Ileal Cavernous Haemangioma Causing Perforation: A Rare Case of Acute Abdominal Pain

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- **Abstract**

- **Keywords:** Haemangiomas, Intestinal perforation, Small intestine hem angioma

- **Background:**
Haemangiomas are benign vascular tumours that can occur in any part of the body. Although they are rare in the gastrointestinal (GI) tract, accounting for approximately 0.05% of all GI tumours, small intestinal haemangiomas are among the rarer benign lesions of the small intestine, representing 7-10% of such tumours. They are most commonly found in the jejunum (60%) and are rarely located in the ileum. Clinical manifestations of small bowel haemangiomas include abdominal pain, gastrointestinal bleeding, intestinal obstruction, intussusception, and, in rare cases, intestinal perforation. We report a case of acute abdominal pain caused by perforation of an ileal cavernous haemangioma.

- **Case Presentation:**
A 31-year-old female with no significant medical history presented with a 3-day history of right lower abdominal pain. She had experienced multiple similar episodes in the past, previously diagnosed and treated as irritable bowel syndrome. The patient also reported weight loss and anaemia over the past two years.

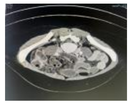
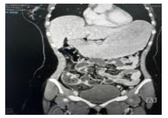
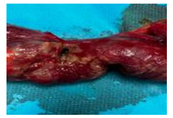
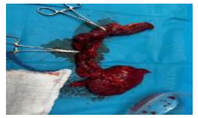
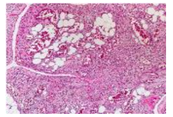
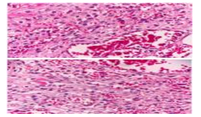
- **CT Scan Findings:**
A contrast-enhanced abdominal CT scan revealed a focal area of wall discontinuity (i.e., perforation) in the distal ileum, approximately 15-20 cm from the ileocecal junction, associated with small amounts of pneumoperitoneum.

- **Surgical Intervention:**
During laparotomy, a perforation was identified 20 cm from the terminal ileum. Resection of the affected bowel segment and anastomosis were performed. Histopathological examination revealed a cavernous haemangioma involving the submucosa and muscularis layers, associated with focal mucosal ulceration and transmural perforation.

- **Conclusion:**
Small intestinal haemangiomas, though rare, should be considered in the differential diagnosis of acute abdominal pain. Although complications such as perforation are uncommon, they highlight the potential severity of these lesions. Diagnosis is often delayed and typically occurs during surgery, with histopathological studies confirming the diagnosis. Clinicians should remain vigilant in identifying such rare causes of gastrointestinal symptoms to prevent delayed diagnosis and complications.

- **CASE PRESENTATION:**

A 31-year-old female with no significant medical history presented with a 3-day history of right lower abdominal pain. She had experienced multiple similar episodes in the past, previously diagnosed and treated as irritable bowel syndrome with the chief complaint of acute abdominal pain She described her pain as generalized progressing cramping abdominal pain that had started suddenly ten days ago and was accompanied by loss of appetite, nausea, and vomiting from 10 hours before she came to the emergency department. There was no pulsatile vomiting. The vomitus was the previously ingested food and yellow-green bile-like substance, and she vomited five times. There was no hematemesis, no fever and no diarrhoea. The abdominal symptoms gradually became aggravated. physical examination revealed an abdominal generalized tenderness and left lower quadrant rebound tenderness. Most laboratory results were normal except something that indicated a leucocytosis (white blood cell (WBC):16830/MCL), and neutrophilia moderated microcytic and hypochromic anaemia (haemoglobin: 7.5 g/dL). The faecal occult blood test was positive. The liver and kidney function, coagulation function, carcinoembryonic antigen, carbohydrate antigen 19-9, and cancer antigen 125 were normal.
Imaging examinations A contrast-enhanced abdominal CT scan revealed a focal area of wall discontinuity (i.e., perforation) in the distal ileum, approximately 15-20 cm from the ileocecal junction, associated with small amounts of pneumoperitoneum Fig 1,2.

TREATMENT:

She was transferred to the operation room for laparotomy with the diagnosis of intestinal perforation. At the operation, a midline incision, in the umbilical region above and below the umbilicus, was performed, and the sign of peritonitis was obvious. While investigating a perforation with the size of 1*1 cm^2 was seen in the small intestine wall at a distance of 20 cm from the ileocecal valve which underwent segmental resection and anastomosis (Fig 3, Fig 4). The resected segment was sent for pathological studies. The pathological studies showed the view of capillary haemangioma (Fig 5, Fig 6) and also revealed that there had been a haemangioma that has ruptured and the haemorrhage occurred, so it caused focal haemorrhagic necrosis and small intestine wall perforation happened due to this necrosis.

OUTCOME AND FOLLOW-UP The patient quickly recovered and had no further episodes of bleeding after the operation. she returned to our hospital for a follow-up visit 1 month after the operation and his haemoglobin levels increased to 10.7 g/dL (normal range: 12-16 g/dL).

INTRODUCTION:

Benign vascular tumours that can be found anywhere in the body are defined as haemangiomas [1,2]. They are histologically classified into cavernous (the most common form which nowadays is called gastrointestinal venous malformations based on ISSVA classification), capillary, or mixed-type tumours, according to the size of the affected vessels [[3], [4], [5]].These tumours usually affect young people and sex is not a risk factor [3]. They are 0.05% of all GI tumours and 7–10% of GI haemangiomas are in the small intestine [1,3]. In the small intestine, the jejunum is the most common site of Haemangioma occurrence, while in 18 years (2000–2018) only 37 cases were reported with small intestine haemangioma [2].The most common clinical manifestation is an obscure GI bleed (OGIB) which also could be massive [[6], [7], [8], [9], [10]]. In 5–10% of GIB, the aetiology of the small intestine is due to small intestine tumours [10]. Other possible signs and symptoms could be Iron deficiency anaemia, intestinal obstruction abdominal pain, intramural hematoma, perforation, platelet sequestration, intussusception or perforation [2,3,11,12].The main diagnostic methods are Computed tomography (CT) and contrast-enhanced computed tomography (CECT), and capsule endoscopy is helpful too [3,13]. The therapeutic plan depends on the disease development and the condition of the patient. In the acute phase, surgical resection is ideal, while in the chronic one medical treatment and clinical follow-up are better choices [3,14].

DISCUSSION: Haemangiomas are congenital benign vascular lesions and they are venous malformations not true tumours [3,15].Haemangiomas are classified by three approaches: The number of masses: Haemangiomas may be solitary or multiple. Multiple haemangiomas usually presented with similar lesions in other organs, like the liver or skin, and also may occur as a part of some syndromes such as Osler-Weber-Rendu syndrome, blue rubber bleb nevus syndrome , Maffucci syndrome , and Klippel-Trenaunay-Weber syndrome [2].**Microscopically: Although** haemangiomas can make invasion to beyond the serosa, they usually involve the intestinal wall from the submucosa to the muscular layer [1,2].**Histologically:** Capillary: a proliferation of small capillaries composed of thin-walled vessels lined by endothelial cells that is more related to bleeding. Cavernous: large sinuses lined by multiple layers of endothelial cells which are the most common type of haemangiomas. Mixed: Have features of both other types [1]. In the pathological studies of our case, her haemangioma was a solitary haemangioma of submucosa with capillary form features. In the GIT, haemangiomas are rare and only 0.05% of intestinal neoplasms are haemangiomas, in this tract, the common site that they could be found is within the small intestine and jejunum in the commonest site (a ratio of 1.6/1 to ileum) and are accountable for 7–10% of all benign masses [10,15]. The haemangiomas are more frequent in the age of 5–25 years and the preference of gender is unclear, the male: female ratio of occurrence was found between 1:1 to 4:1 in different articles [1,3,10]. 90% of haemangiomas, unlike other benign tumours of GI, are symptomatic. The most common sign and symptoms are chronic gastrointestinal bleeding that causes anaemia of unknown origin and sometimes massive bleeding. The non-common clinical manifestations are intestinal obstruction, invagination, or recurrent abdominal pain and rarely platelet sequestration, perforation [1,3,10,15]. Five databases consist of PubMed, Cochrane, Web of Science, ProQuest, and Google Scholar with the time filter of 2012–2022 and no study type filter and with the Mesh terms and Keywords such as “haemangioma”, “perforation”, “rupture”,” necrosis” and “small intestine” were investigated. Our library was revised by excluding criteria of irrelevant studies. As we cannot find any past case of this age in which the rupture and haemorrhage of jejunum solitary mixed haemangioma lead to intestinal wall necrosis and perforation without any evidence of GIB The diagnosis of haemangioma in GIT, especially in the small intestine, before the operation is difficult. Usually, it is suspicious in patients admitted with acute or chronic GIB with normal results of frequent gastroscopy and colonoscopy studies. A simple abdominal X-ray may be useful in the case of phleboliths, obstructions, or perforations due to haemangiomas. CT and CECT could be used even in emergency cases, to diagnose extraintestinal lesions.

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