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A New Treatment Strategy in Basal Ganglia Hemorrhage for Moyamoya Disease: A Retrospective Study

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Abstract

Objective: To analyze the clinical characteristics and explore the surgical treatment strategies of basal ganglia hemorrhage patients with moyamoya disease.

Methods: Follow-up data of all patients were obtained in this retrospective study, and SPSS 21 was used to analysis the statistical significance.

Results: A total of 78 patients (38 males and 40 females) were enrolled, the average age was 43.7 years ± 11.5 years, and the median time from onset to hospitalization was 3 h. Clinically, there were 47 cases mainly presented with dizziness and headache, 19 cases only presented with limb movement disorder and/or language disorder, 12 cases presented with sudden loss of consciousness and 2 cases with epilepsy. Logistic regression analysis showed that hemorrhage broke into the ventricle (OR=12.382, P=0.021<P=0.05) is a risk factor for death. Further analysis by Kaplane-Meier curve (X^2=8.223, P=0.004) and Cox (X^2=29.334, P=0.001) between hematoma broke into the ventricle and non-broke group demonstrated significant statistical difference of survival time. Additionally, 5 patients (5/21) underwent hematoma removal combined Encephalo-Duro-Arterio-Myosynangiosis (EDAMS) indirect bypass in one surgery. DSA showed the patency blood flow and well compensated neovascularization 3 months postoperative.

Conclusion: Clinical characteristics in basal ganglia hemorrhage for MMD are basically consistent with the hemorrhagic Moyamoya disease. Combined hematoma removal and indirect revascularization is a feasible strategy for surgical procedure.

Keywords: Basal ganglia hemorrhage; Moyamoya disease; Clinical features; Surgical strategy

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Introduction

To our knowledge, spontaneous Intracerebral Hemorrhage (ICH) involving the basal ganglia remains a leading cause of stroke-related mortality and morbidity [1]. Basal ganglia is the most common bleeding site of cerebral hemorrhage, followed by the cerebellum and other brain lobes [2,3]. Hypertension is one of the most important reasons for basal ganglia hemorrhage and clinical features of basal ganglia hemorrhage caused by hypertension and Moyamoya disease are different. The former have an average age about 65 years, the average duration from onset of symptoms to admission was about 23 h. Also, there generally was no obvious signs except a disturbance of unconscious, or a severe headache accompanied with nausea, vomiting and neurological deficit suddenly. Basal ganglia cerebral hemorrhage is traditionally considered to be hypertensive cerebral hemorrhage [4], but in recent years, the detection rate of Moyamoya Disease (MMD) in the basal ganglia cerebral hemorrhage patients has been gradually increased. However, the risk factor of MMD has been ignored severely among cerebral hemorrhage. To date, there only a few studies on this topic and the surgical treatment strategies of basal ganglia hemorrhage for MMD have not been provided except just remove the hematoma. Therefore, the clinical features should be identified and appropriate treatment measures should be taken.

This report studied the clinical data of 78 patients with MMD who had basal cerebral hemorrhage from January 2013 to January 2019 retrospectively. The purpose of this study was to investigate the clinical features and surgical treatment strategies of basal ganglia hemorrhage for MMD.

Methods and Analysis

Patients selection

3,555 patients with cerebral hemorrhage were admitted in the First Affiliated Hospital of

Zhengzhou University from January 2013 to January 2019, including 1466 patients (41.2%) with basal ganglia hemorrhage. Among the imaging data of basal ganglia hemorrhage, there were 521,404 and 36 patients underwent CTA, MRA and DSA respectively (Figure 1). According to the diagnostic criteria of MMD [5], we analyzed all the imaging data of basal ganglia hemorrhage. A total of 78 (78/1466, 5.3%) patients were diagnosed with Moyamoya disease or Moyamoya syndrome whose vessel like a puff of smoke in the bottom of the brain. Among them, the imaging data of DSA 36 cases, CTA 36 cases and MRA 6 cases, respectively.

Research methods

The clinic data were collected from patients with Moyamoya Disease (MMD) or Moyamoya Syndrome (MMS), including gender, age, first symptom, time from onset to hospitalization, bleeding side, and the hematoma broke into the ventricles or not. According to whether the patients died or not, all 78 patients were divided into survival and non-survival groups, then the risk factors of death were analyzed between the two groups. The long-term survival status of the patients was evaluated using the mRS score. All patients were followed by telephone combined with medical records.

Statistical analysis

Statistical analyses were performed using the SPSS software (vision 21.0). Medians or the Mean ± Standard Deviation (SD) was used to describe continuous variables, and percentage (%) was used to describe discrete variables. A univariate analysis was performed by chi-square test. A Multivariate analysis was performed by a logistic regression analysis. The test level was set as α=0.05, P<0.05 was considered statistically significant.

Results

Clinical results

We identified 78 cases of basal ganglia hemorrhage patients for MMD totally, including 38 males and 40 females with female/male ratio of 1.05:1. The mean age was (43.7 ± 11.5) years (range 12 years to 76 years). There were 35 cases with basal ganglia hemorrhage on the left and 43 cases on the right side. Furthermore, 26 patients of the basal ganglia hemorrhage for MMD were treated conservatively while 52 patients underwent removal of intracranial hematoma. During the follow-up time, there were 17 cases occurred cerebral hemorrhage once again, accounting for 21.8% (17/78). Meanwhile, among the 78 patients of basal ganglia hemorrhage, there were 47 cases with hematoma broke into the ventricle, 7 cases broke into the subarachnoid, and 1 case occurred both in ventricle and subarachnoid. We also found 21 (21/78, 26.9%) patients with hypertension, the blood pressure was all controlled well with the regular use of drugs. The mean follow up time was 38.8 (range 6-231) months in the retrospective study.

In terms of clinical symptoms, there were 47 cases mainly presented with dizziness and headache, among which accompanied by nausea and vomiting 20 cases, limb movement disorders 14 cases, language disorder 3 cases and visual impairments 2 cases. Also, there were 19 cases only presented with limb movement disorder and/or language disorder, including 10 cases with limb movement disorder, 2 cases with language disorder, and 7 cases with both disorders at the same time. Moreover, there were 12 cases presented with sudden loss of consciousness. Furthermore, there were 2 cases just presented with epilepsy. Finally, the mRS of the survive patients were evaluated during follow-up, 43 (55.1%) cases with mRS of 0-2, 17 cases (21.8%) of 3-4 and 18 cases (23.1%) of 5-6. Up to date of follow-up, 12 patients died of the first basal ganglia hemorrhage and 2 died from postoperative infection or multiple organ failure. The clinical features of the patients are summarized in Table 1.

Table 1: The clinical features of basal ganglia hemorrhage for MMD.

Variable	Number (%)
Death	14 (17.9%)
Follow-up time (M ± SD)	38.8 ± 35.7
Hemorrhage side in left	35 (44.9%)
Female	40 (51.3%)
Average age	43.7 ± 11.5
Median onset to hospital (h)	3
Hemorrhage imaging data	
limited in basal ganglia	24 (30.7%)
accompanied broke into the ventricles	47 (60.2%)
accompanied broke into the subarachnoid	7 (8.8%)
accompanied above two types	1 (1.3%)
Clinical symptom	
headache accompanied dizziness,	45 (57.7%)
limb movement and/or language disorders	19 (24.3%)
acute consciousness disorder	12 (15.4%)
epilepsy	2 (2.6%)
mRS (median)	2
0	6 (7.7%)
1-2	37 (47.6%)
3-4	17 (21.7%)
5-6	18 (23.0%)

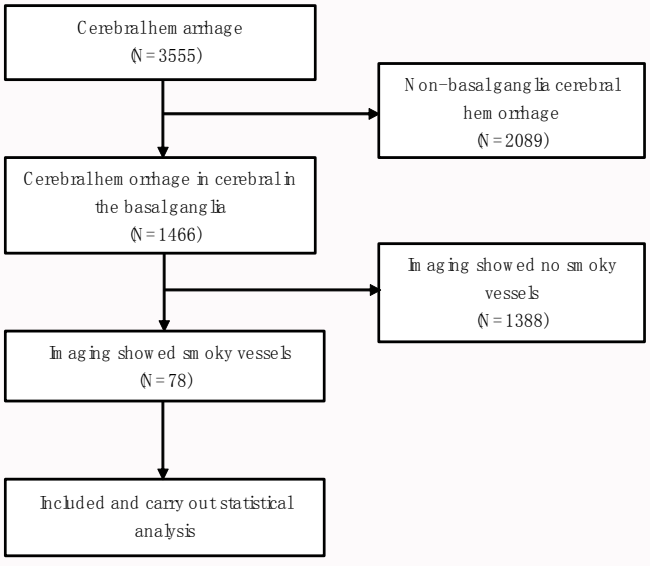

Figure 1: Flowchart of the study.

Surgical results

The patients were divided into the survival (64 cases) and non-survival groups (14 cases) according to survive or not. In the non-survival group, the hematoma broke into the ventricle in the whole group of patients. Between the two groups, logistic regression analysis

Table 2: χ^2 test and logistic regression analysis between survival and non-survival groups.

Variable	χ^2 test		Logistic regression analysis		
	χ^2 value	P	OR	OR 95% CI	P
Sex	0.235	0.628	1.945	0.439-8.630	0.381
Age	1.565	0.211	0.453	0.094-2.170	0.322
The hemorrhage side	0.181	0.67	2.379	0.541-10.458	0.251
Hematoma broke into the ventricle	8.644	0.003	12.382	1.340-114.471	0.027
Rebleeding	5.31	0.021	1.674	0.360-7.787	0.511
State of consciousness	3.791	0.052	0.341	0.071-1.640	0.179
Revascularization	3.393	0.065	8.2	0.707-95.063	0.092

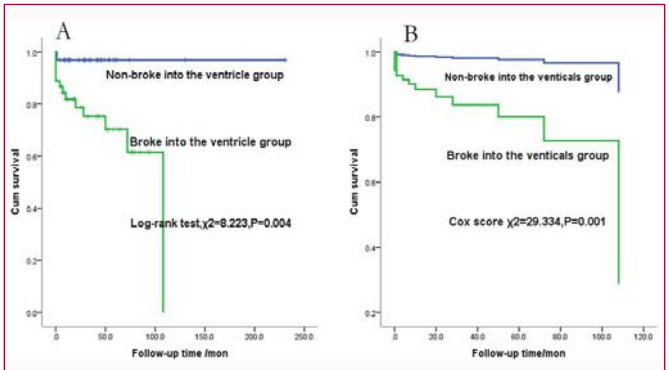

Figure 2: The survival time has a significant difference between hemorrhages broken into the ventricle group and non-broke into the ventricle group.
A: Kaplane-Meier curve (χ^2=8.223, P=0.004); B: Cox regression curve (χ^2=29.334, P=0.001).

showed that hematoma broke into the ventricle (OR=12.382, P=0.021) was a risk factor for death. The statistical analyses are summarized in Table 2. Up to the date of follow-up, we further analyzed the survival time of patients by Kaplane-Meier curve (χ^2=8.223, P=0.004) and Cox (χ^2=29.334, P=0.001) between hematoma broke into the ventricle and non-broke group, significant statistical difference were found by the log-rank test and Cox score (Figure 2).

In this study, 21 patients underwent indirect revascularization after first hemorrhage, including Encephalo Myo Synangiosis (EMS) or Encephalo-Duro-Arterio-Synangiosis (EDAS). The mean age was 39.7 years (range 12-62), and the mean follow-up time was 27.5 months (range 6-53). Noteworthy, 5 of 21 patients underwent combined hematoma removal and EDAS in one surgery. The 5 patients were admitted in our hospital and received head CT scan showing the basal ganglia hemorrhage, and then they were diagnosed as hemorrhagic Moyamoya disease by DSA in preoperative. Intracranial hematoma removal combined with EDAS was performed in the surgery, focusing on protecting the superficial temporal artery and its branches. Intraoperative indocyanine green angiography and DSA in 3 months postoperative showed the vessel all patency, and both clinical manifestation and imaging evidence showed positive effect. The presented patient is a female MMD with hypertension, 43 years, her blood pressure was controlled well with the regularly use of drugs. The imaging data was showed as Figures 3 and 4.

Discussion

Traditionally, it is believed that cerebral hemorrhage in the basal ganglia is hypertensive cerebral hemorrhage, and the cause of Moyamoya Disease (MMD) is ignored. MMD is a chronic occlusive cerebrovascular disorder characterized by bilateral stenosis of the terminal parts of the internal carotid arteries and/or proximal parts of anterior and middle cerebral arteries with the formation of an abnormal vascular network at the base of the brain [6].

DSA is considered to be the criterion standard for diagnosis of MMD and MMS, MRA can diagnose the vascular disease as well when DSA cannot be performed according to diagnostic criteria of MMD [5]. CTA and MRA have been widely accepted for diagnosis and evaluation of MMD and MMS [7], it's reported that CTA was superior to MRA in identification of very small stenotic lesions in the internal carotid artery, middle cerebral artery, and posterior cerebral artery [8]. Therefore, these commonly available modalities can provide clues to make preliminary evaluation and offer the possibility to obtain the early diagnosis of MMD/MMS among basal ganglia hemorrhage. According to the literature [9,10], MMD accounts for 0.3% to 2.24% of cerebral hemorrhage in young people. However, the clinical characteristics and surgical strategies of basal ganglia hemorrhage for MMD have not been reported.

In the study, 78 cases of patents diagnosed with Moyamoya

Figure 3: Preoperative imaging of the patient.
A: CT scan showed hemorrhage in the left basal ganglia with regular morphology; B and **C:** CTA and DSA showed moyamoya vessels in the bottom of skull.

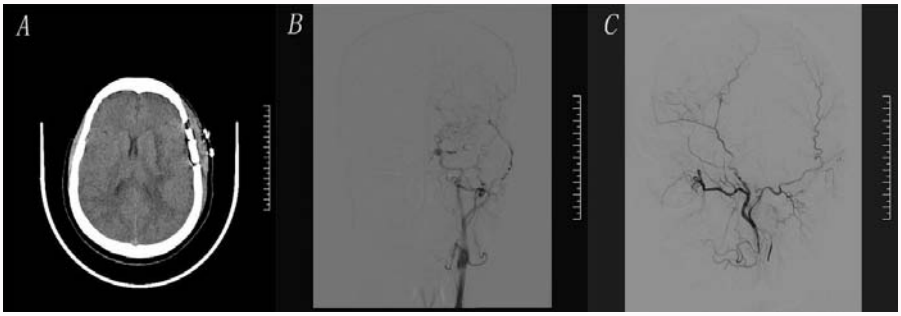

Figure 4: Imaging results in postoperative of the patients underwent combined hematoma clearance and indirect revascularization. A: One week after operation, the head was scanned by CT; B: 6 months after operation, the left side was examined by DSA in coronal position; and C: 6 months after operation, the left side was examined by DSA in sagittal position.

disease or Moyamoya syndrome were enrolled totally, accounting for 5.3% (78/1466) of the initial diagnosis of basal ganglia hemorrhage. Since DSA was not performed routinely for the cerebral hemorrhage patients, the actual proportion is expected to be higher. Clinically, we have found several interesting items as followed. Firstly, the patients were young with the average age 43.7 years and the sub-group age of 40 years to 49 years showed a higher incidence. There was no significant difference in gender; but more hematoma on the right side was observed. Secondly, dizziness and headache were the most common clinical symptoms, usually accompanied by nausea, vomiting, limb movement disorders or language disorders. With the average of 38.8 months, we revealed 17.9% of mortality rate and 26.9% of moderate to severe neurological dysfunction. Thirdly, the imaging data showed that the basal ganglia hemorrhage caused by MMD can easily break into the ventricle. Statistics analysis also found that hematoma broke into the ventricle might be a risk factor for the death. Furthermore, the survival time showed significant difference between broke and non-broke patients. There have also other studies reported that irregular shape of hematoma has been linked to a higher risk of hematoma growth and in turn to a poorer outcome [11,12]. However, a meta-analysis including 2294 patients revealed that its predictive values for hematoma expansion were limited [13]. To date, the epidemiological characteristics of hemorrhagic MMD have gradually been reported [14]. We can see that all of the above findings are basically consistent with the clinical characteristics of hemorrhagic Moyamoya disease. Meanwhile, these clinical manifestations suggested that there were some significant difference of clinical characteristics between the basal ganglia hemorrhage for MMD and hypertension, which can provide reference for clinical manifestation and prognosis.

In hemorrhage MMD, the presence of microaneurysms, as well as an increased blood flow through thin collateral walls during stress, is the probable cause of intracranial hemorrhages [15]. Studies have revealed that rebleeding tends to be common within 5 years or 6 years after the first bleeding for hemorrhagic MMD [14,16]. The proportion of rebleeding up to 21.8% in our study. It suggests that patients who suffer bleeding should receive revascularization surgery as soon as possible. In addition, long-term outcomes demonstrated that surgical treatment of hemorrhagic MMD can improve regional blood flow and have greater efficacy at preventing rebleeding than conservative treatment [17]. Therefore, the diagnosis of MMD in basal ganglia hemorrhage should be taken more seriously. As we all known, evacuation of intracranial hematoma is the main step in the treatment of cerebral hemorrhage especially in hypertensive cerebral hemorrhage. There are different available surgical procedures in clinical practice, such as small bone window craniotomy surgery, directional catheter drainage, neuroendoscopic minimally invasive surgery and so on [18-21]. The most fundamental and chief purpose of various surgical methods is to achieve clearing the hematoma and reducing the intracranial pressure, the superficial temporal artery is cut off throughout the operation. Multicenter studies have reported that revascularization involves Superficial Temporal Artery to Middle Cerebral Artery (STA-MCA) anastomosis, Encephalo-Duro-Arterio-Synangiosis (EDAS) and Encephalo-Duro-Arterio-Myo-Synangiosis (EDAMS) was effective for the hemorrhagic MMD [22-24]. However, it will take a great challenge when the superficial temporal artery is destroyed. Therefore, the superficial temporal artery, as well as the established lateral collateral vascular, should be protected as much as possible in basal ganglia hemorrhage for MMD during the hematoma evacuation. Of course, the premise is that the patients should be diagnosed with MMD. Except EDAS or EDAMS is a convenient and feasible surgical procedure, Encephalo-Myo-Synangiosis (EMS) is also a simple and available surgical method. In brief, the concept of combined hematoma evacuation and indirect revascularization is proposed to guide the surgical procedure. Not only could it achieve the purpose of clearing hematoma, but also reconstructs the intracranial and extracranial blood vessels in one surgery. The surgical strategy avoids the secondary stress of the operation on human body and reduces the cost of treatment, so that patients will benefit more.

This retrospective study had some limitations. First, there were 21 patients with hypertension. Although all the imaging data presented abnormal vascular in the base of the brain, it's really beyond our ability to completely excluded the role of hypertension in basal ganglia hemorrhage. Second, only a few cases underwent hematoma removal combined indirect revascularization in one surgery, and more clinical cases need to be observed.

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