

## ·临床研究·

# 以脑卒中样发病的可逆性胼胝体压部病变综合征Ⅱ型二例

李佩湖 封浑 李吕力 姜炳坚

**【摘要】目的** 报告2例以脑卒中样发病的可逆性胼胝体压部病变综合征(RESLES)Ⅱ型病例,结合文献总结其发病特征及影像学表现,以提高临床医师对RESLES尤其是RESLESⅡ型的认识。**方法与结果** 2例青年男性患者均为广西壮族自治区人民医院2018年7月至2021年6月收治的RESLESⅡ型病例,以言语不清、肢体无力等脑卒中样发作发病,病程中伴头晕、恶心等急性颅内压升高症状;既往有烟酒史或代谢紊乱;体格检查和各项实验室指标无明显异常,头部MRI显示胼胝体压部及周围白质呈对称性、可逆性长T<sub>1</sub>、长T<sub>2</sub>和DWI高信号;经抗炎症、改善循环和营养神经等对症治疗后影像学检查可见病灶消退,症状与体征完全缓解。随访期间病情无复发,恢复日常生活与工作,RESLESⅡ型诊断明确。**结论** 对于脑卒中样发作且不存在动脉粥样硬化危险因素的青年患者,应考虑RESLES可能,可通过MRI特别是DWI序列快速明确诊断。

**【关键词】** 胼胝体; 脑疾病; 磁共振成像

## Reversible splenial lesion syndrome type II with stroke-like onset: two cases report

LI Pei-hu, FENG Hun, LI Lü-li, JIANG Bing-jian

Department of Neurology, Guangxi Academy of Medical Sciences, the People's Hospital of Guangxi Zhuang Autonomous Region, Nanning 530021, Guangxi, China

Corresponding author: LI Pei-hu (Email: lipeihu1992@163.com)

**【Abstract】 Objective** To report 2 cases of reversible splenial lesion syndrome (RESLES) type II presented with stroke-like attacks, and summarize the pathogenesis and imaging findings based on the literature to improve clinicians' understanding of RESLES, especially type II. **Methods and Results** The 2 young male patients were both RESLES type II cases admitted to the People's Hospital of Guangxi Zhuang Autonomous Region from July 2018 to June 2021, presented with stroke-like attacks such as slurred speech and limb weakness, and developed symptoms of acute intracranial hypertension such as dizziness, nausea, and so on. During the course of the disease, both of them had a history of alcohol consumption and smoking or metabolic disorders, physical examination and laboratory indicators were unremarkable, and head MRI examination showed symmetrical and reversible long T<sub>1</sub>, long T<sub>2</sub> and DWI hyperintensity lesions in the splenium of corpus callosum and surrounding white matter. After symptomatic treatment such as anti-inflammatory, improving blood circulation, and nourishing nerves, imaging findings showed the lesions subsided, and the symptoms and signs were completely relieved. There was no recurrence of the disease during the follow-up period, the patients have returned to their daily life and work, and the diagnosis of RESLES type II was clear. **Conclusions** For young patients with stroke-like attacks and no risk factors for atherosclerosis, the possibility of RESLES should be considered, and MRI especially DWI can quickly confirm the diagnosis.

**【Key words】** Corpus callosum; Brain diseases; Magnetic resonance imaging

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作者单位:530021 南宁,广西医学科学院 广西壮族自治区人民医院神经内科

通讯作者:李佩湖,Email:lipeihu1992@163.com

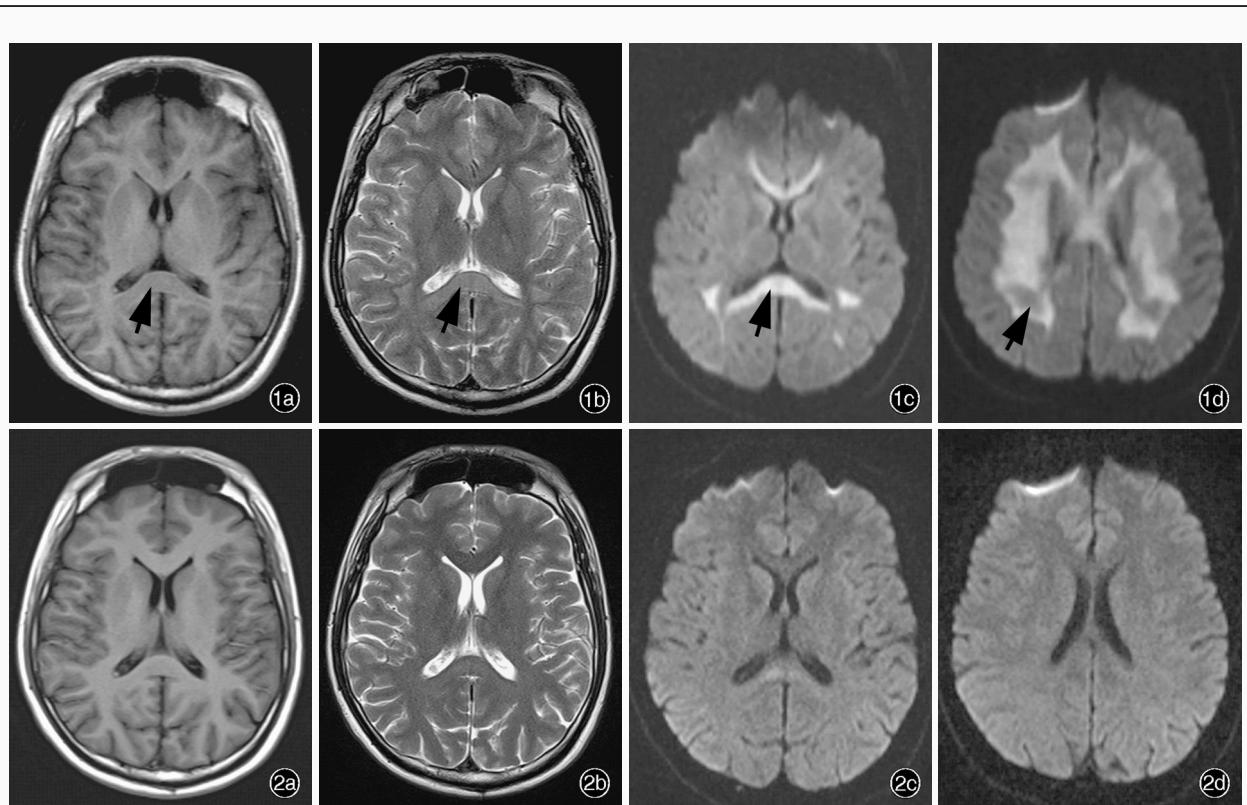
可逆性胼胝体压部病变综合征(RESLES)是一种临床较为少见的炎症性脑病,主要表现为意识改变、言语不清、癫痫发作、谵妄,也可出现头痛、发热、共济失调、呕吐、腹泻等症状,但均缺乏特异性;而以类卒中样发作发病者鲜有报道。据病灶部位可将RESLES分为I型和II型,文献报道以I型居多,II型者罕见。广西壮族自治区人民医院神经内科自2018年7月至2021年6月共收治2例以脑卒中样发病的RESLES II型患者,分别以急性脑血管病和发作性肢体麻木入院,最终经DWI明确诊断。由于RESLES II型患者临床表现复杂多样性,常因误诊或漏诊而导致过度治疗,为增强临床医师对该病的认识,笔者拟对本院收治的2例患者的临床症状与体征以及影像学资料进行回顾分析,并结合文献资料总结其发病特征,以期对临床同道诊断与鉴别诊断有所助益。

### 临床资料

**例1** 男性,28岁。因言语不清、右侧肢体无力7小时,于2018年7月16日急诊入院。患者入院前日晚跑步后出现言语不清,左侧嘴角歪斜,右侧肢体无力、活动受限,伴头晕、恶心,无头痛、呕吐、肢体抽搐,约2小时后症状加重,虽可站立行走但欠平稳,头晕、恶心未缓解并逐渐出现意识模糊,遂至我院急诊。既往史、家族史无特殊。吸烟史10余年,平均20支/d,偶少量饮酒。头部CT检查未见明显异常,考虑“急性脑血管病”收入院。入院后体检:体温36.7℃,脉搏83次/min,呼吸22次/min,血压147/106 mm Hg(1 mm Hg=0.133 kPa)。神志模糊、嗜睡,构音障碍;双侧眼动充分,双侧瞳孔等大、等圆,直径约3 mm,对光反射存在,右侧鼻唇沟变浅,伸舌稍右偏;双侧肌张力正常,右侧肌力5级、左侧5级,双侧腱反射正常;右侧Babinski征可疑阳性,左侧病理征未引出,共济运动不配合,双侧深浅感觉粗测正常,颈部柔软,无抵抗,Kernig征阴性。血清学和脑脊液检测无明显异常。头部MRI显示胼胝体压部及周围白质对称性异常信号影,呈稍长T<sub>1</sub>、稍长T<sub>2</sub>和DWI明显扩散受限(图1);心电图、心脏B超、腹部超声等均未见明显异常。根据患者临床症状、体征及影像学征象诊断为RESLES。入院第2天予以地塞米松20 mg/d静脉滴注以及维生素B<sub>1</sub>0.10 g/d和维生素B<sub>12</sub>0.50 mg/d肌肉注射;入院第5天言语不清、肢体无力及头晕等症状基本消失。

患者共住院8天,出院时神志清楚、言语清晰、四肢肌力正常,改良Rankin量表(mRS)评分为零,1周后头部MRI可见胼胝体压部及周围白质病灶基本消退(图2),最终诊断为RESLES II型;随访期间未复发,已恢复正常工作,mRS评分为零。

**例2** 男性,22岁。主诉头晕伴右侧头面部麻木3天、加重2天,于2021年2月26日急诊入院。患者2021年2月23日晚餐前无明显诱因出现头晕,伴右侧头面部、右上肢麻木,持续数十分钟,餐后自行缓解;次日因“受凉”再次出现头晕及右侧头面部麻木,自觉头晕程度有所加重,伴言语不清,无肢体无力、意识障碍、肢体抽搐等,无畏寒、发热、恶心、呕吐,无胸痛、胸闷、心悸、呼吸困难等症状与体征。至我院急诊就诊,头部CT未见异常,考虑为脑梗死,经验性予阿司匹林100 mg/d联合氯吡格雷75 mg/d、阿托伐他汀20 mg/d口服,治疗后自觉头晕症状缓解,但右侧头面部麻木不适仍存在,且在急诊科候床期间上述症状再次发作,右侧头面部麻木症状加重,并伴有咽喉部麻木、饮水呛咳、流涎、右上肢无力等症状,以“肢体麻木原因待查”收入院。既往史、个人史及家族史均无特殊。入院后各项检查:体温36.5℃,心率82次/min,呼吸20次/min,血压103/62 mm Hg。神志清楚,精神欠佳,言语不清;双侧瞳孔等大、等圆,直径约3 mm,对光反射存在,各向眼动充分,双侧鼻唇沟对称,伸舌居中;四肢肌张力正常,左侧肢体肌力5级,右上肢远端肌力4级、近端5级,右下肢肌力5级,右侧额面部、右上肢麻木,感觉系统粗测正常;腱反射减弱,病理征未引出,共济运动正常;颈部柔软,无抵抗,Kernig征阴性。大便常规肝吸虫卵0~1个/低倍视野,余各项实验室指标均无明显异常。头部CT、头颈部CTA检查无明显异常;头部MRI提示胼胝体压部、双侧放射冠、半卵圆中心异常信号,表现为稍长T<sub>1</sub>、稍长T<sub>2</sub>和DWI明显扩散受限(图3),考虑为RESLES。遂停用调脂、抗血小板和抗菌药物,改用改善循环、营养神经药物,疏血通6 ml/d静脉滴注以及维生素B<sub>1</sub>0.10 g/d和维生素B<sub>12</sub>0.50 mg/d肌肉注射,治疗次日饮水呛咳、流涎及右上肢无力等明显好转,10天后MRI显示胼胝体压部、双侧放射冠、半卵圆中心异常信号和扩散受限均减轻(图4),最终确诊为RESLES II型。患者共住院10天,出院时无头面部、肢体麻木、吞咽困难等不适,mRS评分为零;出院3个月后电话随访无复发。



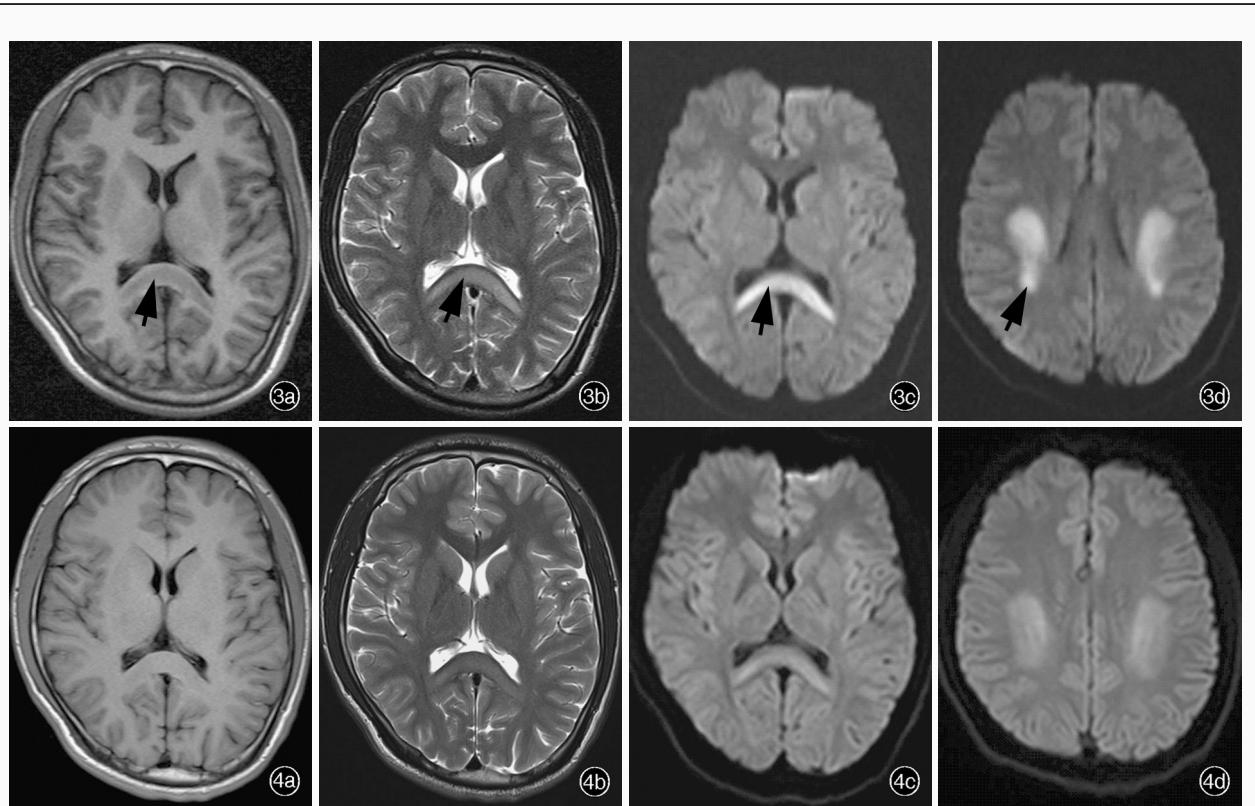
**图1** 例1患者治疗前MRI所见 1a 基底节层面横断面T<sub>1</sub>WI显示胼胝体压部、膝部和侧脑室旁稍低信号影(箭头所示) 1b 基底节层面横断面T<sub>2</sub>WI显示胼胝体压部、膝部和侧脑室旁稍高信号影(箭头所示) 1c 基底节层面横断面DWI显示胼胝体压部、膝部和侧脑室旁病变呈片状高信号(箭头所示) 1d 放射冠层面横断面DWI显示侧脑室周围、胼胝体干呈大片状高信号影(箭头所示) **图2** 例1患者治疗后1周复查MRI显示胼胝体压部及周围白质病灶基本消退 2a 基底节层面横断面T<sub>1</sub>WI 2b 基底节层面横断面T<sub>2</sub>WI 2c 基底节层面横断面DWI 2d 放射冠层面横断面DWI

**Figure 1** Head MRI findings of Case 1 before treatment. Axial T<sub>1</sub>WI of basal ganglia level showed slight hypointensity in the splenium of corpus callosum, genu of the corpus callosum and paraventricular white matter (arrow indicates, Panel 1a). Axial T<sub>2</sub>WI of basal ganglia level showed hyperintensity in the splenium of the corpus callosum, genu of the corpus callosum and paraventricular white matter (arrow indicates, Panel 1b). Axial DWI of basal ganglia level showed pieces of hyperintensity in the splenium of the corpus callosum, genu of the corpus callosum and paraventricular white matter (arrow indicates, Panel 1c). Axial DWI of corona radiata level showed large pieces of hyperintensity in periventricular white matter and trunk of corpus callosum (arrow indicates, Panel 1d). **Figure 2** After one week of treatment of Case 1, MRI showed the lesions of the splenium of corpus callosum and surrounding white matter disappeared. Axial T<sub>1</sub>WI of basal ganglia level (Panel 2a). Axial T<sub>2</sub>WI of basal ganglia level (Panel 2b). Axial DWI of basal ganglia level (Panel 2c). Axial DWI of corona radiata level (Panel 2d).

## 讨 论

RESLES为罕见临床影像学综合征,Tada等<sup>[1]</sup>于2004年提出“伴胼胝体压部可逆性病变的轻度脑炎/脑病(MERS)”定义;2011年,Garcia-Monco等<sup>[2]</sup>在既往研究的基础上将此类疾病定义为“RESLES”。RESLES多呈急性发病,临床表现多样,无明显特异性,MRI是确诊的“金标准”。根据病灶部位可将RESLES分为两种类型<sup>[3,4]</sup>: I型者病灶仅见于胼胝体压部,基本为圆形或椭圆形; II型除累及胼胝体压部,膝部、体部也可有明显病理改变,甚至可累及深部或皮质下白质。文献报道以 I型多见,本文报告2例患者均为RESLES II型,较为罕见,典型MRI

特征为T<sub>1</sub>WI等或稍低信号,T<sub>2</sub>WI、FLAIR成像为高信号,DWI呈明显高信号,ADC值降低,增强后病灶无强化。影像学表现具有可逆性是RESLES的特征性表现。因此,Garcia-Monco等<sup>[2]</sup>提出RESLES诊断标准:存在神经功能受损;头部MRI可见胼胝体病变,增强后无强化,随访过程中病灶可完全消失或显著改善;伴或不伴胼胝体以外病变。本文2例患者均符合RESLES II型诊断标准,但需注意与原发性胼胝体变性(MBD)、胼胝体肿瘤、胼胝体梗死、可逆性后部白质脑病综合征(PRES)、多发性硬化(MS)、急性播散性脑脊髓炎(ADEM)、肾上腺白质营养不良(ALD)、弥漫性轴索损伤、中毒性脑病等疾病相鉴别。其中,值得一提的是原发性胼胝体变



**图3** 例2患者治疗前MRI所见 3a 基底节层面横断面T<sub>1</sub>WI显示胼胝体压部稍低信号影(箭头所示) 3b 基底节层面横断面T<sub>2</sub>WI显示胼胝体压部高信号影(箭头所示) 3c 基底节层面横断面DWI显示胼胝体压部病变呈片状高信号(箭头所示) 3d 放射冠层面横断面DWI显示侧脑室周围白质大片状高信号影(箭头所示) **图4** 例2患者治疗后4天复查MRI显示胼胝体压部及周围白质病灶基本消退 4a 基底节层面横断面T<sub>1</sub>WI 4b 基底节层面横断面T<sub>2</sub>WI 4c 基底节层面横断面DWI 4d 放射冠层面横断面DWI

**Figure 3** Head MRI findings of Case 2 before treatment Axial T<sub>1</sub>WI of basal ganglia level showed slight hypointensity in the splenium of corpus callosum (arrow indicates, Panel 3a). Axial T<sub>2</sub>WI of basal ganglia level showed hyperintensity in the splenium of corpus callosum (arrow indicate, Panel 3b). Axial DWI of basal ganglia level showed pieces of hyperintensity in the splenium of corpus callosum (arrow indicates, Panel 3c). Axial DWI of corona radiata level showed large pieces of hyperintensity in periventricular white matter (arrow indicates, Panel 3d). **Figure 4** After 4 days of treatment of Case 2, MRI showed the lesions of splenium of corpus callosum and surrounding white matter disappeared Axial T<sub>1</sub>WI of basal ganglia level (Panel 4a). Axial T<sub>2</sub>WI of basal ganglia level (Panel 4b). Axial DWI of basal ganglia level (Panel 4c). Axial DWI of corona radiation level (Panel 4d).

性,该病主要见于慢性、严重酒精中毒和多种维生素缺乏症患者<sup>[5]</sup>,胼胝体常表现为严重坏死或进行性脱髓鞘改变,亦可累及胼胝体以外区域,病理改变多不可逆,DWI高信号消退不完全,可资鉴别。

RESLES病因尚不明确,文献报道的病因可以分为以下几类:(1)与感染相关,以病毒感染最为常见,包括轮状病毒、腺病毒、腮腺炎病毒、水痘-带状疱疹病毒、甲型肝炎病毒、乙型流感病毒等;细菌感染以大肠杆菌、链球菌、金黄色葡萄球菌等居多<sup>[6]</sup>。(2)与癫痫发作或应用抗癫痫药物相关,癫痫发作时神经元异常放电并向周围扩散,胼胝体游离水分子扩散程度降低,形成细胞毒性水肿<sup>[7]</sup>,常见抗癫痫药物包括卡马西平、苯妥英钠和拉莫三嗪等。(3)与代谢紊乱相关,常见诱发因素有低血糖<sup>[8]</sup>、维生素B<sub>12</sub>缺乏、高/低钠血症<sup>[9]</sup>、酒精依赖<sup>[10]</sup>以及高乳酸血

症<sup>[11]</sup>。(4)其他原因,如偏头痛<sup>[12]</sup>、脑静脉系统血栓形成(CVT)<sup>[13]</sup>、抗电压门控性钾离子通道(VGKC)自身抗体综合征<sup>[14]</sup>、房间隔缺损<sup>[15]</sup>等病因。目前关于RESLES的病理生理学机制尚不清楚,推测其DWI可逆性扩散受限可能与髓鞘内水肿、血-脑屏障破坏、可逆性脱髓鞘、精氨酸加压素释放和炎性细胞浸润致细胞毒性水肿有关<sup>[2,5]</sup>。RESLES病灶选择性发生于胼胝体及其两侧对称脑白质的机制可能是病毒抗原或其所诱导的抗体对胼胝体轴突受体具有特异性亲和力,抑或是胼胝体对代谢性疾病中兴奋性氨基酸毒性损伤不具有抵抗力<sup>[16-17]</sup>。本文例1患者既往有烟酒史,二者均可使胼胝体微结构受损<sup>[3]</sup>,且酒精依赖为RESLES的高危因素<sup>[9]</sup>,患者于运动后发病,可能存在高/低钠血症、高乳酸血症等代谢紊乱,故推测代谢紊乱为诱因,遗憾的是,急

诊入院时未及时监测影响内环境紊乱的指标。例2患者晚餐前发病,自觉有饥饿感,进餐后症状好转,且症状复发及加重时亦有饥饿感,推测病因与糖代谢紊乱有关,入院后虽血糖和胰岛素水平均于正常值范围,但不能排除发病时存在低血糖状态致脑细胞能量衰竭、细胞膜离子泵活性降低和细胞外液水分子转移至细胞内,形成细胞毒性水肿<sup>[18]</sup>。由于未在发病第一时间检测患者血糖,入院后也未行24小时动态血糖监测,以及急诊未予MRI动态观察病灶变化,故无法获得病因诊断。

RESLES临床表现以意识障碍、言语不清、癫痫发作和谵妄常见,病程中也可出现头痛、发热、共济失调、呕吐、腹泻等症状,但均缺乏特异性<sup>[19]</sup>。而单纯表现为类脑卒中样发作的RESLES鲜有文献报道,Dong等<sup>[20]</sup>于2016年报告1例14岁女性患儿,表现为3次发作性右上肢无力及言语不清,每次持续数小时,发病2周前有轻微的上呼吸道感染症状,食欲不振,疲倦,头痛,无发热,未予特殊治疗,10天后头部MRI显示胼胝体及周围白质异常信号消退,病因考虑为病毒感染引起的胼胝体髓鞘弥漫性肿胀。姚棉璇等<sup>[21]</sup>报告1例28岁女性,以言语模糊伴右上肢乏力首发,静脉溶栓后症状改善,但次日复发,与本文例2患者相似,发病前有饥饿感,指尖血糖为4.10 mmol/L,约10分钟后症状自行缓解,出院30天后头部MRI显示胼胝体病灶消退,但病因不明。本文例1患者表现为言语不清、肢体无力及意识模糊急性发病特点,类似脑卒中样发作,极为罕见,极易被误诊为脑梗死而施以血管内治疗;但该患者为青年,无脑卒中危险因素,可排除脑血管病可能,经MRI检查确诊为RESLESⅡ型;例2类似短暂性脑缺血发作(TIA),症状波动,急诊时考虑急性脑卒中采取二级预防治疗,最后经头部MRI检查方确诊为RESLESⅡ型。结合文献报道及本文2例患者发病特点,提示对无动脉粥样硬化危险因素,呈脑卒中样发作的年轻(<30岁)患者,应考虑RESLES可能。

RESLES的治疗尚无特异性方案。一般认为,考虑与感染有关者,采取控制感染与免疫抑制剂或静脉注射免疫球蛋白联合治疗以减少感染后免疫介导的病理损伤。涂琪和吴成斯<sup>[22]</sup>认为,RESLES患者存在不同程度的血清白蛋白水平降低,可能是胼胝体水肿的高危因素,建议尽早行血清学检查,及时补充白蛋白有助于缩短病程。本文例1患者确诊后予以地塞米松症状明显好转;例2患者急诊入

院时所予以的抗血小板、调脂及改善循环治疗具有降低微栓子形成、抑制细胞毒性水肿和抗炎症作用,对改善病情亦有裨益。然而作为一种自限性疾病,是否需要采取上述治疗尚存争议<sup>[23]</sup>。研究显示,伴严重意识障碍、胼胝体以外病变或脑电图呈弥漫性慢波的RESLES患者预后不良<sup>[24]</sup>,合并脑静脉窦血栓或抗VGKC自身抗体综合征患者多为预后不良高危人群。本文2例患者病变均累及半卵圆中心深部白质,由于为青年,且无感染及其他合并症,影像学检查显示病灶和症状于发病后数小时至数周内即完全消失,总体预后良好。

本文2例患者均为临床罕见的以脑卒中样发作发病的RESLESⅡ型病例,提示急诊科医师对类脑卒中样发作且无动脉粥样硬化危险因素的青少年患者,应考虑RESLES可能,通过MRI特别是DWI快速明确诊断,使患者避免不必要的检查或治疗;入院后尽可能查找病因,以降低复发风险。

利益冲突 无

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**【点评】** 可逆性胼胝体压部综合征(RESLES)临床少见,本文报告2例RESLESⅡ型典型病例,临床资料完整,尤其是提供了较为详尽、治疗前后对比的MRI和DWI影像学资料。虽然2例患者病因不甚明确,但既往史中分别存在烟酒史或代谢紊乱等危险因素,其中酒精依赖为RESLES的高危因素,例1于运动后发病,可能存在高钠血症或低钠血症、高乳酸血症;例2于晚餐前发病,进餐后症状好转且症状复发及加重均与“饥饿”有关。虽然2例患者诱发因素与糖代谢紊乱有关,但遗憾的是,入院后均未行动态血糖和内环境相关指标监测以及MRI动态观察胼胝体病灶变化,无法明确病因。虽然如此,本文作者仍结合文献对各种可能的病因进行阐述,诸如感染、抗癫痫药物、代谢紊乱等,可供读者诊断与治疗该病时进一步探讨。由于RESLES病因尚未明确,《中国现代神经疾病杂志》于本期报告2例典型病例,希望临床医师今后多加关注。

(吉林大学第一医院神经内科 刘明利教授)

## · 小词典 ·

### 中英文对照名词词汇(八)

- 中国罕见病联盟 China Alliance for Rare Disease(CHARD)  
中性粒细胞/淋巴细胞比值 neutrophil to lymphocyte ratio(NLR)  
肿瘤基因组学图谱计划 The Cancer Genome Atlas(TCGA)  
肿瘤微环境 tumor microenvironment(TME)  
肿瘤治疗电场 tumor-treating fields(TTFields)  
重症肌无力 myasthenia gravis(MG)

- 自身免疫性肝炎 autoimmune hepatitis(AIH)  
自身免疫性胶质纤维酸性蛋白星形胶质细胞病 autoimmune glial fibrillary acidic protein astrocytopathy (GFAP-A)  
自身免疫性脑炎 autoimmune encephalitis(AE)  
自体造血干细胞 hematopoietic stem cells(HSCs)  
组织转染法 tissue-based assay(TBA)