Trauma Associated Ectopic Ileal Varices

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ABSTRACT Ectopic varices (EcV) are portosystemic collaterals that can be observed anywhere in the gastrointestinal tract (GIT) except stomach and esophagus. It accounts for 1-5% of all variceal hemorrhages.\textsuperscript{1} The most common cause of the etiopathogenesis is portal hypertension. Ileum is the rarest localization of ectopic varices in the gastrointestinal tract (GIT).\textsuperscript{2} There are no clear treatment guidelines. In the literature, invasive radiological and surgical methods have been tried for the treatment.

Here, we have reported a case who was diagnosed with EcV localised in the ileum and treated with angiographic coil embolization. In this case, thrombus in the inferior vena cava (IVC) due to penetrating trauma and associated systemic venous pressure increase was responsible for the pathogenesis unlike other EcV in the literature.\textsuperscript{2}

CASE REPORT

A 37-year-old male patient presented to our clinic with complaint of intermittent black color defecation since 2011. The patient underwent multiple esophagogastroduodenoscopies (EGD) and colonoscopies previously. Computed tomography (CT) angiography scan did not demonstrate a clear identification of the source of bleeding. Capsule endoscopy was also performed once, but the origin of hemorrhage was not detected. The patient had 3 episodes of syncope in the past and had a total of ten units of blood transfusion over the years. The patient did not have comorbid diseases and drug use. He had a history of stabbing in 2004. On physical examination, his skin was pale but his vital signs were stable. In the abdominal examination, a linear scar area of 3 cm in diameter was observed in the hypogastric area. The rectal examination was compatible with melena. In his laboratory values, hemoglobin: 7.1 g/dl, platelet count: 284,000/mm\textsuperscript{3}, PT INR: 1. After initial evaluation, four units of erythrocyte suspension were replaced. EGD and colonoscopy were repeated in our clinic, but bleeding was not observed. In biphasic abdomen computed tomographic angiograms (CTA), EcV protruding into the lumen of the ileal segment was observed at the right paramedian portion of
lower quadrant (at the position of penetrating trauma scar) (Figure 1). In addition, IVC was finely calibrated at bifurcation level and more distal to thrombosis. Numerous pelvic and intraabdominal venous collateralization was observed. Doppler ultrasonography showed a chronic complete occlusion in the caudal section of the IVC and the bilateral main iliac vein. It was found that both lower extremities and pelvic veins were drained through deep pelvic venous collaterals. The patient was consulted to the Department of Interventional Radiology and angiography was performed. The venographies were obtained through the right femoral vein; the right iliac vein and IVC were occluded. Portosystemic varicose collateralization was observed on the right side of the midline (Figure 2). The late phase images showed that ileal varicose veins drained into the superior mesenteric vein (Figure 3). Fistulosis was embolized with 0.018 inch pushable coils (VortX 0.018 Fibered Platinum Coils, Boston Scientific Corporation, Natick, MA) after venous catheterization of the EcV with 2.8 Fr microcatheter (Progreat microcatheter; Terumo, Tokyo, Japan) through the right femoral vein. There was no filling in EcV in control venography (Figure 4). The patient having stable hemogram values during 3-day follow-up was discharged and had been followed up for about 1 year had no GIT bleeding again.

**DISCUSSION**

Variceal hemorrhage is a common and serious complication of portal hypertension. In esophagogastric varices, 6-week mortality rate varies between 10-20%. The risk of EcV hemorrhage and mortality is 4 fold higher and approximately 40%. According to the results of the survey conducted by the Japan Portal Hypertension Association about EcV, localisation was most common in rectum (44.5%) and most rare in ileum (2%) in GIT. In Western countries, intestinal EcV are more common than rectal localisation. In a study, asymptomatic patients with portal hypertension were screened by capsule endoscopy, intestinal EcV were observed in 8.1% (3/37) of the patients.

The most common cause of EcV is cirrhosis. In addition, the history of abdominal surgery in patients with portal hypertension is an important risk factor for small intestine varicose veins. Postoperative adhesions between abdominal wall and small intestine is also thought to be important in the pathogenesis.

The pathophysiology of varicose veins should be known when planning the treatment of EcV. Portosystemic flow is provided by collaterals developed due to portal hypertension because of various reasons. Magnetic resonance and CT angiography can be used for varicose veins hemodynamics. In our patient, the systemic circulation drained into the portal circulation as a result of the systemic circulation occlusion unlike in other EcV. Abdominal CT angiography revealed total occlusion in the iliac vein and
IVC due to penetrating trauma and ectopic varices protruding into the lumen in ileal segment. In the venogram taken during the angiography, an ectopic anastomosis was found between the inferior epigastric vein and the ileal branches of the superior mesenteric vein.

Numerous guidelines have been suggested for the management of esophagogastric variceal bleeding. However, there is no clear guidelines for the treatment of EcV. Endoscopic treatments (Endoscopic band ligation (EBL) and endoscopic injection sclerotherapy (EIS)) are very limited especially in EcV hemorrhage of the small intestine. Endoscopic treatment have been performed successfully especially for duodenal varices. In few reported cases in the literature, invasive radiological treatments (Balloon-occluded retrograde transvenous obliteration (B-RTO), transjugular intrahepatic portosystemic shunt (TIPS)) and surgical resection are among the applied treatments for the treatment of EcV. In our case, embolization was performed in the treatment of EcV in the ileum for the first time unlike in other cases in the literature.

In conclusion, EcV should be considered during the investigation of the etiology of gastrointestinal hemorrhage, especially in the patients who has the history of abdominal surgery or trauma even if there is no portal hypertension. Because its diagnosis and treatment is different from the esophagogastric varices, multidisciplinary approach (gastroenterology, interventional radiology and general surgery) is necessary.

Informed Consent
Informed consent was taken from the patient.

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Conflict of Interest
No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions
Idea/Concept: Abdullah Murat Buyruk; Design: Abdullah Murat Buyruk; Control/Supervision: Galip Ersöz; Data Collection and/or Processing: Abdullah Murat Buyruk; Analysis and/or Interpretation: Abdullah Murat Buyruk; Literature Review: Abdullah Murat Buyruk; Writing the Article: Abdullah Murat Buyruk, Celal Çınar; Critical Review: Galip Ersöz, Ömer Öztüeniz; References and Fundings: Abdullah Murat Buyruk.
REFERENCES

1. Norton ID, Andrews JC, Kamath PS. Management of ectopic varices. Hepatology. 1998;28(4):1154-8. [Crossref] [PubMed]

2. Watanabe N, Toyonaga A, Kojima S, Takashimizu S, Oho K, Kokubu S, et al. Current status of ectopic varices in Japan: results of a survey by the Japan Society for Portal Hypertension. Hepatol Res. 2010;40(8):763-76. [Crossref] [PubMed]

3. de Franchis R, Bavero VI Faculty. Expanding consensus in portal hypertension: report of the Bavero VI Consensus Workshop: stratifying risk and individualizing care for portal hypertension. J Hepatol. 2015;63(3):743-52. [Crossref] [PubMed]

4. Saad WE, Lippert A, Saad NE, Caldwell S. Ectopic varices: anatomical classification, hemodynamic classification, and hemodynamic-based management. Tech Vasc Interv Radiol. 2013;16(2):108-25. [Crossref] [PubMed]

5. Norton ID, Andrews JC, Kamath PS. Management of ectopic varices. Hepatology. 1998;28(4):1154-8. [Crossref] [PubMed]

6. De Palma GD, Rega M, Masone S, Persico F, Siciliano S, Patrone F, et al. Mucosal abnormalities of the small bowel in patients with cirrhosis and portal hypertension: a capsule endoscopy study. Gastrointest Endosc. 2005;62(4):529-34. [Crossref] [PubMed]

7. Masuda K, Takenaga S, Ashida H, Ojiri H. Variceal bleeding in the small intestine successfully treated with balloon-occluded retrograde transvenous obliteration using N-butyl-2-cyanoacrylate: a case report. Radiol Case Rep. 2016;13(2):400-3. [Crossref] [PubMed] [PMC]

8. Watson GA, Abu-Shanab A, O’Donohoe RL, Iqbal M. Enteroscopy management of ectopic varices in a patient with liver cirrhosis and portal hypertension. Case Reports Hepatol. 2016;2016:2016642. [Crossref] [PubMed] [PMC]

9. Angell P, Bernardi M, Villanueva C, Francoz C, Mookerjee RP, Trebicka J, et al. EASL Clinical Practice Guidelines for the management of patients with decompensated cirrhosis. J Hepatol. 2018;69(2):406-92. [Crossref] [PubMed]

10. Garcia-Tsao G, Abraldes JG, Berzigotti A, Bosch J. Portal hypertensive bleeding in cirrhosis: risk stratification, diagnosis, and management: 2016 practice guidance by the American Association for the study of liver diseases. Hepatology. 2017;65(1):310-35. [Crossref] [PubMed]

11. Sato T, Yamazaki K, Toyota J, Karino Y, Ohmura T, Akaike J. Ileal varices treated with balloon-occluded retrograde transvenous obliteration. Gastroenterology Res. 2009;2(2):122-5. [Crossref] [PubMed] [PMC]

12. Guth E, Katz MD, Hanks SE, Teitelbaum GP, Ralls P, Korula J. Recurrent bleeding from ileal varices treated by transjugular intrahepatic portosystemic shunt: value of Doppler ultrasonography in diagnosis and follow-up. J Ultrasound Med. 1996;15(1):67-9. [Crossref] [PubMed]

13. Masuda K, Takenaga S, Ashida H, Ojiri H. Variceal bleeding in the small intestine successfully treated with balloon-occluded retrograde transvenous obliteration using N-butyl-2-cyanoacrylate: a case report. Radiol Case Rep. 2018;13(2):400-3. [Crossref] [PubMed] [PMC]

14. López-Benítez R, Seidensticker P, Richter GM, Stampfl U, Haitscheidt P. [Case report: massive lower intestinal bleeding from ileal varices: treatment with transjugular intrahepatic portosystemic shunt (TIPSS)]. Radiologie. 2007;47(5):407-10. [Crossref] [PubMed]

15. Ohtani T, Kajiwara E, Suzuki N, Kawasaki A, Sadowshima S, Sakata H, et al. Ileal varices associated with recurrent bleeding in a patient with liver cirrhosis. J Gastroenterol. 1999;34(2):264-8. [Crossref] [PubMed]

16. Kobayashi K, Yamaguchi J, Mizoe A, Isomoto I, Kishiishi T, Izawa K, et al. Successful treatment of bleeding due to ileal varices in a patient with hepatocellular carcinoma. Eur J Gastroenterol Hepatol. 2001;13(1):53-6. [Crossref] [PubMed]

17. Ueda J, Yoshida H, Mamada Y, Tanai N, Mizuguchi Y, Shimmizu T, et al. Successful emergency enteroectomy for bleeding ileal varices in a patient with liver cirrhosis. J Nippon Med Sch. 2006;73(4):221-5. [Crossref] [PubMed]

18. Mashimo M, Haraj J, Nita A, Oohira M, Yamaguchi S, Yamada S, et al. [A case of ruptured ileal varices associated with alcoholic liver cirrhosis]. Nippon Shokakibyo Gakkai Zasshi. 2007;104(4):561-7. [PubMed]