Isolated Jejunal Angiodysplasia: A Rare Cause of Massive Lower Gastrointestinal Bleeding Requiring Surgical Resection with an Unexpected Localization

Mehmet Tolga Kafadar^I, Metin Yalaza², Gürkan Değirmencioğlu³, Aydın İnan⁴

¹Clinic of General Surgery, Mehmet Akif İnan Training and Research Hospital, Şanlıurfa, Turkey ²Clinic of General Surgery, Division of Surgical Oncology, Numune Training and Research Hospital, Ankara, Turkey ³Department of General Surgery, Turgut Özal University School of Medicine, Ankara, Turkey ⁴Clinic of General Surgery, Ankara Umut Hospital, Ankara, Turkey

Angiodysplasias are most commonly observed in the cecum and ascending colon. The number of cases with angiodysplasias in the jejunum is very few. The endovascular transcatheter embolization method can be used in cases of lower gastrointestinal tract bleeding when endoscopy is not effective. According to the nature of lesion and source of bleeding, embolization is performed using the appropriate embolic agent. A surgical approach is used when endoscopic treatment methods and mesenteric angiography attempts fail. Here we present a rare case of jejunal anjiodysplasia with massive lower gastrointestinal bleeding treated with selective arterial embolization followed by urgent surgery because of a failed attempt to gain hemodynamic stability using both conservative and endoscopic methods.

Keywords: Lower gastrointestinal bleeding, angiodysplasia, jejunum

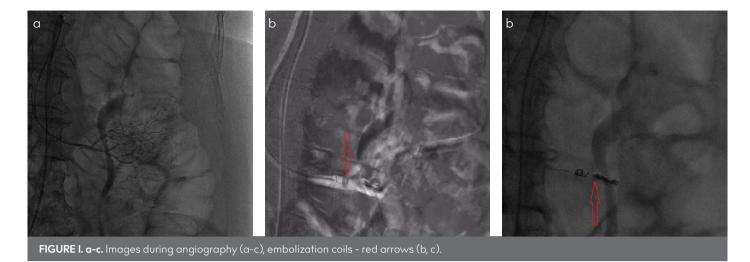
INTRODUCTION

Gastrointestinal (GI) tract bleeding is associated with a group of diseases that have high mortality rates and for which it is difficult to make diagnosis and differential diagnosis. The most common causes of lower GI bleeding in adults older than 60 years of age are vascular ectasias (angiodysplasias) and diverticulosis (I). Here we present a case of isolated jejunal angiodysplasia treated with selective arterial embolization followed by urgent surgery.

CASE PRESENTATION

A 70-year-old female patient with fatigue and anal bleeding was admitted to the gastroenterology clinic. Hematochezia was detected on rectal examination. The patient's hemoglobin level on admission was 7.3 g/dL. The hemoglobin level continued to drop as a result of bleeding. The active source of bleeding was not detected on endoscopy and colonoscopy. Extravasation located at the segment about 50–60 cm away from Treitz ligament was observed by IV contrast tomography screening. The decision to perform angiography was made because no clear focus of bleeding was detected by endoscopic examination. Superselective embolization was performed by placing embolic agents (coils) in the feeding artery (branch of the superior mesenteric artery) of the bleeding bowel segment accompanied by angiography screening at the interventional radiology department (Figure I). She was administered 6 units of erythrocyte suspension within 24 h prior to the urgent surgery. An urgent surgical approach was considered because of deterioration of the general condition and a decrease in the hemoglobin level of the patient. Approximately 60 h passed between admission to the gastroenterology clinic and the surgery. With the guidance of jejunal segment, where the coil was preoperatively placed, jejunotomy was performed approximately 30 cm away from Treitz ligament, targeting the segment observed on tomography. Many hemorrhagic foci were observed in the mucosal surfaces of the jejunotomy region and were considered to be angiodysplasias. The ends of these lesions were resected, both in the proximal and distal segments (Figure 2). In total, a 40-cm-long jejunal segment was resected beginning from 25 cm to 65 cm of the jejunum, and end-to-end anastomosis was performed. No other gross pathology observed in the ongoing exploration. The patient was discharged with full recovery after 7 days of hospitalization as no

This study was presented at the 20th National Surgical Congress, 13-17 April 2016, Antalya, Turkey





 $\ensuremath{\textbf{FIGURE 2.}}$ Hemorrhagic foci at the mucosa of the resected jejunal segment

problems or complications were observed. Follow-ups over 3 months after the surgery were uneventful. The postoperative pathological examination of the resected material indicated angiodysplasia (Figure 3). Informed consent was obtained from patient who participated in this case.

DISCUSSION

Angiodysplasias are the most common vascular abnormalities of the GI tract and are caused by intestinal submucosal vein dilatations. They are often observed in the cecum and ascending colon but are unlikely to occur in the jejunum (I). Endoscopic methods for treating lower GI bleedings are the most commonly used and have been found to have steadily increasing success rates in recent years. Angiography, technetium-99m-labeled erythrocyte scintigraphy, and small bowel screening should be performed when bleeding, which cannot be diagnosed by colonoscopy alone, is suspected (2). Selective superior mesenteric artery (SMA) angiography can show all parts of the small bowel and right colon (3).

Since the advent of capsule endoscopy, a noninvasive, safe, and well-tolerated technique, in 2001 it has become possible to directly visualize the small intestine. The diagnostic accuracy of capsule endoscopy (CE) for GI system bleeding of unknown etiology (GSBUE) has been found to range between 41% and 92% in several studies. Pennazio et al. (4) performed CE in 100 patients with GSBUE and found the diagnostic accuracy to be 92.3%. Embolization treatment is associated with up to 20% risk of intestinal infarcts; thus, distal embolization should be performed as much as possible using temporary blocking agents and superselective catheterization techniques. Complications depending on the contrast agent, including arterial dissection and occlusion, renal failure, and bowel infarction, and on vasopressin, including myocardial infarction, may develop. Gordon et al. (5) examined I7 patients with angiographically detected bowel or colon bleeding. Of these, 14 patients had superselective catheterization, and embolization was successfully performed in 13 of them without any necessities of additional treatment; further, bowel necrosis was not observed among the patients. In our case, superselective embolization was performed because endoscopic treatment methods are not effective enough, and we failed to obtain the desired success. We think that embolization failed because of the embolization of the minor branches of the major vessel instead of branch of the superior mesenteric artery because of technical problems. No complications related to embolization were detected in our patient. There were no signs of ischemia of the bowels during the intraoperative exploration. To avoid risking the patient's life, surgery was performed immediately without a plan of second embolization because of the active bleeding aftermath of the first embolization, deterioration of general the condition of the patient, and concern about re-experiencing the technical problems that occurred during the first embolization. Poultsides et al. (6) reported that a waiting period before surgery is unnecessary for the patients with a deteriorating general condition, active bleeding, 6 or more units of blood exchange.

Urgent surgery is essential in 5%–10% of patients with lower Gl bleeding, which cannot be treated using nonsurgical methods, require repeated blood transfusions, or are associated with continuous bleeding (7, 8). For an optimal surgical process, the patient should be as stable as possible for the hemodynamics and the bowel part to be resected should be identified previously. Searching the lesion by inspection and palpation during surgery is recommended in patients without a certain source of bleeding, and if the lesion cannot be detected in this manner, it is recommended to detect the source

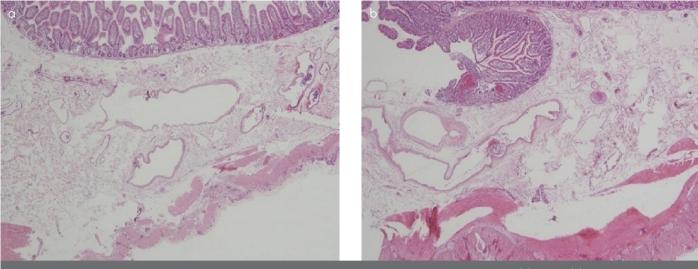


FIGURE 3. a, b. Submucosal, irregularly shaped, dilated, and thin-walled vessels in the jejunal tissue- HEx20 (a), HEx40 (b)

of bleeding by patting and clamping the bowel segments. Blind resections were associated with re-bleeding rates of 75% and mortality rates as high as 57%. In case of massive bleeding, the source of bleeding can be detected by ileotomies and/or colotomies (9).

Intraoperative enteroscopy, on the other hand, is a technique in which small intestinal examination can be performed by advancing an endoscope from the enterotomy site, mouth, or rectal opening during surgery. It has a diagnostic accuracy of 60%–88% and recurrent bleeding rates ranging between 13% and 60%. Hartmann et al. (10) compared CE and intraoperative enteroscopy in 42 patients and found that CE has a sensitivity of 95%, specificity of 75%, positive predictive value of 95%, and negative predictive value of 86%. A very good correlation has been observed between CE and intraoperative enteroscopy. CE or intraoperative enteroscopy could not be used for our patient because of lack of equipment.

In our case, jejunotomy was performed on the segment where the embolization agents had been placed preoperatively, and the sources of bleeding were observed. The postoperative period was uneventful. There are very few reported cases with isolated jejunal angiodysplasia requiring surgical resection because of a failed attempt to control bleeding using nonoperative methods (II).

CONCLUSION

Angiodysplasias are rare in the jejunum. Although angiodysplasias are generally located in the cecum and ascending colon, isolated jejunal angiodysplasia should be kept in mind during the differential diagnosis of massive lower GI bleeding, and surgeons must be aware of these rare cases and the methods of managing them.

Informed Consent: Written informed consent was obtained from patient who participated in this study. Peer-review: Externally peer-reviewed.

Author contributions: Concept - M.T.K., A.İ.; Design - M.T.K., G.D., A.İ.; Supervision - M.T.K., M.Y., G.D.; Resource - M.T.K., M.Y.; Materials - M.T.K., G.D.; Data Collection and/or Processing - M.T.K.; Analysis and/or Interpretation - M.T.K., A.İ.; Literature Search - M.T.K., M.Y.; Writing - M.T.K., M.Y.; Critical Reviews - M.T.K., A.İ.

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

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

REFERENCES

- I. Farrell JJ, Friedman LS. Gastrointestinal bleeding in the elderly. Gastroenterol Clin North Am 2001; 30: 377-407. [CrossRef]
- Rossini FP, Ferrari A, Spandre M, Cavallero M, Gemme C, Loverci C, et al. Emergency colonoscopy. World J Surg 1989; 13: 190-2. [CrossRef]
- Fiorito JJ, Brandt LJ, Kozicky O, Grosman IM, Sprayragen S. The diagnostic yield of superior mezenteric angiography: correlation with the pattern of gastrointestinal bleeding. Am J Gastroenterol 1989; 84: 878-81.
- Pennazio M, Santucci R, Rondonotti E, Abbiati C, Beccari G, Rossini FP, et al. Outcome of patients with obscure gastrointestinal bleeding after capsule endoscopy: report of 100 consecutive cases. Gastroenterology 2004; 126: 643-53. [CrossRef]
- Gordon RL, Ahl KL, Kerlan RK, Wilson MW, LaBerge JM, Sandhu JS, et al. Selective arterial embolization for the control of lower gastrointestinal bleeding. Am J Surg 1997; 174: 24-8. [CrossRef]
- Poultsides GA, Kim CJ, Orlando R 3rd, Peros G, Hallisey MJ, Vignati PV. Angiographic embolization for gastroduodenal hemorrhage: safety, efficacy, and predictors of outcome. Arch Surg 2008; 143: 457-61. [CrossRef]
- Çolak B, Erdal H, Arhan M, İbiş M, Ünal S. A rare cause of massive lower gastrointestinal bleeding; jejunal Dieulafoy's lesion. Turk J Gastroenterol 2014; 25 Suppl I: 268-9. [CrossRef]
- Andrei GN, Popa B, Gulie L, Diaconescu BI, Martian BV, Bejenaru M, et al. Highlighted steps of the management algorithm in acute lower gastrointestinal bleeding - case reports and literature review. Chirurgia 2016; III: 74-9.
- 9. Szold A, Katz LB, Lewis BS. Surgical approach to occult gastrointestinal bleeding. Am J Surg 1992; 163: 90-3. [CrossRef]

- Hartmann D, Schmidt H, Bolz G, Schilling D, Kinzel F, Eickhoff A, et al. A prospective two-center study comparing wireless capsule endoscopy with intraoperative enteroscopy in patients with obscure GI bleeding. Gastrointest Endosc 2005; 6I: 826-32. [CrossRef]
- II. Assenza M, Ricci G, Clementi I, Antoniozzi A, Simonelli L, Modini C. Gastrointestinal bleeding in emergency setting: two cases of intestinal angiodysplasia and unusual conservative surgical treatment. Clin Ter 2006; 157: 345-348.