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An Unusual Culprit: Rare Case of Duodenal Hemangioma in a Middle-Aged Man Presenting with Anemia

Anand Bhandary Panambur[1], Anand Peter Ignatius[2]

[1]Assistant Professor, A.J. Institute of Medical Sciences & Research Centre, Mangalore, India
[2]Professor, Department of General Surgery, A.J. Institute of Medical Sciences & Research Centre, Mangalore, India

Corresponding Author: Anand Bhandary Panambur

Abstract

Hemangiomas are benign vascular tumors that typically arise from the submucosal blood vessels. Duodenal hemangiomas, caused by abnormal vascular growth in the duodenal wall, are rare but can lead to severe, rapidly progressing GI bleeding requiring urgent intervention. Diagnosis usually involves endoscopic or surgical visualization, along with histological confirmation. These tumors make up less than 0.05% of all GI tumors and 7- 10% of small bowel neoplasms, making them an exceptionally rare cause of upper GI bleeding. Vascular lesions should be considered in the differential diagnosis of unexplained gastrointestinal bleeding. Gastrointestinal hemangiomatosis is associated with conditions such as Blue Rubber Bleb Nevus Syndrome, Proteus Syndrome, Maffucci's Syndrome, Diffuse Neonatal Hemangiomatosis, and Klippel-Trénaunay-Weber Syndrome. It is characterized by diffuse infiltration of the intestinal wall, mesentery, and occasionally the retroperitoneum. Options for treatment include observation, sclerotherapy, and, in certain cases, open or laparoscopic surgery. Minimally invasive procedures like endoscopy - laser coagulation, endoscopic resection are preferred for diagnosing and treating duodenal hemangiomas in select patients.

We report a 40 year old man with upper abdominal pain, frequent vomiting, and intermittent melena for the past 5 - 6 months. After routine blood workup, patient was subjected for Upper GI endoscopy which revealed a 2 cm bluish swelling near the ampulla with intact mucosa and no active bleeding, consistent with duodenal hemangioma. MRCP identified it to be duodenal D2 hemangioma. This case highlights the need to consider duodenal hemangioma in upper GI bleeding and underscores the role of endoscopy in diagnosis and treatment, along with the potential need for surgery.

Keywords: Upper gi endoscopy, duodenal hemangioma, melena, upper gi bleeding, mrcp, diagnostic and therapeutic endoscopy, gastrointestinal hemangiomatosis.

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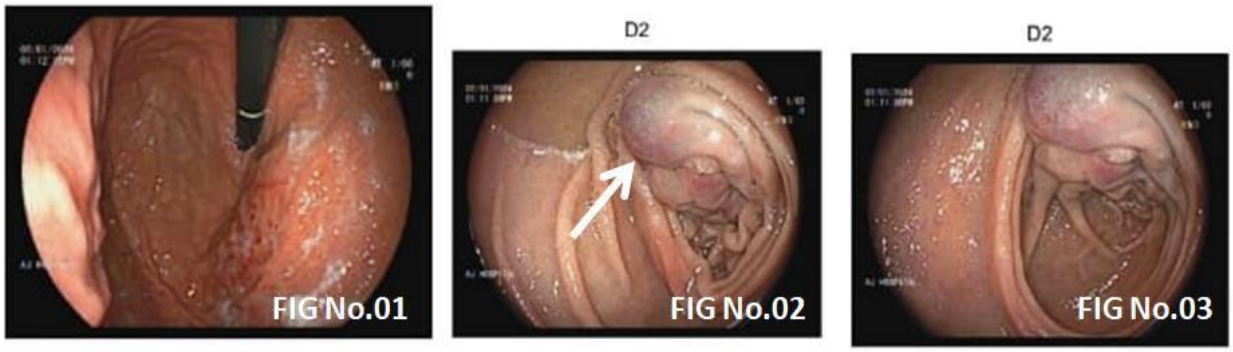
Introduction:

Hemangiomas of the small intestine account for 5% to 10% of benign tumors[1] and roughly 0.3% of all neoplasms in this segment of the gastrointestinal tract (GIT). Among them, those found in the duodenum are particularly rare, comprising just 3.4% of all GIT hemangiomas.[2] These benign vascular growths originate from dysembryoplastic alterations in mesenchymal tissue and manifest as capillary, cavernous, or mixed structural formations within the submucosal layer and mucous membranes when occurring in a hollow organ.[3] While some upper gastrointestinal hemangiomas remain asymptomatic, others may lead to complications such as bleeding, abdominal pain, and even life-threatening conditions like intestinal intussusception, obstruction, or perforation.[1-3] It can occur in one or more locations, with the jejunum being the most frequently affected[4]. Originating from the submucosal vascular plexuses, these lesions can extend into the muscular layer or beyond.[5]

Histologically, hemangiomas are congenital benign vascular anomalies that are classified as capillary, cavernous, or mixed-type, depending on the size of their vascular channels.[6] With the advent of capsule endoscopy (CE) and balloon-assisted enteroscopy (BAE), it is now possible to conduct a thorough investigation of the small bowel, markedly enhancing preoperative diagnostic accuracy.[7] While recent advances in endoscopic techniques have paved the way for successful endoscopic interventions, the majority of larger lesions still require surgical management. It is not regarded as a frequent cause of GI bleeding due to its rarity. Previously, the preoperative diagnosis of this disease was difficult and nearly all instances were discovered during or following the procedure[8]. These days, the small intestine can be targeted thanks to the development of CE and BAE in recent decades[9].

Case Report

A 40-year-old male presented to the outpatient surgical department with upper abdominal pain, frequent vomiting, and intermittent melena persisting for the past 5–6 months. Routine blood investigations revealed a hemoglobin level of 7.2 g/dL, and a peripheral smear demonstrated microcytic hypochromic anemia. Further evaluation showed decreased serum iron levels, elevated total iron-binding capacity (TIBC), reduced transferrin saturation, and low serum ferritin, confirming a diagnosis of iron deficiency anemia. The patient underwent upper gastrointestinal (GI) endoscopy, which revealed pan gastritis(FIG No.01) with a 2 cm bluish swelling near the ampulla with intact mucosa and no active bleeding, consistent with a duodenal hemangioma (FIG No.02, FIG No.03)and Rapid Urease Test (RUT) being positive. Given the classical endoscopic appearance and to avoid the risk of re-bleeding, a biopsy was not performed.



(FIG No.01: UGI Endoscopy showing mucosal hyperemia with features consistent with gastritis and RUT positive) (FIG No.02, 03: UGI Endoscopic images showing a 2 cm bluish swelling near the ampulla with intact mucosa and no active bleeding,)

Magnetic resonance cholangiopancreatography (MRCP) was conducted for academic purposes, further confirming the presence of a ill defined heterogeneous enhancing lesion in the duodenal 26 x 14mm intraluminal region in the medial wall of duodenum D2 distal to ampulla hemangioma.((FIG No.03, FIG No.04,FIG No.05)

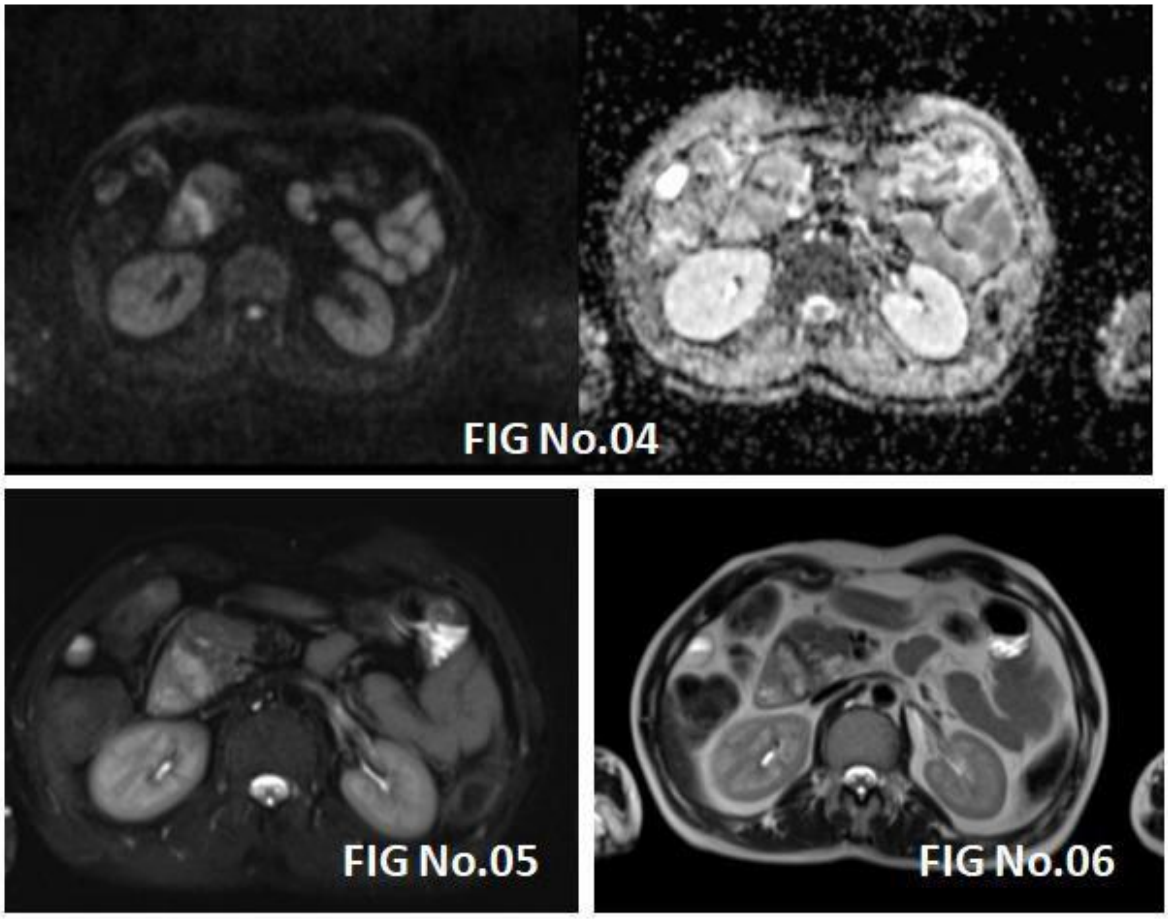


FIG No.04: MRCP images showing hypointense lesion on T1, hyperintense on T2 and shows diffusion restriction. FIG No.05: Image showing ill defined signal intensity measuring 26 x 14 mm noted in the intramural medial wall of D2 distal to ampulla. FIG No.06: Post contrast study shows delayed heterogenous enhancement.

Two units of packed red blood cells (PRBC) were transfused, raising the hemoglobin level to 10.2 g/dL. Other potential causes of anemia were ruled out. The patient was managed conservatively with a proton pump inhibitor (Rabeprazole), antacids, a triple antibiotic regimen for Helicobacter pylori, Albendazole 400 mg, and antispasmodics. Once clinically stable and symptom-free, he was discharged with advice for periodic follow-up with endoscopic evaluation. Considering the anatomic location and the risk of rebleeding due to the size of the D2 hemangioma, an endoscopic approach or laparoscopic and endoscopic cooperative surgery (LECS) was deferred.

This case underscores the importance of considering duodenal hemangioma as a differential diagnosis in cases of upper GI bleeding. It highlights the role of endoscopy in both diagnosis and management and emphasizes the potential necessity for surgical intervention in select cases.

DISCUSSION

Gastrointestinal hemangiomas account for ~0.05% of all GI neoplasms and typically present with GI bleeding and iron deficiency anemia. Due to their

rarity, they are an uncommon cause of GI bleeding. Historically, preoperative diagnosis was challenging, with most cases identified intraoperatively or postoperatively.[8] Capsule endoscopy (CE) and Balloon-assisted enteroscopy (BAE), have made targeting the small intestine possible in recent decades[9] . Intraluminal GI bleeding can be life-threatening, but in cases of obscure GI bleeding (OGIB), this is rare if EGD and colonoscopy with proper preparation are performed, often leading to a diagnostic challenge.[10,11] In 75% of OGIB patients, the lesion is located in the small intestine and may produce recurrent or persistent bleeding.[11]

Small bowel bleeding is rare, accounting for approx. 5% of GI bleeding, with causes varying by age.[12] In patients <40 yrs, inflammatory bowel disease is most common, followed by Dieulafoy's lesions, neoplasms, Meckel's disease, and polyposis syndromes. In those >40 yrs, angioectasia, including AVMs and hemangiomas, is most common, followed by Dieulafoy's lesions, neoplasms, and NSAID-induced ulcers. Angioectasia-related bleeding accounts for approx 20% of cases.[13] Small bowel hemangiomas, comprising approx.10% of benign small bowel tumors, primarily occur in the jejunum and ileum.[14] Iron-deficiency anemia, pain, and intussusception are the most prevalent early signs of small intestinal hemangioma, accounting for 41%, 31%, and 13%, respectively[14].

Endoscopically, they frequently present as submucosal, purple to red, soft, and occasionally pedunculated lesions.[15] Hemangiomas are classified histologically according to vascular size as capillary, cavernous, or mixed, with cavernous hemangiomas being the most prevalent. These are often composed of numerous dilated, irregular blood-filled spaces or sinuses bounded with endothelial cells.[16]

Recent advancements in endoscopic techniques, notably video capsule endoscopy (VCE) and double-balloon enteroscopy (DBE), have significantly enhanced the preoperative diagnosis of small bowel hemangiomas.[16] VCE, a non-invasive procedure, allow for comprehensive visualization of the entire small intestine and is recommended in the routine evaluation of patients with obscure gastrointestinal bleeding, provided there is no intestinal stenosis. However, VCE's diagnostic accuracy may be limited in certain cases, such as those involving vascular sources of bleeding. DBE offers both diagnostic and therapeutic capabilities, including tissue biopsy, lesion localization, and interventions like coagulation and hemostasis through clipping.[15] Despite these advantages, the success rate of achieving total enteroscopy via the antegrade approach remains relatively low, with some studies reporting rates as low as 1.6%. Combining VCE and DBE has been shown to improve the detection rates of bleeding sources in the small bowel, highlighting the complementary roles of these modalities in the management of small bowel diseases.[17]

Endoscopic treatment of small bowel hemangiomas carries risks such as intestinal perforation and lesion persistence. Due to their rich vascularity and potential submucosal involvement, there's an increased risk of significant bleeding during endoscopic procedures, especially if the lesion's depth is underestimated.[15] To mitigate these risks, Laparoscopic and Endoscopic Cooperative Surgery (LECS) has been introduced as a novel approach for tumor resection. LECS combines the visual and therapeutic advantages of both laparoscopic and endoscopic techniques, allowing for precise tumor localization and resection while minimizing invasiveness. This collaborative method enhances surgical accuracy and reduces potential complications associated with traditional endoscopic or open surgical interventions. Recent studies have demonstrated the safety and efficacy of LECS in resecting hemangiomas located in challenging areas, such as the third portion of the duodenum.[18]

Consclusion

Duodenal hemangiomas, though rare, should be considered in cases of obscure gastrointestinal bleeding, particularly in patients presenting with anemia and melena. Advances in endoscopic modalities, including video capsule endoscopy and

double-balloon enteroscopy, have improved preoperative diagnosis, reducing the need for invasive surgical exploration. However, given the risks associated with endoscopic management, including bleeding and lesion persistence, laparoscopic and endoscopic cooperative surgery (LECS) offers a promising alternative for precise and minimally invasive resection. This case highlights the importance of early recognition and a multidisciplinary approach in managing duodenal hemangiomas to optimize patient outcomes.

Author Contributions

Collection and/or assembly of data: Anand Bhandary Panambur

Manuscript writing and approval: Anand Bhandary Panambur & Anand Peter Ignatius.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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