abdomen. An intussusception had occurred involving the proximal ileum and the tumor, and the length of the intussusception was approximately 18 cm. The proximal intestine was significantly dilated, and a large amount of bloody stool was seen in the lumen of the distal intestine (Fig. 2). Reduction of the intussusception and resection of the ileal tumor were performed. The resected tumor was a finger-like projection approximately 4 cm \times 2 cm \times 1.5 cm in size. Ulceration and necrosis were seen at the top of the tumor (Fig. 3). The surgery was successful, and the patient recovered well. Postoperative pathology showed that the small intestinal mucosa protruded, and proliferative and dilated veins were noted in the protrusion. Many capillaries were seen on the tumor surface, and proliferative local smooth muscle tissue and some atypical glands were observed, consistent with a vascular hamartoma polyp of the small intestine (Fig. 4).

2.2. Case 2

A 54-year-old man was initially referred to our department on December 27, 2017, due to intermittent hematochezia for 1 day. The patient had intermittent hematochezia twice 1 day previously, with a blood volume of approximately 400 mL each time. The patient was initially admitted to a local hospital, and blood tests showed that the hemoglobin level was 97 g/L. The patient was given a blood transfusion of 3 units of red blood cells, and the hemoglobin level was 71 g/L the next morning. Because the patient still had intermittent hematochezia, he was referred to our hospital. The patient was in good health in the past and denied having smoking and drinking habits. An abdominal

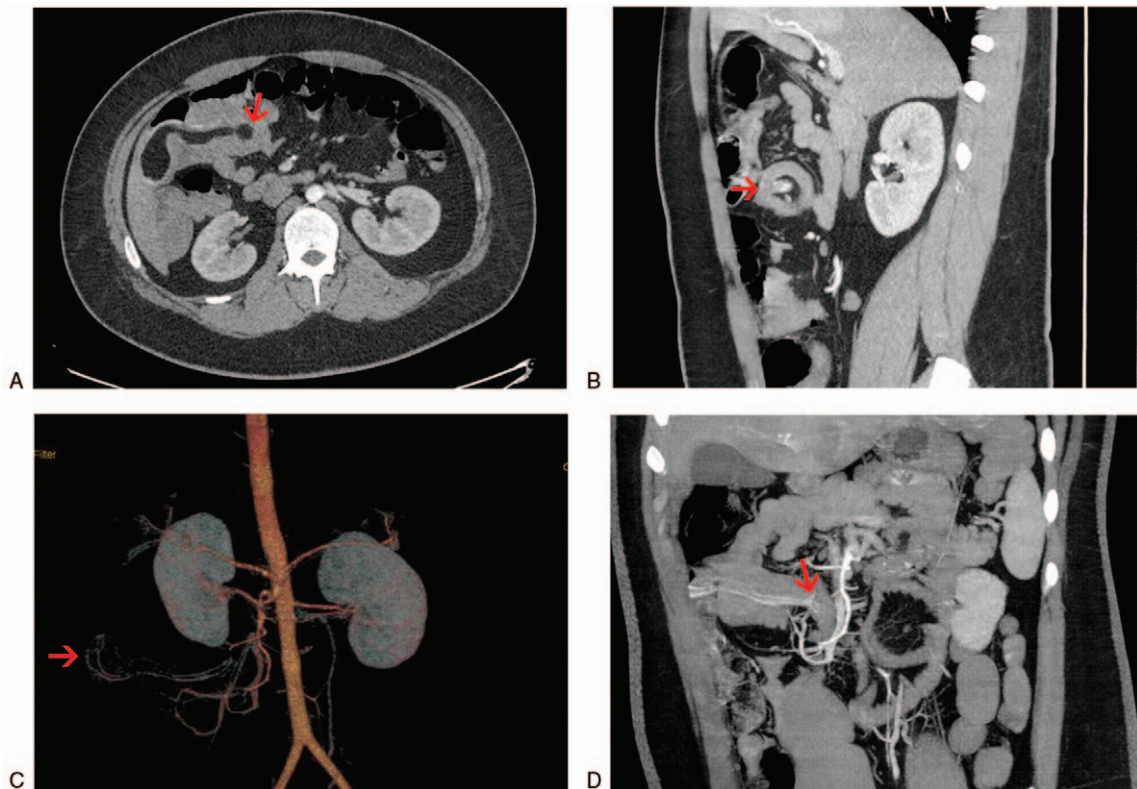


Figure 1. Abdominal CTA showing (A) the tortuous intestine and its mesentery in the right upper abdomen with a ring-shaped appearance (arrow) of the end of the mesentery; (B) the target sign of the intestinal canal (arrow); (C) the tortuous mesenteric vessels after vascular reconstruction; and (D) the suspicious bleeding point (arrow). CTA=computed tomography angiography.

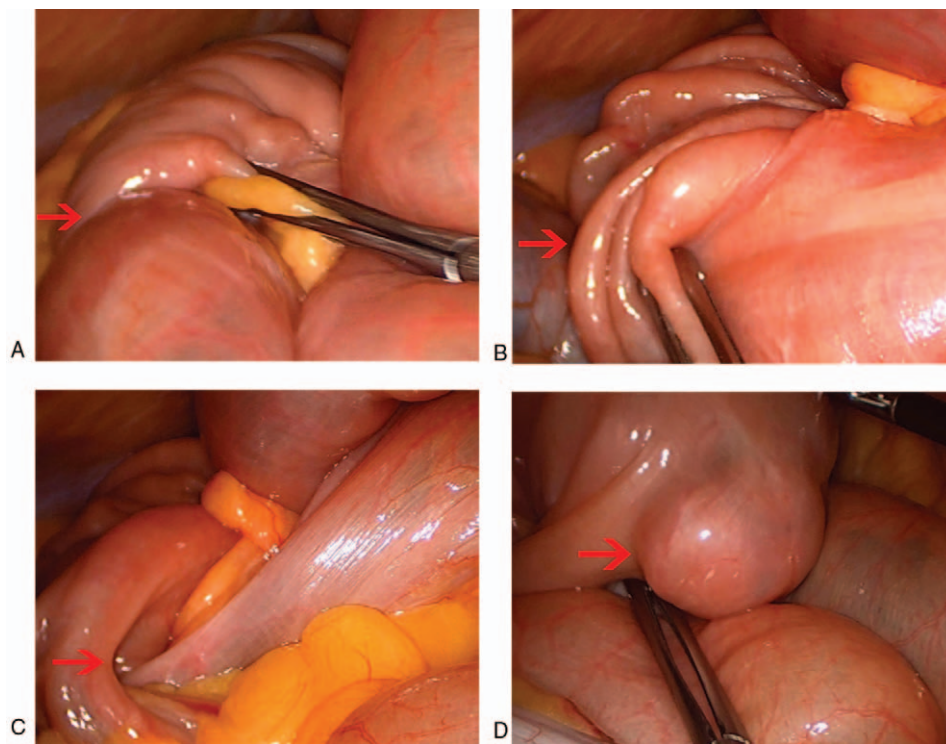


Figure 2. Laparoscopic exploration revealing (A) an enteroenteric intussusception in the right upper abdomen (arrow); (B) the intussuscepted small intestine (arrow); (C) the distal end of the intussusception (arrow); and (D) a tumor in the ileum (arrow).

examination also showed no positive signs. Blood tests showed that the patient's red blood cell count was $2.93 \times 10^{12}/L$, the hemoglobin level was 85 g/L, the PT was 16.8 seconds, the APTT was 48.1 seconds, and the INR was 1.37. An abdominal computed tomography (CT) examination showed that part of the small intestine and its surrounding mesentery in the left lower left abdomen were telescoped into the enteric cavity and had a concentric circular appearance, suggesting a high possibility of an intussusception (Fig. 5). The patient was given a blood transfusion of 3.5 units of red blood cells and 600 mL of plasma. However, he still had intermittent hematochezia, and blood tests showed that the hemoglobin level was 49 g/L 8 hours after admission. A laparoscopic exploration was performed immediately. An ileal tumor approximately 5 cm \times 3 cm \times 2 cm in size was found 20 cm from the ileocecal region in the left lower

abdomen. An intussusception had occurred involving the proximal ileum and the tumor, the length of which was approximately 10 cm (Fig. 6). The tumor was thought to be benign based on its appearance (Fig. 7), and reduction of the intussusception and resection of the ileal tumor were performed. Postoperative pathology showed that the tumor was a small intestinal lipoma with an intussusception and that ulcers had formed on the tumor mucosal surface (Fig. 8). The patient recovered well and was discharged 5 days after surgery.

3. Discussion

Intussusception was first reported by Barbette of Amsterdam in 1674^[13] and was later reported by John Hunter in more detail in 1789.^[14] In children, approximately 95% of intussusceptions

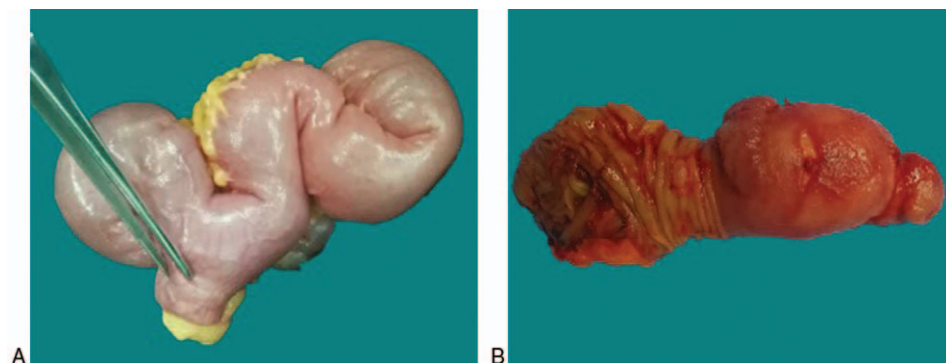


Figure 3. (A) The ileal tumor that caused the intussusception. (B) The resected tumor was a finger-like projection. Ulceration and necrosis were seen at the top of the tumor.

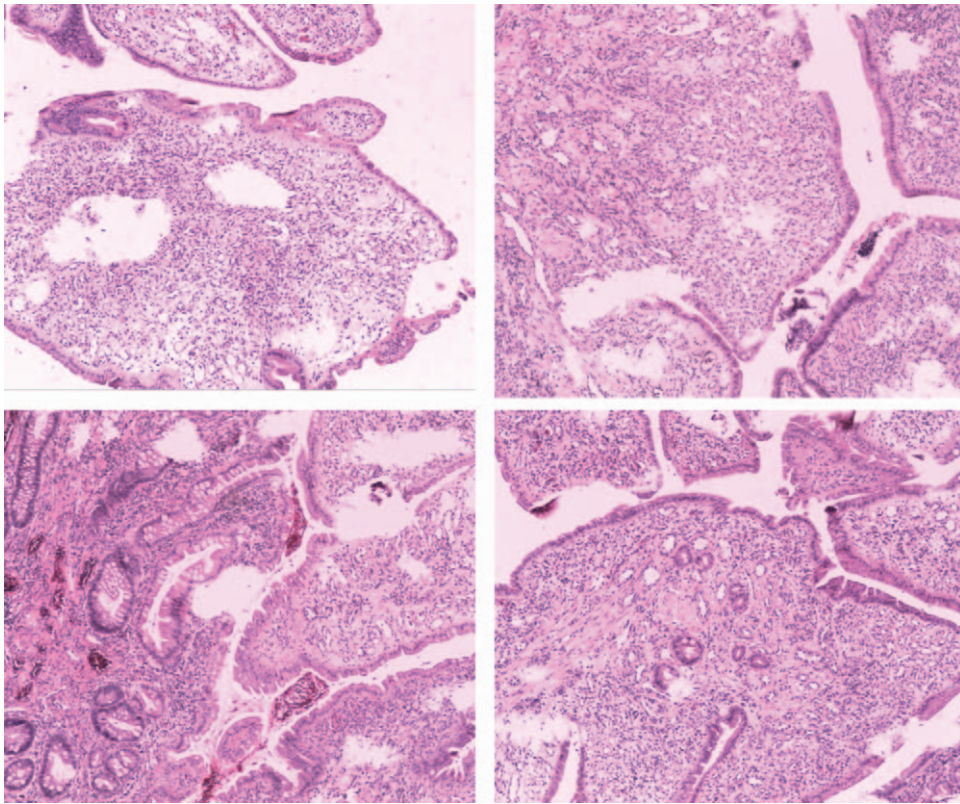


Figure 4. Postoperative pathology showed that the tumor was a vascular hamartoma of the small intestine.

are idiopathic and are not associated with organic lesions and thus can be reduced by an air enema or a barium enema. Conversely, approximately 80% of adult intussusceptions are secondary, with a clear cause that often requires surgical treatment.^[15] A clear sex difference occurs in intussusceptions in children, with the incidence of intussusception in boys more than twice that in girls.^[16,17] In adults, no significant difference in morbidity is found between men and women, and the average age of onset is 50 years.^[18,19] Both patients in this report were adults, and the intussusceptions were caused by small intestinal tumors.



Figure 5. Abdominal CT showing the target sign of the intestinal canal (arrow). CT=computed tomography.

Intussusception can be divided into 4 types according to the leading point (the enteroenteric, ileocolic, ileocecal, and colocolic types). In adult intussusceptions, the enteroenteric type is the most common, possibly due to the relatively high activity of the small intestine and the relatively high incidence of small intestinal tumors in adults. Wang et al^[18] analyzed 41 cases of adult intussusceptions and found that 45.5% were the enteroenteric type and 34.1% were the ileocolic type. The study by Onkendi et al^[20] showed that the enteroenteric type accounted for 70% of all intussusceptions. Small intestinal intussusceptions are mainly caused by lesions in or outside the intestine, including adenomatous polyps, lipomas, stromal tumors, lymphoma, intestinal inflammation, Michael diverticulum, postoperative intestinal adhesions, adenocarcinoma, and metastatic carcinoma. Some rare lesions also occur, such as a neuroendocrine tumor,^[9] angiolipoma,^[21] myoepithelial hamartoma,^[22] and appendiceal mucinous cystadenoma,^[23] which have only been described in case reports. Adenocarcinoma accounts for approximately 30% of small intestinal intussusceptions, whereas colonic intussusceptions are mainly caused by malignant tumors.^[15,24] Barussaud et al^[19] found in 44 adult patients with intussusception that 37% of the small bowel lesions were malignant tumors and 58% of the colonic lesions were malignant tumors. An older age of the patient is associated with a higher probability of the primary disease being a malignant tumor. In this report, the 2 cases of intussusception were caused by benign small intestinal neoplasms; one case was an isolated hamartoma polyp, and the other case was a lipoma, and both intussusceptions were of the enteroenteric type. Hamartoma polyps are mainly found in hamartoma polyp syndromes, including Peutz-Jeghers syndrome, juvenile polyposis, Cronkhite-Canada syndrome, and Gardner

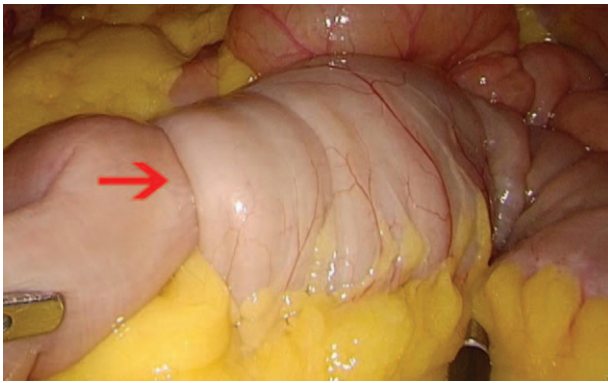


Figure 6. Laparoscopic exploration revealing an enteroenteric intussusception in the left lower abdomen (arrow).

syndrome. Among these syndromes, Peutz-Jeghers syndrome mainly manifests as pigmented spots on the skin and mucous membranes, multiple polyps in the gastrointestinal tract, and a family history, which easily leads to intussusception.^[2,5,26] Based on the medical history, family history, physical examination, auxiliary examination, surgical findings, and postoperative pathology, we considered the first case to be an isolated hamartoma polyp. Intussusceptions caused by solitary hamartoma polyps are very rare, with few reports in the literature.^[8,27] Lipomas can occur in any part of the gastrointestinal tract, of which the colon is the most common site, and are common benign tumors that lead to intussusception. Taraneh Azar study of 58 cases of adult intussusception found 3 cases of intestinal intussusception and 3 cases of colonic intussusception caused by lipomas.^[2] In another study that included 196 adult intussus-



Figure 7. The resected ileal tumor that caused the intussusception. Ulcers had formed on the tumor mucosal surface.

ceptions, 10 primary lesions were intestinal lipomas, accounting for 21% of all benign tumors.^[20] Therefore, when adult intussusception is suspected, the clinician should enquire about the medical history in detail, examine the body carefully, perform related examinations and identify the cause of the disease to provide timely and effective treatment.

Intussusceptions in children are mostly acute and manifest as a triad of cramping abdominal pains, bloody stool, and a palpable tender mass, whereas the clinical manifestations of adult intussusception are very atypical; adult patients who present with the same triad of symptoms only account for approximately 2% to 10% of cases.^[18,20] Most adult intussusceptions are chronic and are characterized by incomplete intestinal obstruction. The clinical symptoms include abdominal pain, nausea, vomiting, hematochezia, change in bowel habits, and abdominal distension.^[2,18] The most common symptom is abdominal pain, which occurs in 70% to 90% of all cases,^[5,6,18] whereas the incidence of hematochezia is relatively low. The study by Wang et al^[6] of 24 patients with adult intussusception found only 2 patients (8.3%) with hematochezia. Another study involving 46 Asian adults with intussusceptions showed that the incidence of hematochezia was 17.4% (8/46).^[28] However, patients presenting only with hematochezia and persistent decreases in hemoglobin levels are very rare. The symptoms of intussusception in adults may be related to the tumor size and the intussusception length and location.^[28] For example, gastrointestinal lipomas with a diameter <1 cm are usually asymptomatic. When the diameter is >4 cm, intussusception, intestinal obstruction, and gastrointestinal hemorrhage are prone to occur.^[12] Hemorrhage may be associated with a larger tumor diameter and excessive surface pressure on the intestinal mucosa, which leads to mucosal necrosis and ulceration. In this report, 2 patients presented with intermittent bloody stool. Their hemoglobin levels continued to decrease, and conservative treatment was not effective. The tumor diameter was found to be >4 cm during surgery, and the top ulceration of the tumor had eroded a small artery in the intestinal wall, resulting in persistent hemorrhage. For patients with gastrointestinal bleeding in the clinic, the possibility of hemorrhage from a small intestinal tumor should be considered after excluding common causes, such as gastric ulcers, esophageal and gastric varices, and gastric cancer.

Due to the various clinical manifestations of adult intussusception, diagnosis is difficult, and the preoperative diagnostic rate is quite low. Most cases can be identified only during surgery. According to earlier reports, the preoperative diagnostic rate of adult intussusception ranges between 30% and 70%.^[2,29] In the past, colonic intussusception was diagnosed mainly by an air-barium double contrast examination, for which the diagnostic accuracy was only approximately 20%. In recent years, with the application of ultrasound, abdominal CT and other auxiliary examinations, the preoperative diagnostic rate has significantly improved. Chang et al^[28] showed that the preoperative diagnostic rate of adult intussusception was 89.1%. In the study by Honjo et al,^[7] the preoperative diagnostic rate increased to 95.5%. Currently, the most widely used diagnostic methods include ultrasound, abdominal CT, abdominal magnetic resonance imaging (MRI), angiography, and colonoscopy. Abdominal CT is currently recognized as the most sensitive and effective method for diagnosing intussusceptions, with an accuracy of 58% to 100%.^[24] The typical imaging features of intussusceptions on abdominal CT include a target sign, banana sign, kidney shape sign, comet tail sign, and double intestine sign. Abdominal CT not only helps diagnose the presence of an intussusception but

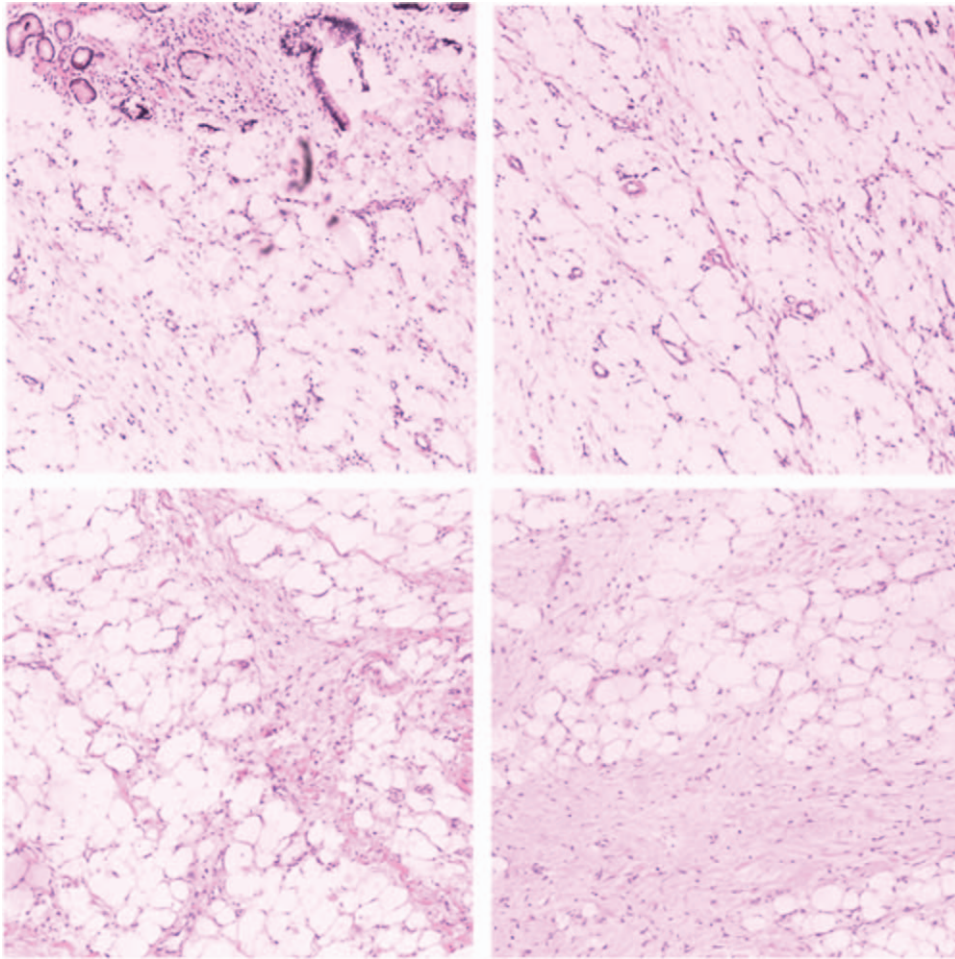


Figure 8. Postoperative pathology showed that the tumor was a small intestinal lipoma.

also helps reveal the lesion as the leading point or cause of the intussusception. Kim et al^[30] found that CT could identify whether the intussusception had a leading point, which helped determine whether surgery was needed. In the present report, the 2 patients both underwent abdominal CT examinations before surgery, and the results suggested that intussusception might have occurred. The difference was that the first patient underwent an abdominal CTA, and the diagnosis of an intra-abdominal hernia or an intussusception was made based on the circular characteristics of the small mesenteric vessels in the right lower quadrant. This finding suggests that when diagnosing the cause of gastrointestinal bleeding is difficult, abdominal CTA can be chosen as the next step.

At present, treatment of adult intussusception is controversial. Most studies suggest that surgical treatment should be performed, because adult intussusception is often caused by pathological factors. Onkendi et al^[20] suggested that surgical treatment should be performed for patients with adult intussusception who had palpable abdominal masses, intestinal obstruction, gastrointestinal hemorrhage, and leading points identified on an abdominal CT. However, the extent to which bowel resection and reduction of the intussuscepted bowel should be performed during surgery remains controversial. A reduction surgery performed before resection may permit a more limited resection, but reduction of the intussuscepted bowel during surgery may result in seeding of the tumor in the abdominal

cavity, perforation, abdominal infection, and anastomotic leakage.^[24,31] Therefore, for patients with ileocolic, ileocecal, and colocolic intussusceptions, particularly those >60 years of age, standard radical surgery using appropriate oncologic techniques is recommended due to the high risk of bowel malignancy as the underlying etiologic factor. However, when intussusception is considered to be caused by benign lesions and there are no obvious signs of intestinal inflammation or ischemia, the surgeon can first reduce the intussusception by milking it out in a distal to proximal direction and then perform the localized resection.^[32] Some studies have evaluated laparoscopic surgery for patients with intussusception under general conditions and have found that laparoscopic surgery is a safe and effective surgical method.^[10,33] Laparoscopic surgery helps with accurate identification of the position and causes of the intussusception and is associated with minimal trauma and a quick recovery. In this report, 2 patients underwent laparoscopic intussusception reduction and ileal tumor resection. The laparoscopic surgery was very successful. The patients recovered well without complications and were discharged <1 week after surgery. Therefore, laparoscopic surgery may be considered for patients in good condition who have a preoperative suspicion of intussusception and a high probability of having benign lesions.

In summary, the 2 reported cases had the following characteristics. The cases were adult intussusceptions with hematochezia as the main symptom. The physical examination signs were

negative or not obvious. The hemoglobin levels progressively declined, and conservative treatment was ineffective. Preoperative abdominal CT examinations indicated intussusceptions, and laparoscopic explorations were performed. During surgery, the intussusceptions were identified as the enteroenteric type, and the underlying causes were identified as ileal tumors. The tumors were fingerlike projections with a diameter >4 cm. The patients recovered well after surgery, and postoperative pathology showed that the tumors were benign.

Some limitations in this study should be addressed. Firstly, this study was a retrospective study and a case report, and the number of cases and clinical data were limited. Secondly, due to the acute conditions of the 2 patients and the lack of bowel preparation, colonoscopy was not performed before surgery. Thirdly, considering the results of this study, surgical exploration is necessary for patients with intussusception presenting with gastrointestinal hemorrhage when conservative treatment is ineffective. However, controversies still exist about the surgical indications and extent of resection of adult intussusception, which require further study. Finally, laparoscopic surgery for adult intussusception was successful in this study, but no comparison with open surgery was performed. Additionally, whether laparoscopic surgery for intussusception caused by malignant tumors is appropriate is uncertain. In the future, we hope to conduct a prospective randomized controlled study comparing laparoscopic and open surgery for adult intussusception, which should provide a basis for the surgical treatment of adult intussusception.

Author contributions

Investigation: Zhongmin Li.

Methodology: Zhongmin Li, Bin Song, Zhenbo Shu.

Resources: Zhongmin Li.

Software: Mingze Sun, Bin Song.

Supervision: Mingze Sun, Bin Song.

Validation: Mingze Sun.

Writing – original draft: Zhongmin Li.

Writing – review & editing: Zhenbo Shu.

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