Spontaneous healing and complete disappearance of a ruptured posterior inferior cerebellar artery dissecting aneurysm

Case report

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A 7-month-old baby presented with a 4-day history of drowsiness and vomiting after a falling accident. Magnetic resonance imaging demonstrated diffuse subarachnoid hemorrhage, intraventricular hemorrhage, and variable stages of subdural hematoma in bilateral occipital and left temporal subdural spaces. A partially thrombosed aneurysm was noted in the right craniocervical junction. Ophthalmological examination revealed bilateral retinal petechial hemorrhages. Conventional cerebral angiography revealed a dissecting aneurysm in the right posterior inferior cerebellar artery (PICA). Endovascular embolization was suggested, but the family refused. After conservative treatment, follow-up MRI revealed that the PICA aneurysm had remodeled and ultimately disappeared completely at the 10th month. This case illustrates the relatively plastic nature of intracranial aneurysms in pediatric patients. More studies are necessary to clarify the natural history of spontaneously thrombosed aneurysms to assist in their overall management. (http://thejns.org/doi/abs/10.3171/2014.1.PEDS13412)

Key Words • dissecting aneurysm • thrombosis • trauma • posterior inferior cerebellar artery • vascular disorders

The incidence and clinical presentation of intracranial aneurysms in the 1st year of life remain unknown, although published reports have shown that aneurysms in the pediatric age group are different from those in their adult counterparts with respect to clinical presentation, location, morphology, and outcome.1,4,6,9 Elgamal and colleagues reported male predominance, more seizure attacks at presentation, preferential location in the middle cerebral artery, and better outcome in infantile intracranial aneurysms.6 The incidence of the posterior inferior cerebellar artery (PICA) aneurysm in infantile intracranial aneurysms is rare. In the report by Elgamal et al.,5 aneurysms (5.9%) were located in the PICA.6 Buis and colleagues noted an incidence of traumatic aneurysms among 131 intracranial aneurysms in children younger than 1 year. The anterior cerebral artery was the most common location, and only one aneurysm was located in the PICA. Although spontaneous thrombosis and repair of intracranial aneurysms in children are not rare,3,9 the exact mechanisms and factors responsible for them remain elusive. Moreover, spontaneous healing and complete disappearance of an aneurysm are very unusual.10

Herein, we describe a 7-month-old baby with a PICA dissecting aneurysm that spontaneously healed and disappeared completely on follow-up images. The possible etiology and treatment strategy are discussed.

Case Report

History and Examination. A 7-month-old baby was sent to our emergency room because she was sleepy and vomited repeatedly after feeding. Her father claimed that she accidentally fell off a sofa 4 days earlier. On the child’s arrival, physical examination revealed a bruise over the occipital scalp and sluggishly reactive pupils. The anterior fontanel was not bulging. Laboratory examination demonstrated no bleeding tendency. A brain CT scan showed diffuse subarachnoid hemorrhage (SAH), intraventricular hemorrhage (IVH), and dilation of the ventricular system. There was a hyperdense lesion in the right craniocervical junction. Two days later, MRI revealed diffuse SAH, IVH, and variable stages of subdural hematoma (SDH) in bilateral occipital and left temporal subdural spaces. A partially thrombosed aneurysm was noted in the right craniocervical junction (Fig. 1). Ophthalmological examination revealed bilateral retinal petechial hemorrhages. No other obvious wound or bruise...
was observed. Subsequent cerebral angiography revealed a characteristic dissecting aneurysm in the proximal segment of the right PICA (Fig. 2). Diffuse vasospasm was also noted in bilateral internal carotid artery territories. Endovascular embolization of the right PICA was suggested, but the family refused.

Treatment. Two weeks after admission, the patient underwent placement of a ventriculoperitoneal shunt because she became drowsier and a CT scan showed marked hydrocephalus.

Posttreatment Course. Gradual improvement in the patient’s clinical condition was observed. She was discharged uneventfully 3 weeks after shunt insertion. One month after discharge, follow-up MRI demonstrated increased size and partial thrombosis of the right PICA dissecting aneurysm (Fig. 3). Compression of the cervicomедullary junction had become more severe. Further angiographic study with possible endovascular embolization was strongly recommended at that time, but the family refused again. A third MRI study performed 4 months after the first study revealed regression of the aneurysm size and decreased mass effect on the right cervicomедullary junction. Ten months later, a fourth MRI study demonstrated complete disappearance of the right PICA dissecting aneurysm (Fig. 4). During the past 7 years of follow-up, the child was neurologically intact but had some developmental delay.

Discussion

Subdural and retinal hemorrhages are markers of shaken baby syndrome. Because of the reported accident and the constellation of bilateral SDH in various stages, IVH, and retinal hemorrhage, shaken baby syndrome was highly suspected in our case. However, repeated rupture of an intracranial aneurysm could be an alternative etiology, because retinal hemorrhage associated with SAH is common in patients with ruptured intracranial aneurysms.

Fig. 1. Axial T1-weighted (A–C) and T2-weighted (D–F) MR images at corresponding levels demonstrating the variable stages of SDHs (white arrowheads) over bilateral occipital and left temporal regions and the IVH (black arrowheads). An aneurysm (white arrows) can also be seen in the right craniocervical junction.

Fig. 2. Conventional angiograms with the catheter placed in the brachiocephalic artery, Towne’s projection (left) and oblique projection (right), revealing a characteristic dissecting aneurysm (arrows) in the right PICA.
The pathogenesis of the PICA dissecting aneurysm in our case is an interesting issue. Multiple intrinsic and environmental factors are associated with aneurysm formation, such as congenital, degenerative, infectious, or traumatic origins, which can affect structural changes of cerebral vessels and hemodynamic stress. In our case, definite evidence of infectious origin was not found. Although no congenital disorder was noted in our patient, the PICA aneurysm could be congenital and could have ruptured repeatedly, causing the falling accident and the presenting neuroradiographic findings. Alternatively, the aneurysm could have developed from the falling accident. If so, this case serves as a useful reminder that intracranial vessels are at risk for injury from head trauma and should be routinely included in the imaging studies.

Spontaneous thrombosis and repair of intracranial aneurysms in children are not rare. However, our case is unique because the aneurysm not only thrombosed spontaneously, but also remodeled and disappeared completely on serial images. The exact mechanisms and factors responsible for spontaneous thrombosis of the intracranial aneurysm have not been fully elucidated. Black and German demonstrated that the volume/orifice ratio of the aneurysm is the major contributing factor to the balance between thrombogenesis and thrombolysis. In aneurysms with a relatively small neck, intraluminal thrombosis can occur. Antifibrinolytic agents have been related to spontaneous thrombosis of ruptured aneurysms. Local inhibition of plasminogen activators in and around the aneurysm wall can cause spontaneous aneurysm thrombosis during treatment with antifibrinolytic drugs. Schubiger and colleagues thought that a congenital defect in the tunica media of the vessel results in a change in hemodynamics and that turbulent flow causes clotting and deposition of onion-skin thrombotic layers. These intraluminal clots can reduce or, in extreme cases, completely obliterate the lumen of the aneurysm. Lasjaunias and colleagues thought the arterial wall should be considered a complex organ with a multitude of pathways leading to aneurysm formation and repair. These mechanisms of vessel wall injury and repair in turn do not occur in isolation from the immediate perivascular environment. Rather, luminal and abluminal relationships should be seen as important biological, not just physical, factors in intracranial aneurysm vasculopathies. Although the exact factors causing spontaneous healing and complete disappearance of the dissecting PICA aneurysm are unknown, our case illustrates the relatively plastic nature of intracranial aneurysms in pediatric patients.

No consensus exists regarding the management of intracranial aneurysm with spontaneous thrombosis at such a young age. Intraaneurysmal thrombosis does not guarantee protection against further bleeding. Moreover, the thrombosed aneurysm can recanalize and rupture. Because of the risk of catastrophic bleeding, aggressive treatment for a partially thrombosed aneurysm should be administered via a surgical or endovascular approach whenever possible. However, conservative treatment with close surveillance is an alternative choice in cases in which there is a high risk for aggressive treatment, or if the patient is asymptomatic and serial images show progressive intraneuralysmal thrombosis.

Conclusions

In summary, we describe a baby with a PICA dissecting aneurysm that spontaneously healed and disappeared completely on follow-up images. This case illustrates the relatively plastic nature of intracranial aneurysms in pediatric patients. More studies are necessary to clarify the natural history of spontaneously thrombosed aneurysms to assist in their overall management.

Disclosure

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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