

Pediatric Hemorrhagic Stroke Complicates Interventions for Congenital Heart Disease: Experiences from Two Centers

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To the Editor: Pediatric stroke refers to the rupture and/or occlusion of arteries or veins. It is one of the highest causes of pediatric preterm death and long-term disability. Health-care providers find dealing with pediatric stroke a challenge because of the difficult diagnosis, the lack of standard guidelines, the rapid progress, and the high mortality. In this study, we observed and analyzed the characteristics of six pediatric hemorrhage strokes complicated with interventions for congenital heart disease (CHD). We found that vascular malformation, hemodynamic fluctuation, and postoperative anticoagulant medicines were risk factors for this condition and that there was an urgent need for awareness and acknowledgment.

The case records of six children who had been discharged from Guangdong General Hospital (Guangzhou) and Taihe Hospital (Shiyan) between December 2007 and June 2017 and with a diagnosis of a postoperative hemorrhagic stroke were obtained and reviewed. Written informed consent was obtained from the patients or the children's parents. The study protocol was conducted in accordance with the *Helsinki Declaration* and was approved by the Institutional Review Board of Guangdong General Hospital.

Six children were diagnosed with hemorrhage stroke after an interventional procedure for CHD: five cases with congenital ventricular septal defect (VSD) and one case with aortic stenosis (AS). The incidence of hemorrhage stroke was 0.055%. The basic clinical characteristics are summarized in Table 1.

Five patients underwent transcatheter VSD occlusion, in which four patients had successful device placement and one patient accepted percutaneous balloon aortic valvuloplasty. In brief, all the patients accepted heart catheterization and cardiac angiography with the aim of defining the characteristics of cardiac lesions. An arterial-defect-vein loop or transfemoral artery approach was used, and a delivery system was created. Available devices or inflatable balloons were accurately positioned.

During the procedure, two patients (one VSD and one AS) were found to have bradycardia. They accepted thoracotomy immediately, but no cardiac injury was found. Three patients presented with coma, irritability, and powerless limbs after recovery from anesthesia. One child suffered a sudden headache on the 6th day postdischarge.

The time from the appearance of symptoms, such as headache, comatose, and bradycardia, to a clear diagnosis of neural hemorrhage was 10.5 h (5–22 h). All patients received multidisciplinary synthetic therapy. The etiology was clear and hematoma removal was performed in two cases, one with a middle cerebral artery M4 aneurysm and another with an arteriovenous fistula of the cervical spinal cord. Regarding the prognosis, only one patient with a mild subarachnoid hemorrhage was cured; three patients recovered but with permanent nervous dysfunction, one patient died from multiple organ failure, and one denied ongoing therapy.

The morbidity of pediatric stroke is 0.13 and 11.3 per 10,000 per year, and hemorrhagic stroke accounts for 30–45%. It is one of the predominant reasons for pediatric death and has a mortality as high as 34%.^[1] The etiological factors, such as vascular malformation, brain trauma, and cardiac disease, are different from adults. In addition, cerebral vascular malformation, such as arteriovenous malformations, cavernous malformation, and aneurysms, is the most common cause, occurring in approximately 40–90% of pediatric hemorrhagic stroke.^[2] The prognosis of pediatric stroke is poorer than with adults.

CHD is an independent risk factor for pediatric stroke. Previous studies reported that the risk of pediatric stroke with CHD is 19-fold higher than the normal.^[3] Among children with ischemic stroke, CHD is present in 25–30%. The incidence of pediatric ischemic stroke in children with CHD who have undergone surgery is 0.54%.^[4] The risk factors associated with CHD for stroke are hypoxemia, hyperhemoglobinemia, and paradoxical embolus. However, previous studies have focused mainly on ischemic stroke; the data on hemorrhage stroke are scarce.^[3] It is commonly believed that patients with CHD have an increased likelihood of peripheral vascular malformation. We found six cases

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Table 1: Clinical data of six cases of hemorrhagic stroke after interventional treatment for CHD

Items	Case 1	Case 2	Case 3	Case 4	Case 5	Case 6
Gender	Female	Male	Male	Female	Male	Male
Age (months)	61	19	6	40	26	37
Height (cm)	110	79	71	87	80	96
Weight (kg)	16	10	8	14	10	16
Diagnosis	mVSD	mVSD	AS	mVSD	mVSD	mVSD
Size (mm)	3.0	4.2	Severe	3.5	3.2	4.0
Operation	A	A, C	B, C	A	A	A
Implantation	Lifetech.VSD occluder (6 mm)	No	No	Huayi.VSD occluder (5 mm)	Lifetech.VSD occluder (5 mm)	Lifetech.VSD occluder (6 mm)
Location	B, C	A, B, C	A, B, C	B, C	B, C	B, D
Treatment	A	A, B	A, B	A, B	A, B, C	A, B, C
Prognosis	Cured	Abandoned	Died	Recovered	Recovered	Recovered

Operation - A: Percutaneous transcatheter occlusion of mVSD; B: PBAV; C: Emergency thoracotomy; Location of hematoma - A: Intracerebral hemorrhage; B: SAH; C: Intraventricular hemorrhage; D: Intraspinal hemorrhage; Treatment - A: Medical treatment; B: Advanced life support; C: Evacuation of hematoma; CHD: Congenital heart disease; PBAV: Percutaneous balloon aortic valvuloplasty; SAH: Subarachnoid hemorrhage; VSD: Ventricular septal defect; AS: Aortic stenosis; mVSD: Muscular VSD.

of hemorrhagic stroke secondary to intervention for CHD and two patients with a clear diagnosis of neurovascular malformation. Therefore, we speculate that CNS vascular malformation combined with hemodynamic fluctuation and anticoagulant medicine may be the cause of hemorrhage stroke. However, the diagnosis of vascular malformations, such as an underlying aneurysm and arteriovenous fistula, is very difficult before surgery.

Strokes associated with CHD always occur during the perioperative period, and a cardiac procedure is an important predisposing factor for a stroke. In this study, one case of hemorrhagic stroke occurred on the 6th day postprocedure and five cases happened during the operation. Interestingly, there was no hemorrhagic stroke secondary to the transcatheter occlusion for atrial septal defect or patent ductus arteriosus at our centers. We attributed this phenomenon to the fluctuation of the hemodynamics during the procedure and the complexity of the operation.

A definitive diagnosis of pediatric stroke is always delayed. The delay at our centers was also significant. The reasons for the delayed diagnosis of stroke are as follows: first, anesthesia may cover the early symptoms of stroke. Second, children are too young to describe the symptoms of a stroke. Third, a lack of knowledge of this rare critical complication is the most important. What is worse is that the lack of standard guidelines for pediatric stroke worsens an already complicated situation.^[5] Given the present difficulties, hemorrhagic stroke after procedures for CHD should attract more attention and needs more research because there were limited cases in this study.

Pediatric stroke is an unpredictable and life-threatening complication of interventional therapy for CHD. Early and accurate diagnosis is the precondition for improving prognosis, and more attention, acknowledgment, and recognition should be given to this fatal complication.

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Conflicts of interest

There are no conflicts of interest.

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