Case Reports

Journal of INTERNATIONAL MEDICAL RESEARCH

Journal of International Medical Research
50(8) 1–6

DOI: 10.1177/03000605221115386
journals.sagepub.com/home/imr
SAGE

A rare case of adult colocolic intussusception secondary to splenosis

Xuemei Tang[1], Peng Gu[1] and Wenming Lu[2]

Abstract

Intussusception is the invagination of a segment of bowel (intussusceptum) into the lumen of an adjacent segment (intussuscipiens). Adult intussusception is rare and typically asymptomatic, although bowel obstruction can be a predominant symptom, making it difficult to diagnose. Splenosis is an uncommon and benign disease, arising from the self-implantation of splenic tissue elsewhere in the body after splenectomy or splenic trauma. Colocolic intussusception secondary to splenosis is rare. We report a case of colon intussusception with a mass in the intussusception detected by ultrasound. Abdominal ultrasound identified the intussusception location but failed to distinguish its pathological properties. Colonoscopy revealed the exudation of necrotic and fibrous tissue. Surgery was performed because of suspicions of a malignant tumor.

Keywords

Splenosis, ultrasound, intussusception, colon, abdominal ultrasound, colonoscopy

Date received: 27 April 2022; accepted: 4 July 2022

Introduction

Adult intussusception (AI) is rare and its clinical presentations differ from those of intussusception in children.[1] AI was previously found in 1% of adult patients with bowel obstruction, and the incidence of ileocolic or colocolic intussusception is low in adults (0.3/100,000 inhabitants per year).[2] AIs can present with acute, intermittent, or chronic symptoms.[3] Despite

[1]Department of Ultrasound, Sichuan Key Laboratory of Medical Imaging, Affiliated Hospital of North Sichuan Medical College, Nanchong 637000, People's Republic of China
[2]Department of Ultrasound, Huzhou First People's Hospital, Huzhou 313000, People's Republic of China

Corresponding author:
Wenming Lu, Department of Ultrasound, Huzhou First People's Hospital, Huzhou 313000, People's Republic of China.
Email: 13957242240@163.com

advances in imaging procedures, intussusception is difficult to preoperatively diagnose.[4,5] Colocolic intussusception is more commonly associated with malignant lead points (~65%) than small bowel intussusception.[6] There have been a few reports of splenosis in the colon,[7–9] but no documentation of colocolic intussusception secondary to splenosis. The present study reports an adult case of colocolic intussusception secondary to splenosis, and reviews the current literature.

Case report

A Chinese man in his early 30s visited our hospital with severe paroxysmal left upper abdominal pain that had persisted for 2 days with no obvious cause. He had undergone splenic surgery 10 years previously. His weight had not altered significantly during the past 3 months. His C-reactive protein levels and white blood cell count were increased at 14.30 mg/L and 11.0/L, respectively. Carbohydrate antigen 125, carbohydrate antigen 199, and prostate-specific antigen levels were all normal.

Ultrasound examination revealed an intussusception mass of approximately 6.2×5.9 cm in the left colon (Figure 1a). Within this mass, a nodule of approximately 4.4×3.8 cm (Figure 1b) was noted. The invagination depth was approximately 3.7 cm, and a star-shaped blood flow signal was seen in ultrasonic color Doppler (Figure 1c). Under ultrasound guidance, the intussusception was relieved with a normal saline enema. Colonoscopy was performed by inserting the lens 40 cm into the anus, which revealed a mass accounting for more than four-fifths of the intestinal cavity (Figure 2a). Biopsy results showed a small amount of necrotic and fibrous tissue exudation from the colon. Surgically, the transverse colon and descending colon were cut off 10 cm above and below the tumor, and side-to-side anastomosis of the distal and proximal bowels and transverse closure were performed. The diameter of the tumor was 4.5 cm (Figure 2b, 2c).

Hematoxylin and eosin staining of tumor sections showed red and white pulp, and the splenic sinus was dilated and extravasated (Figure 3a). An intramedullary hemorrhage (Figure 3b) and ulcers were observed (Figure 3c). Immunoenzyme labels 140083 and P53 were negative, and cluster of differentiation (CD)20, CD79a, CD34, CD15, CD30, and Ki-67 were not obviously expressed. Lesions were detected in the liver (Figure 4a), the space between the liver and stomach (Figure 4b), and the abdominal cavity (Figure 4c). All lesions were well-defined, round, or oval, and had a homogeneously hypoechoic echotexture identical to that of the spleen on ultrasound.

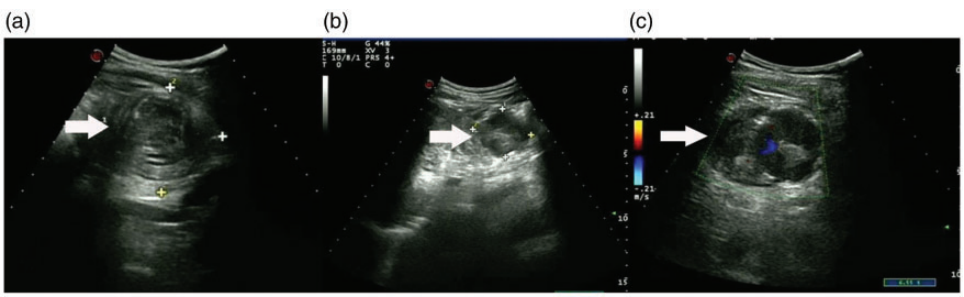

Figure 1. (a) Ultrasound examination showing an intussusception mass approximately 6.2×5.9 cm in the left colon. (b) A nodule approximately 4.4×3.8 cm was seen in the mass and (c) A star-shaped blood flow signal was shown in color Doppler of the nodule.

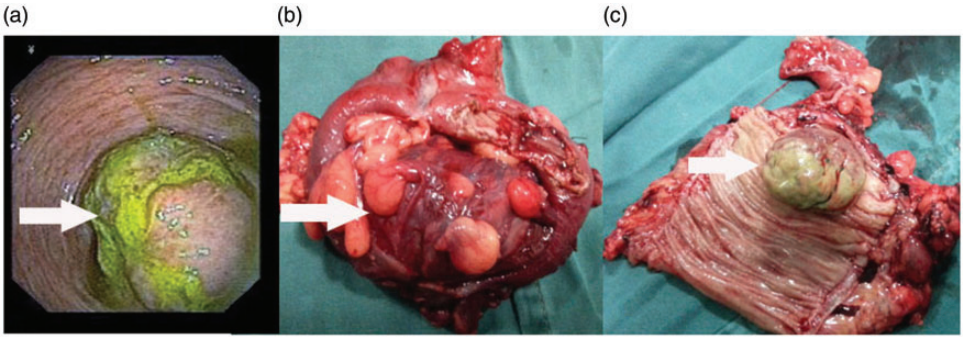

Figure 2. (a) Colonoscopy showing a mass accounting for greater than 4/5 of the intestinal cavity. (b) Diseased bowel covered with intestinal fat and (c) A mass of about 4.5 cm in diameter on the intestinal wall.

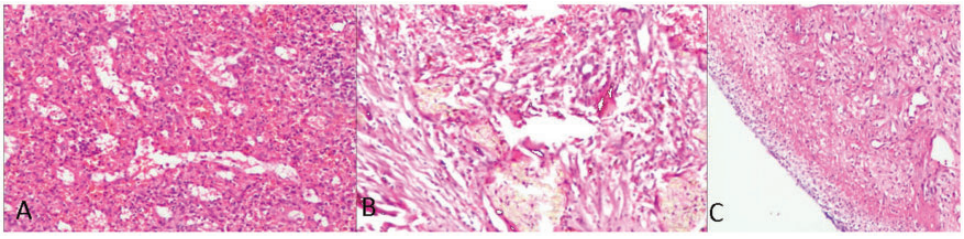

Figure 3. (a) Hematoxylin and eosin staining of red and white pulp in the tumor, showing a dilated and extravasated splenic sinus. (b) Intramedullary hemorrhage, hemosiderin deposition, and connective tissue hyperplasia with a small amount of multinucleated giant cell reaction and (c) Surface exudation necrotic granulation tissue, and the formation of superficial ulcers.

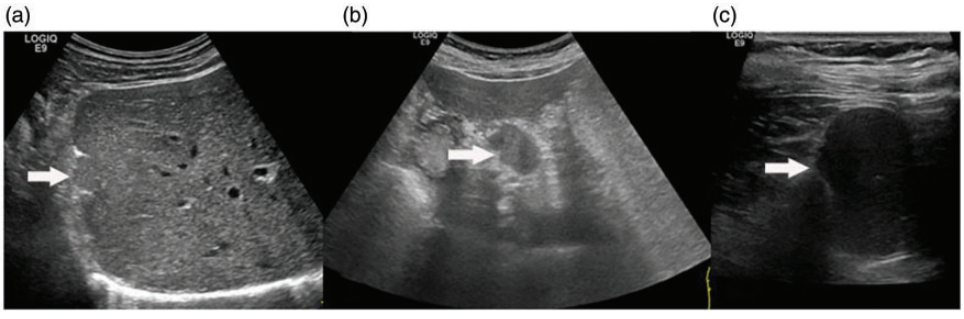

Figure 4. Splenosis located in the liver (a), the space between the liver and stomach (b), and the abdominal cavity (c).

Discussion

AIs typically involve a pathologic lead point, and up to 57% of reported cases are attributable to a malignant tumor.[10,11] The main cause of colonic intussusception was reported to be primary adenocarcinoma, followed by lymphoma and metastatic

arcinoma.[12] However, a few benign cases have been documented, such as those arising from adenoma[13]and ediculated lipoma.[14]

Splenosis is an acquired benign condition resulting from the heterotopic transplantation of splenic tissue after splenic trauma or splenectomy. Its diagnosis is usually an incidental finding.[15] The incidence of splenosis development was reported to be <0.3% after splenectomy.[16] Frequent areas of splenosis include the peritoneum, omentum, and mesentery, but other locations such as the liver, colon, pericardium, subcutaneous tissue, and brain tissue have also been described.[7,15,17]

Patients often present with nonspecific, varied symptoms which differ depending on the location of ectopic splenic tissue.[18] Kwok et al.[19] reported the migration of erythrocytic progenitor cells via the portal vein following traumatic splenic rupture and the local induction of erythropoiesis by hypoxia. Our case had colonic splenosis rather than intrahepatic splenosis, and we have encountered three other splenosis cases in the past 10 years. Maillard et al.[20] detected similar ultrasound features to those seen in our patient.

Ultrasound images can be masked by gas-filled loops of bowel. This makes diagnosis difficult because most AIs present with intestinal obstruction.[21] The preoperative diagnosis accuracy using ultrasonography is 60.0%, but this increases to 91.7% in cases of palpable abdominal masses.[5] Coloscopy can provide information about the benign or malignant nature of the lesion, potentially avoiding unnecessary surgery, although abdominal computed tomography was found to be the most accurate preoperative diagnostic method of diagnosing intussusception compared with ultrasonography, small bowel series, barium enema, and colonoscopy, with a pooled accuracy of 77.8%.[12] However, it has limited value in discriminating whether a lead point is malignant, benign, or idiopathic.

Splenic trauma or splenectomy are key events to recognize in the diagnosis of splenosis. In recent years, 99m-tagged heat-damaged erythrocyte scintigraphy and technetium-99m sulfur colloid scintigraphy have been developed to provide greater specificity in the identification of splenic tissue.[22,23]

Patients with asymptomatic splenosis do not require surgical treatment, although related therapies can be performed to treat corresponding symptoms in other locations. Because most AIs have underlying pathological lesions, laparotomy is usually agreed to be necessary.[24] However, it remains controversial whether intussusception should be reduced before resection.

In conclusion, when patients with abdominal tumors experience splenic trauma or splenectomy, it is important for clinicians to be aware of the possibility of splenosis. In patients with intussusception, ultrasound can help with diagnosis and relief, while coloscopy can provide pathological information about large bowel intussusception with tumors. Additionally, appropriate imaging and scintigraphy can be performed to recognize key features and avoid unnecessary invasive diagnostic procedures and surgery.

Declaration of conflicting interest

The authors declare no conflicts of interest.

Ethics statement

This case report was not required to be reviewed by the ethics review committee because it discusses the diagnosis and pathogenesis of a rare disease; no exploratory treatment was performed. Written informed consent was obtained from the patient for the publication of this case report and any accompanying images. A copy this consent is available for review by the Editor-in-Chief of this journal. We have

de-identified all patient details. The reporting of this study conforms to CARE guidelines.[25]

Funding

The authors disclosed receipt of the following financial support for the research, authorship, and/or publication of this article: This study was supported by the Youth Innovation Project of Sichuan Medical Association (No. Q20032).

ORCID iDs

Xuemei Tang https://orcid.org/0000-0003-0748-2201
Wenming Lu https://orcid.org/0000-0002-5600-0398

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