Small Bowel Cavernous Hemangioma Complicated with Intussusception: Report of an Extremely Rare Case and Review of Literature

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Abstract Cavernous hemangioma of the small intestine is a rare disease in adult patients. Intussusception caused by small bowel hemangioma is extremely rare. We report a 20-year-old female presenting with lower abdominal pain and post-prandial bloating. Laboratory studies showed leukocytosis with left shifting and prominent anemia. Computed tomography of the abdomen revealed small bowel intussusception with bowel ischemia. Segmental resection of the intussuscepted jejunum was performed, and the diagnosis of cavernous hemangioma causing intussusception and anemia simultaneously was made.

Keywords Anemia · Cavernous hemangioma · Intussusception · Small bowel hemangioma

Introduction

Cavernous hemangioma of the small intestine is a rare disease in adult patients and is usually located in the jejunum [1]. The usual clinical features include abdominal pain, bleeding, and obstruction. Acute intestinal bleeding leading to anemia is the most common presentation in most patients [2]. Intussusception and even perforation caused by cavernous hemangioma are extremely rare [3]. Herein we report an adult

patient with small bowel cavernous hemangioma presenting as intussusception and anemia.

Case Report

This 20-year-old woman presented at the emergency room with sudden onset of lower abdominal pain accompanied with post-prandial bloating for hours. She did not have fever, nausea, hematemesis, hematochezia, or weight loss except for melena during 3 weeks. Lower abdominal tenderness and pale conjunctiva were detected in physical examination. Laboratory studies revealed leukocytosis with left shifting (WBC 11.04 k; segment form 94 %) and prominent anemia (Hb 7.4 g/dL). Computed tomography of the abdomen was arranged to identify the nature of hemorrhage of the intestinal tract and lower abdominal tenderness. Target sign of small bowel with poor contrast enhancement of the bowel wall and intra-luminal hematoma formation were noted over the left lower quadrant of the abdomen (Fig. 1). These findings were consistent with intussusception with bowel ischemia. The patient underwent emergent laparotomy. On exploration, an intussusception, causing dilatation of the proximal bowel, was found at the jejunum, 70 cm distal to the ligament of Treitz. Segmental resection of the intussuscepted jejunum was done due to apparent ischemic change. Fifteen centimeters away from the proximal cut margin, a polypoid lesion, measuring 5.6×4.6×1.5 cm, was noted in the lumen of the resected small bowel and was proposed to be the leading point of intussusception. Histologically, this unencapsulated polypoid lesion consisted of multiple various blood-filled spaces which were lined by thin endothelial cells and separated by scant connective tissue stroma (Fig. 2). The final diagnosis is cavernous hemangioma causing intussusception of the jejunum. This patient was discharged without immediate complications on the sixth day. This patient has remained well and symptom-free.

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Fig. 1 Abdominal computed tomography demonstrated target sign of small bowel with poor contrast enhancement of the bowel wall (*black arrow*) and intra-luminal hematoma formation (*red arrow*) over the left lower quadrant of the abdomen

Discussion

Intussusception is defined as invagination of a proximal portion of the bowel and its mesentery (intussusceptum) into a more distal segment (intussusciens) [4]. Intussusception can result in impaired peristalsis, obstruction, and even vascular compromise [5]. Adult intussusception represents 5–16 % of all cases of intussusception and only 1–5 % of all cases of intestinal obstruction. Primary or idiopathic adult intussusception accounts for only 10 % of cases and is more likely to develop in the small intestine. Up to 90 % of occurrences in adults have a leading point, which is defined as a well-definable pathological abnormality [6].

Fig. 2 Cavernous hemangioma. The lesion is unencapsulated and composed of blood-filled spaces of variable size and shape. Foci of thrombi and hemorrhagic necrosis are present. The overlying mucosa shows hemorrhagic necrosis. The lumens are filled with blood cells and lined by thin endothelial cells and separated by scant connective tissue stroma (H&E, ×200)

Small bowel hemangioma accounts for 5 to 10 % of all benign neoplasms of the small intestine [7]. It is thought to be one of the congenital benign vascular lesions which are venous malformations and not true tumors [2, 8]. Hemangiomas are classified as cavernous, capillary, or mixed type, and the cavernous type is the most common [1]. Malignant change is quite unusual. As the previous literature reported, most patients having small bowel hemangioma presented with abdominal pain and intestinal bleeding with acute or chronic anemia. As for this case, lower abdominal pain and post-prandial bloating were noticed at triage. According to the age of this patient and the laboratory result of anemia, rupture of the corpus luteum, endometriosis, or ectopic pregnancy was the first suspicion needed to be ruled out. However, post-prandial bloating and leukocytosis with left shifting implied bowel obstruction and the progression of infectious process. Further computed tomography of the abdomen helped us make an accurate diagnosis. Very few cases have been described in the English-language literature about small bowel hemangioma being responsible for intussusception [3]. This case provides a message to all practitioners that one entity can cause small bowel obstruction and bleeding simultaneously. Early diagnosis and appropriate intervention could provide good outcomes. Diagnostic and therapeutic measures should be taken to avoid further complications.

Contribution Details Liang-Wei Kuo carried out literature search, data acquisition, and manuscript preparation. Hao-Wen Chuang performed data acquisition, and Yu-Chia Chen performed manuscript editing and manuscript review.

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