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Case Report Open Access

Recurrent Obscure Gastrointestinal Bleeding: Intestinal Lymphangiohemangioma

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Abstract

Obscure gastrointestinal bleeding (OGIB) is thought to be a diagnostic and therapeutic challenge for the clinicians and radiologists. A 46-year-old woman complaining of intermittent melena for 3 months was admitted. After extensive investigations, a vascular lesion located in the ileum, diagnosed as intestinal lymphangiohemangioma pathologically, was found by CT angiography (CTA) and selective arterial angiography. The value of CTA to the diagnosis of OGIB is showed.

Keywords: Gastrointestinal bleeding; CT angiography

Introduction

Obscure gastrointestinal bleeding (OGIB) is defined by the American Gastroenterological Association (AGA) as bleeding of unknown cause after upper or lower endoscopy [1]. OGIB may be categorized into obscure overt bleeding and obscure silent bleeding. Overt OGIB is high-volume bleeding manifesting as hematemesis, melena, or hematochezia.

Silent OGIB may manifest as amounts of blood detectable only with chemical tests in the stool or as iron deficiency anemia [2,3]. Due to noninvasiveness and sensitivity for low rates of bleeding (0.5 mL/min), CTA is used to find the cause of OGIB more and more [4]. A case of recurrent obscure gastrointestinal bleeding diagnosed by CT angiography (CTA) is presented to illustrate the value of CT angiography to diagnose OGIB caused by vascular lesions.

Case Report

A 46-year-old woman was admitted to our institution complaining of intermittent melena for the past 3 months. On admission, physical examination was negative for abdominal palpable mass. The patient denied non-steroidal anti-inflammatory drug assumption and past bleeding tendency. Except for the iron deficiency anemia and decreases of the hemoglobin values down to 7.5 g/L, all the laboratory values were in the normal limit. The investigation of anemia included repeated colonoscopies, gastroscopy, abdominal ultrasound of the abdomen, which were all normal. CT angiography and selective arterial angiography for the patient showed an obvious vascular lesion in the distal ileum (Figure 1).

The patient underwent a laparoscopy with segmental ileum resection because of non-candidate for transcather arterial embolization. At laparoscopy, a purple polypoid lesion measuring 1.5 cm × 1.8 cm was found in the ileum about 40 cm proximal to the ileocecal valve. Final pathology was compatible with lymphangiohemangiomas with cavernous sinuses and lymphangiectasis, involving submucosa (Figure 2). The patient had uneventful recovery and was discharged home in postoperative 7th day. On 3-month follow-up, the patient's hemoglobin was 14 g/dL suggestive of no further evidence of gastrointestinal bleeding.

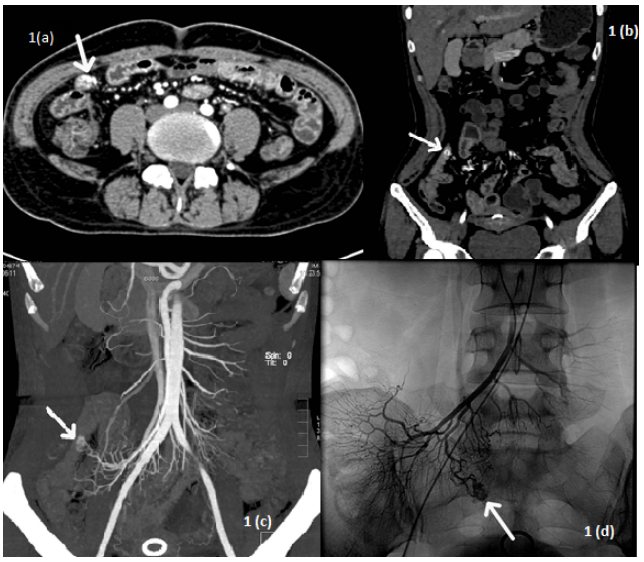

Figure 1: CT angiography and selective arterial angiography for the patient showed the following finding in the distal ileum.

Disscussion

Vascular lesions are the most common cause of OGIB associated with small intestine, and the most common pathologic lesions are angiodysplasias, found in 30% to 60% of this group [5]. Small bowel tumors are the next most common source [6]. Other causes include small bowel ulcers (eg., Crohn disease, use of nonsteroidal anti-inflammatory drugs, Meckel diverticulum), vasculitis, small bowel diverticula, aortoenteric fistulas, and caliber-persistent arteries of the stomach (Dieulafoy lesion) [7]. Intestinal lymphangiohemangiomas,

which are usually solitary and may vary in size from nodules of few millimeters to large lesions up to 11 cm projecting into the bowel lumen, is composed of large blood-filled sinuses lined by endothelial cells. Lymphangiohemangioma of the small intestine are rare benign tumors, but important source of OGIB [8].

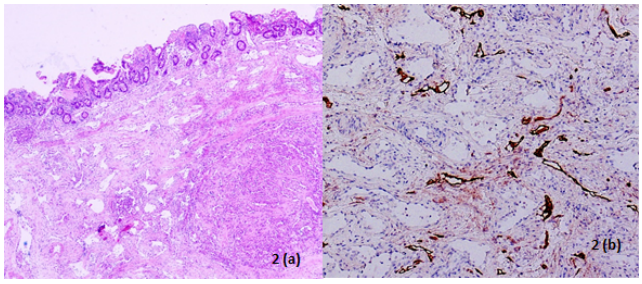

Figure 2: Final pathology showed lymphangiohemangiomas with cavernous sinuses and lymphangiectasis, involving submucosa.

Because of their rarity, they are frequently not considered as a cause of gastrointestinal bleeding. Furthermore, lymphangiohemangiomas of the small bowel are difficult to differentiate from other more common entities clinically owing to lacking specific symptoms. The most common symptoms are occult bleeding and iron deficiency anemia, while intestinal intussusception and bowel obstruction rarely occur. Investigations for recurrent digestive tract bleeding have developed many approaches, including barium meal, endoscopy, CT, radionuclide imaging, digital subtraction angiography, CT angiography and capsule endoscopy.

All these examinations manifest variable clinic implications in the relevant literatures. Selective arterial angiography is commonly thought to be most useful in the setting of active bleeding; noninvasive CT angiography is consistent with the diagnosis from angiography in this case. Capsule endoscopy is a noninvasive tool for imaging the entire small intestine and used especially for recurrent episodes of digestive bleeding in current literature [9], however, our case highlights the clinical value of CT angiography in the diagnostic approach to those vascular lesions, which defines the precise localization of the lesion and is convenient for the surgeon planning surgery.

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