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Familial Idiopathic Pan-Colonic Varices Found Incidentally in a Young Patient with a Hepatic Flexure Tumor

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Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
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Conflict of interest: None declared

Patient: Male, 24-year-old
Final Diagnosis: Colon cancer
Symptoms: —
Medication: —
Clinical Procedure: Laparoscopic right hemi-colectomy
Specialty: Surgery

Objective: Rare co-existance of disease or pathology

Background: Colonic varices are rare entity that often results from portal vein hypertension and hepatic cirrhosis. In the absence of underlying pathology, they are termed "idiopathic colonic varices". They are usually an incidental finding; however, they can present with varying degrees of lower gastrointestinal bleeding. There is only one reported case in the literature of colonic varices with a concomitant colonic tumor; our patient is the second one with such a presentation. We report a case of this rare combination with the outcomes of the elected surgical management and review the literature.

Case Report: A 24-year-old male was referred to our hospital with a 1-month history of colicky abdominal pain. His family history is remarkable of 2 relatives with colonic varices. A computed tomography scan of the abdomen and pelvis showed a hepatic flexure colonic mass. Colonoscopy revealed pancolonic varices. Biopsy from the lesion revealed adenocarcinoma. Options were discussed with the patient to undergo only a right hemicolectomy for his cancer or a total colectomy to include the colonic segment involved with varices, and he elected the first option, with no complications upon 1 year follow up.

Conclusions: Idiopathic pan-colonic varices are rare pathology. Their presence with colonic tumor presents a dilemma as to whether a subtotal/total colectomy is needed on the premise that a limited resection may carry the risk of subsequent bleeding. In the literature, the only similar case to ours had brisk postoperative bleeding, while ours did not experience such a complication.

MeSH Keywords: Colon, Ascending • Colonic Neoplasms • Colorectal Neoplasms

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Background

Colonic varices are very rare entity, and in the majority of cases result from portal vein hypertension and hepatic cirrhosis [1]. In the absence of underlying pathology, these lesions are termed "idiopathic colonic varices", and if more than 1 family member is affected, they are referred to as "familial idiopathic varices" [1,2]. Fewer than 50 cases of idiopathic colonic varices have been reported in the literature [2–4]. Varices are usually an incidental finding during investigations for other reasons; however, they can present acutely with lower gastrointestinal bleeding or chronically with iron deficiency anemia [5–7]. There is only 1 reported case in the literature of colonic varices with a concomitant diagnosis of colon cancer; our patient is the second one with such a presentation. The authors report a case of this rare combination with the outcomes of the elected surgical management and review the literature.

Case Report

A 24-year-old male was referred to our hospital with a 1-month history of colicky abdominal pain, dizziness, palpitations, exertional dyspnea, and long-standing constipation. There was no history of nausea or vomiting, and there had been subjective weight loss. He denied rectal bleeding. Family history was positive for breast cancer and G6PD deficiency. Two of his second-degree relatives were known to have colonic varices and were on regular follow-up with our gastroenterology unit. On examination, he was comfortable at rest, but he looked pale. Laboratory tests including inflammatory markers were all within normal except for iron deficiency anemia. Chest and abdominal x-rays were unremarkable. An ultrasound scan of the abdomen showed a normal appearance of the liver, spleen, and pancreas. No signs of thrombosis of the portal axis on Doppler examination. A computed tomography (CT) scan of the abdomen and pelvis was obtained and showed a hepatic flexure colonic mass that measures 2.5×7 cm, with adjacent lymph node of 1.8×1.3 cm in size (Figure 1). Also, a low attenuation lesion of 4 mm was seen in the right posterior hepatic segment that likely represented a hemangioma. Colonoscopy revealed a large polypoid tumor at the level of the hepatic flexure (Figure 2). Exaggerated vasculature of the colon (varices) was noted along its entire walls (Figure 3A, 3B). Biopsies were taken from the mass, and histopathology examination revealed a well-differentiated adenocarcinoma. The case was discussed at our multidisciplinary colorectal tumor board and decision was made for surgery. Options were discussed with the patient to undergo only a right hemicolectomy for his cancer or a total colectomy to include the colonic segment involved with varices. The former option was agreed upon, and the patient underwent a laparoscopic right hemicolectomy with side-to-side ileo-transverse anastomosis. Intraoperatively, an ultrasound

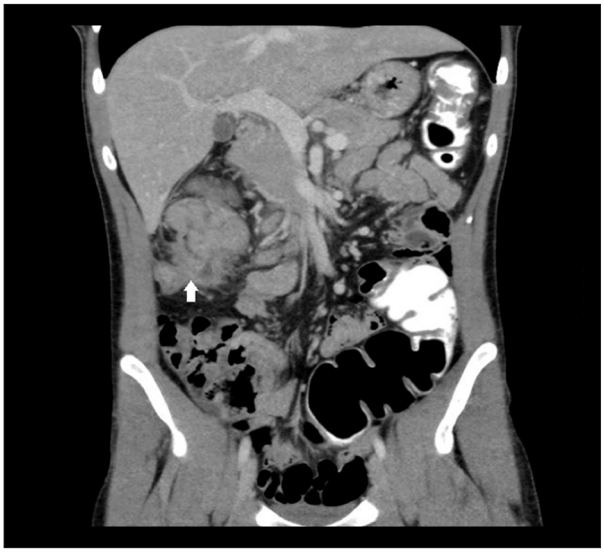

Figure 1. Contrast enhanced computed tomography of the abdomen and pelvis, coronal section, shows hepatic flexure colonic mass with surrounding fat stranding (arrow).

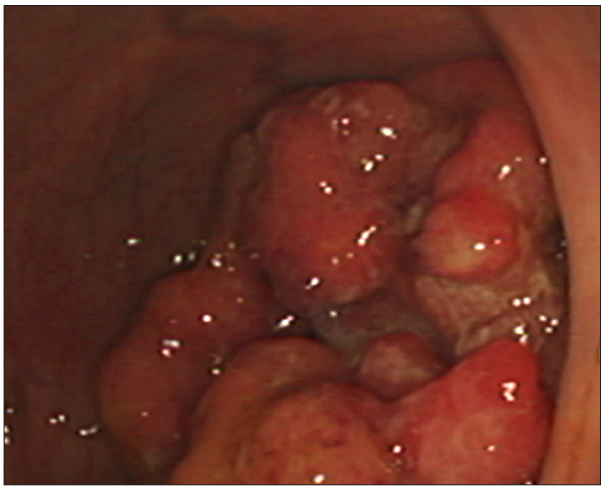

Figure 2. Colonoscopy image shows the right hepatic flexure mass.

scan of the liver was performed to assess the lesion shown on CT. Findings were again consistent with hemangioma and no treatment was carried out. The patient made an uneventful recovery and was discharged on the 6th postoperative day. Final histopathology examination revealed a well to moderately differentiated adenocarcinoma arising from a tubulo-villous adenoma, pT3 pN2a M0, with free resection margins. Molecular tests including BRAF, NRAS, and MSI were negative. The patient received adjuvant chemotherapy and at 1-year follow-up was free of disease as evidenced by follow-up colonoscopy; the colonic varices were noted again and unchanged. Our patient did not experience any episode of lower gastrointestinal tract bleeding after his surgery.

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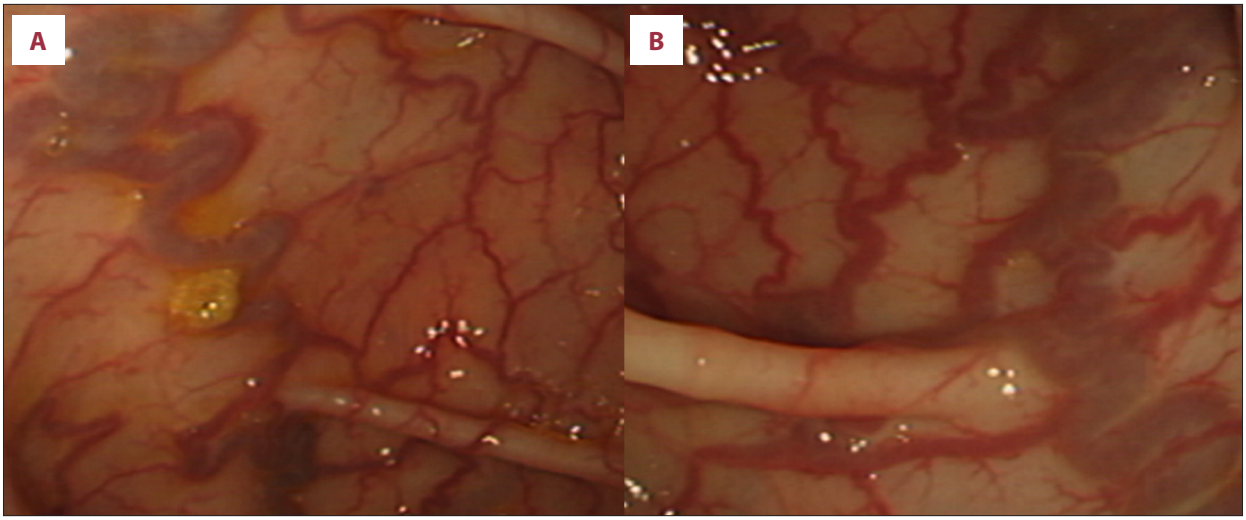

Figure 3. (**A, B**) Colonoscopy images show exaggerated vasculature of the colon (varices).

Discussion

Colonic varices a rare condition; their incidence is still unknown, but it is estimated to be 0.07% as reported by Feldman et al. in an autopsy-based study of 2912 patients presenting with lower gastrointestinal bleeding (LGIB) [3]. It usually develops on the background of portal hypertension and hepatic cirrhosis [1–4]. However, other conditions have been associated with colonic varices including chronic pancreatitis, congestive heart failure, distant malignancies, postoperative mesenteric adhesions, and mesenteric or splenic vein obstruction. Idiopathic colonic varices (ICVs) are even rarer entity; and as of 2014, a literature review of the previously reported cases, conducted by Spechier et al., identified only 39 such cases, 26 of which were pan-colonic, and 15 only had familial component [2–4]. We identified 10 more cases, apart from our case, since the Spechier review, shown in Table 1 [2,3,6–41]. The etiology of these lesions is unknown, but congenital anomalies of the porto-systemic anastomosis or vascular wall abnormalities have been considered. Also, the mode of inheritance in familial cases is not yet understood, but it is thought to be autosomal recessive [1,5,6]. It is very important to rule out underlying causes before labeling the diagnosis as idiopathic [1,6].

Based on the analysis of the existing cases in the literature, Spechier et al. found that the median age at diagnosis of idiopathic colonic varices was 41 years old, ranging from 14 to 81 years old; male to female ratio was 25: 15, and as compared to non-idiopathic cases, which tend to have segmental distribution, idiopathic varices are often pan colonic, and in those cases where segmental distribution was seen, the right and the left sides of the colon were equally affected [3,7,8]. It is also noted that those particular patients have a familial tendency [2,5,9].

Most of the reported cases in the literature presented with LGIB ranging from recurrent minimal per rectal bleeding with anemia to massive and life threatening-bleeding [2,9–11]. Bleeding in these cases may be precipitated by trauma caused by the passage of hard stool or by mucosal ulceration due to ischemia induced by varices enlargement [9,12].

Gudjonsson et al. noted that colonic varices were generally diagnosed only during exploratory laparotomy or at autopsy before the 1970s owing to the fact that most of the patients presented with recurrent massive bleeding before the correct diagnosis was made, which can be explained by their rarity. However, since their invention, endoscopy and angiography emerged as the diagnostic modalities of choice [9]. Colonoscopy allows direct visualization of the varices; they appear as serpiginous, bluish structures with superficial venules. However, they can be easily missed during insufflation or in a hypotensive state when their volume is reduced. Biopsy is not advised as this may result in heavy bleeding [5,12]. Colon capsule endoscopy was also used in some reports, especially in cases where the varices disappeared during colonoscopy. Some authors also advocated its use in evaluating the entire small bowel shall terminal ileum involvement be noted during colonoscopy [8,13]. On the other hand, some authors cited mesenteric angiography as a better and more accurate diagnostic tool and the varices are best seen during the venous phase [4,5,8,14]. It allows visualization and mapping of the abnormal vessels, outlines their distribution, and identifies the source of active bleeding with a sensitivity of 95%. It also has a therapeutic utility as it can be used for injection of pharmacologic or embolic materials to control active bleeding [4]. Care must be taken also when interpreting the radiological and endoscopy findings as these lesions can be misconceived sometimes as tumor; this was seen in 3 reported cases, 2 of which had barium enema that showed multiple filling defects, initially

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Table 1. Summary of the published cases of idiopathic colonic varices, clinical presentation, management and status upon follow-up.

Number	Author (year)	Patient's age (years)	Familial component	Extent of the varices	Presentation	Other pathology	Management	Follow-up
1	Solis-Herruzo (1977) [17]	18	Yes	Pan-colonic	Recurrent LGIB	None	Exploratory laparotomy with dissection of the inferior mesenteric vein till the splenic vein; No bowel resection done	Unreported
2	Solis-Herruzo (1977) [17]	26	Yes	Pan-colonic	Recurrent LGIB	None	Conservative	Unreported
3	Weingart (1982) [18]	70	None	Pan-colonic	Recurrent LGIB; profuse bleeding post biopsy of the varix as it was thought to be polyp	None	Conservative	Unreported
5	Hawkey (1985) [19]	71	Yes	Pan-colonic; starting from the jejunum	Recurrent LGIB	None	Surgical resection done; no further information available	Unreported
6	Hawkey (1985) [19]	66	Yes	Pan colonic	Recurrent massive LGIB	None	Conservative	The patient passed away after 7 years due to LGIB
7	Beermann (1988) [20]	65	Yes	Pan-colonic	Rectal mass	Rectal polyp	Severe LGIB post polypectomy; managed conservatively	Unreported
8	Beermann (1988) [20]	65	Yes	Pan-colonic	Recurrent LGIB	Colon cancer; the varices were not present at time the patient was investigated and managed for colon cancer	Conservative	Unreported
9	Isbister (1989) [21]	14	None	Hepatic flexure and sigmoid colon	LGIB	Cecal arteriovenous malformation	Right hemi-colectomy	No evidence of bleeding post op
10	Nikolopoulos (1990) [14]	22	None	Pan-colonic	Recurrent LGIB	None	Subtotal colectomy with ileorectal anastomosis	No evidence of bleeding post op
11	Iredale (1992) [6]	58	None	Pan colonic	Recurrent LGIB	None	Conservative	Unreported
12	Atin (1992) [7]	23	None	Pan-colonic	Recurrent massive LGIB	Colonic hamarto-matous polyps	Segmental resection; right hemi-colectomy and sigmoidectomy	No evidence of bleeding at 2 years post op

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Table 1 continued. Summary of the published cases of idiopathic colonic varices, clinical presentation, management and status upon follow-up.

Number	Author (year)	Patient's age (years)	Familial component	Extent of the varices	Presentation	Other pathology	Management	Follow-up
13	Morini (1993) [22]	67	Yes	Pan-colonic; entire small bowel	Recurrent LGIB	Adenomatous polyp	Conservative	Unreported
14	Morini (1993) [22]	65	Yes	Pan-colonic;	Massive LGIB	Adenomatous polyp; Crohn's disease	Conservative	Unreported
15	Villarreal (1995) [23]	41	None	Descending colon	Recurrent massive LGIB	None	Left hemi-colectomy	No evidence of bleeding at 6 months post op
16	Shrestha (1995) [24]	33	None	Pan-colonic	Massive LGIB	None	Conservative	Further episodes of bleeding at 2 and 3 months managed conservatively
17	Detry (1996) [25]	32	None	Sigmoid and rectum	Recurrent LGIB	None	Anterior resection with coloanal anastomosis	No evidence of bleeding post op
18	Loffeld (1996) [26]	81	None	Pan-colonic	History of LGIB	None	Observation	Unreported
19	Bernardini (1998) [27]	61	Yes	Pan-colonic; terminal ileum	Incidental	None	Observation	Unreported
20	Bernardini (1998) [27]		Yes	Pan-colonic; terminal ileum	Incidental	None	Observation	Unreported
21	Bernardini (1998) [27]	43	Yes	Pan-colonic; terminal ileum	Recurrent LGIB	None	No data available	Unreported
22	Bernardini (1998) [27]	17	Yes	Pan-colonic; terminal ileum	Recurrent LGIB	None	Conservative	Unreported
23	Schmidt (1998) [28]	34	None	Pan colonic	Recurrent massive LGIB	None	Conservative	Further episodes of massive LGIB at 8 months and 3 years after the first encounter

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Table 1 continued. Summary of the published cases of idiopathic colonic varices, clinical presentation, management and status upon follow-up.

Number	Author (year)	Patient's age (years)	Familial component	Extent of the varices	Presentation	Other pathology	Management	Follow-up
24	Kori (2000) [10]	20	Yes	Splenic flexure till the rectum	Abdominal pain, bloody diarrhea	None	Conservative	Recurrent minor bleeding at 12 months follow up; 6 months later he was asymptomatic
25	Kori (2000) [10]	15	Yes	Hepatic flexure till sigmoid colon	One episode of massive LGIB	None	Conservative	No evidence of bleeding at 3 months after the first encounter
26	Place (2000) [29]	27	None	Pan-colonic	LGIB	None	Conservative	No evidence of bleeding at 48 months after the first encounter
27	Lopez (2000) [30]		Yes	Pan-colonic	Recurrent LGIB	None	Conservative	Unreported
28	Lopez (2000) [30]		Yes	Pan-colonic	Recurrent LGIB	None	Conservative	Unreported
29	Gossum (2000) [31]		None		Recurrent LGIB			
30	Igwe (2002) [16]	37	None	Pan-colonic	Intermittent LGIB	Sigmoid cancer	Sigmoid colectomy	Two episodes of brisk LGIB at 3 months and 7 years post op; no evidence of bleeding over 18-year follow up
31	Mehta (2004) [32]	30	None	Hepatic flexure	LGIB	None	Conservative	Unreported
32	Vuilemin (2004) [33]	39	None	Pan-colonic	Recurrent LGIB	None	Conservative	No evidence of recurrence 2 years after the second episode
33	Keren (2005) [34]	17	None	Pan colonic	Recurrent massive LGIB	None	Subtotal colectomy with ileo-rectal anastomosis	No evidence of bleeding at 4 years post op
34	Han (2006) [35]	24	None	Descending colon till the rectum	Recurrent LGIB	None	Anterior resection with coloanal anastomosis	No evidence of bleeding post op
35	Lopes (2006) [36]	64	None	One large varix extending from the cecum till the rectum	Recurrent LGIB; acute episode of massive LGIB	None	Subtotal colectomy with ileo-rectal anastomosis	No evidence of bleeding post op

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Table 1 continued. Summary of the published cases of idiopathic colonic varices, clinical presentation, management and status upon follow-up.

Number	Author (year)	Patient's age (years)	Familial component	Extent of the varices	Presentation	Other pathology	Management	Follow-up
36	Simvoulakis (2006) [37]	74	None	Pan-colonic	Recurrent LGIB	None	Conservative	No evidence of bleeding 10 months after first encounter
37	Zaman (2008) [15]	61	Yes; Daughter of patient #6	Pan-colonic	History of recurrent LGIB from the age of 17 till 23; investigated for intermittent diarrhea.	Tubular adenoma	The patient had significant bleeding post polypectomy, managed endoscopically; Otherwise, she only needed observation	No evidence of bleeding upon follow up
38	Krishna (2010) [8]	21	None	Pan-colonic; terminal ileum	Recurrent LGIB	None	Subtotal colectomy with ileo-rectal anastomosis	Unreported
39	Grasso (2012) [38]	33	None	Terminal ileum; ascending colon; descending colon till rectum	Recurrent LGIB	None	Subtotal colectomy with ileo-colic anastomosis	Unreported
40	Gentilli (2012) [39]	21	None	Pan-colonic	Massive LGIB	None	Subtotal colectomy with ileo-rectal anastomosis	Unreported
41	Dina (2014) [3]	38	None	Pan-colonic	Incidental finding on colonoscopy while investigating the patient for indigestion	None	Observation	Unreported
42	Akkucuk (2014) [12]	44	None	Pan colonic	Recurrent LGIB	None	Conservative	No evidence of bleeding after 6 months since the admission
43	Speicher (2014) [3]	30	None	Pan-colonic; terminal ileum	Abdominal pain and diarrhea	None	Observation	Unreported
44	Kawasaki (2016) [13]	37	None	Pan-colonic; terminal ileum	Recurrent LGIB	None	Managed with Losartan for 3 months	The varices disappeared after trial of angiotensin II receptor blocker

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Table 1 continued. Summary of the published cases of idiopathic colonic varices, clinical presentation, management and status upon follow-up.

Number	Author (year)	Patient's age (years)	Familial component	Extent of the varices	Presentation	Other pathology	Management	Follow-up
45	Zizzo (2016) [40]	27	None	Pan-colonic	Recurrent massive LGIB	Arteriovenous malformation of the mesocolon; presumed to be the primary cause	Right hemicolectomy	Unreported
46	Peixoto (2016) [41]	54	None	Pan-colonic	Incidental	None	Observation	Unreported
47	Chowdhury (2016) [9]	65	None	Multiple colonic varices, 30 cm distal to the anus	Recurrent, mild LGIB	None	Conservative	No evidence of bleeding after 1 month since the first encounter
48	Kahl (2017) [11]	71	None	Ascending colon till the rectum	Recurrent LGIB	Splenic artery aneurysm	Conservative	Unreported
49	Sunkara (2018) [2]	71	None	Pan-colonic	Incidental finding on screening colonoscopy; had a few episodes of mild LGIB	Adenomatous polyps	Conservative management	Unreported
50	Present case (2019)	24	Yes	Pan-colonic	Incidental finding while being investigated for colonic lesion seen previously on CT scan	Right-sided colon cancer	Right hemicolectomy with primary anastomosis	No evidence of bleeding at 1-year post op

The cases summarized in the table are of patients who were found to have colon varices, without hepatic dysfunction or portal hypertension, thus certain cases were not included. LGIB – lower gastrointestinal bleeding; CT – computed tomography.

interpreted as multiple polyposes, but later confirmed by endoscopy to be varices [7], and in another patient the varices looked like an ulcerated, necrotic mass on colonoscopy [15].

Due to the paucity of the cases, and for being an unexpected cause of LGIB, definitive management recommendations are lacking for both, idiopathic and non-idiopathic colonic varices. However, initial management follows the same approach as with other cases of LGIB. Correction of the underlying cause in the non-idiopathic case is also needed [1–12]. As far as ICVs are concerned, many authors opted to reserve surgical management to those patients presenting with hemodynamically significant or recurrent and intractable bleeding, while conservative therapy, with observation, iron supplements, laxatives or periodic blood transfusion, is a preferred option in the less severe cases [4,6,9–14]. Interestingly, pharmacological therapy has been used with success. Kawasaki et al. managed a middle-aged patient with idiopathic pan-colonic varices with losartan (angiotensin II receptor blocker) for 3 months; the patient was then re-evaluated using capsule endoscopy and disappearance of the varices was noted, but no further evidence about the success rate of this modality exists [13].

As to which surgical option is valid to patients with colonic varices, Gudjonsson et al. (1986), reviewed a group of patients with bleeding colonic varices, idiopathic and non-idiopathic, who underwent surgical management, and found that 9 out of 16 patients who were managed with segmental, subtotal,

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or total colectomy passed away, whereas for the 9 patients who had porto-systemic shunts, all but 1 survived. The high mortality rate in the first group was seen more or less in those patients with the non-idiopathic type of varices and could be explained in part the complex surgery in light of the presence of portal hypertension. Thus, they concluded that in the settings of massive LGIB from colonic varices in patients with portal hypertension, portal decompression is the option of choice, whereas colonic resection is reserved for patients not known to have portal hypertension [9].

There is no recent comparative study exploring the outcomes of medical versus surgical treatment of the already reported cases, but in 1992, Iredale et al. reviewed 9 cases of ICV, 6 of which presented with recurrent LGIB and managed conservatively with no reported mortality; on the other hand, 3 patients had surgical management, 1 patient needed subtotal colectomy, 1 patient underwent segmental resection, and the third patient had exploratory laparotomy with no further details mentioned. The 2 aforementioned patients did not have further episodes of bleeding upon follow-up. It is also worth mentioning that Iredale et al. observed a more benign presentation in patients older than 50 years of age, necessitating conservative management as compared to those patients who presented in their 20s who often had significant hemorrhage, thus concluding that this particular group of patients benefit more from surgical management [6]. Other reported modalities in the management include endoscopic ligation and sclerotherapy, with success reported in the literature for the actively bleeding varices [1,2,9]. We reviewed some of the available cases of ICV and compared the outcomes between the medically and surgically treated patients, and the clinical characteristics of the affected patients in Table 1. The available data reveals that surgical resection in patients with pancolonic varices doesn't carry further risk of bleeding as theoretically believed; however, it should be kept in mind that the time period during which the patients were followed was not specified in most of the reports, thus some data about the actual risk is still missing.

Our patient not only presented with a rare case of familial idiopathic pancolonic varices, but he presented with a colon carcinoma at a young age with no known family history of such a tumor. Our literature review revealed only 1 similar case (published in 2002 by Igwe and Patel [16]) of a 37-year-old male who was found to have a concomitant presence of colonic varices and sigmoid carcinoma on a background of intermittent LGIB, managed with sigmoid colectomy. An 18 year-follow up revealed 2 episodes of brisk per rectal bleeding, 3 months and 7 years after the surgery, both necessitated blood transfusion and conservative management. The source of bleeding could not be ascertained during the 2 episodes: neither by colonoscopy nor by mesenteric angiography [16].

As mentioned earlier, our patient elected to have a right hemicolectomy; and since his operation, he has not had any episodes of per rectal bleeding.

Conclusions

In conclusion, an ICV is a rare pathology that can be detected incidentally or present with LGIB ranging from mild and recurrent episodes to life-threatening hemorrhage necessitating surgical resection of the affected segment. The presence of a tumor in a colon that is carpeted with non-bleeding varices presents a dilemma as to whether a subtotal/total colectomy is needed on the premise that a limited resection may carry the risk of subsequent bleeding or not. Out of the 15 patients, including our patient, that were treated surgically in the literature, only 1 patient had 2 episodes of brisk bleeding postoperatively; however, this does not allow us to conclude that surgery eliminates the risk of subsequent bleeding in such patients due to the limitation of the data available.

Conflict of interest

None.

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