DOI: 10.4274/tjcd.galenos.2021.2021-8-10
Turk J Colorectal Dis 2022;32:154-156

CASE REPORT

A Rare Cause of Gastrointestinal Bleeding: Jejunal Angiodysplasia

Kasım Çağlayan[1], Tamer Sağıroğlu[1], Nurten Türkel Küçükmetin[2], Sibel Özkan Gürdal[1], Hadi Sasani[3]

[1]Tekirdağ Namık Kemal University Faculty of Medicine, Department of Surgery, Tekirdağ, Turkey
[2]Tekirdağ Namık Kemal University Faculty of Medicine, Department of Gastroenterology, Tekirdağ, Turkey
[3]Tekirdağ Namık Kemal University Faculty of Medicine, Department of Radiology, Tekirdağ, Turkey

ABSTRACT

In this article, the diagnosis and treatment process of a case examined for gastrointestinal system (GIS) bleeding and found to have jejunal angiodysplasia (JA) is presented. A 65-year-old female was examined due to intermittent melena persisting for about one year. Laboratory findings were unremarkable, except for iron deficiency anemia. Upper GIS endoscopy, colonoscopy, computed tomography (CT) and CT angiography were normal. Capsule endoscopy revealed multiple lesions consistent with angiodysplasia in the proximal jejunum. Intra-operative enteroscopy revealed multiple JAs between 10 and 20 cm from the Ligament of Trietz. The patient underwent segmental resection and was discharged on the seventh postoperative day.

Keywords: Jejunal angiodysplasia, capsule endoscopy, intraoperative enteroscopy

Introduction

Although intestinal vascular malformations are rare, they should be kept in mind in the differential diagnosis of gastrointestinal system (GIS) bleeding and chronic anemia. Approximately 4% of all GIS bleeds originate from the small intestine.[1] Although angiodysplasia is a common pathology in the gastrointestinal tract, only 15% of the lesions are seen in the small intestine and 77% in the cecum or ascending colon. Angiodysplasia results from ectasia of submucosal vessels and although the mechanism of formation is not fully understood, it is thought to be caused by chronic venous obstruction.[2] Endoscopic examinations, such as gastroscopy, colonoscopy, enteroscopy, and intraoperative enteroscopy, multidetector computed tomography (CT), magnetic resonance angiography, and capsule endoscopy are the methods used for diagnosis.[1,2]

Case Report

A 65-year-old female was admitted with complaints of persistent weakness and palpitation for about one year. Iron deficiency anemia was detected in the blood tests performed elsewhere and iron replacement therapy was started. She was referred to our hospital for examination due resumption of complaints a few months after the medical treatment and a decrease in the routine follow-up hematocrit. On physical examination, the skin and conjunctiva were pale, S1 and S2 sounds were tachycardic (104/min/rhythmic). Melena was detected on rectal examination. Laboratory results were: hematocrit 24.1% (normal range: 35-47%); hemoglobin 7.41 g/dL (normal range: 12.5-16.0 g/dL); ferritin 15.89 ng/mL (normal range: 13-150 ng/mL); iron level: 16.0 µg/dL (normal range: 37-145 µg/dL); iron binding capacity: 435 µg/dL (normal range: 135-392 µg/dL). Iron deficiency due to GI bleeding was considered and etiological investigation was performed. No pathology was detected on gastroscopy and colonoscopy and combined intravenous and oral contrast-enhanced CT (portal phase) was evaluated as normal. Capsule endoscopy was performed with a Pillcam II capsule whose angiography was performed and no bleeding focus could be detected. The capsule passed to the duodenum at approximately 28 minutes and to the colon at the fourth hour. At about 67-69 minutes of capsule transit multiple

Address for Correspondence: Kasım Çağlayan MD,
Tekirdağ Namık Kemal University Faculty of Medicine, Department of Surgery, Tekirdağ, Turkey
E-mail: kasimcaglayan@hotmail.com **ORCID ID:** orcid.org/0000-0001-8856-1969
Received: 24.08.2021 **Accepted:** 04.10.2021

lesions consistent with angiodysplasia were detected in the proximal jejunum (Figure 1). Other regions of the GIS were evaluated as normal. Despite medical treatment, the patient's blood values did not normalize and surgery was decided. Preoperatively, two erythrocyte suspensions were transfused. Laparotomy was performed under general anesthesia. On exploration, edema and dilatation were detected in the jejunal segment approximately 40 cm from the Ligament of Trietz, and enterotomy was performed just distal to the dilated segment and enteroscopy was performed intra-operatively. On enteroscopy, multiple angiodysplastic lesions were seen between 10 and 20 cm from the Ligament of Trietz (Figure 2). Segmental jejunal resection to include the lesions was performed and end-to-end anastomosis was used. The patient was discharged on the seventh postoperative day. At the six-month follow-up, hemoglobin (12.5 g/dL) and hematocrit (36.5%) values were within normal limits. Informed consent was obtained.

Discussion

Gastrointestinal angiodysplasia (GIAD) is an acquired vascular lesion, which typically presents as bright red, irregular, round, slightly raised lesions on visualization. Intestinal angiodysplasias are frequently seen in patients over 60 years of age and are usually localized to the cecum and ascending colon. GIAD was defined in 1974 as a single or multiple vascular superficial lesion that develops from the mucosa or submucosa of the gastrointestinal tract. Today, these lesions are generally referred to as arterio-venous malformation, telangiectasia, angiectasia, or vascular ectasia. This disease increases at similar rates in both genders and after the age of 60, usually in predisposing conditions, such as aortic stenosis, chronic renal failure and von Willebrand's disease, atherosclerosis, cirrhosis or pulmonary disease.[3,4] Although there are some hypotheses about the etiological mechanism, there is as yet no consensus. It has been suggested that the etiology is multifactorial and may involve mild chronic venous obstruction, chronic mucosal ischemia, and potential underlying causes such as comorbidities.

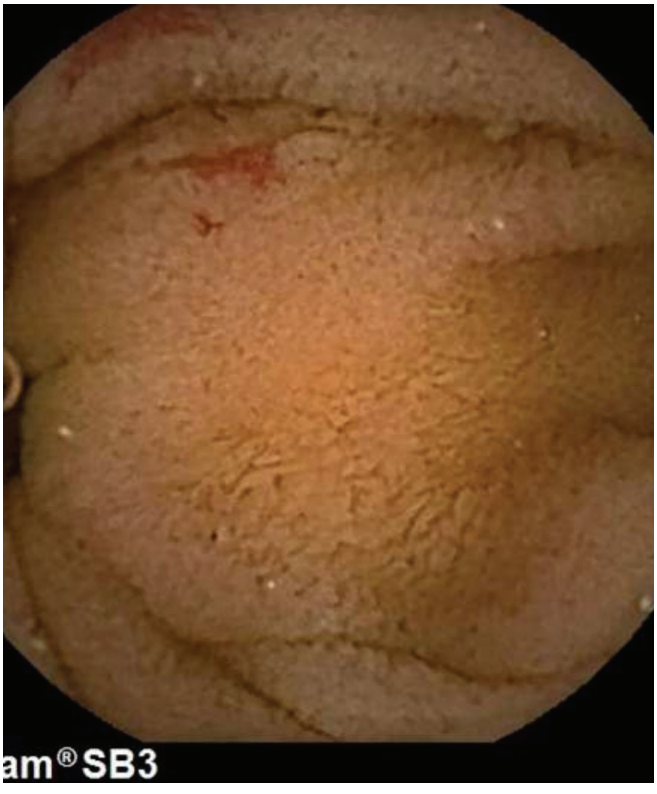

Figure 1. Capsule endoscopy shows multiple angiodysplastic lesions in the jejunal loops (arrows)

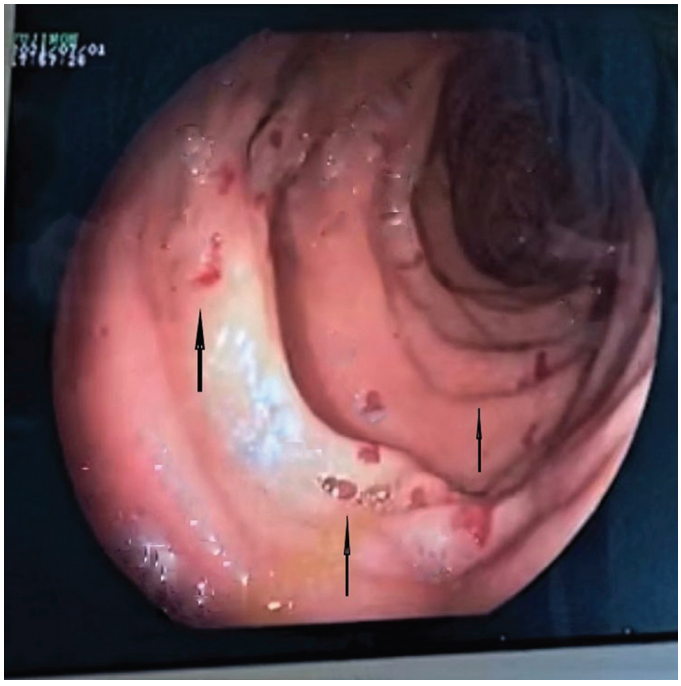

Figure 2. Multiple lesions are seen on intra-operative enteroscopy (arrows)

Since GIADs are mostly asymptomatic, the true prevalence is not known, and they are usually diagnosed in investigations due to unrelated symptoms. Symptomatic GIADs can cause life-threatening heavy bleeding from occult bleeding. Depending on the location of the lesions, the findings may present as hematochezia, melena, or hematemesis. Although the first bleeding stops spontaneously in approximately 90% of cases, there is a tendency to re-bleed. Factors leading to rebleeding include previous bleeding history, history of heart disease, valve abnormalities, arrhythmias, chronic kidney disease, anticoagulant use, cirrhosis, and the presence of multiple angiodysplasia.[5,6] In the current case, melena was detected as a sign of GIS bleeding. In the laboratory findings of the patient, iron deficiency anemia was detected due to

of the patient, iron deficiency anemia was detected due to chronic blood loss, but acute bleeding attacks that could threaten the patient's life were not observed.

Direct endoscopic imaging is the gold standard in the diagnosis of GIAD. On endoscopy, the lesions appear as thin-walled, dot-shaped, mucosal lesions, usually 5-10 mm in size, with a cherry red color and pale surrounding mucosa. Approximately 10-15% of GIADs are located in the small intestine. Small intestines are difficult to investigate due to their anatomically long and curved nature. However, it is possible to overcome this difficulty with a non-invasive method, such as capsule endoscopy. Although there is a high probability of false negatives in capsule endoscopy, there better accuracy with the combined use of capsule and balloon endoscopy. CT angiography, MRI, and radionucleotide scans are potential imaging options for location of acute hemorrhages.[6,7] In the present case, gastroscopy and colonoscopy were performed primarily for diagnosis and CT angiography was performed when the lesion could not be detected. However capsule endoscopy was performed as the diagnosis could not be made. On capsule endoscopy, multiple angiodysplastic lesions were detected in the jejunal proximal segments.

Although there is a tendency to rebleed from multiple lesions, the treatment options for GIADs are still controversial, as bleeding stops spontaneously in approximately 90% of cases. In clinical practice, an individual decision is made about the treatment approach according to the patient's condition. In the literature, hormones (estrogen and progesterone), somatostatin analogs and antiangiogenic drugs are included in the treatment as pharmacological agents.[8] In general, conservative treatments, such as correction of anemia can be performed in asymptomatic cases, but pharmacological treatments are not appropriate in cases that need transfusion.[6] GIAD treatment options, other than pharmacological agents, include endoscopic treatments (argon plasma coagulation, mechanical clip placement, multipolar electrocoagulation, laser photoablation, sclerotherapy, and band ligation), angiographic embolization, and surgical resection. Despite the rates of recurrent bleeding, endoscopic methods are effective in the initial treatment of GIADs.[5] Surgical interventions may be considered in cases where endoscopic or radiological hemostasis cannot be achieved.[6,9] Examination of the entire small intestine with intra-operative enteroscopy during surgery is an appropriate approach. In the current case, surgical treatment was chosen because the patient needed continuous blood transfusions and curative treatment was technically unfeasible with endoscopic or radiological methods. After multiple angiodysplastic foci were detected in the proximal jejunum by capsule endoscopy, resection margins were determined to include all angiodysplastic foci, aided by intra-operative enteroscopy, and a segment of approximately 30 cm was resected.

JA(s) constitutes a small proportion of the etiologies of GIS bleeds, and angiodysplasias should be considered in patients without pathology in the upper and lower endoscopic examinations. Capsule endoscopy is an important diagnostic method for location in cases without acute bleeding but may be combined with balloon endoscopy. Intra-operative enteroscopy has an important place in determining the exact location and extent of the lesions and determining the resection margins in cases undergoing surgical resection.

Ethics

Informed Consent: It was obtained.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: T.S., K.Ç., N.T.K., Concept: K.Ç., T.S., S.Ö.G., Design: K.Ç., T.S., H.S., S.Ö.G., Data Collection or Processing: K.Ç., N.T.K., H.S., T.S., Analysis or Interpretation: K.Ç., H.S., N.T.K., Literature Search: K.Ç., N.T.K., S.Ö.G., Writing: K.Ç., T.S., N.T.K.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study received no financial support.

References

1. Goel V, Verma AK, Sethi S, Puri SK. MDCT demonstration of jejunalangiodysplasia: a nun common cause of lower gastrointestinal haemorrhage. BMJ Case Rep 2014;2014:bcr2013203414.
2. Tiwary SK, Hakim MZ, Kumar P, Khanna AK. Jejunal angiodysplasia causing recurrent gastrointestinal bleeding presenting severe anaemia and melena. BMJ Case Rep 2015;2015:bcr2015212798.
3. Becq A, Rahmi G, Perrod G, Cellier C. Hemorrhagic angiodysplasia of the digestive tract: pathogenesis, diagnosis, andmanagement. Gastrointest Endosc 2017;86:792-806.
4. Grassi R, diMizio R, Romano S, Cappabianca S, del VecchioW, Severini S. Multiple jejunal angiodysplasia detected by enema-helical CT. Clin Imaging 2000;24:61-63.
5. Jackson CS, Strong R. Gastrointestinal Angiodysplasia: Diagnosis and Management. Gastrointest Endosc Clin N Am 2017;27:51-62.
6. Beg S, Ragunath K. Review on gastrointestinal angiodysplasia throughout the gastrointestinal tract. Best Pract Res Clin Gastroenterol 2017;31:119-125.
7. Runza G, Barbiera F, Maffei E, Punzo B, Cavaliere C, Cademartiri F. Multi-Dedector CT Enterography to detect jejunal angiodysplasia: challenging cause of gastrointestinal bleeding. Acta Biomed 2012;92:e2021144.
8. Nardone G, Compare D, Martno A, Rocco A. Pharmacological teratment of gastrointestinal bleeding due to angiodysplasias: A position paper of the Italian Society of Gastroenterology. Dig Liver Dis 2018;50:542-548.
9. Dray X, Camus M, Coelho J, Ozenne V, Pocard M, Marteau P. Treatment of gastrointestinal angiodysplasia and unmet needs. Dig Liver Dis 2011;43:515-522.