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Clinical Article

Spontaneous Intracranial Hemorrhage in Children : Analysis of Clinical Characteristics

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Objective : Spontaneous intracranial hemorrhage in children is not common and very different compared to adults. We analyze the etiology, hemorrhagic type, clinical features, and outcome of spontaneous intracranial hemorrhage in children.

Methods : Twenty-nine patients under 17years of age with nontraumatic intracranial hemorrhage were analyzed retrospectively. Neonates were excepted. We reviewed their medical records in regard to their age, symptoms, radiologic findings, treatment, and prognosis.

Results : Among 29 patients, there were 17boys and 12girls. The average age was 10.2years. The most common presenting symptom was mental deterioration, and the most common cause was arteriovenous malformation. Spontaneous intracranial hemorrhage in children showed a better prognosis than in adults.

Conclusion : Spontaneous intracranial hemorrhage in children resulted mainly from vascular malformation and the prognosis is relatively good. More careful follow-up studies and active management are needed for better outcomes.

KEY WORDS : Nontraumatic · Spontaneous ICH · Children.

Introduction

In pediatric patients, similar to adult patients, cerebrovascular disease can be classified into two types : ischemic stroke and hemorrhagic stroke. The causes of ischemic stroke are congenital heart disease, hematologic disorder, and so on. The causes of hemorrhagic stroke, on the other hand, have been reported as arteriovenous malformation, cerebral aneurysm, brain tumor, and infection[5,6,10]. Incidence of cerebrovascular disease in children is significantly lower than in adults. The percentage of ischemic infarction in adults is 80~90%[4]. In contrast, the incidence of ischemic infarction in children is comparable to the incidence of hemorrhagic stroke, or more frequently presented as spontaneous intracranial hemorrhage[1,5,17]. In such cases, compared to adults, hemorrhagic disorder is more prevalent in children and the cause is frequently structural disorder. However, clinical studies of spontaneous intracranial hemorrhage in pediatric patients have been rarely reported, particularly in Korea. In addition, despite of the development of radiology, the underlying disease and mechanism of bleeding in spontaneous intracranial hemorrhage in pediatric patients were not characterized in many cases. The analysis of outcomes has also been rare. So, we report the result of analysis about clinical symptoms, the cause, and the outcomes of spontaneous intracranial hemorrhage in children.

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Materials and Methods

Among 2,443 patients with spontaneous intracranial hemorrhage who were treated in the department of neurosurgery at our hospital from 1990 to 2002, 29 patients (1.2%) were examined retrospectively. Neonates younger than 1 month and patients over 17years old were excluded from the analysis. Neonates were excluded from this study because birth trauma could not be ruled out. Also, the neonates' birth weight and the mother's condition could have an influence on the hemorrhage. The diagnosis of intracranial hemorrhage had been confirmed by computed tomography (CT) or magnetic resonance imaging (MRI). In all patients, past history and coagulation disorder were examined. In 19 of 29 patients (65%), hemorrhage or underlying disease had been treated by surgery. In the remaining 10 patients, surgery had not been performed because of the small amount of hemorrhage, hopeless state of the patient, coagulation disorder, or refusal of the guardians. Clinical analysis was performed based on medical records and radiographic images. Age, sex, symptoms at admission, type of intracranial hemorrhage shown in the CT, cause of hemorrhage, therapeutic modalities, and the outcome were

examined. The status of the patients at the final follow-up after discharge was considered as the outcome. The follow-up period ranged from 3 to 42months with an average of 13.9months. Four patients were excluded because they had transferred to another hospital or had expired. The outcome was evaluated according to the Glasgow Outcome Scale (GOS). Good recovery and moderately disabled patients were classified into the good outcome group. Severely disabled, vegetative state, and dead patients were classified into the poor outcome group. Statistical analysis was performed by applying the paired t-test using SPSS version 1.10.

Results

Age and sex

The average age of patients was 10.2years, and the age ranged from 1.5months to 16years. Two patients were under one year old, nine patients were between one and ten years old, and 18 patients were over ten years old. The ratio of female to male patients was 1:1.4. The incidence of spontaneous intracranial hemorrhage was higher in males(Table 1).

Symptoms and signs

On admission, 15 patients (52%) had a mental deterioration, and ten patients had scored lower than eight points on the Glasgow Coma Scale(GCS). Fourteen patients (48%) had Symptoms of increased intracranial pressure such as headache, nausea, or vomiting. Eight patients (28%) had hemiparesis. Three patients (10%) had seizure. One patient had hemianopsia.

Type of hemorrhage

Intracerebral hemorrhage was detected in 20 patients, and five patients had intraventricular hemorrhage concurrently. One patient had subarachnoid and intraventricular hemorrhage concurrently. Five patients had only intraventricular hemorrhage. Four patients had only subarachnoid hemorrhage(Table 1).

Coagulation disorder

Two patients had a history of coagulation disorder. One had vitamin K deficiency. The other had thrombotic thrombocytopenic purpura(TTP). In the vitamin K-deficient patient, disseminated intravascular coagulation(DIC) was also detected.

Cause of hemorrhage

In 22 patients, cerebral angiography was performed. The cause of hemorrhage was arteriovenous malformation in nine cases, cerebral aneurysm in three cases, moyamoya disease in two cases, and cerebral aneurysm with moyamoya disease in one case. In seven patients, abnormal findings were not detected in the cerebral angiogram(Table 2).

MRI was performed in four patients. In one patient, similarly in the cerebral angiogram, arteriovenous malformation was detected. In two patients whose cerebral angiogram was normal, the MRI showed brain tumors.

Cerebral angiography was not performed in seven patients. Among them, one patient had a history of coagulation disorder. In another patient, a cerebral angiogram was not required as he was suspected of having a brain tumor. In the remaining five patients, cerebral angiography was not performed because their guardians refused or the test for assessing the cause could not be performed due to their unimproving condition. In regards to the hemorrhagic type, in six patients with arteriovascular malformation, hemorrhage was detected in the intracerebral region. In three patients, only hemorrhage in the intraventricular space was detected. In patients with aneurysm, hemorrhage in the subarachnoid space was detected in two of them and intracerebral hemorrhage was detected in another two patients.

Table 1. Outcome of spontaneous intracranial hemorrhage in children

	Good outcome*	Poor outcome**	Total	P-value
Age				0.019
≤10	4	7	11	
>10	17	1	18	
Initial GCS				0.032
≤8	2	8	10	
>8	19	0	19	
Hemorrhagic type				0.362
ICH (č/š SAH,IVH)	12	8	20	
SAH only	4	0	4	
IVH only	5	0	5	
Operation				0.573
done	14	5	19	
not done	7	3	10	

*Good recovery & moderative disable in Glasgow outcome scale **Severe disable, vegetable state, and death in Glasgow outcome scale

Table 2. Cause and treatment in pediatric spontaneous intracranial hemorrhage

Cause	Craniotomy	Burr-hole trephination	Conservative*
Aneurysm	3	0	1
AVM	5	2	2
Moyamoya	0	2	0
Tumor	1	0	2
Hematologic disorder	0	2	0
Unknown origin	0	4	5
Total	9	10	10

*conservative teatment without operation

Treatment

Craniotomy with nidus removal was performed in seven of the nine patients with artriovascular malformation. Aneurysmal neck clipping was performed in three patients with aneurysm only. In two patients with moyamoya disease, burr-hole trephination for hematoma evacuation was performed. However additional treatment was not performed due to the refusal of the patients' guardians. In two of seven patients with normal MRI, burr-hole trephination was performed to remove hematoma. In one patients with suspicious brain tumors, craniotomy with mass removal was performed. In the pathologic finding of this patient, primitive neuroectodermal tumor (PNET) was confirmed. In four of seven patients who had not without performed a MRI, burr-hole trephination was performed (Table 1).

Outcome

All patients were evaluated according to the Glasgow outcome scale(GOS). The outcome of 16 patients was classified as good recovery, five patients as moderately disabled, four patients as severely disabled, and four patients had expired. The good outcome group consisted of 21 patients and the poor outcome group consisted of eight patients. In patients under 10years of age, the outcome of four patients was good and seven patients was poor. In patients over 10years of age, the outcome of 17 patients was normal and one patient was poor. Thus, the outcome of patients over 10years was generally good. The difference was statistically significant (p=0.016). In all patients with an initial GCS over 8 points, the outcome was normal, and it was statistically significant (p=0.032). The difference of patients' outcome treated with and without surgery, or with different hemorrhage sites, was not statistically significant (Table 1).

Discussion

In children with combined ischemic and hemorrhagic stroke, the incidence of cerebrovascular disease is estimated to be approximately 2.5~2.7/100,000[5,10,17]. The incidence is on the rise as diagnostic medicine improves. Broderick, et al. reported that in pediatric patients, the incidence of hemorrhagic stroke is higher than that of ischemic hemorrhage[5]. Schoenberg, et al., on the other hand, reported that the incidence of ischemic stroke and hemorrhagic stroke are comparable[17].

Song, et al. reported that the incidence of cerebrovascular disease in adults is higher, 1.48~2.24/1000, and found hemorrhagic disease in 34% of patients[20]. Inagawa, et al. reported that the incidence of hemorrhagic stroke is 47~52/100,000[9]. In regards to the ratio of hemorrhagic stroke to ischemic stroke, Rathore, et al. reported that approximately 15% of cases are hemorrhagic stroke[14]. Song, et al., on the other hand, reported that the incidence is higher, approximately 34%[20]. The reports indicate that although the incidence of cerebrovascular disease in children is lower than in adults, the proportion of hemorrhagic disease is higher. Regarding the ratio of males to females, the incidence is higher in male. Al-Jarallah, et al. reported the mean age as 7.1years[1], and Broderick, et al. reported as 10years[5]. We were unable to estimate the incidence in our study, but we can report that the incidence was higher in males and the mean age was 10.2years.

Clinical symptoms

Al-Jarallah, et al. reported that the most frequent symptoms are signs of increased intracranial pressure such as headache or nausea[1]. Other symptoms include seizure, hemiplegia, and decreased mental state, in order of frequency. Our data showed that 15 patients (52%) showed decreased mental state and 14 patients (48%) showed signs of increased intracranial pressure. Hemiplegia, seizure, hemianopsia followed in order of frequency. In adults, mental deterioration was seen in 50% of patients and headache in 40% of patients. Thus the frequency of symptoms is similar in both groups[4].

Cause of hemorrhage

In children, the cause of spontaneous intracranial hemorrhage is frequently a structural vascular disorder such as arteriovenous malformation, cerebral aneurysm, moyamoya disease, etc. In many cases, however, the cause is hard to identify[10,15,17]. This is different from adults in whom the most frequent cause of cerebral hemorrhage is hypertension and amyloid angiopathy[4].

To characterize the cause of hemorrhage, angiography was performed in 22 patients; among these, MRI was also performed in three patients. In one patient, only MRI was performed. In this study, vascular malformation and brain tumor were confirmed in 18 patients (78%). In many cases, the cause were arteriovenous malformation. Brain tumor was confirmed in three patients.

Ruiz-Sandoval, et al. reported that in patients under 35years old, the cause of cerebral hemorrhage was arteriovenous malformation and hypertension, in that order[16]. This shows that as patients' age increases, hypertension increases. In five patients, abnormalities could not be detected in the study. In such cases, angiographically occult vascular malformation (AOVM), which cannot be detected by current diagnostic tools, may be considered to be a cause. AOVM refers to a cerebrovascular abnormality that is confirmed as a vascular

abnormality histologically but cannot be detected by angiography. AOVM is most prevalent in patients with their 20s and 30s. AOVM is also known to be frequently found in children[7,10,19]. Furthermore, AOVM is well-known to have a tendency to cause hemorrhage[7,13]. In addition, as patients with AOVM have a risk of rebleeding, surgical treatment may be recommended.

In pediatric patients, cerebral aneurysm is reported to be most frequently found in the internal carotid artery. In addition, in comparison with adults, giant, mycotic, traumatic, and multiple aneurysms are more frequent in children[10]. We have detected cerebral aneurysms in four patients, including patients with moyamoya disease. The incidence was not high. However, one case of multiple aneurysms and one case of mycotic aneurysm were detected, excluding combined case with moyamoya disease.

Moyamoya disease is most prevalent in early teenagers, and adults in their 30s and 40s. Moyamoya disease is the most frequent cause of pediatric stroke[11,17]. The tendency of stroke in pediatric patients is primarily ischemic stroke, whereas hemorrhagic stroke in adult patients[11,17]. In our study, in patients diagnosed by angiography, moyamoya disease was identified as the cause of hemorrhage in only three patients (14%), including patients who also had cerebral aneurysm. Among them, moyamoya disease occurred in two children over 15years of age. This shows that the hemorrhage caused by moyamoya disease occurs in relatively old children. The type of hemorrhage was intracerebral and/or intraventricular hematoma. In pediatric patients, moyamoya disease is known to be the primary cause of ischemic stroke. We observed hemorrhage in three moyamoya patients. This shows that even in pediatric cerebral hemorrhagic patients, moyamoya disease cannot be ignored and attention should be paid.

In brain tumor patients, hemorrhage occurs in 5~10% of them, particularly those with metastatic tumors[12,18]. Intracerebral hemorrhage is most frequent. In our study, three patients who were suspected to have brain tumors also had intracerebral hemorrhage. Among them, one patient was diagnosed by a pathologic finding and was confirmed to have a primitive neuroectodermal tumor(PNET). But we could not confirm the pathologic diagnosis in the other patients.

Type of hemorrhage

In adults with intracranial hemorrhage, the incidence of intracerebral hemorrhage was twice more frequent than subarachnoid hemorrhage, particularly in the putamen and the thalamus[2,4,9]. In pediatric patients, intracerebral hemorrhage was most frequently observed. However, hemorrhage in the putamen or thalamus was rare. But intraventricular hemorrhage was relatively high[1,2]. This is due to the fact that the most frequent cause of hemorrhage in adults is hypertension whereas arteriovenous malformationin pediatric patients. In aneurysm patients, subarachnoid hemorrhage is less frequent in children than in adults[8]. Similarly, we had observed that in two of four confirmed aneurysm patients, and intracerebral hemorrhage without subarachnoid hemorrhage was detected. Thus special attention should be paid in such cases.

Outcome

Analyzing the overall outcome, 16 patients (55%) showed good recovery, and five patients (17%) were moderately disabled. Thus 21 patients (72%) were in the good outcome group. The poor outcome group consisted of 8 patients, four of which (14%) were severely disabled and four in expired. In adults, the mortality rate within one month of hemorrhage was reported to be 30~50%. The recovery rate that allows the patient to lead an independent life within one month was low(12~20%)[2,4,9]. In contrast, the outcome of spontaneous intracranial hemorrhage in pediatric patients may be considered to be better than adults. Such discrepancy may be due to the hemorrhage in the putamen and thalamus caused by hypertension in adults.

The benefit of surgical treatment is controversial. Broderick, et al. reported that the mortality rate in patients treated by surgery is low but the outcome is poor[3]. This may not be significant because patients treated by surgery were younger and hemorrhage was more severe. We did not detect any statistically significant differences in the outcomes of patients who were treated by surgery and those who were not. In patients treated with surgery, the operations were not chosen randomly, so the significance was not feasible to confirm.

Conclusion

In pediatric patients, the overall incidence of spontaneous intracranial hemorrhage was lower than in adults, and the incidence of cerebral hemorrhage was higher than that of ischemic stroke. In regards to the causes, structural abnormalities, such as arteriovenous malformation, were more prevalent, which is in contrast to the causes in adults. The outcome was influenced by the state of consciousness on admission and the age of the patient. Overall outcome of pediatric patients was much better than that of adult patients. Taken together, to improve the outcome of spontaneous intracranial hemorrhage in pediatric patients, we recommend analyzing the causes thoroughly and to select the treatment modality appropriately.

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