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共同主编

党双锁, 博士, 教授, 研究员, 主任医师, 710004, 陕西省西安市, 西安交通大学医学院第二附属医院感染科

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Editorial Board Member of *World Chinese Journal of Digestology*, Bo-Yu Xue, Professor, PhD, Nanjing University of Traditional Chinese Medicine, No. 2301 Building 9, Langshi Linglong Yu, Tianyuan Middle Road, Jiangning District, Nanjing 211199, Jiangsu Province, China. xueboyu9502@sina.com

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Baishideng Publishing Group Inc

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蓝色橡皮泡痣综合征消化道大出血并肺栓塞1例

程发辉

程发辉, 湖南省脑科医院(湖南省第二人民医院)微创外科 湖南省长沙市 410011

程发辉, 主治医师, 研究方向为普外科疾病临床及基础研究.

作者贡献分布: 本论文由程发辉写作完成.

通讯作者: 程发辉, 主治医师, 410011, 湖南省长沙市芙蓉中路三段427号, 湖南省脑科医院(湖南省第二人民医院)微创外科. 455131290@qq.com

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Blue rubber bleb nevus syndrome with massive gastrointestinal hemorrhage and pulmonary embolism: A case report

Fa-Hui Cheng

Fa-Hui Cheng, Minimally Invasive Surgery Department, Hunan Provincial Brain Hospital (The Second People's Hospital of Hunan Province), Changsha 410011, Hunan Province, China

Corresponding author: Fa-Hui Cheng, Attending Doctor, Minimally Invasive Surgery Department, Hunan Provincial Brain Hospital (Hunan Second People's Hospital), No. 427 Section 3 Furong Middle Road, Changsha 410011, Hunan Province, China. 455131290@qq.com

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Abstract

BACKGROUND

Blue rubber bleb nevus syndrome is a rare disease. The main cause of death in these patients is massive hemorrhage of the digestive tract. When the hemorrhage is

combined with pulmonary embolism, greater treatment challenges are posed.

CASE SUMMARY

A man was admitted to the hospital because of repeated black stools for more than 20 years and aggravation for 10 d. A diagnosis of sigmoid colon and rectal blue rubber bleb nevus syndrome with massive hemorrhage due to nevus rupture was made. Conservative treatment including medical hemostasis and blood transfusion had a poor effect. Hemorrhagic shock occurred. The sigmoid colon and rectum were totally resected in the emergency setting. After operation, acute pulmonary embolism developed. Low molecular weight heparin anticoagulation and subsequent oral warfarin anticoagulation were given. The patient's bleeding stopped, and pulmonary thrombosis disappeared. He has recovered well and is now alive.

CONCLUSION

In patients with preoperative massive hemorrhage due to blue rubber bleb nevus syndrome, acute pulmonary embolism may occur after operation, and the risk of intravenous thrombolytic hemorrhage is very high. In such cases, low-molecular-weight heparin + warfarin oral bridging treatment can be chosen. During the treatment, the coagulation function should be closely monitored to avoid massive hemorrhage. The dosage should be adjusted according to the prothrombin time and international normalized ratio (INR) to maintain the INR between 2-3, and the patients should be subject to whole process management. D-dimer and INR should be reviewed regularly to monitor the complications of bleeding, and patients can benefit when they are treated properly.

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Key Words: Blue rubber bubble nevus syndrome; Pulmonary

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摘要

背景

蓝色橡皮泡痣综合征(blue rubber bleb nevus syndrome, BRBNS)是一种罕见病,其死亡的主要原因是消化道大出血,当出血同时合并肺栓塞则严重增加了患者的治疗难度。

病例简介

本例患者因反复黑便20余年,再发加重便鲜血10 d入院,诊断为乙状结肠、直肠蓝色疱痣破裂大出血,予以内科止血、输血等保守治疗后效果不佳,失血性休克,急诊行开腹乙状结肠、直肠全切除,术后合并急性肺栓塞,予以低分子肝素抗凝及后续口服华法林抗凝治疗,患者出血停止,肺血栓消失,恢复良好,目前存活。

结论

蓝色橡皮疱痣术前大出血患者,术后第一天发生急性肺栓塞,静脉溶栓出血风险极高,在此情况下我们可以选择予以选择低分子肝素+华法林口服桥接治疗,治疗期间严密监测凝血功能,避免大出血的发生,依据凝血酶原时间INR调整用量,使INR维持2-3之间,并对患者实行全程化管理,定期复查D-二聚体,INR注意出血得并发症,治疗得当时患者可以获益。

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关键词: 蓝色橡皮泡痣综合征; 肺栓塞; 消化道出血

核心提要: 蓝色橡皮泡痣综合征是一种罕见病,其死亡的主要原因是消化道大出血,在急性消化道大出血时并发肺栓塞治疗严重棘手,因此值得临床医生的关注和研究。

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0 引言

本文报道了湖南省第二人民医院收治的一例女性蓝色橡皮泡痣综合征消化道大出血并肺栓塞患者,以提高该疾病出血时合并血栓性疾病的认识。

1 病例简介

患者女性,53岁,因“反复黑便20余年,再发加重便鲜血10天”入院。既往:有肝硬化、门静脉高压、门静脉海绵样变”病史;于2015-01在湖南省人民医院行“降结肠造瘘术、脾切除术、肠系膜下动脉结扎术”,有“血管瘤、有多次血制品输注史。查体:贫血貌,急性病容,皮肤黏膜色泽苍白,腹部可见2条相互垂直的7 cm-8 cm陈旧性瘢痕;左侧腹部可见结肠造瘘口,全腹柔软,腹部无压痛,无反跳痛,右侧大腿上1/3及右小腿中部皮肤可见血管畸形治疗后手术疤痕,右小腿后侧2 cm×2 cm血管瘤肿块,右侧臀部可见3 cm×4 cm血管瘤肿块(图1)。入院后检查,血常规:红细胞 $1.77 \times 10^{12}/L$,红细胞压积16.0%,血红蛋白49.0 g/L,平均血红蛋白浓度306 g/L,血小板计数 $216 \times 10^9/L$ 。腹部CT:(1)降结肠、乙状结肠、直肠管壁弥漫性增厚并多发致密灶,周围多发迂曲血管影,右侧臀部皮下软组织密度灶同前,符合蓝色橡皮-疱痣综合征影像改变,请结合临床;(2)肝硬化,门脉高压,门静脉海绵样变;(3)降结肠造瘘术、脾脏切除术后改变,肝脏多发血管瘤。患者反复出血不止,色素下降至49 g/L,导致失血性休克。

2 最终诊断

(1)蓝色疱痣综合征;(2)消化道大出血;(3)失血性休克;(4)急性肺栓塞;(5)肝硬化、门静脉高压、门静脉海绵样变;(6)降结肠造瘘术后。

3 治疗

患者出血原因为乙状结肠及直肠蓝色疱痣破裂,因病变部位广泛,出血量大,肠镜无法完成,经内科止血药物+输血保守治疗无效,遂急诊在全麻下行剖腹探查,肠粘连松解,乙状结肠+直肠全切术(图2)手术顺利。术后第1天患者突发右侧胸痛及气促,急查血浆D-二聚体为29.8 mg/L(图3),考虑急性肺梗可能。急诊行肺动脉血管造影CT示:右侧肺动脉主干、右下肺动脉干、右上、中、下肺叶动脉及其分支栓塞(图4)确诊急性肺栓塞。因患者术前大出血并且为术后第一天,静脉药物溶栓风险极高,因患者生命体征稳定,无休克表现,经反复权衡后决定先予以低分子肝素抗凝5000 u皮下注射Q12 h,后予以口服华法林3.75 mg抗凝桥接3 d,国际标准化比值(international normalized coagulation ratio, INR)达到2.53后停用肝素;出院后继续口服华法林,根据INR调整剂量2.5 mg,维持INR 2-3之间(图5)。

4 结果和随访

随访11 mo后患者未再次出血,复查血常规提示:红细胞



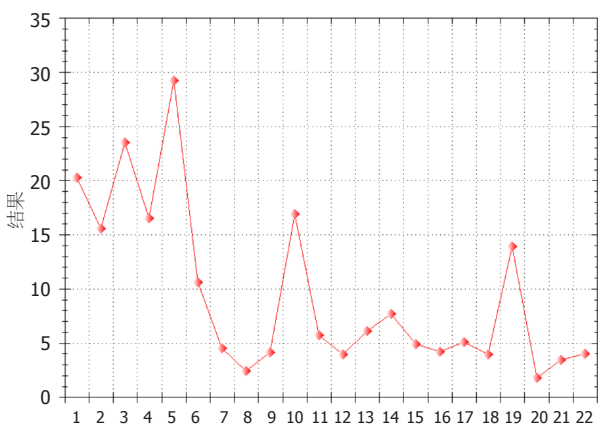
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图 1 臀部蓝色血管瘤。



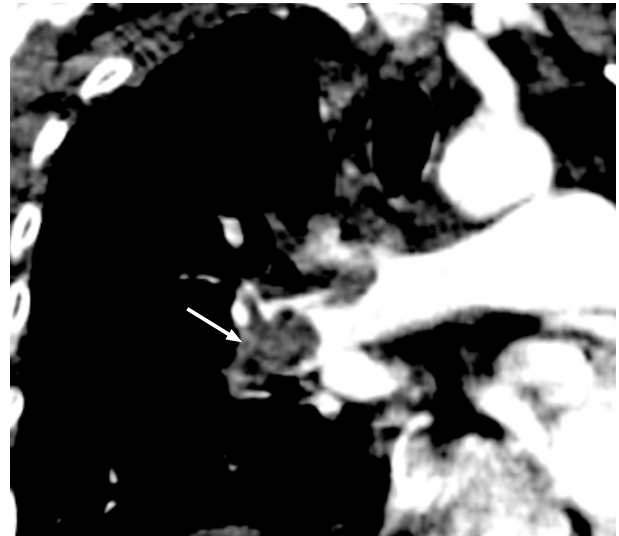
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图 2 术中乙状结肠、直肠呈瘤样改变。



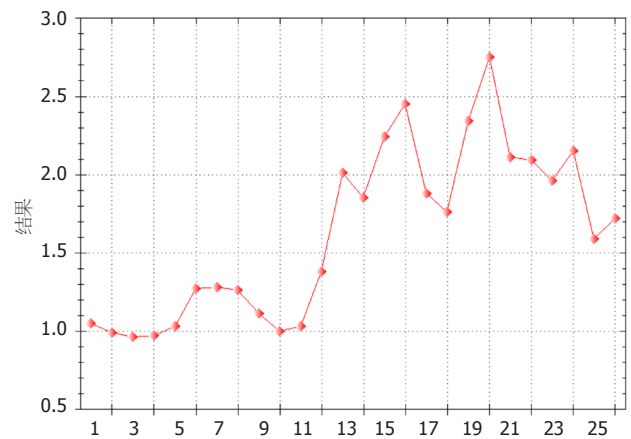
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图 3 血浆D-二聚体结果趋势图。



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图 4 肺动脉血管造影CT. 箭头: 肺动脉血栓。



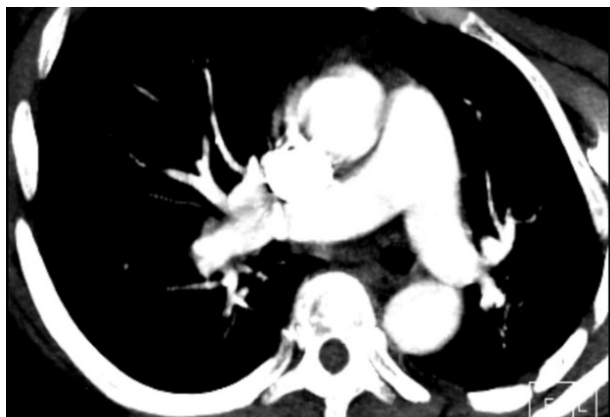
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图 5 INR结果趋势图. INR: 国际标准化比值。

$4.86 \times 10^{12}/L$, 红细胞压积42.5%, 血红蛋白145.0 g/L, 平均血红蛋白浓度342 g/L, 血小板计数 $308 \times 10^9/L$; 复查肺动脉血管造影CT提示血栓消失(图6), 无咳嗽、胸闷、胸痛呼吸困难等表现, 预后良好, 未再次出血, 目前存活良好。

5 讨论

BRBNS属罕见病, 发病率为1:140000, 主要特征是皮肤和内脏多发血管瘤, 临床表现为消化道出血及不明原因贫血^[1]。BRBNS原因不明^[1], 少数与9号染色体短臂基因突变、染色体显性遗传相关^[2]。BRBNS还可累及全身各个组织和脏器^[3,4]。不同脏器的血管瘤可以引起不同症状^[5-9]。消化道血管瘤是BRBNS常见表现, 常引起不同程度的消化道出血。多数患者为少量间断的出血, 而



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图 6 复查肺动脉血管造影CT显示血栓消失。

急性消化道大出血, 却是患者死亡的最主要原因, 需要积极抢救。内科治疗不能控制的消化道大出血或并发肠扭转、肠套叠、肠穿孔则需要外科手术^[1-3]。该患者由于消化道大出血后不可避免的使用止血药物, 以及后来的手术创伤、禁食、卧床、高凝等综合因素的影响, 从而导致肺栓塞疾病的发生。80%肺栓塞患者起病时无任何临床症状, 约2/3肺栓塞患者死亡在2 h内发生, 发病急骤、死亡率极高, 因此在消化道大出血及手术患者诊治过程中需注意血栓性疾病的防治^[10,11], 如患者出现气促、胸痛、胸闷等症状需高度警惕肺栓塞可能, 需及时复查血浆D-二聚体, 下肢静脉彩超以及肺动脉CT血管造影。在出血同时合并肺栓塞治疗时需权衡出血风险来选择溶栓或者抗凝治疗, 因患者术前大出血, 术后第一天发生急性肺栓塞, 静脉溶栓出血风险极高, 故未采用静脉溶栓治疗, 予以选择低分子肝素+华法林口服桥接治疗, 治疗期间严密监测凝血功能, 避免大出血的发生, 依据凝血酶原时间INR调整用量, 使INR维持2-3之间, 并对患者实行全程化管理, 定期复查D-二聚体, INR并且注意出血得并发症^[12,13]。

6 结论

综上所述, BRBNS临床较罕见, 但关于其并发的急性消化道大出血和肺栓塞的治疗较棘手, 因此值得临床医生

的关注和研究。

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