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特发性肠系膜静脉肌内膜增生症临床病理特征

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摘　要

目的：探讨特发性肠系膜静脉肌内膜增生症(idiopathic myointimal hyperplasia of mesenteric venules, IMHMV)的临床病理特征与鉴别诊断，提高对该罕见病的认识。方法 收集1例50岁男性结肠IMHMV患者的临床资料、采用HE染色及弹力纤维染色等进行病理学观察，并复习相关文献。**结果** 患者临床表现为反复腹痛、血便1年余，临床疑诊溃疡性结肠炎。肠镜显示乙状结肠黏膜充血、糜烂伴溃疡形成，因症状持续并出现肠狭窄，行腹腔镜下左半结肠切除术。巨检见肠壁大片不规则溃疡，伴粘膜颗粒样增生及肠腔狭窄。镜检示肠系膜内静脉管壁呈偏心性增厚，致管腔狭窄，内膜下平滑肌细胞显著增生；弹力纤维染色证实病变血管为静脉，伴行动脉正常；周围背景粘膜呈慢性缺血损伤模式，此外，灶性区域粘膜固有层见毛细血管壁增厚(“动脉化”现象)。术后患者未予药物治疗，随访2年无复发。**结论** IMHMV是一种罕见的肠道缺血性疾病，确诊依赖于手术标本的病理检查，其特征为静脉肌内膜增生而无其他血管病变。病理医师熟悉其关键形态学特征，给出提示性诊断，避免误诊及指导临床治疗至关重要。

关键词

特发性肠系膜静脉肌内膜增生症，肠系膜血管病，缺血性结肠炎，炎症性肠病

Clinicopathological Features of Idiopathic Myointimal Hyperplasia of Mesenteric Venules

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Abstract

Objective: This study aimed to examine the clinicopathological features and differential diagnosis of idiopathic myointimal hyperplasia of the mesenteric venules (IMHMV), with a view to improving recognition of this rare condition. Methods: Clinical data from a 50-year-old male patient with colonic IMHMV were collected. Histopathological examination was conducted using hematoxylin-eosin (HE) staining and elastic fiber staining, and relevant literature was reviewed. Results: The patient presented with recurrent abdominal pain and bloody stools for over one year and was clinically suspected of having ulcerative colitis. Colonoscopy revealed mucosal congestion, erosion, and ulcer formation in the sigmoid colon. Due to persistent symptoms and the development of intestinal stenosis, laparoscopic left hemicolectomy was performed. Gross examination showed extensive irregular ulcers in the intestinal wall, accompanied by mucosal granular hyperplasia and luminal stenosis. Microscopic examination revealed eccentric thickening of the mesenteric venous walls, leading to luminal narrowing, with significant hyperplasia of subendothelial smooth muscle cells. Elastic fiber staining confirmed the affected vessels were veins, while accompanying arteries appeared normal. The surrounding background mucosa exhibited a pattern of chronic ischemic injury. Additionally, focal areas showed thickening of the capillary walls in the lamina propria ("arteriolization"). No postoperative medication was administered, and the patient remained recurrence-free during a 2-year follow-up. Conclusion: IMHMV is a rare ischemic bowel disease, and its diagnosis relies on the pathological examination of surgical specimens. It is characterized by myointimal hyperplasia of the venules in the absence of other vascular pathologies. Familiarity with its key morphological features is crucial for pathologists to provide a suggestive diagnosis, which can help prevent misdiagnosis and guide appropriate clinical management.

Keywords

Idiopathic Myointimal Hyperplasia of Mesenteric Venules, Mesenteric Vasculopathy, Ischemic Colitis, Inflammatory Bowel Disease

1. 引言

特发性肠系膜静脉肌内膜增生症(idiopathic myointimal hyperplasia of mesenteric venules, IMHMV)是一种罕见的肠道血管性疾病，临床常表现为慢性腹痛、便血，易误诊为炎症性肠病(inflammatory bowel disease, IBD)。多数患者在术前被误诊，并因此接受了无效的治疗[1] [2]。更有文献报道，不当使用免疫抑制剂或皮质类固醇甚至可能引发肠穿孔等严重并发症[3] [4]。本文报道一例以反复血便为主要表现 IMHMV 病例，并结合文献探讨其临床病理特征、诊断要点及鉴别诊断，旨在提高临床及病理医师对该病的认识。

2. 材料与方法

2.1. 病例资料

患者男性，50 岁，因“反复血便 1 年余”入院。患者 1 年多前无明显诱因出现便血，外院多次肠镜提示“溃疡性结肠炎”，但经相应对症治疗(具体方案不详)后症状无明显缓解。本次入院后复查结肠镜：

进镜至距肛缘 15 cm 处见肠腔狭窄，镜身无法通过；狭窄近端肠黏膜充血、糜烂伴溃疡形成，管腔内可见大量血凝块(图 1①)。盆腔增强 CT 显示乙状结肠区肠壁弥漫性增厚，强化明显，周围脂肪间隙内见多发迂曲血管影及絮状渗出(图 1②)。因患者合并肠道狭窄，临床行腹腔镜下左半结肠切除术。

2.2. 方法

活检和手术切除组织常规 4%中性缓冲甲醛液固定，石蜡包埋，HE 染色，光镜观察。采用罗氏 Verhoeff 改良弹力纤维染色试剂盒(Elastic Stain Core Kit)与配套的 Special Stain Van Gieson CS 试剂盒，在罗氏全自动特殊染色仪上进行 Verhoeff-van Gieson (VVG)染色。所有染色步骤中使用的缓冲液均购自罗氏公司。

3. 结果

3.1. 肠镜活检病理

镜下见大量肉芽组织，少量黏膜腺体结构扭曲，但黏膜固有层内淋巴细胞、浆细胞浸润轻微(图 1③)，不支持溃疡性结肠炎病理改变。

3.2. 手术标本病理检查

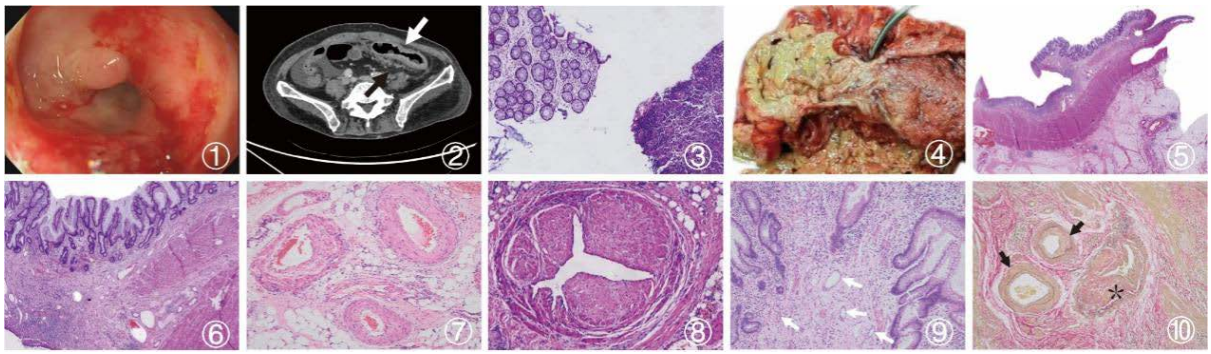
巨检：送检结肠肠管一段，长 21.0 cm，距一侧切缘 7.0 cm 处见一大小约 7.5 cm × 4.5 cm 的不规则溃疡区，周围黏膜粗糙，局部肠壁增厚、管腔狭窄(图 1④)。镜检：溃疡处黏膜及黏膜下层缺失，底部见肉芽组织，邻近黏膜肌层增生肥厚(图 1⑤)。溃疡周边黏膜隐窝结构紊乱，但固有层内淋巴浆细胞浸润不明显(图 1⑥)。肠系膜脂肪组织中见多处静脉管壁呈偏心性增厚，管腔重度狭窄(图 1⑦，图 1⑧)。此外，黏膜固有层内部分毛细血管壁因平滑肌增生而增厚，呈现“动脉化”现象(图 1⑨)。

3.2. 特殊染色

弹力纤维染色(VVG)显示病变血管内弹力膜存在，管壁因平滑肌增生而显著增厚(图 1⑩，星号示病变静脉)，伴行动脉结构未见异常(图 1⑩，黑色箭头示正常动脉)。

病理诊断：(乙状结肠) IMHMV。

随访情况：术后患者未予药物治疗，随访 2 年无复发。



① 内镜下黏膜糜烂、溃疡及狭窄；② CT 见肠壁增厚(白色箭头)；③ 活检组织中见肉芽组织，周边黏膜固有层内淋巴浆细胞浸润轻微(HE × 40)；④ 大体标本见肠道大片溃疡；⑤ 镜下见溃疡(HE × 20)；⑥ 溃疡周围粘膜隐窝结构扭曲，固有层内淋巴浆细胞浸润轻微(HE × 40)；⑦ 肠系膜内多处静脉管管壁增厚(HE × 100)；⑧ 个别管腔几乎完全闭塞(HE × 100)；⑨ 黏膜固有层中见毛细血管平滑肌增生(白色箭头) (HE × 100)；⑩ VVG 染色见静脉肌内膜壁明显增生(黑色星号)，伴行动脉未见明显异常(黑色箭头) (VVG × 100)。

Figure 1. Endoscopic, radiological, gross and microscopic pathological images of IMHMV
图 1. IMHMV 的内镜、影像、大体和镜下病理图片

Table 1. Summary of clinical and pathological characteristics of IMHMV cases reported in the literature
表 1. 文献报道 IMHMV 病例临床与病理特征汇总

性别	年龄	发病部位	临床症状	大体	病理	预后
男[5]	63	乙状结肠	急性水样腹泻、体重明显减轻	肠壁增厚，大面积溃疡	大静脉显示肌内膜增生	术后恢复良好
男[6]	63	左侧结肠	下腹痛和非血性腹泻	缺血、硬化的肠壁	静脉结构变化，管腔几乎闭塞	预后良好
男[7]	55	直肠、乙状结肠	持续性腹痛、食欲不振、体重减轻	肠道见暗色斑块，存在穿孔	静脉中平滑肌细胞增生	无
女[8]	81	末端回肠	腹痛、恶心和呕吐	肠壁增厚、环形溃疡伴瘢痕	慢性结肠炎，存在肌内膜增生	术后状况良好
男[9]	71	乙状结肠、直肠	腹痛、便秘	肠壁溃疡、瘢痕	明显肌内膜增厚和管腔闭塞	未见复发
男[10]	67	乙状结肠、降结肠	进行性血性腹泻	管腔狭窄伴硬化的肠系膜	静脉腔水肿和闭塞	无
男[11]	66	降结肠、乙状结肠及直肠	左下腹绞痛、发热、盗汗和腹泻	无	浅表粘膜缺血改变、厚壁毛细血管	因误吸心肺骤停死亡
男[12]	44	左侧结肠	便秘数周	肠壁水肿伴局灶性肠壁坏死	静脉管腔狭窄，EVG 染色不清	术后恢复良好
男[13]	37	降结肠、乙状结肠和直肠	腹痛、便秘、腹泻，近期便血	结肠僵硬，脂肪出血性粘连	出血性坏死、透壁坏死	术后恢复良好
男[13]	49	降结肠、乙状结肠和直肠	顽固性疼痛、痉挛和血性腹泻	浆膜见粘连，管腔狭窄	内膜平滑肌增生伴管腔闭塞	术后恢复良好
男[14]	82	降结肠至直肠	排便不稳定、腹胀，体重下降	无	厚壁静脉，动脉正常	术后恢复良好
男[14]	59	乙状结肠	腹泻，里急后重	无	静脉肌内膜增生、管腔狭窄	术后恢复良好
男[15]	68	左侧结肠	持续便秘、粘液便和腹胀	节段性溃疡，严重狭窄	静脉明显增厚	无
男[1]	64	末端回肠	腹痛、腹胀、体重减轻	管壁增厚、管腔狭窄	静脉壁增厚平滑肌增生，管腔狭窄	术后恢复顺利
女[16]	30	盲肠	间断性右下腹痛	盲肠溃疡，脂肪组织攀爬	静脉管壁增厚，肌内膜增生	术后恢复良好
男[17]	29	小肠	腹胀、腹痛、呕吐	系膜侧肠壁增厚，狭窄和穿孔	静脉管壁、肌内膜增生、管腔狭窄	术后恢复良好
男[18]	25	空肠	脐周痛、恶心、呕吐	局部溃疡、管腔狭窄	静脉内膜平滑不规则增生，管腔狭窄甚至闭塞	术后恢复良好

4. 讨论

IMHMV 是一种罕见的慢性肠道缺血性疾病，自 Genta 等[19]首次报道以来，目前报道的文献仍十分

有限。因其临床症状和肠镜下表现与 IBD 相似，易造成误诊。IMHMV 好发于中老年男性，既往文献报道平均年龄约 54 岁，病变多累及左半结肠[1] [20]。本研究汇总文献报道 17 例病例(见表 1)，显示平均发病年龄为 54 岁，男性占比高达 88.2% (15/17)，且 12 例(70.6%)病变位于左半结肠。本病例为 50 岁男性，病变位于乙状结肠，与文献报道特征相符。

IMHMV 确诊依赖于手术标本的病理检查。镜下特征性的肠系膜静脉偏心性或同心性肌内膜增生是诊断的“金标准”[1]。本病例手术标本 VVG 染色清晰地显示静脉壁明显增厚，而伴随动脉正常。然而，IMHMV 的术前活检诊断极具挑战性[3]。本病例术前活检未能确诊，其原因在于活检组织镜下仅见非特异性的肉芽组织，未能显示血管改变。但活检组织中黏膜固有层缺乏显著的慢性炎细胞浸润，这些组织学特征提示临床存在缺血性肠病的可能，不支持 IBD 的诊断。此外，文献报道黏膜固有层毛细血管的“动脉化”(即管壁平滑肌增生)是另一个有重要提示作用的形态学指标[1] [16] [21]。尽管有文献报道在活检中可见毛细血管“微血栓”等提示本病[20]，但本例并未观察到此类改变，这表明这些特征并非普遍存在，在诊断时需谨慎对待。

IMHMV 主要需与以下疾病相鉴别：首先，与溃疡性结肠炎鉴别，后者以黏膜固有层弥漫性慢性炎细胞浸润和隐窝结构扭曲为特征[22]，而 IMHMV 的背景炎症轻微，其黏膜改变本质上是缺血所致。其次，需与克罗恩病鉴别，克罗恩病通常表现为多节段性病变、透壁性炎症以及出现非干酪样肉芽肿[23]，这些特征在 IMHMV 中通常缺失[1]。最后，还需与肠系膜动脉缺血性疾病鉴别，后者病因多为动脉栓塞或血栓，通过弹力纤维染色可明确地将 IMHMV 的静脉病变与动脉疾病区分开来[21]。

IMHMV 的病因目前尚不明确，部分病例中的患者存在如高血压、糖尿病等基础疾病，然而其与血管异常是否存在明确关联，目前仍不清楚[20]。此例患者既往并无确切的血管相关疾病病史。该病在病程上可能存在“活动期”与“静止/纤维化期”之分[24] [25]：在活动期，病变以淋巴细胞性静脉炎为主要特征，同时伴有血管壁的炎症细胞浸润；随着病程的进展，当进入晚期静止期或纤维化期时，炎症会逐渐消退，纤维化阶段特有的静脉肌内膜增生与纤维化便成为主要的病理改变。本病例的手术标本中未发现淋巴细胞浸润，这表明患者可能处于疾病的“静止/纤维化期”。然而，文献报道显示，多数病例的病变静脉壁缺乏明确的淋巴细胞浸润[6] [8]，这也提示 IMHMV 的发病机制可能存在多种途径，可能与淋巴细胞静脉炎是相对独立的疾病。

手术切除是当前最主要且有效的治疗手段[2]。绝大多数患者术后预后良好，无需辅助药物治疗，且罕见复发[19]。本例患者行左半结肠切除术后，随访 2 年状况良好，与文献报道一致。

总之，IMHMV 是一种易误诊的罕见结肠缺血性疾病，临床及病理医生对该病的认识，避免将其误诊为 IBD，早期识别并采取正确的手术干预，防止患者接受长期无效药物治疗，对于改善患者预后至关重要。

声　明

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