Intracranial vasospasm with subsequent stroke after traumatic subarachnoid hemorrhage in a 22-month-old child

Case report

Brian V. Nahed, M.D., Manuel Ferreira Jr., M.D., Ph.D., Matthew R. Naunheim, A.B., Kristopher T. Kahle, M.D., Ph.D., Mark R. Proctor, M.D., and Edward R. Smith, M.D.

Department of Neurosurgery, Massachusetts General Hospital; and Department of Neurosurgery, Children’s Hospital Boston, Harvard Medical School, Boston, Massachusetts

Clinical and radiographic evidence of subarachnoid hemorrhage (SAH)-related vasospasm is rare in children and has not been reported in infants. In this report the authors present the case of a 22-month-old child who developed clinically symptomatic, radiographically identifiable vasospasm after traumatic SAH. To the authors’ knowledge, this is the first report of vasospasm associated with SAH in a child this young. This 22-month-old boy fell and had a dense SAH. He had a history of surgically corrected craniosynostosis and nonsymptomatic ventriculomegaly. The boy was evaluated for occult vascular lesions using imaging; none were found and normal vessel caliber was noted. Ten days later, the child developed left-sided weakness and a right middle cerebral artery infarct was identified. Evaluation disclosed significant intracranial vasospasm. This diagnosis was supported by findings on CT angiography, transcranial Doppler ultrasonography, MR imaging, and conventional angiography. The child was treated using intraarterial verapamil with a good result, as well as with conventional intensive care measures to reduce vasospasm. This report documents the first known case of intracranial vasospasm with stroke after SAH in a patient under the age of 2 years. This finding is important because it demonstrates that the entity of SAH-associated vasospasm can affect the very young, widening the spectrum of ages susceptible to this condition. This case is also important because it demonstrates that even very young children can respond to conventional therapeutic interventions such as intraarterial verapamil. Thus, clinicians need to be alert to the possibility of vasospasm as a potential diagnosis when evaluating young children with SAH. (DOI: 10.3171/2008.12.PEDS08206)

Key Words • angiogram • stroke • subarachnoid hemorrhage • traumatic brain injury • vasospasm

It is that vasospasm has been reported. No cases of vasospasm in infants have been reported to date. The youngest patient to suffer from posthemorrhagic vasospasm noted in the literature was 9 years of age. It is unclear whether this is a function of the rarity of aneurysmal SAH in children or an intrinsic biological property of developing vessels. It is also possible that vasospasm in children is not diagnosed because it is not often considered in this age group and therefore not as thoroughly investigated using diagnostic studies as in adults.

Although aneurysmal SAH in children is rare, traumatic SAH is often found on imaging studies of the 475,000 children who suffer traumatic brain injuries annually in the US. Given the large number of children with traumatic SAH, it would be useful to know if children are capable of developing traumatic SAH-related vasospasm and if conventional treatments are effective.
in this age group. Awareness of the possibility of vasospasm is important because if present and left untreated, cerebral ischemia and stroke may occur. Triple-H therapy (hypertension, hypervolemia, and hemodilution) and intraarterial local delivery of vasodilatory agents (such as verapamil) have been used successfully in adults, but the efficacy of these treatments for vasospasm in children remains largely unknown.

In this report, we describe the youngest case of clinical vasospasm resulting from traumatic SAH and describe the effectiveness of the administration of antivasospasm therapies in an infant.

Case Report

History and Presentation. This 22-month-old boy sustained a fall from a high chair, subsequently losing consciousness for 20 seconds. When the patient awoke, he was crying and irritable but neurologically intact. The child had a medical history significant for Cri du Chat syndrome (partial deletion of chromosome 5) and surgical correction of metopic synostosis. The patient was also known to have asymptomatic ventriculomegaly.

Examination and Treatment. At presentation, the patient was awake and alert with normal neurological examination results. Strength in all of his limbs was full and symmetric. Computed tomography imaging of the head was performed, revealing dense SAH (Fig. 1). A CT angiogram was then performed to exclude the presence of vascular lesions. The initial CT angiogram was unremarkable, with no vascular lesions noted and no vasospasm identified.

Given the degree of hemorrhage, the patient was initially admitted to the intensive care unit for observation. After several days with stable examination results, the child was transferred out of the intensive care unit.

The family of the patient had come to our institution from a small island country without access to neurosurgical care; because of the combination of the injury and the distance traveled by the family, the child remained in the hospital for an extended period of time. His examination results remained unremarkable until posthemorrhage Day 10, when he was noted to have a new onset of left-sided weakness involving the face, arm, and leg. The patient underwent a CT scan of the head that revealed new hypodensity in the right MCA distribution, suggestive of stroke (Fig. 2). Magnetic resonance imaging confirmed a right MCA distribution infarct. In addition, MR angiography was performed, revealing markedly decreased vessel caliber of both MCAs, consistent with vasospasm (Fig. 3). The patient was also found to have experienced worsening of his ventriculomegaly and an external ventricular drain was inserted on an emergency basis. The findings indicating vasospasm were further substantiated by the finding of elevated velocities using TCD ultrasonography.

The patient underwent a conventional digital subtraction angiogram for further diagnosis and treatment. The angiogram corroborated the significant vasospasm noted previously on MR and CT angiography (Figs. 3 and 4). The child was treated using 1 mg of intraarterial verapamil injected bilaterally into the ICAs (resulting in a total dose administration of 2 mg), which resulted in dilation of the previously narrowed vessels (Fig. 5). The patient tolerated the angiography and intraarterial treatment without complication and was transferred into the intensive care unit in which triple-H therapy was initiated. Another head CT angiogram obtained 4 days after treatment demonstrated resolution of vasospasm (Fig. 4). Neurologically, the patient’s left hemiparesis improved after intraarterial treatment and continued to improve throughout his hospital stay, although worsening hydrocephalus necessitated the insertion of a ventriculoperitoneal shunt. The boy was discharged with closely monitored follow-up and outpatient physical therapy to assist with the rehabilitation of his left arm and leg strength.

Discussion

Subarachnoid hemorrhage is classified as either traumatic or spontaneous (including aneurysmal) in origin,
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and is estimated to have an incidence of 6–15 per 100,000 person-years. Traumatic brain injury often results in traumatic SAH and remains one of the leading causes of death for children between the ages of 1 and 14 years old. Vasospasm is a leading cause of morbidity and death following SAH, usually developing 5–15 days posthemorrhage. Vasospasm after SAH, however, is much less common in children than in adults. The youngest among these rare cases were reported by Aryan et al. (a 9 year old) and by Sert et al. (a 12 year old). Danchaivijitr and colleagues noted radiological features suggestive of vasospasm in a 7 month old with postinfectious SAH, but imaging results were inconclusive and intraarterial therapy was not instituted.

There is a strong correlation between vasospasm onset and the amount of subarachnoid blood in the basal cisterns. Pasqualin and associates reported that 51% of adult patients with aneurysms and a thick cisternal blood layer on CT develop vasospasm, whereas only 2% of patients with a thin or absent cisternal blood layer suffered similar complications. Interestingly, the authors noted that thick cisternal blood layers were rare in CT analysis of children posthemorrhage, which has been suggested as a potential explanation for the decreased incidence of vasospasm in children.

The mechanism of vasospasm following SAH remains incompletely understood. Aberrant mechanisms of smooth muscle contraction, immune-mediated responses, and inflammatory reactions have all been implicated in the onset of vasospasm. Once initiated, vascular cell proliferation and vessel wall thickening may contribute to progression of

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Fig. 3. Magnetic resonance images (upper) reveal restricted diffusion in the right MCA distribution indicative of infarct, whereas MR angiography (lower) shows focal vasospasm of the right and left M1 branch of the MCA (arrows).

Fig. 4. Computed tomography angiograms of the patient reveal vasospasm at the junction (arrow) of the right ICA and the M1 branch of the MCA (left), and resolution of vasospasm and return to normal vessel caliber (arrow) after treatment with intraarterial verapamil (right).

Fig. 5. Conventional angiograms of right ICA injection demonstrating vasospasm of the M1 branch of the MCA and distal carotid artery (left), and dilation of the M1 branch of the MCA and the A1 branch of the anterior cerebral artery after intraarterial delivery of verapamil (right).
the process. Regardless of the mechanism, vasospasm can cause cerebral ischemia, making identification and treatment of this condition imperative to prevent stroke. For patients with SAH, various methods of screening for vasospasm can be used including CT angiography, conventional angiography, and TCD ultrasonography. Transcranial Doppler ultrasonography is commonly used in the US after aneurysmal SAH to screen for vasospasm. These studies are noninvasive and have been shown to be reliable in detecting vasospasm with elevated flow velocities suggestive of the presence of vasospasm.

Once identified, there are a number of potential therapeutic treatments for vasospasm. Triple-H therapy is widely used, although its clinical efficacy, particularly hypervolemia, has been assumed rather than proven. Magnesium, statins, endothelin antagonists, and intrathecal fibrinolytic therapy have shown promise in animal models but remain largely investigational. A recent review of randomized controlled trials of vasospasm treatment identified intraarterial delivery of calcium channel blockers, such as nimodipine and verapamil, as the treatment with the strongest evidence supporting efficacy. Unfortunately, few studies have examined SAH-induced vasospasm and its treatment in children.

Our patient, who developed vasospasm after traumatic SAH, is notable for his young age (22 months) and his response to intraarterial therapy. Interestingly, the patient developed vasospasm during the expected time interval in adults (5–15 days posthemorrhage) and the radiographic data confirmed our clinical suspicions. Such findings are rare in children and this is the youngest documented case of SAH-induced vasospasm. Furthermore, administration of 1 mg of intraarterial verapamil, a calcium channel blocker, visibly improved vasospasm on subsequent angiography. This therapy, although conventional in adults, is novel in children.

It is possible that our patient is unique and his physiological responses may not be generalized to the pediatric population. He does have a chromosomal abnormality and Cri du Chat syndrome, the effects of which are completely unknown in this cascade. In addition, he had undergone a prior craniotomy for frontoorbital advancement that increased his intracranial volume, giving him an increase in extraxial space and perhaps making his brain more susceptible to rotational forces and injuries.

The role of hydrocephalus and increased intracranial pressure is also unclear. It is possible that vasospasm, in and of itself, would be necessary but not sufficient to produce symptomatic ischemia or an infarct in a pediatric patient and this is why the incidence of symptomatic vasospasm is very low in children. However, the combination of vasospasm and hydrocephalus is likely a more potent cause of ischemic complications.

We report the youngest case of intracranial vasospasm with stroke after traumatic SAH in the pediatric literature. This case illustrates that SAH-associated vasospasm can affect very young children and therefore should be included in the list of differential diagnoses by pediatricians, neurologists, and neurosurgeons. We have also demonstrated that vasospasm in a very young child responds to intraarterial verapamil, a treatment used effectively in adults for SAH-associated vasospasm. Although this is likely a rare phenomenon, we hope to raise awareness that infants can develop symptomatic vasospasm after traumatic SAH and we call for further investigation into the use of intraarterial therapies in children with vasospasm.

Disclaimer
The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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Address correspondence to: Edward R. Smith, M.D., Department of Neurosurgery, Children’s Hospital Boston, Harvard Medical School, 300 Longwood Avenue, Boston, Massachusetts 02115. email: edward.smith@childrens.harvard.edu.