

Intracranial arterial aneurysm due to birth trauma

Case report

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✓ The authors present what is believed to be the first description of an intracranial arterial aneurysm attributable to birth trauma. A male neonate, the product of a precipitous, instrumented, footling breech delivery, exhibited seizures at the age of 18 hours. A computerized tomography scan of the head showed hemorrhage along the tentorium with a globular component at the incisura. Transfontanel Doppler ultrasound examination detected pulsatile arterial flow within the globular mass. Cerebral angiography demonstrated a 1.5-cm saccular aneurysm arising from a small distal branch of the superior cerebellar artery. The pathogenesis of aneurysms in children is obscure and controversial. Birth trauma may be responsible for some pediatric aneurysms that are currently classified as idiopathic or congenital, particularly aneurysms in the region of the tentorial incisura.

KEY WORDS • birth injury • traumatic aneurysm • neonate • tentorial incisura

INFANTS and children are subject to the development of infectious and traumatic intracranial aneurysms just like adults, and, similar to adults, they occasionally exhibit aneurysms with an apparently hemodynamic pathogenesis in association with conditions such as arteriovenous malformations and moyamoya syndrome. Nevertheless, most pediatric intracranial arterial aneurysms are unexplained, and the etiology of these lesions remains controversial. Despite speculation that certain pediatric aneurysms may be attributable to birth trauma,^{27,36,41} a review of the literature discloses no previous examples of this phenomenon.

Case Report

This infant was born to a 36-year-old woman after an unremarkable full-term pregnancy. Presentation was by footling breech, the delivery was precipitous, and the after-coming head was delivered with forceps. The Apgar scores were 8 and 9 at 1 and 10 minutes, respectively. The infant was well until the age of 18 hours when opisthotonic posturing was observed, followed shortly by two tonic-clonic seizures. A computerized tomography (CT) scan of the head demonstrated hemorrhage layered over the surface of the tentorium with a discrete globular hematoma at the edge of the incisura in the ambient cistern on the left (Fig. 1).

At the time of transfer, the patient appeared well. The head circumference was 37 cm and the fontanel was slack. The lambdoid sutures were overriding. Transfontanel Doppler ultrasound examination of the head demonstrated pulsatile arterial flow at the center of the globular incisural mass (Fig. 2); there was no hydrocephalus. At the age of 5 days, a cerebral angiogram was performed. The left vertebral artery injection demonstrated a 1.5-cm saccular aneurysm arising from a small branch of the left superior cerebellar artery (Fig. 3). Conservative management was recommended, and the patient was discharged home at the age of 6 days.

Repeat angiography at 40 days of age was normal. The aneurysm was not visualized. Repeat transfontanel Doppler ultrasound examination was also normal. Pulsatile arterial flow was no longer detected at the site of the hematoma, and there was no hydrocephalus. The patient remained well at 6 months of age.

Discussion

Intracranial Hemorrhage in the Neonate

Intracranial hemorrhage has long been recognized as a major source of morbidity and mortality in the neonate. In the modern era the most common form of neonatal intracranial hemorrhage is the germinal matrix hemorrhage of extreme prematurity; however, in



FIG. 1. Computerized tomography scan of the head without intravenous contrast enhancement showing fresh hemorrhage layered over the surface of the tentorium. At the edge of the tentorial incisura on the left is a discrete globular hematoma with central low density (arrow).

the first part of this century birth trauma was the predominant etiology, and the mechanisms and gross pathology of traumatic neonatal intracranial hemorrhage received intensive examination.^{5,6,8,11,14,15,17,28} Births with vertex or breech presentation subject the head to compressive fronto-occipital foreshortening during passage through the birth canal. Actual flexion of the skull base about a transverse axis through the spheno-occipital synchondrosis has been demonstrated.²⁸ Because the contents of the cranial cavity are non-

compressible, the volume of the cranial cavity must remain constant and, as the distance from the nasion to theinion diminishes, the distance from the skull base to the vertex must increase.^{15,17,28} The tentorium and the posterior portion of the falx resist this deformation and are placed under tension. Abrupt fronto-occipital compression in the course of a precipitous delivery may cause laceration of the falx or the free edge of the tentorium. Rupture of the great vein of Galen and its tributaries has been observed in this setting.^{5,6,8,11,15,17}

Neonatal hemorrhage in the region of the tentorial incisura has generally been considered to be venous in origin.^{5,6,8,15,17,45} We have found only two examples of intracranial arterial hemorrhage related to birth trauma, and neither of these cases involved the incisura.^{22,35} Nevertheless, there are at least two plausible mechanisms for arterial hemorrhage at this location during the birth process: 1) the deformation to which the region of the incisura is subject during the birth process might lead to forcible apposition of the posterior cerebral artery against the sharp, hard free edge of the tentorium; 2) as the superior cerebellar and posterior cerebral arteries both give off small branches that pass through the perimesencephalic cistern to irrigate the free edge of the tentorium,³⁰ severe deformation of the tentorium might rupture or avulse one of these branches.

Aneurysms in Infancy and Childhood

Compelling circumstantial evidence suggests that the aneurysm in the case reported here was of traumatic etiology. Breech presentation, precipitous delivery, and the use of forceps are all risk factors for neonatal intracranial hemorrhage that suggest that sudden fronto-occipital compression of the head occurred dur-

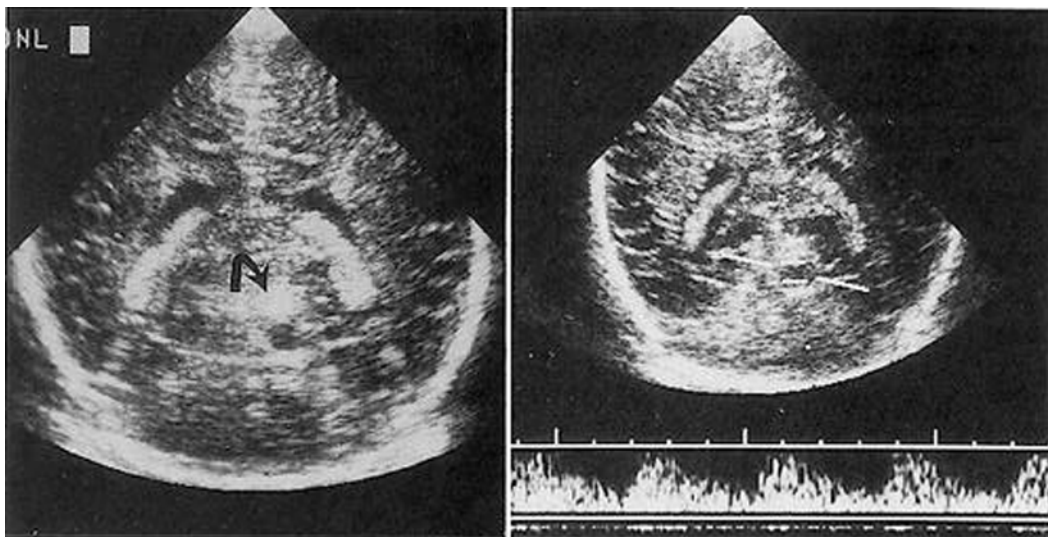


FIG. 2. *Left:* Transfontanel ultrasound study in a coronal plane demonstrating an echogenic mass (arrow) with a hypoechoic center at the level of the tentorial incisura. *Right:* The Doppler ultrasound cursor has been positioned overlying the hypoechoic center of the incisural mass. A pulsatile blood velocity signal is recorded below.

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ing the birth process in this case. The CT appearance of the hematoma was consistent with hemorrhage during delivery or at some time during the first 18 hours after birth. The location of the aneurysm, although not characteristic of traumatic aneurysms in older age groups, was at a site critically affected by the deformations of intracranial structures associated with fronto-occipital compression. We believe that fronto-occipital compression of the head in the course of a precipitous delivery caused stretching of the tentorium with injury of a small distal branch of the superior cerebellar artery. Whether the aneurysm that developed was a true or a false aneurysm could not be determined because thrombosis and healing of the lesion occurred in the interval between angiograms and no operation was performed. Other explanations seem very improbable. The situation of the aneurysm at the end of a small vessel distal to the circle of Willis argues against a degenerative, hemodynamic pathogenesis. The clinical setting and the patient's complete recovery without specific treatment allow dismissal of an infectious etiology. Although birth trauma as a cause for intracranial arterial aneurysm has not been described before, there is no other plausible explanation for the observations in this case.

Intracranial arterial aneurysms in the neonatal period are rare occurrences. Defining the neonatal period as the first 4 weeks after birth, we have identified only 13 other cases in the literature.^{4,10,19,20,23,24,29,34,39,43,44,47} The aneurysms were located on the middle cerebral artery in six cases, on the internal carotid artery in two cases, on the posterior cerebral artery in two cases, on the posterior inferior cerebellar artery in one case, and at the basilar bifurcation in one case; in one case the precise location was not described. In none of these reports was the lesion suspected to be mycotic, and no patient had a systemic disease associated with the development of intracranial arterial aneurysms. Bremer⁴ described a basilar bifurcation aneurysm in a 42-mm fetus, and Wierdis, *et al.*,⁴⁷ described a middle cerebral

artery aneurysm in a stillborn infant. These cases aside, our patient had the earliest reported presentation of aneurysm. No other patient had a history of difficult delivery, and in no case was the possibility of a traumatic etiology entertained. Thus, in every other case the development of the aneurysm was believed to be prenatal, and only in the case of Wierdis, *et al.*, did there appear to be rupture of a pre-existing lesion during delivery.

The peak incidence of aneurysmal subarachnoid hemorrhage (SAH) is in the fifth and sixth decades of life, and the diagnosis of intracranial arterial aneurysm in childhood is uncommon.²⁵ In the Cooperative Study only 0.2% of patients with aneurysmal SAH presented in the first decade of life.²⁵ Infection, trauma, hemodynamic disturbances such as arteriovenous malformation and moyamoya disease, postirradiation vasculopathy, intracranial neoplasms, and a variety of systemic diseases such as arterial hypertension, polycystic kidney disease, fibromuscular hyperplasia, coarctation of the aorta, disorders of collagen metabolism, and tuberous sclerosis have all been identified as settings for pediatric intracranial arterial aneurysms,^{1-3,9,12,13,31,46,48} but no more than one-third of cases have evident etiologies.^{12,18,32,42,48}

If the large fraction of pediatric aneurysms that cannot be attributed to identifiable vascular diseases cannot alternatively be attributed to the mechanisms that generate aneurysms in adults, the etiology of aneurysms in childhood is obscure indeed. That most aneurysms found in adults are acquired during adult life is manifest by the virtual nonexistence of incidental aneurysms in pediatric cerebral angiography and in pediatric autopsy material.^{16,26,37,42} The great majority of aneurysms in adulthood are now attributed to degeneration of the internal elastic lamina at sites of hemodynamic stress.^{38,40} Stehbens,⁴¹ the major proponent of the acquired degenerative theory of arterial aneurysm formation, has presented a critical review of reports of congenital arterial aneurysms and has argued that typical berry

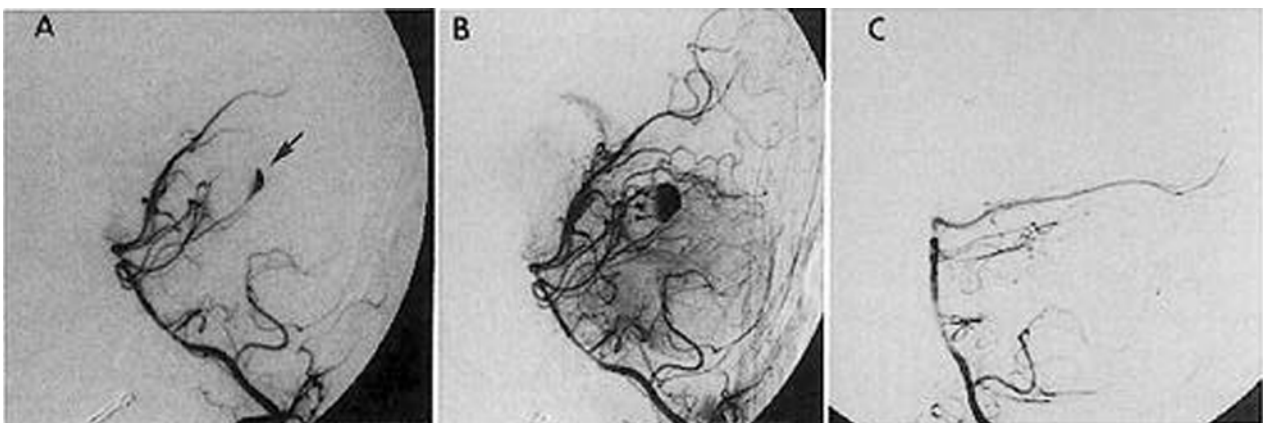


FIG. 3. Left vertebral angiograms, lateral projection. A: An early image demonstrating a 1.5-cm aneurysm arising from a distal vermian branch of the left superior cerebellar artery. A jet of contrast material is seen entering the aneurysm (arrow). B: Later image showing a blood contrast level (arrows). C: This follow-up angiogram obtained 5 weeks later was normal.

aneurysms of adulthood do not occur in early childhood. The possibility that birth trauma may be responsible for pediatric aneurysms has been entertained,^{27,36,41} but we have been unable to find previous documentation of this phenomenon. Our observations suggest that some pediatric aneurysms that have been categorized in the past as idiopathic or congenital, particularly aneurysms in the region of the tentorial incisura, are due to birth trauma.

Interestingly, several reviews of intracranial arterial aneurysms in infancy and early childhood have reported a higher prevalence of posterior circulation aneurysms among infants as compared to adults.^{7,44} The higher prevalence is especially pronounced for posterior cerebral artery lesions, which account for 11% of aneurysms among infants and young children as compared to no more than 2% among adults.^{7,33,44} Childhood posterior cerebral artery aneurysms tend to develop distal to the circle of Willis;²¹ the proximity of this artery to the free edge of the tentorium and the deformation to which the region of the incisura is subjected during the birth process suggest that birth trauma may account for the excess of lesions at this location.

Conclusions

Our experience suggests that noninvasive imaging modalities, such as transfontanel Doppler ultrasonography or magnetic resonance angiography,³⁶ may be indicated in neonates with intracranial hemorrhage, particularly hemorrhage in the region of the tentorial incisura, and may lead to more frequent detection of intracranial arterial aneurysms attributable to birth trauma.

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