CASE REPORT

Idiopathic Ileocolonic Varices Coexisting with a Colon Polyp Treated Successfully by Endoscopy: A Case Report and Literature Review

Takao Miwa, Takashi Ibuka, Noritaka Ozawa, Tomohiko Sugiyama, Masaya Kubota, Kenji Imai, Hiroyasu Sakai, Koji Takai, Hiroshi Araki and Masahito Shimizu

Abstract:
Colonic varices are usually associated with portal hypertension. Idiopathic colonic varices are extremely rare. A 68-year-old man with a positive fecal occult blood test result underwent colonoscopy. We detected idiopathic ileocolonic varices and a coexisting ascending colon polyp. While reviewing the literature, we found cases of biopsies and polypectomies resulting in significant bleeding. We herein report a case of idiopathic ileocolonic varices coexisting with a colon polyp treated successfully by endoscopy. The coexistence of colonic varices and a colorectal lesion that requires endoscopic treatment may lead to significant bleeding. During management, the development of a treatment strategy and obtaining informed consent are necessary.

Key words: Colon polyp, Colonic varices, Endoscopic mucosal resection, Idiopathic, Ileocolonic varices


Introduction
Colonic varices are rare and usually associated with portal hypertension. Idiopathic colonic varices are very rare. In most cases, the major complement is hematochezia. Colon polyps are typically treated by endoscopy, and bleeding is the most common serious complication occurring after endoscopic mucosal resection (1). The coexistence of colonic varices and colorectal lesions that require endoscopic procedures may cause significant bleeding. When reviewing case reports of previous idiopathic colonic varices cases (2-30), we found cases wherein significant bleeding was caused by a biopsy or polypectomy (20, 31).

We herein report the first case of idiopathic ileocolonic varices coexisting with a colon polyp treated successfully by endoscopy.

Case Presentation
A 68-year-old man was referred to our hospital because he had positive fecal occult blood screening test results. He had a history of duodenal ulcer and operation for sinusitis in his 30s. During the examination of the duodenal ulcer, no esophageal varices were detected. He also had a history of hematochezia twice in his 50s. He had never been found to have any liver function abnormality or liver disease, portal hypertension, colon malignancy, pancreatitis, or congestive heart failure. He had no history of abdominal operation, blood transfusion or a family predisposition. He was a social drinker and never drank more than 60 g alcohol per day.

Total colonoscopy revealed the presence of ileocolonic varices in the terminal ileum and throughout the colon, extending from the ascending colon to the rectum (Fig. 1A-F). In addition to ileocolonic varices, the patient had a 12-mm sessile polyp with a clear margin, slightly uneven surface, and faded color without reddish area in the ascending colon on the colonic varices. The Japan Narrow-band imaging Expert Team (JNET) classification (32) was type 2A (Fig. 2A-C).

Endoscopic ultrasonography (EUS) using a small-caliber ultrasonic probe showed 3- to 4-mm colonic varices in the submucosa under the polyp (Fig. 2D). Considering the risk of bleeding after resection, we decided to further continue...
Figure 1. Total colonoscopy revealed the presence of ileocolonic varices at the terminal ileum and throughout the colon, extending from the ascending colon to the rectum, A: Terminal ileum, B: Ascending colon, C: Transverse colon, D: Descending colon, E: Sigmoid colon, F: Rectum.

Figure 2. A: Conventional endoscopy in the ascending colon shows a 12-mm polyp, existing on the colonic varices, B: Narrow-band imaging, C: Chromoendoscopy, D: Endoscopic ultrasound with a small-caliber ultrasonic probe showed 3- to 4-mm colonic varices in the submucosa.

with the investigation of the portal hypertension and hemodynamics of the ileocolonic varices. The laboratory test results revealed a history of HBV infection, but no other data supported the suspicion of a present viral hepatitis, autoimmune disease, endocrine disease, or other liver disease. Mac-2 binding protein glycan isomer (M2BPGi) was within nor-
Table 1.

<table>
<thead>
<tr>
<th>&lt;Peripheral blood&gt;</th>
<th>ALT</th>
<th>13 U/L</th>
<th>&lt;SeroLOGY&gt;</th>
</tr>
</thead>
<tbody>
<tr>
<td>WBC 5,580 /μL</td>
<td>LDH</td>
<td>173 U/L</td>
<td>IgG 1,197 mg/dL</td>
</tr>
<tr>
<td>RBC 466x10⁴ /μL</td>
<td>ALP</td>
<td>151 U/L</td>
<td>IgA 226 mg/dL</td>
</tr>
<tr>
<td>Hb 14.6 g/dL</td>
<td>γ-GTP</td>
<td>19 U/L</td>
<td>IgM 54 mg/dL</td>
</tr>
<tr>
<td>Ht 42.8 %</td>
<td>ChE</td>
<td>310 U/L</td>
<td>CRP 0.03 μg/dL</td>
</tr>
<tr>
<td>Plt 21.6x10⁴ /μL</td>
<td>T-chol</td>
<td>248 mg/dL</td>
<td>Ferritin 49.8 μg/dL</td>
</tr>
<tr>
<td>&lt;Coagulation&gt;</td>
<td>TG</td>
<td>100 mg/dL</td>
<td>M2BPGi 0.39 COI</td>
</tr>
<tr>
<td>PT 111 %</td>
<td>UA</td>
<td>5.4 mg/dL</td>
<td>&lt;Viral markers&gt;</td>
</tr>
<tr>
<td>PT-INR 0.98</td>
<td>BUN</td>
<td>13.6 mg/dL</td>
<td>HBs-Ag (-)</td>
</tr>
<tr>
<td>APTT 23 sec</td>
<td>Cr</td>
<td>1.1 mg/dL</td>
<td>HBs-Ab (-)</td>
</tr>
<tr>
<td>FIB 258 mg/dL</td>
<td>NH3</td>
<td>42 μg/dL</td>
<td>HBc-Ab (+)</td>
</tr>
<tr>
<td>FDP &lt;2.0 μg/mL</td>
<td>Na</td>
<td>139 mEq/L</td>
<td>HBc-Ab (-)</td>
</tr>
<tr>
<td>D dimer 0.8 μg/mL</td>
<td>K</td>
<td>4.2 mEq/L</td>
<td>HBc-Ab (+)</td>
</tr>
<tr>
<td>AT III 94 %</td>
<td>Cl</td>
<td>106 mEq/L</td>
<td>HBV-DNA (PCR) (-)</td>
</tr>
<tr>
<td>&lt;Biochemistry&gt;</td>
<td>Fe</td>
<td>139 μg/dL</td>
<td>HCV-Ab (-)</td>
</tr>
<tr>
<td>TP 6.5 g/dL</td>
<td>UIBC</td>
<td>179 μg/dL</td>
<td>CMV pp65 (C10/C11) (-)</td>
</tr>
<tr>
<td>Alb 4.2 g/dL</td>
<td>Cu</td>
<td>89 μg/dL</td>
<td>EBV VCA-IgM Ab &lt;10 x</td>
</tr>
<tr>
<td>T-Bill 1.9 mg/dL</td>
<td>Zu</td>
<td>86 μg/dL</td>
<td>EBV VCA-IgM Ab &lt;10 x</td>
</tr>
<tr>
<td>D-Bill 0.2 mg/dL</td>
<td>BTR</td>
<td>8.46</td>
<td>EBNA 40 x</td>
</tr>
<tr>
<td>AST 19 U/L</td>
<td>AFP</td>
<td>2.3 ng/mL</td>
<td></td>
</tr>
</tbody>
</table>


Table 1.

- **Peripheral blood**
  - WBC: 5,580 /μL
  - RBC: 466x10⁴ /μL
  - Hb: 14.6 g/dL
  - Ht: 42.8 %
  - Plt: 21.6x10⁴ /μL

- **Coagulation**
  - TG: 100 mg/dL
  - PT: 111 %
  - PT-INR: 0.98
  - APTT: 23 sec
  - FIB: 258 mg/dL
  - FDP: <2.0 μg/mL
  - D dimer: 0.8 μg/mL

- **Biochemistry**
  - TP: 6.5 g/dL
  - Alb: 4.2 g/dL
  - T-Bill: 1.9 mg/dL
  - D-Bill: 0.2 mg/dL
  - AST: 19 U/L

- **SeroLOGY**
  - IgG: 1,197 mg/dL
  - IgA: 226 mg/dL
  - IgM: 54 mg/dL
  - CRP: 0.03 μg/dL

Discussion

Varices are frequently associated with portal hypertension resulting from liver cirrhosis and other less-common causes. Colonic varices are rare, and it is reported that the incidence of colonic varices is 0.07% (2 of 2912 cases) (34), whereas that of rectal varices is 3%-7% in cirrhotic patients on endoscopy (35, 36). Idiopathic ileocolonic varices with no under-

Discussion

Varices are frequently associated with portal hypertension resulting from liver cirrhosis and other less-common causes. Colonic varices are rare, and it is reported that the incidence of colonic varices is 0.07% (2 of 2912 cases) (34), whereas that of rectal varices is 3%-7% in cirrhotic patients on endoscopy (35, 36). Idiopathic ileocolonic varices with no under-
lying causes of portal hypertension are extremely uncom-
mon. The most common cause of portal hypertension is
liver cirrhosis. Other causes of portal hypertension, such as
pre-hepatic abnormalities (e.g. portal or splenic vein throm-
bosis, malignancy, pancreatitis, postsurgical state), post-
hepatic abnormalities (e.g. Budd-Chiari syndrome, congest-
tive heart disease), or intrahepatic non-cirrhotic abnormali-
ties (37, 38), can induce colonic varices. Varices develop as
venous collaterals when the portal pressure remains higher
than the hepatic venous pressure, a condition called portal
hypertension. Reopening of collapsed embryonic channels
and reversal of the flow of existing adult veins is caused by
portal hypertension, which can lead to the development of
varices (39). The diagnosis of idiopathic colonic varices is
made by detecting varices and excluding portal hypertension
as a cause. In our case, laboratory testing revealed a history
of HBV infection, but no other data suggested viral hepati-
tis, autoimmune disease, endocrine disease, or any other
liver disease. M2BPGi, an accurate, reliable, and reproduc-
ible marker for the assessment of liver fibrosis (40), was
within normal levels (Table 1). Shear wave measurement
(Hitachi, Ltd.), an ultrasonographic modality for assessing
liver cirrhosis (33), showed no evidence of liver cirrhosis.
There was no portal or splenic vein thrombosis, according to
contrast-enhanced CT and angiography of the abdomen and
pelvis.

To our knowledge, there have been 29 reports on 33 pa-
tients with idiopathic colonic varices published since 1986
(Table 2) (2-30). On reviewing these cases, the average age
was 44.8 years old, and 17 (77.3%) of the 22 patients were
men (4, 6-10, 12, 13, 18, 19, 21, 23, 24, 27, 29). A family
tendency was evaluated in 16 cases, and 6 cases had familial
tendency (9, 20, 25). Occurrence site of varices was men-
tioned in 23 cases, and 6 cases (26.1%) were ileocolonic
varices (6, 16, 21, 22, 27, 28) and 12 (52.2%) were total
colic varices (2, 3, 5, 17, 19, 20, 23, 24, 29, 30). The en-
tire colon was involved in almost 80% of cases. A major
complement was hematochezia in most cases (3, 4, 6-8, 10,
12, 13, 16-24, 28, 29), and some cases presented with diar-
rhrea (20, 24, 26). Among the 19 cases of hematochezia, 7
(36.8%) required an operation as treatment (3, 7, 9, 12, 18,
21, 22). Endoscopic treatment of active bleeding from idi-
opathic ileocolonic varices has not been reported yet. There
were only two asymptomatic cases (27, 30), and both were
detected by screening colonoscopy.

Screening colonoscopy is an effective method of reducing
the risk of death due to colon cancer (41), and its spread
might be the reason for these recent asymptomatic cases. We
detected colonic varices by conducting screening colono-
scopy and established the diagnosis by EUS, digital subtrac-
tion angiography, and CT angiography. CT angiography
might be the most reliable method for making a diagnosis,
and digital subtraction angiography might be the only
method for determining the blood flow in the varices. As we

Figure 3. A, B: Digital subtraction angiography of the superior mesenteric artery showed delayed
venous pooling in the ascending colon. C, D: Computed tomography during superior mesenteric an-
giography showed the development of colonic varices in the ascending colon.
Table 2.

<table>
<thead>
<tr>
<th>No.</th>
<th>Age</th>
<th>Sex</th>
<th>Location</th>
<th>Family tendency</th>
<th>Symptom</th>
<th>Treatment</th>
<th>Prognosis</th>
<th>Ref.</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>71</td>
<td>F</td>
<td>colonic</td>
<td>ND</td>
<td>asymptomatic</td>
<td>none</td>
<td>stable</td>
<td>[30]</td>
</tr>
<tr>
<td>2</td>
<td>76</td>
<td>M</td>
<td>colonic</td>
<td>ND</td>
<td>hematochezia</td>
<td>preserved</td>
<td>stable</td>
<td>[29]</td>
</tr>
<tr>
<td>3</td>
<td>37</td>
<td>M</td>
<td>ileocolonic</td>
<td>ND</td>
<td>hematochezia</td>
<td>preserved (ARB)</td>
<td>disappeared</td>
<td>[28]</td>
</tr>
<tr>
<td>4</td>
<td>54</td>
<td>M</td>
<td>ileocolonic</td>
<td>none</td>
<td>asymptomatic</td>
<td>none</td>
<td>stable</td>
<td>[27]</td>
</tr>
<tr>
<td>5</td>
<td>38</td>
<td>F</td>
<td>colonic</td>
<td>none</td>
<td>diarrhea</td>
<td>none</td>
<td>stable</td>
<td>[26]</td>
</tr>
<tr>
<td>6</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>(+)</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
</tr>
<tr>
<td>7</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>(+)</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
</tr>
<tr>
<td>8</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>(+)</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
</tr>
<tr>
<td>9</td>
<td>30</td>
<td>M</td>
<td>colonic</td>
<td>none</td>
<td>hematochezia, diarrhea</td>
<td>preserved</td>
<td>stable</td>
<td>[24]</td>
</tr>
<tr>
<td>10</td>
<td>44</td>
<td>M</td>
<td>colonic</td>
<td>none</td>
<td>hematochezia</td>
<td>preserved</td>
<td>stable</td>
<td>[23]</td>
</tr>
<tr>
<td>11</td>
<td>20</td>
<td>F</td>
<td>ileocolonic</td>
<td>ND</td>
<td>hematochezia</td>
<td>operation</td>
<td>stable</td>
<td>[22]</td>
</tr>
<tr>
<td>12</td>
<td>21</td>
<td>M</td>
<td>ileocolonic</td>
<td>none</td>
<td>hematochezia</td>
<td>operation</td>
<td>stable</td>
<td>[21]</td>
</tr>
<tr>
<td>13</td>
<td>61</td>
<td>F</td>
<td>colonic</td>
<td>(+)</td>
<td>hematochezia, diarrhea</td>
<td>preserved</td>
<td>stable</td>
<td>[20]</td>
</tr>
<tr>
<td>14</td>
<td>43</td>
<td>M</td>
<td>colonic</td>
<td>none</td>
<td>hematochezia</td>
<td>preserved (propranolol)</td>
<td>decreased</td>
<td>[19]</td>
</tr>
<tr>
<td>15</td>
<td>24</td>
<td>M</td>
<td>rectum-descending</td>
<td>none</td>
<td>hematochezia</td>
<td>operation</td>
<td>stable</td>
<td>[18]</td>
</tr>
<tr>
<td>16</td>
<td>74</td>
<td>F</td>
<td>colonic</td>
<td>ND</td>
<td>hematochezia</td>
<td>preserved</td>
<td>stable</td>
<td>[17]</td>
</tr>
<tr>
<td>17</td>
<td>64</td>
<td>ND</td>
<td>ileocolonic</td>
<td>ND</td>
<td>hematochezia</td>
<td>operation</td>
<td>stable</td>
<td>[16]</td>
</tr>
<tr>
<td>18</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
</tr>
<tr>
<td>19</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
</tr>
<tr>
<td>20</td>
<td>30</td>
<td>M</td>
<td>hepatic flexure</td>
<td>ND</td>
<td>hematochezia</td>
<td>preserved</td>
<td>ND</td>
<td>ND</td>
</tr>
<tr>
<td>21</td>
<td>37</td>
<td>M</td>
<td>sigmoid</td>
<td>ND</td>
<td>hematochezia</td>
<td>operation</td>
<td>stable</td>
<td>[12]</td>
</tr>
<tr>
<td>22</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
</tr>
<tr>
<td>23</td>
<td>27</td>
<td>M</td>
<td>ND</td>
<td>ND</td>
<td>hematochezia</td>
<td>preserved</td>
<td>stable</td>
<td>[10]</td>
</tr>
<tr>
<td>24</td>
<td>56</td>
<td>M</td>
<td>ND</td>
<td>(+)</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
</tr>
<tr>
<td>25</td>
<td>28</td>
<td>M</td>
<td>ND</td>
<td>(+)</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
</tr>
<tr>
<td>26</td>
<td>81</td>
<td>M</td>
<td>ND</td>
<td>ND</td>
<td>hematochezia</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
</tr>
<tr>
<td>27</td>
<td>32</td>
<td>M</td>
<td>rectum-sigmoid</td>
<td>none</td>
<td>hematochezia</td>
<td>operation</td>
<td>stable</td>
<td>[7]</td>
</tr>
<tr>
<td>28</td>
<td>25</td>
<td>M</td>
<td>ileocolonic</td>
<td>ND</td>
<td>hematochezia</td>
<td>preserved</td>
<td>stable</td>
<td>[6]</td>
</tr>
<tr>
<td>29</td>
<td>ND</td>
<td>ND</td>
<td>colonic</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
</tr>
<tr>
<td>30</td>
<td>58</td>
<td>M</td>
<td>rectum-sigmoid, caecum</td>
<td>none</td>
<td>hematochezia</td>
<td>preserved</td>
<td>stable</td>
<td>[4]</td>
</tr>
<tr>
<td>31</td>
<td>ND</td>
<td>ND</td>
<td>colonic</td>
<td>none</td>
<td>hematochezia</td>
<td>operation</td>
<td>stable</td>
<td>[3]</td>
</tr>
<tr>
<td>32</td>
<td>ND</td>
<td>ND</td>
<td>colonic</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
</tr>
<tr>
<td>33</td>
<td>ND</td>
<td>ND</td>
<td>colonic</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
<td>ND</td>
</tr>
</tbody>
</table>

ND: Not detected; ARB: Angiotensin receptor blocker

showed in our case, EUS or transrectal Doppler sonography have also been reported to be useful for making a diagnosis (18, 19). Capsule endoscopy may also be a reliable tool for investigating the location of varices (28). A barium enema can reportedly depict colonic varices as tumor-like serpiginous lesions (20). However, it is difficult to exclude polyps, cancer, or other colorectal lesions using a barium enema.

Colonic varices may be misinterpreted as polyps, cancer, air bubbles, or fecal material (2, 20). There are cases of a biopsy or polypectomy resulting in a significant bleeding (20, 31). In asymptomatic ileocolonic varices, as in our case, a careful examination using a variety of techniques is required in order to avoid significant complications of endoscopy.

The treatment of idiopathic colonic varices remains unclear. While most cases with hematochezia show no active bleeding during colonoscopy, others require emergency operation (16) or extensive colectomy (3, 7, 21, 22). There have been no reports of endoscopic treatment for active bleeding caused by idiopathic ileocolonic varices. The short-term prognosis seems good, but the long-term prognosis is unclear. A family history of death from gastrointestinal hemorrhaging was reported in one case (20). Very few attempts have been made to treat colorectal lesions (e.g. polyps, carcinoma) accompanied by colonic varices. Depending on the bleeding status, an operation seems to be the safest method, but it is highly invasive. As mentioned above, there are cases of significant bleeding arising from a biopsy or a polypectomy (20, 31). Such cases show that endoscopic treatment can cause significant complications related to colorectal varices. Generally, active lower gastrointestinal bleeding is first treated by endoscopic hemostasis therapy, including mechanical, thermal, and injection therapy or a combination thereof (42). The majority of bleeding events after a colonoscopy are managed successfully by endoscopy, whereas very few cases require operation (43). When colorectal lesions coexist with colonic varices and require treat-
ment, an endoscopic approach should be considered from the point of invasiveness. In our case, an interventional approach to stop the bleeding was considered difficult because the blood flow of the colorectal varices was from the mesenteric artery to the mesenteric vein. Therefore, we planned endoscopic clipping or endoscopic injection sclerotherapy to manage the bleeding, and informed consent was obtained, including consent regarding the operative method. At the time of EMR, we shortened the electricity time as much as possible. We proceeded with clip closure in order to prevent post-EMR bleeding, and no immediate or delayed bleeding was noted.

Cold snare polypectomy has a lower risk of post-polypectomy bleeding than does hot snare polypectomy (44). This shows that a longer time of exposure to electricity might increase the risk of post-polypectomy bleeding. Prophylactic clipping is reportedly unnecessary for preventing post-polypectomy bleeding with polyps smaller than 2 cm in diameter (45). The polyp in our case was smaller than 2 cm, but we proceeded with clip closure to prevent post-EMR bleeding, as this case had coexistent colonic varices.

To our knowledge, this is the first report of idiopathic ileocolonic varices with a colon polyp treated successfully by endoscopy. The diagnosis of idiopathic colonic varices requires a careful examination using a variety of techniques. During management, the development of a treatment strategy and obtaining informed consent are required.

The authors state that they have no Conflict of Interest (COI).

Acknowledgement
This study did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

References