POST-CLIPPING CEREBROVASCULAR COMPLIANCE IN AN INFANT WITH BILATERAL ANTERIOR CIRCULATION ANEURYSMS

Christopher Jones, Jacob Fairhall, Venkataraman Balakrishnan, Agadha Wickremesekera

Department of Neurosurgery, Wellington Hospital, Wellington, New Zealand

ABSTRACT

We present an uncommon case of an infant aged 12 months, presenting with a seizure due to subarachnoid hemorrhage secondary to an anterior circulation aneurysm. Following angiography, a right A1/A2 junction aneurysm was clipped by a right pterional approach. Angiography 5 months after clipping revealed loss of flow in the right anterior cerebral artery and a de novo L A1/A2 aneurysm, which was electively clipped by a left pterional approach. Follow up angiography 5 months later showed loss left anterior cerebral artery flow and the anterior cerebral artery territory perfused by posterior cerebral arteries. Cerebral angiography 8 months later did not show further aneurysms and demonstrated the posterior circulation vessels perfusing the anterior circulation vascular territory The child remains neurologically intact.

***Key words**: pediatric cerebral aneurysm, de novo aneurysm*

CASE REPORT

A 12 month old female infant presented with a week of a flu-like illness and a generalized tonic clonic seizure lasting 5 minutes. She was born at term by normal vaginal delivery, and she did not have a past medical history of intracranial hemorrhage or a family history of cerebrovascular disease. On initial examination she was afebrile, with generalized hypotonia, reduced responsiveness and normal blood pressure, but moving all limbs to stimuli. Fundoscopy was unremarkable. She was reusable maintaining a good airway.

Lumbar puncture revealed bloodstained cerebrospinal fluid with xanthochromia. No organisms were seen on gram stain and culture. The serum white cell count was elevated, but c-reactive protein and coagulation profile were normal. Intravenous fluid and nimodipine were commenced.

Correspondencia: Agadha Wickremesekera MBChB, FRACS, Department of Neurosurgery, Wellington Hospital, Capital and Coast District Health Board, Riddiford St, Private Bag 7902, Wellington South, New Zealand.
Email: agadha.wickremesekera@ccdhb.org.nz

Computed tomography (CT) of the brain revealed extensive subarachnoid hemorrhage particularly in the interhemispheric cistern, extending into the ventricular system with moderate hydrocephalus (Fig. 1).

Cerebral angiography demonstrated tortuosity of anterior cerebral (A1) and pericallosal (A2) arteries. The angiogram revealed a 5 mm A1/A2 junction aneurysm; the anterior communicating segment was hypoplastic (Fig. 2).

At operation, a right pterional craniotomy exposed a partially thrombosed 15 mm right A1/A2 junction aneurysm, which was clipped with a Sugita bayonet clip. A 2-3 mm loculus/aneurysm on the distal right A1, not seen on angiography, was also clipped with a curved Sugita miniclip. Postoperatively, on day 2 she suffered focal seizures with left-sided twitching, but no loss of consciousness. She recovered with no neurological deficit and was discharged on day 14.

Check cerebral angiography 5 months later confirmed absence of the right A1/A2 aneurysm, but also showed loss of the right A1 (Fig. 3). The same angiogram also showed a de novo 5 mm left A1/A2 junction aneurysm (Fig. 4), not visualized on the initial angiogram (Fig. 5). When comparing

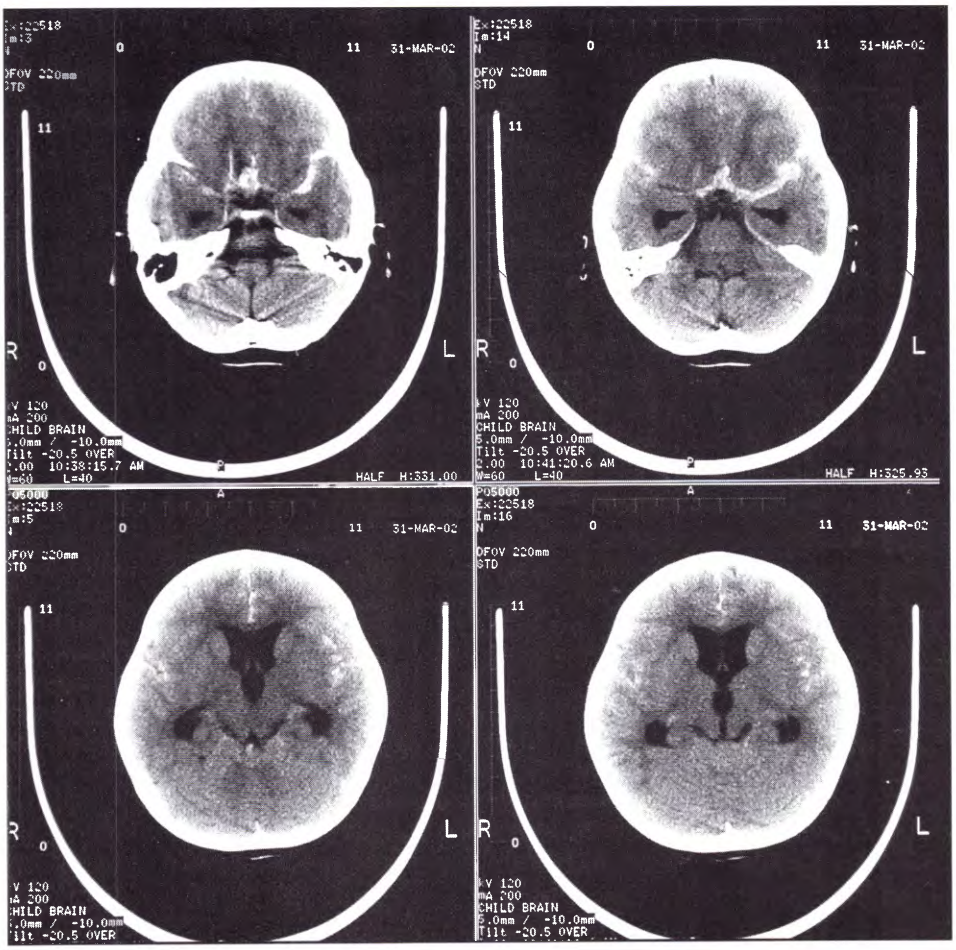

Fig 1. Admission CT: subarachnoid hemorrhage and ventriculomegaly

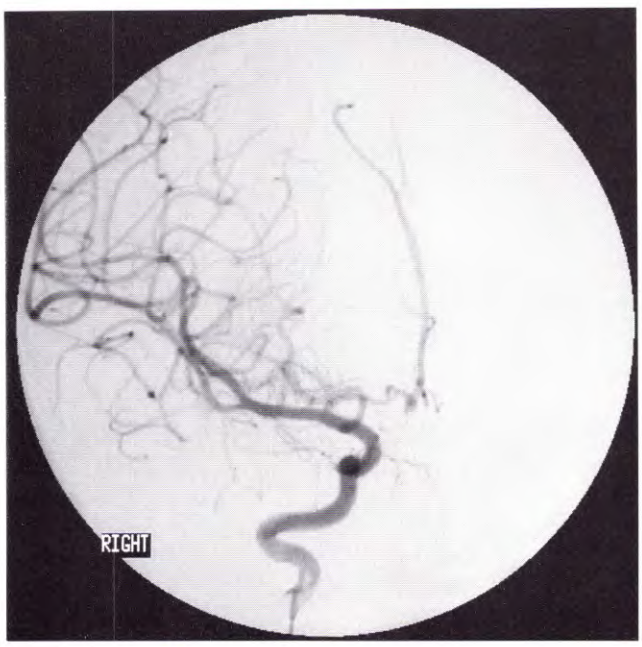

Fig 2. Right carotid angiogram on admission demonstrating 5 mm A1/A2 junction aneurysm

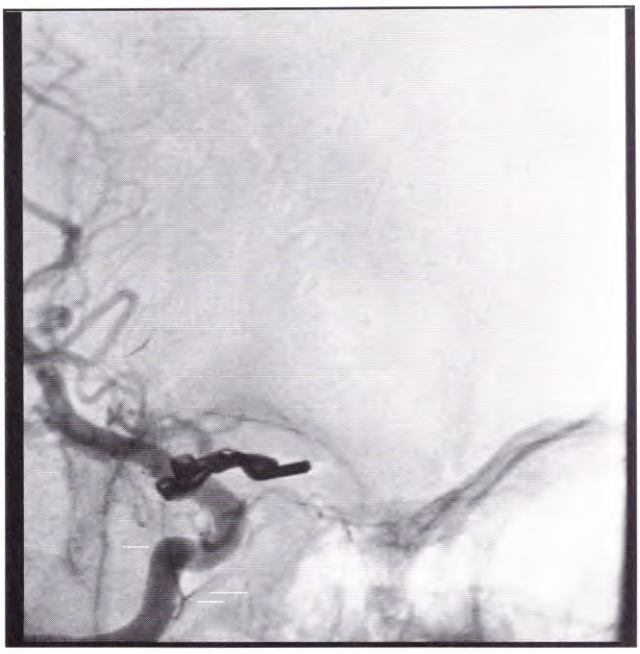

Fig 3. Post-operative angiogram following clipping of the right A1/A2 aneurysm, showing absence of right anterior cerebral artery

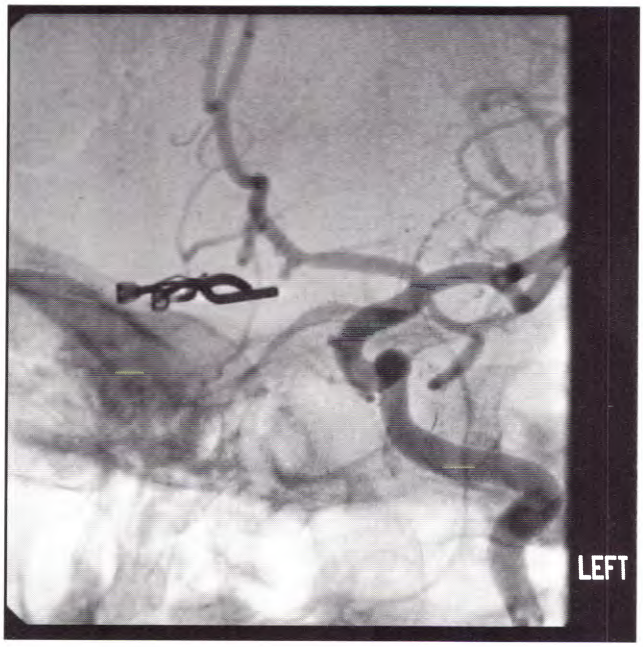

Fig 4. Post-operative left internal carotid injection showing de novo *left A1 aneurysm*

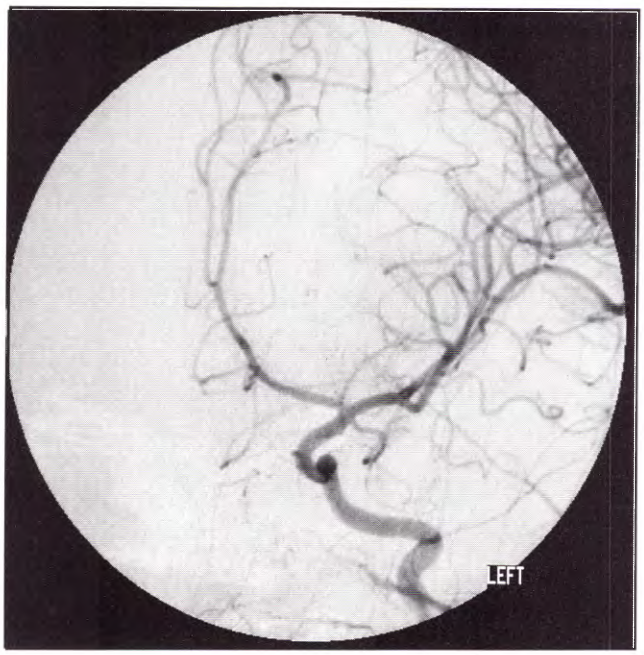

Fig 5. Left carotid angiogram on admission: no aneurysm seen

similar projections of the interval angiograms, the left sided aneurysm clearly appeared to be at a de novo site, be it adjacent to the anterior cerebral complex.

The de novo aneurysm was clipped electively by a left pterional approach. At operation a 10 mm

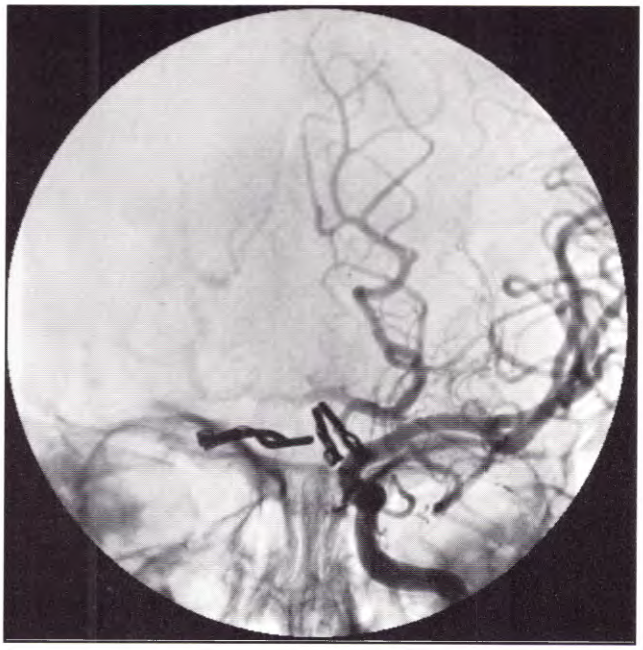

Fig 6. Left internal carotid injection demonstrating collateral flow to the posterior circulation via the posterior communicating and posterior cerebral arteries.

partially thrombosed aneurysm was seen, which was separate from the site of the right aneurysm and its clip. The postoperative period was uneventful and she was discharged on day 5.

Repeat check cerebral angiography 5 months following the second operation showed no aneurysms, continued loss of flow in the right A1 and significantly reduced flow in the left A1. A further four vessel cerebral angiogram after 12 months showed no aneurysms and no flow in either A1 segments, with collateral flow via the posterior communicating and the posterior cerebral arteries, especially on the left side (Fig. 6). The patient has continued to develop normally. She has had a normal cardiac and renal ultrasound. Follow up is planned with cerebral CT angiography.

DISCUSSION

Aneurysmal subarachnoid hemorrhage in the pediatric population is rare, with an incidence of one to three cases per one million populations. Pediatric aneurysms are reported to represent 0.5 to 4.6% of all cerebral aneurysms[1]. Due to their rarity, it is uncommon for any one center to have a large series, but a recent review found 63 of 85 infants with cerebral aneurysms presented with subarachnoid hemorrhage[2]. Some report, that pediatric aneurysms usually present as subarachnoid hemorrhage rather than mass effect, most

arising from the anterior circulation[3], but others suggest that aneurysms in the pediatric population commonly occur in the posterior circulation[4-5]. Delayed diagnosis of subarachnoid hemorrhage in infants, especially neonates is associated with a high mortality and morbidity[6]. Aneurysmal subarachnoid hemorrhage in infancy also has a significant mortality. Postmortem histological studies following a right middle cerebral artery hemorrhage in an infant revealed disruption of the internal elastic lamina of both middle cerebral arteries, reinforcing the theory of dysembryogenesis and the congenital nature of aneurysms in infancy[7]. Pediatric cerebral aneurysms differ from those that occur in adults in several ways. Pediatric aneurysms appear more common in the posterior circulation[4-5], although due to small numbers this varies markedly between series[8] overall 21% in children compared with 7% in adults. Male predominance of pediatric aneurysms is converse to the trend in adults[1].

Children appear to be either less susceptible to or more tolerant of vasospasm[3]. One series, noting severe angiographic vasospasm in 6 symptomatic children, concluded that vasospasm seemed to be well tolerated in children[8].

Pediatric cerebral aneurysms are recognized to be associated with a number of conditions, including but not limited to aortic coarctation, arteriovenous malformation, birth trauma, cardiac myxoma, fibromuscular dysplasia, head injury, tuberous sclerosis, polycystic kidney disease and other vascular anomalies[5].

The incidence of adult de novo aneurysms, may be calculated as 1.8% per annum, and more common with a history of multiple aneurysms[9]. Adult de novo aneurysms are more common in the anterior cerebral circulation, and are often seen some years after initial diagnosis of the first aneurysm, but have been detected 3 months after initial presentation[10-11]. The incidence of childhood de novo aneurysms remains uncertain.

Spontaneous thrombosis of an aneurysm has been attributed to hemodynamic flow factors, ratio of neck to fundus diameter, and the age of the aneurysm. Clipping of a posterior cerebral aneurysm in a child was suggested to lead to spontaneous thrombosis of a second superior cerebellar artery aneurysm. The mechanism was unclear and there was no parent artery thrombosis[12].

In our case, we are unable to attribute any specific cause for the partial thrombosis of both aneurysms. Tortuous abnormal vessels and post-subarachnoid hemorrhage vasospasm may conceivably explain partial thrombosis within the aneurysms, as well as the loss of A1s postoperatively. In our case, the absence of a left A1/A2 junction aneurysm on the initial angiogram raised the possibility of recanalization and/or enlargement of a thrombosed aneurysm. The disappearance and reappearance of a cerebral aneurysm in an infant after 6 months, has been described recently[13]. Another possible mechanism may be that, cessation of flow in the right A1 increased flow in the left A1 contributing to enlargement of a thrombosed aneurysm, in a similar fashion to compaction of coils within an aneurysm. On the other hand the left A1/A2 junction aneurysm may be de novo, as described above[11], but the true mechanism of formation of short term de novo aneurysms remains unclear. In this reported case both aneurysms were partially thrombosed, but complete spontaneous thrombosis occurs in infants, in particular the larger cerebral aneurysms[14].

Another explanation is that the left aneurysm is part of the same complex of the right aneurysm. Nevertheless, in comparing interval angiograms, it is compelling that the left aneurysm was not seen on the first angiogram, and has developed since the first operation.

In our case, despite the loss of A1s, anterior cerebral infarction was prevented by sustained collateral perfusion by the posterior cerebral arteries. Children are considered to have superfluous cerebrovascular compliance and greater tolerance to surgery. These features may be attributed to minimal vasospasm after subarachnoid hemorrhage, absence of systemic hypertension and/or the absence of endothelial changes seen in adults.

CONCLUSIÓN

This case presented with abnormal anterior cerebral arteries, seen to be tortuous with irregularities seen at angiography and surgery. There was no anterior communicating artery seen despite cross compression. The anterior cerebral arteries were prone to aneurysm formation and thrombosis. The post-operative loss of A1 flow could be explained by clip obliteration of the parent A1s, although this was clearly not the case at surgery, or collapse of pre-existing abnormal arteries with/without vasospasm, enhanced by surgical manipulation. If the anterior cerebral arteries were normal and there was no collateral perfusion, one would expect cerebral infarction. The presence of well-developed posterior circulation collaterals with

angiographic loss of the A1s after clipping of the aneurysms, suggest a pre-existing abnormality of the anterior cerebral arteries. Such cerebrovascular compliance without the development of neurological sequelae in this case, implicates not only the hypothesis of abnormal neurovascular embryogenesis of the anterior cerebral/anterior communicating arteries, but also the pre-existence of an easily accessible protective collateral perfusion.

References

1. Wojtacha M, Bazowski P, Mandera M, Krawczyk I, Rudnik A. Cerebral aneurysms in childhood. **Childs Nerv Syst** 2001; 17: 37-41.
2. Elgamal EA, Murshid WR, Abu-Rahma HM, Samir D. Aneurysmal subarachnoid hemorrhage in the first year of life: case report and review of the literature. **Childs Nerv Syst** 2004.
3. Young WF, Pattisapu JV. Ruptured cerebral aneurysm in a 39-day-old infant. **Clin Neurol Neurosurg** 2000; 102: 140-3.
4. Kasahara E, Murayama T, Yamane C. Giant cerebral arterial aneurysm in an infant: report of a case and review of 42 previous cases in infants with cerebral arterial aneurysm. **Acta Paediatr Jpn** 1996; 38: 684-8.
5. Khoo LT, Levy ML. Intracranial aneurysms. In: Albright A.L, Adelson P.D., Pollack I.F.(eds) Principles and Practice of Pediatric Neurosurgery; Thieme Medical Publishers. New York, Stuttgart, pp 973-1001.
6. Maroun F, Squarey K, Jacob J, Murray G, Cramer B, Barron J, Weir B. Rupture of middle cerebral artery aneurysm in a neonate: case report and review of the literature. **Surg Neurol** 2003; 59:114-9.
7. Hulsmann S, Moskopp D, Wassmann H. Management of a ruptured cerebral aneurysm in infancy. Report of a case of a ten-month-old boy. **Neurosurgery** 1998; 21:161-6.
8. Proust F, Toussaint P, Garnieri J, Hannequin D, Legars D, Houteville JP, Freger P. Pediatric cerebral aneurysms. **J Neurosurg** 2001; 94:733-9.
9. David, CA, Vishteh AG, Spetzler RF, Lemole M, Lawton MT, Partovi S. Late angiographic follow-up review of surgically treated aneurysms. **J Neurosurg** 1999; 91: 396-401.
10. Sim JH, Kim SC, Kim MS. Early development and rupture of de novo aneurysm—case report. **Neurol Med Chir** (Tokyo) 2002; 42: 334-7.
11. Matheus MG, Castillo M. Development of de novo intracranial aneurysm in three months: case report and literature review. **AJNR** 2003; 24(4): 709-10.
12. Nanda A, Drury BT. Posterior circulation aneurysms in a child: clipping of one leads to spontaneous thrombosis of the other. **Pediatr Neurosurg** 1997; 26: 41-7.
13. Motohashi O, Kameyama M, Imaizumi S, Mino M, Naganuma H, Ishii K, Onuma T. *A distal anterior cerebral artery aneurysm in infant: disappearance* and reappearance of the aneurysm. **J Clin Neurosci** 2004; 11: 86-8.
14. Russegger L, Grunert V. A thrombosed giant MCA aneurysm in a ten-week-old infant. Neurochirurgia (Stuttg) 1987; 30: 186-9.

RESUMEN

Se presenta un caso inusual de un niño de 12 meses de edad que consulta por convulsiones debidas a hemorragia subaracnoidea secundaria al sangrado de un aneurisma de la circulacion anterior. Por un abordaje pterional derecho se clipa un aneurisma localizado en la unión de A1 y A2 de la arteria cerebral anterior derecha. Una angiografía de control, efectuada 5 meses después, mostró un aneurisma "de novo" de la unión A1 - A2 izquierda que se clipó a través de un abordaje pterional izquierdo. La angiografía de control, 5 meses después, mostró una pérdida del flujo sanguíneo de la arteria cereral anterior izquierda, cuyo territorio pasa a ser irrigado por las arterias cerebrales posteriores. La angiografía de control 8 meses después, mostró el patrón angiográfico antes descripto sin nuevos aneurismas. El niño evolucionó sin déficit neurológico alguno.

Palabras clave: *aneurisma cerebral en pediatría, aneurisma de novo*