Case Report

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Jejunal angiodysplasia: surgery can be life-saving – a case report

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Introduction: Angiodysplasia, a rare cause of gastrointestinal (GI) bleeding, presents a spectrum of clinical manifestations from anemia to life-threatening hemorrhage. This case study emphasizes the significance of considering intestinal vascular malformations as a differential diagnosis, especially in the context of chronic anemia and GI bleeding. Jejunal angiodysplasia, though infrequent, poses diagnostic challenges due to the hidden nature of the small bowel in the GI system.

Case presentation: A 23-year-old male presented with acute hematochezia and melena, necessitating prompt intervention. Despite a normal esophagogastroduodenoscopy, colonoscopy was hindered, CT angiography could not be performed due to the patient's unstable condition, leading to a planned exploratory laparotomy. Surgical exploration revealed a mucosal vascular lesion in the jejunum, prompting resection, and anastomosis. The patient's postoperative course was uneventful, reinforcing the importance of swift diagnosis and intervention.

Clinical discussion: Angiodysplasia's pathogenesis remains unclear, with hypotheses implicating vascular endothelial growth factor and submucosal changes. Challenges in management revolve around lesion localization and stabilizing hemodynamics, necessitating a multidisciplinary approach. While endoscopy is often diagnostic and therapeutic, advanced modalities such as CT angiography may be required. Literature review highlights diverse presentations and successful interventions, including embolization and surgical resection.

Conclusion: Jejunal angiodysplasia demands a comprehensive diagnostic and therapeutic strategy. The presented case underscores the pivotal role of endoscopy, embolization, and surgery in managing this condition. Timely diagnosis and intervention are crucial for mitigating the impact of angiodysplasia, necessitating further research and collaborative efforts for improved management of this rare condition.

Keywords: angiodysplasia, case report, GI bleeding, jejunal angiodysplasia, upper GI surgery

HIGHLIGHTS

- Angiodysplasia presents diverse manifestations, from anemia to life-threatening hemorrhage.
- Jejunal angiodysplasia diagnosis requires a multidisciplinary approach due to hidden small bowel location.
- Surgery, including enterotomy and resection, remains crucial for both diagnosis and treatment.
- A case study emphasizes successful surgical intervention in identifying and treating jejunal angiodysplasia.
- Prompt diagnosis and intervention are essential, emphasizing the need for research and collaborative efforts.

Introduction

Angiodysplasia constitutes an etiological factor in gastrointestinal (GI) bleeding, exhibiting a spectrum of clinical presentations. These manifestations encompass indicators of iron deficiency anemia to concealed bleeding, and in severe cases, life-threatening hemorrhage[1]. Being a rare condition, intestinal vascular malformations should be ruled out as a differential diagnosis for both chronic anemia and GI bleeding[2]. It is considered a degenerative disease of previously normal blood vessels. It is most frequently found in the ascending colon and cecum (about 75%), with 15% located in the jejunum and ileum, and the remaining 10% scattered throughout the rest of the alimentary tract[3].

In alignment with the SCARE (Surgical CAse REport) guidelines, this case report has been prepared to ensure comprehensive and standardized reporting of the clinical case[4].

Case presentation

A 23-year-old nonsmoker male, with a clear medical and family history, presented with the acute onset of hematochezia and melena within the preceding 12 h. On physical examination, he

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appeared pale, with a pulse rate of 128 per minute, a respiratory rate of 33 per minute, and a blood pressure of 80/55 mmHg. Abdominal examination revealed a soft, nondistended abdomen with no collateral venous circulation, hepatomegaly, or splenomegaly.

The patient was resuscitated by inserting two wide cannulas, received 2 l of fluid (normal saline and Ringer lactate), and a nasogastric tube was inserted to exclude upper GI bleeding. Blood samples were taken for laboratory investigations. Laboratory findings included a hemoglobin level of 6.4 g/dl, platelet count of 440 × 10^9 /l, and normal renal and liver function tests.

The patient was then prepared for esophagogastroduodenoscopy (OGD) and colonoscopy. The OGD was normal, but the colonoscopy could only pass up to the splenic flexure due to a poor view. Therefore, CT angiography was planned, but the patient became unstable; consequently, he received blood and fresh frozen plasma. As a result, surgical intervention was deemed necessary.

The patient was taken to the operating room and underwent laparotomy. Gross examination revealed that both the small and large bowel were loaded with blood, raising suspicion that the bleeding originated from the proximal small bowel. A multiple clamps test was performed on the small bowel after milking the small bowel lumen. After waiting for 10 min, a highly suspicious area was identified 40 cm from the duodenojejunal flexure. Enterotomy of the jejunum was performed, revealing a small mucosal vascular lesion measuring 1.5 cm with an active bleed (Figs 1, 2). Resection and anastomosis were performed, and the patient was transferred to the ICU. He remained fasting for 48 h, stayed in the hospital for 5 days, and was discharged in good condition. A follow-up CT angiography on day 4 postoperation showed normal findings.

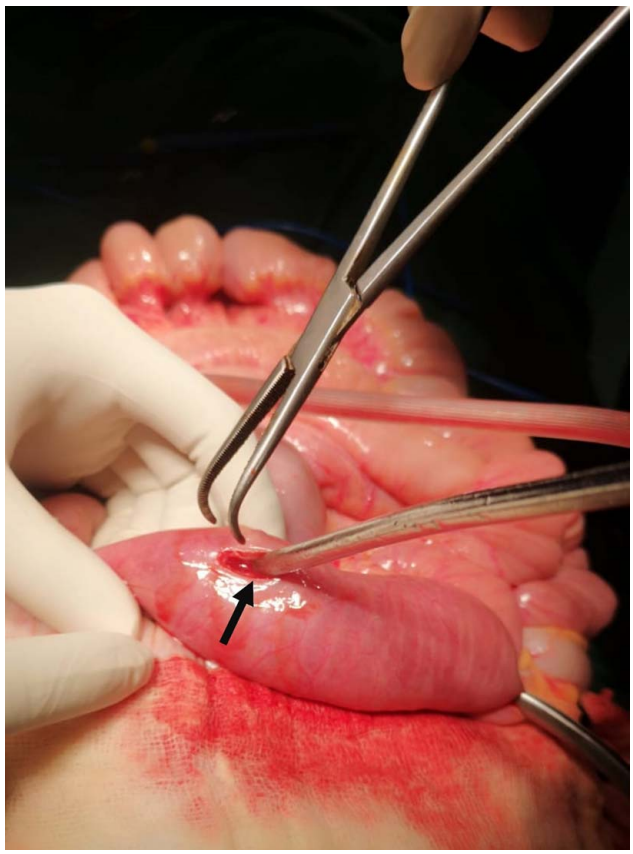

Figure 1. Intraoperative finding of suspicious area 40 cm from Deudojejunal flexure. The black arrow indicates the suspected area of the enterotomy.

Discussion

Intestinal vascular malformations are an infrequent occurrence and should be considered as part of the range of potential differential diagnoses for GI bleeding and chronic anemia[5]. Various hypotheses have been posited to elucidate the pathogenesis of angiodysplasia. One theory proposes an association between the vascular nature of angiodysplasia and elevated levels of vascular endothelial growth factor, resulting in mucosal hypoxia[6]. Another perspective suggests that chronic obstructive changes within the submucosa could induce pressure on the submucosal vessels, leading to their dilation[7].

Diagnosing the source of bleeding in jejunal angiodysplasia (AD) may require a multidisciplinary team of gastroenterologists, interventional radiologists, and surgeons, due to the small bowel's hidden nature in the GI system. While endoscopy remains the gold standard for diagnosis and treatment of AD, CT angiography, radiolabeled red blood cells, and surgery may be necessary in some cases[8].

The macroscopic diagnosis of AD depends on the presence of small (0.5–1 cm), red mucosal lesions on the mesenteric border. Meanwhile, the histological diagnosis of angiodysplasia is based on the presence of mucosal vascular ectasias associated with prominently dilated capillaries penetrating the muscularis mucosae[9].

Angiodysplasia has been reported in patients with certain predisposing conditions, such as liver disease, renal disease, Von Willebrand disease, and aortic stenosis[10].

Challenges in managing angiodysplasia cases primarily revolve around lesion localization and stabilizing the patient's hemodynamics[11]. While endoscopy serves as a valuable tool for both diagnosis and treatment in many instances, more advanced diagnostic modalities such as CT angiography with embolization might be required for hemodynamically stable patients[11].

In considering potential differential diagnoses, it is imperative to address peptic ulcer disease, given its association with upper GI bleeding. Although our patient's OGD was normal, acknowledging the possibility of peptic ulcer disease presenting with similar symptoms.

In 2021, Ali TA *et al*., reported a case involving a 71-year-old woman presenting with hypovolemic shock due to lower GI bleeding. Clinical examination revealed severe anemia and melena, with laboratory tests indicating low hemoglobin and impaired renal function. CT imaging identified a 1.2 × 1.5 cm vascular lesion along the duodenum, consistent with duodenal angiodysplasia. Subsequent angiogram and coil embolization successfully treated the lesion, preventing further arterial extravasation. The patient experienced clinical improvement with no complications reported during the 6-month follow-up, highlighting the efficacy of the interventional approach[11]. Additionally, in 2020, Alghamdi T *et al*., reported a case involving a 68-year-old male presenting with pallor, hematochezia,

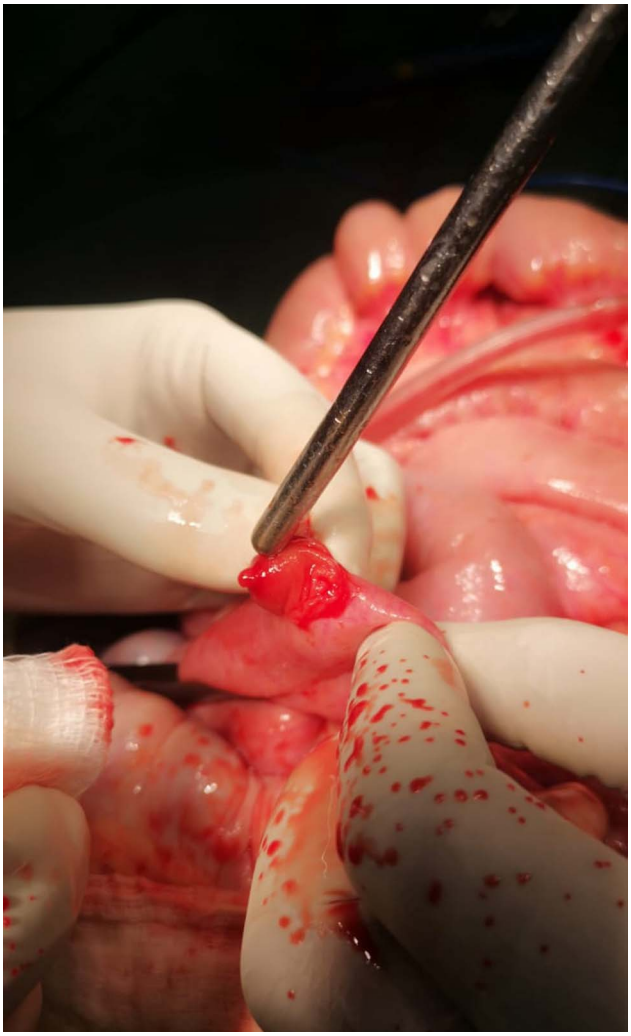

Figure 2. After enterotomy antimesentric vascular lesion with active bleeding source (angiodysplasia).

and iron deficiency anemia. Despite declining colonoscopy, abdominal CT revealed abnormal contrast flocculation in the small bowel and rectum, indicating angiodysplasia. Subsequent surgical resection (20 cm) and histopathological examination confirmed mucosal vascular ecstasies[12].

In another case of angiodysplasia reported in 2012, Olokoba AB *et al*., documented two instances of colonic angiodysplasia. The initial case involved an 85-year-old man experiencing constipation, left-sided abdominal pain, and weight loss. A colonoscopy revealed dilated tortuous blood vessels in the descending colon, situated ~45 cm from the anal verge, indicating angiodysplasia. The second case featured a 30-year-old female trader presenting with massive hematochezia and shock. Following stabilization, colonoscopy uncovered an area of erosion with mucosal blood clot about 27 cm from the anal verge, confirming the diagnosis of angiodysplasia[13]. It is worth noting that in our case, the colonoscopy could only progress up to the splenic flexure due to a limited field of view.

In 2007, Coral RP *et al*., reported a case of a 65-year-old woman with recurrent melena and severe anemia due to duodenal angiodysplasia. Despite three episodes of acute upper GI bleeding, electrocoagulation successfully controlled the bleeding. Further investigation revealed left renal artery obstruction and atherosclerotic changes. Surgical exploration, including antrectomy and duodenectomy, followed by reconstruction with gastroenterostomy, effectively addressed the angiodysplasia[14].

Surgery, encompassing both open and laparoscopic approaches, remains integral to the diagnostic and therapeutic aspects of angiodysplasia management. Surgical interventions include enteroscopy or segmental clamping of the bowel to directly identify the source of bleeding. In complex cases, extensive bowel resection might be necessary if the identification of the bleeding source proves challenging. Considering the segmental or multifocal nature of angiodysplasia, a comprehensive examination of the entire bowel should be conducted during exploration[1].

Furthermore, certain medical interventions, such as long-acting somatostatin analogs, have demonstrated efficacy in reducing rebleeding in patients with refractory small bowel angiodysplasia[15]. Suppression of vascular endothelial growth factor has also shown promise in reducing bleeding episodes[16].

The presented case involved surgical intervention due to the unstable condition of the patient. However, it is essential to acknowledge a notable limitation in our study—the short follow-up period. While the immediate postoperative course and findings were reported, a more extended follow-up could provide valuable insights into the long-term efficacy of the intervention and potential recurrence of angiodysplasia-related complications.

Conclusion

Jejunal angiodysplasia presents with diverse clinical manifestations, ranging from anemia to life-threatening bleeding. This study underscores the importance of a comprehensive diagnostic and therapeutic approach. Endoscopy, embolization, and medical therapy play key roles, with surgery as a crucial option for failed endoscopic management and unstable patients. The presented case illustrates the significance of prompt diagnosis and intervention in mitigating the impact of angiodysplasia, highlighting the need for further research and collaborative efforts in managing this rare condition.

Ethical approval

Ethical approval for this was provided by the Ethical Committee Ibn Sina Specialized Hospital, Khartoum, Sudan on 15 August 2023.

Patient consent

Written informed consent was obtained from the patient for publication and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Author contribution

A.D., A.N., H.K., M.A., and A.R.: writing the paper; A.D., H.K., and A.R.: data collection and final approval of the report.

Conflicts of interest disclosures

Not applicable.

Research registration unique identifying number (UIN)

'Cannot be registered because they are not clinical studies'.

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Data availability statement

No additional data are available.

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