· 论著 临床诊治 ·

# 联合脑血运重建术治疗儿童烟雾病的疗效

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【摘要】 **目的** 观察联合脑血运重建术治疗儿童烟雾病的临床疗效。**方法** 回顾性分析2020-01—2021-12河南省人民医院烟雾病诊疗中心收治的儿童烟雾病(年龄8~15岁)14例,收集和分析患儿的一般资料、临床特征、围术期并发症和改良Rankin量表(mRS)评分。**结果** 14例儿童烟雾病共行25次手术,其中21次联合脑血运重建术,1次手术后患儿出现一过性癫痫发作,1次手术后患儿出现头皮局部坏死,2次手术后患儿出现小梗死,无大梗死和脑出血等严重并发症。术后中期随访DSA显示,颈外动脉系统通过桥血管和颞中、深动脉向脑皮层代偿供血,PWI显示脑灌注缺血情况不同程度改善;术后4例(28.57%)患儿神经功能缺损症状得到改善,9例(64.29%)患儿症状稳定,未再出现新发的神经功能缺损症状,1例(7.14%)患者出现新的神经功能缺损症状。**结论** 联合脑血运重建术治疗儿童烟雾病较安全,可以明显改善脑灌注情况,中期随访效果好。

【关键词】 烟雾病;儿童;联合脑血运重建术;脑灌注;神经功能缺损
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## Efficacy of combined cerebral revascularization in the treatment of children moyamoya disease

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【**Abstract**】 **Objective** To investigate the clinical effect of combined cerebral revascularization in the treatment of children moyamoya disease. **Methods** Fourteen children moyamoya disease patients in Henan Provincial People's Hospital from January 2020 to December 2021 were analyzed retrospectively. The clinical features, score of modified Rankin Scale(mRS)and perioperative complications were collected. **Results** A total of 25 operations were performed in 14 cases of children moyamoya disease, of which 21 were combined cerebral revascularization. Transient epileptic seizure occurred after 1 operation, local scalp necrosis occurred after 1 operation, and minor infarction occurred after 2 operations, without major infarction or cerebral hemorrhage and other serious complications. DSA showed that the external carotid artery system compensated blood supply to the cerebral cortex, and PWI showed that cerebral perfusion ischemia improved to varying degrees. After surgery, 4 children (28.57%) had improved neurological deficits, 9 children (64.29%) had stable symptoms without new neurological deficits, and 1 patient (7.14%) had new neurological deficits. **Conclusion** Combined cerebral revascularization is safe in the treatment of children with moyamoya disease, can significantly improve cerebral perfusion, with good mid-term follow-up results.

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【Key words】 Moyamoya disease; Children; Combined cerebral revascularization; Cerebral perfusion; Neurological deficits

烟雾病由日本两位学者SUZUKI和TAKAKU在1969年首次报道[1]，国内是在1976年由河南医学院李树新教授首次报道[2]。烟雾病病因目前尚不明确，临床表现为双侧颈内动脉末端（包括大脑中动脉和大脑前动脉起始部）进行性狭窄或闭塞，继发颅底异常血管网形成（在脑血管造影图像上形似“烟雾”）为特征的一种脑血管病[1,3]。烟雾病具有种族差异，在东亚国家（中国、韩国和日本等）发病率相对较高，而在欧美国家发病率非常低，且烟雾病有一定的家族聚集性，表明遗传因素参与疾病的发生[4]。烟雾病的发病年龄呈特征性的“双峰”分布，一个高峰是5～9岁的儿童，另一个高峰是40岁左右的成人[5-7]。成人烟雾病缺血型和出血型发病率无明显差别，儿童烟雾病主要为缺血型，表现为短暂性脑缺血发作（transient ischemic attack，TIA）、脑梗死、头痛头晕，出血型较少见[8]。近年来随着影像技术的进步和对烟雾病认识的加深，手术理念也在不断更新，但国内对于联合脑血运重建术治疗儿童烟雾病效果的研究报道较少。本研究通过河南省人民医院单中心临床诊疗经验，分析联合脑血运重建术治疗儿童烟雾病的临床效果。

## 1 资料与方法

**1.1 一般资料** 回顾性分析2020-01—2021-12河南省人民医院神经外科收治的14例儿童烟雾病临床资料，患儿均为缺血型烟雾病，其中男6例，女8例，年龄8～15（11.93±2.3）岁。

纳入标准：（1）烟雾病的诊断符合2017版《烟雾病和烟雾综合征诊断与治疗中国专家共识》[3]；（2）年龄≤16岁；（3）由同一名术者主刀完成手术操作。排除标准：（1）遗传性和自身免疫性疾病以及头颈部放射治疗后等可能引起脑血管损伤者；（2）伴颅内动脉瘤和脑血管畸形等其他脑血管病的患儿；（3）临床资料不完善的患儿。

首发症状：头痛头晕3例，脑梗死4例，TIA 7例。患者术前mRS评分（0.64±0.63）分；Suzuki分期：Ⅱ期3侧，Ⅲ期18侧，Ⅳ期5侧，Ⅴ期1侧。PWI表现均为前循环灌注减低。

**1.2 手术治疗** 采用额颞开颅，将颞肌完整剥离，保护好颞中、深动脉，分离颞浅动脉顶支或额支，将颞浅动脉从颞肌底部穿过作为供体血管，取下骨瓣，剪开硬脑膜并将其四周悬吊，反转硬膜贴敷在脑表面，将颞浅动脉与大脑中动脉远端血管行端侧吻合，最后将颞肌表面止血，贴敷在脑表面，其边缘固定，骨瓣塑形后还纳。

**1.3 随访** 术后中期[6个月和（或）12个月]对患儿行临床随访，行DSA检查评估颅内血管的重建情况，术侧新生血管评估参照Matsushima分级标准[9]；行PWI检查评估脑灌注改善情况；采用改良Rankin量表（mRS）评分评估患儿的临床症状改善情况。

**1.4 统计学方法** 采用SPSS 26.0对数据进行统计学分析，计量资料以均数±标准差（$\bar{x}\pm s$）表示，2组间比较采用$t$检验。以$P<0.05$为差异有统计学意义。

## 2 结果

14例儿童烟雾病共行25次手术，其中21次联合脑血运重建术，1次手术后患儿出现一过性癫痫发作，1次手术后患儿出现头皮局部坏死，2次手术后患儿出现小梗死，无大梗死和脑出血等严重并发症。术后中期随访DSA显示，颈外动脉系统通过桥血管和颞中、深动脉向脑皮层代偿供血，PWI显示脑灌注缺血情况不同程度改善（图1）。术后4例（28.57%）患儿神经功能缺损症状得到改善，9例（64.29%）患儿症状稳定，未再出现新发神经功能缺损症状，1例（7.14%）患者出现新的神经功能缺损症状。患儿神经功能缺损症状改善或稳定认为手术是有效的，本研究中手术有效率为92.86%，至末次随访，14例患儿的mRS评分为（0.43±0.76）分。见表1。

## 3 讨论

烟雾病是一种慢性进展性疾病，目前没有方法能够减缓或阻止这一过程的进展。对于儿童烟雾病来说，开始有时是单侧病变，但随着年龄的增长，另一侧才逐渐出现病变，单侧病变常常发展为双侧病变，因此，发生于儿童的单侧病变也被认为是烟雾病[10]。

血管内皮细胞增生与迁移导致管腔进行性狭窄或闭塞是烟雾病的病理基础[3]。不稳定的脑血管系统是烟雾病的另一个特点，脆弱的侧支血管是潜在的出血原因，脉络膜新生血管是出血型烟雾病患者再次出血的诱因，病理性新生血管的形成导致的血流动力学改变是引起侧支血管病理变化继而出血的

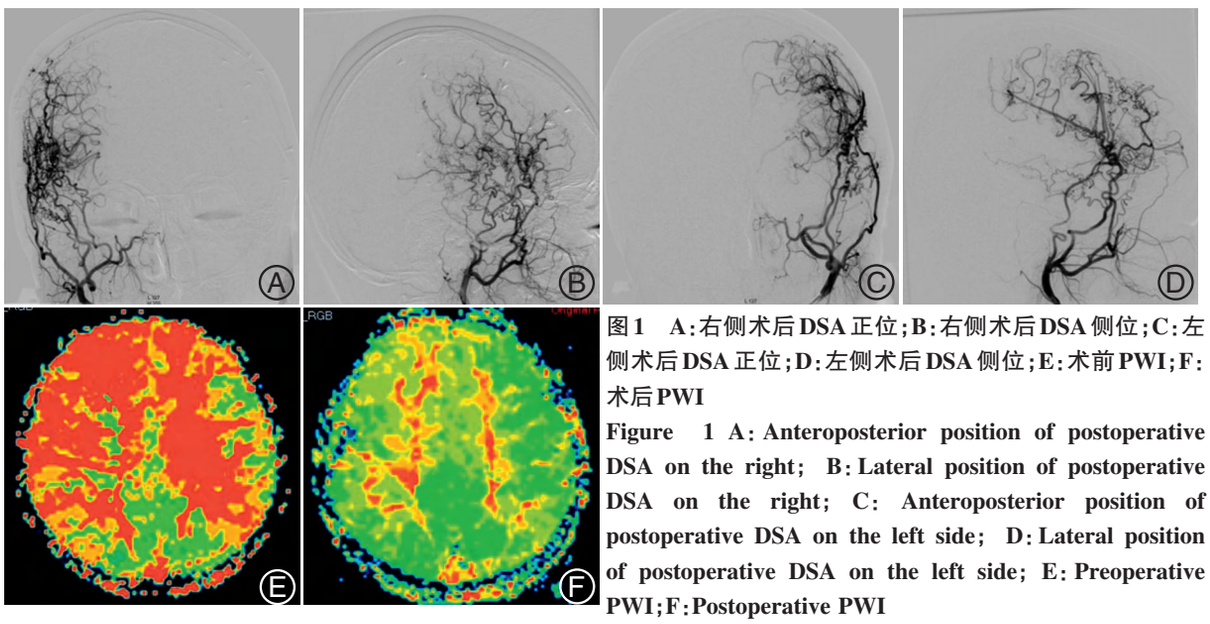


**图1 A:右侧术后DSA正位;B:右侧术后DSA侧位;C:左侧术后DSA正位;D:左侧术后DSA侧位;E:术前PWI;F:术后PWI**

**Figure 1 A: Anteroposterior position of postoperative DSA on the right; B: Lateral position of postoperative DSA on the right; C: Anteroposterior position of postoperative DSA on the left side; D: Lateral position of postoperative DSA on the left side; E: Preoperative PWI; F: Postoperative PWI**

**表1 14例MMD患儿的临床资料**

**Table 1 Clinical data of 14 children with MMD**

| 编号 | 年龄/岁 | 性别 | 首发症状 | Suzuki分期 | 手术方式 | 术后并发症 | 术前mRS评分/分 | 术后早期mRS评分/分 | 术后半年mRS评分/分 | Matsushima分级 |
|---|---|---|---|---|---|---|---|---|---|---|
| 1 | 8 | 女 | TIA | 左Ⅲ 右Ⅲ | 右侧EDMS，左侧联合 | 无 | 1 | 1 | 0 | 左2<br>右2 |
| 2 | 11 | 男 | 头痛、头晕 | 右Ⅲ 左Ⅳ | 左侧EDMS，右侧联合 | 无 | 1 | 1 | 0 | 左2<br>右3 |
| 3 | 11 | 女 | TIA | 单侧(左Ⅲ) | 左侧联合 | 无 | 1 | 1 | 0 | 左3 |
| 4 | 15 | 女 | 脑梗死 | 左Ⅱ右Ⅴ | 右侧联合，左侧保守 | 无 | 2 | 2 | 2 | 右2 |
| 5 | 10 | 男 | 脑梗死 | 左Ⅲ 右Ⅲ | 左侧联合，右侧保守 | 第1次术后局灶性脑梗死 | 1 | 3 | 1 | 左2 |
| 6 | 13 | 男 | 脑梗死 | 左Ⅲ 右Ⅲ | 左侧联合，右侧EDMS | 第2次术后局灶性脑梗死 | 1 | 3 | 2 | 左3<br>右3 |
| 7 | 13 | 男 | TIA | 左Ⅲ 右Ⅲ | 右侧联合，左侧联合 | 第1次术后头皮局部坏死 | 0 | 0 | 0 | 左2<br>右2 |
| 8 | 12 | 女 | TIA | 左Ⅳ 右Ⅲ | 右侧联合，左侧联合 | 无 | 0 | 0 | 0 | 左2<br>右2 |
| 9 | 15 | 女 | TIA | 左Ⅲ 右Ⅲ | 右侧联合，左侧联合 | 无 | 0 | 0 | 0 | 左2<br>右2 |
| 10 | 11 | 女 | 脑梗死 | 左Ⅳ 右Ⅳ | 左侧EDMS，右侧联合 | 无 | 1 | 1 | 1 | 左3<br>右3 |
| 11 | 12 | 女 | 头痛 | 左Ⅱ右Ⅲ | 右侧联合，左侧联合 | 无 | 0 | 0 | 0 | 左3<br>右3 |
| 12 | 13 | 女 | 头晕 | 左Ⅲ 右Ⅲ | 左侧联合，右侧联合 | 无 | 1 | 1 | 0 | 左2<br>右2 |
| 13 | 8 | 男 | TIA | 左Ⅱ右Ⅲ | 左侧联合，右侧联合 | 无 | 0 | 0 | 0 | 左3<br>右3 |
| 14 | 15 | 男 | TIA | 左Ⅳ 右Ⅲ | 右侧联合，左侧联合 | 第1次术后发生癫痫 | 0 | 1 | 0 | 左2<br>右1 |

注:EDMS为脑-硬膜-肌肉-血管融合术,联合为联合脑血运重建术

原因[11,12]。但对于儿童烟雾病来说,绝大多数是缺血型,出血型较少见,随着年龄增长出血型比例才开始增加。

烟雾病外科手术治疗优于保守治疗,可以减少卒中的复发[13]。儿童烟雾病的发展速度很快,而且预后较差。因此,对于儿童烟雾病,建议尽早行脑血运重建术[14]。

目前手术方式可分为单纯直接、单纯间接和联合血运重建三种手术方案,均可用于治疗儿童烟雾病[15]。直接血运重建术的一个主要的方式是颞浅动脉-大脑中动脉(STA-MCA)。颞浅动脉–大脑前动脉(STA-ACA)或枕动脉–大脑后动脉(OA-PCA)吻合也可以根据缺血情况使用[1]。最近的一项Meta分析证实了直接血运重建术在降低脑卒中风险方面优于间接血运重建术[16],同时直接搭桥也可减少出血性烟雾病再出血的风险[17]。STA-MCA也可以通过增加脑血流缓解烟雾病患者严重的头痛情况[18]。直接搭桥术后最常见的并发症之一是高灌注综合征,其特点是脑血流灌注增加,可能导致癫痫、高灌注出血和神经功能缺损[19]。间接血运重建手术的中心原则是利用带血管蒂的组织放置于脑表面,使其形成新生血管[1]。间接血运重建依赖于新血管的形成,这可能需要几个月时间的形成。间接血运重建也取决于年龄,儿童术后效果比成人表现得更好。直接血运重建提供了即刻的脑灌注,而间接血运重建提供了持续的血管再生情况[20]。但对于儿童来说,实施直接血运重建术存在一定程度困难,因儿童脑皮层动脉血管较成人更纤细、脆弱[21]。我中心部分儿童烟雾病采用的是联合脑血运重建术,术后中期随访证实取得良好的临床效果,脑灌注缺血情况有不同程度的改善。烟雾病术后癫痫发生率为10.9%~18.9%[22],病变侧别、铃木分期、术后新发脑出血、脑梗死及高灌注综合征等与术后癫痫发生相关[23],烟雾病术后癫痫发作大多数为一过性,极少出现癫痫持续状态和癫痫长期存在的情况。儿童烟雾病术后癫痫发生率高于成人,这可能与儿童神经系统发育尚不完善有关[24]。直接血管搭桥术引起的血流动力学改变,不仅可能引起高灌注综合征,而且可能发生分水岭迁移,导致新发脑梗死[25-30]。本研究中有2次联合脑血运重建术后早期患儿出现小梗死,但后期经过康复训练以及新生血管的建立,丧失的神经功能得以恢复。手术过程中颞浅动脉的分离使得头皮血供变差,患儿头皮又较薄,所以术后容易出现头皮坏死,研究的病例中有1次手术后出现局部头皮坏死情况,经过加强换药后愈合。此外,有的临床中心在成人烟雾病术后会使用抗血小板药物以减少吻合口微血栓形成,但这也存在一定的出血风险,对于儿童烟雾病术后应用与否目前尚无定论[31-35]。

本研究显示,联合脑血运重建术治疗儿童烟雾病安全有效,中期随访临床效果良好[36-42]。本研究仅为单中心研究,且样本量有限,后续需要扩大样本量以及开展多中心的研究证实。

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