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A L Thrush and G D Marano

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Infantile Intracranial Aneurysm: Report of a Case and Review of the Literature

Ann L. Thrush¹
Gary D. Marano

The CT and angiographic diagnosis and successful surgical treatment of a ruptured cerebral aneurysm in a 1-month-old girl are described. The incidence of intracranial aneurysms in infancy is less than 1% [1]. A review of intracranial aneurysms in the neonate and infant is presented to define clearly the incidence and clinical characteristics of aneurysms in this age group. We have reviewed 43 cases of neonatal and infantile aneurysms reported since 1916.

Cerebral artery aneurysms are extremely rare in childhood. McDonald and Korb [2] reviewed 1125 cases of saccular cerebral aneurysms. Only 17 (1.5%) of these aneurysms occurred in children. The youngest patient in this series was 1½ years old. Laitinen [3] reported a similar childhood incidence of 1.3% in 688 patients. Matson [4] identified 13 children with intracranial aneurysms in a 12-year period. The youngest patient in his series was 16 years old. Patel and Richardson [5] reviewed 3000 cases of subarachnoid hemorrhage in which the youngest patient was 8 years old. Locksley [6], in the Cooperative Study in 1966, reviewed 2637 cases of cerebral aneurysms and found only one patient younger than 4 years old.

Our patient represents the eighth reported case of an intracranial aneurysm in a neonate. In accordance with *Dorland's Medical Dictionary* [7] we have defined the neonatal period as the first four weeks of life; infancy is defined as a period that extends to the end of the first 24 months.

Case Report

A normal term female child, with no history of trauma or illness, presented with a history of increasing irritability followed by decreasing alertness over a 24-hour period. She was taken to another hospital where she became increasingly lethargic and developed agonal respirations. At this time, she was intubated and an unenhanced CT scan of the head was obtained. This demonstrated a large, left frontotemporal intracranial hematoma. She was then transferred to West Virginia University Hospital where repeat unenhanced and enhanced CT scans of the head showed enlargement of the hematoma with an enhancing central mass (Figs. 1 and 2). At this time it was noted that her pupils were unreactive and unequal. She was immediately taken to the arteriography suite where part of the temporal blood clot was aspirated as an emergency procedure. A subsequent arteriogram showed a large, slowly filling aneurysm arising from the distal portion of the left middle cerebral artery (Fig. 3). The aneurysm measured approximately 6 mm. The patient's condition stabilized 3 days later and the aneurysm was clipped without complication. She was discharged from the hospital with no apparent sequelae. A 20-day postoperative clinic follow-up showed no appreciable neurologic deficit.

Discussion

A review of the literature reveals seven cases of cerebral aneurysms in the neonatal period. The youngest patient was a 3-day-old twin who was cyanotic at birth and died 64 hr after delivery [8]. His twin sister also died shortly after birth

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¹ Both authors: Department of Radiology, West Virginia University Medical Center, Morgantown, WV 26506. Address reprint requests to A. Thrush.

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Fig. 1.—Unenhanced head CT shows large intracranial hemorrhage.



Fig. 2.—Enhanced head CT shows enhancing aneurysm within hemorrhage.

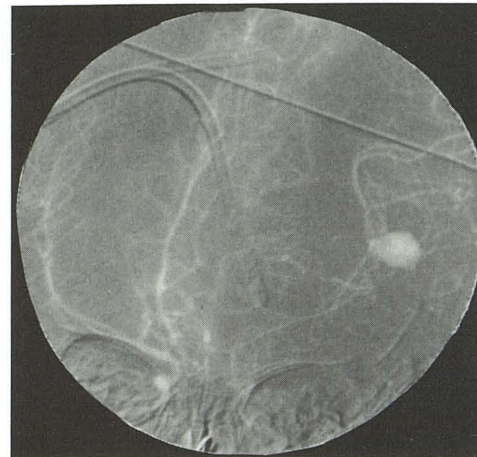


Fig. 3.—Digital subtraction angiogram shows middle cerebral aneurysm.

but an autopsy was not performed. Newcomb and Munns [8] reported a 23-day-old infant who presented with cyanosis after a minor head trauma. The patient died 4 hr after admission, and an autopsy revealed a ruptured aneurysm arising from the circle of Willis. The exact location could not be determined. Table 1 summarizes the pertinent facts concerning the seven neonatal cases listed in the references [8–13]. Several studies have cited a neonatal case reported by Thompson [14]. We did not include this case in our neonatal data since a definitive diagnosis of the aneurysm was not made until the child was 9 months old. We also did not include two fetal cases reported by Bremer [15].

A more extensive review of the literature covering the incidence of cerebral aneurysms in infancy revealed 43 cases including our own. Batnitzky and Muller [16] reported 12 cases of infantile and juvenile cerebral aneurysms in patients whose ages ranged from 6 months to 17 years. We did not include their data, since the number of cases that involved children under 1 year old was not specified.

Presenting Symptoms

The presenting symptoms ranged from an increase in irritability and vomiting to seizures and coma. An increase in irritability was a common symptom (10%). Extreme irritability in neonates and infants is most commonly associated with meningitis but the differential diagnosis should include a possible ruptured aneurysm. Five of the cases presented after head trauma [17–19]. Eight cases (19%) presented with seizure [14, 20–26]. A 9-month-old girl with a large posterior cerebral artery aneurysm presented with cerebellar and cranial nerve symptoms [27]. Thompson and Pribram [14] reported a 9-month-old girl with a large internal carotid artery aneurysm who developed ophthalmoplegia and quadriplegia. Four patients presented with hydrocephalus [27–30].

Gender

Of the 43 cases including our own, 24 were male and 12 were female. Our review demonstrates a male predominance

with a two to one ratio. Data concerning the gender of seven of the cases could not be obtained.

Multiple Lesions

There were 47 aneurysms in the 43 reported cases. Ferry et al. [31] reported a 3-month-old boy with a large proximal basilar artery aneurysm associated with two daughter aneurysms and multiple ectatic cerebral vessels. The case reported by Arai et al. [25] had two aneurysms arising from the distal middle cerebral artery as well as an associated arteriovenous malformation. The 8-month-old boy reported by Lemmen and Schneider [28] had bilateral posterior cerebral artery aneurysms.

Site

Reports of 45 aneurysms were used to compile Table 2 [1, 4, 8, 9, 12–25, 27–39]. The sites of two aneurysms could not be identified. Thirty of the aneurysms arose from the anterior circulation, which is an occurrence of 67% compared with an adult incidence of 85% from this location. Three arose from the anterior communicating artery [22, 32, 34] and two arose from the pericallosal artery [17, 27]. Nineteen (42%) arose from the middle cerebral artery. Seven were described as distal middle cerebral artery aneurysms and two arose from the anterior parietal branch. Three arose from the internal carotid artery. Ventureyra et al. [1] reported a large anterior choroidal aneurysm. Shucart and Wolpert [37] reported a large aneurysm at the internal carotid bifurcation.

Fifteen (33%) of the 45 aneurysms arose from the posterior circulation. The incidence of aneurysms arising from the posterior circulation in adults is 15%.

It is clear from Table 2 that infantile aneurysms tend to be more peripheral in location and occur more frequently in the posterior fossa.

Size

Unfortunately, the size of the aneurysm was not reported in 16 cases. The reported sizes ranged from 1–6 cm. Ten of

TABLE 1: Data on Seven Neonatal Cases

Reference	Age	Gender	Site	Size	Presentation	Diagnostic Study	Outcome
Newcomb and Munns [8]	64 hr	M	PCA	2 mm	Cyanotic at birth	Autopsy	Died 64 hr after birth
Newcomb and Munns [8]	23 days	M	Circle of Willis	DNA	Minor head trauma, cyanosis	Autopsy	Died 4 hr after admission
Lee et al. [9]	13 days	F	Basilar bifurcation	3 cm	Coma, hypotonia	Angiography	Died day 37
Hungerford et al. [10]	28 days	F	Peripheral MCA	1.5 cm	Vomiting, irritable	CT	Good surgical angiography outcome
Jones and Shearburn [11]	4 weeks	F	MCA	DNA	Extremely irritable, seizure	Angiography	Good surgical outcome
Pickering et al. [12]	1 month	M	Distal PICA	2.5 cm	Apnea with cyanosis	Autopsy	Died 24 hr after admission
Wierdis et al. [13]	Newborn	DNA	MCA	DNA	SAH	Autopsy	Died

Note.—PCA = posterior cerebral artery; DNA = data not available; MCA = middle cerebral artery; PICA = posterior inferior cerebellar artery; SAH = subarachnoid hemorrhage.

TABLE 2: Comparison of the Sites and Incidence of Aneurysms in Infancy and Adulthood (n = 45)

	% of Cases	
	Adults	Infants
Aneurysms arising from anterior circulation (n = 30)		
ACA [17, 22, 27, 32–34] ^a	30	16
MCA [13, 14, 16–18, 20, 21, 24, 25, 30, 35, 36]	25	42
ICA [1, 9, 23, 37]	30	9
Total	85	67
Aneurysms arising from posterior circulations (n = 15)		
PCA [1, 8, 28]	2	11
Basilar artery [12, 31, 38, 39]	10	13
Vertebral/PICA [4, 15, 19, 29]	3	9
Total	15	33

Note.—ACA = anterior cerebral artery; MCA = middle cerebral artery; ICA = internal carotid artery; PCA = posterior cerebral artery; PICA = posterior inferior cerebellar artery.

^a Numbers in brackets refer to references at end of article.

the aneurysms were reported as “large.” Nine measured between 1 and 4 cm. The aneurysm in our case measured approximately 6 mm.

Associated Abnormalities

Three of the cases had other associated anomalies. An 11-month-old reported by Pool and Potts [30] died from obstructive hydrocephalus caused by stenosis of the sylvian aqueduct. Other congenital anomalies included hydromyelia of the spinal cord, a malformed fourth ventricle, hypoplasia of the cerebellum, and a suboccipital encephalocele. An autopsy on a 7-week-old girl reported by Garcia-Chavez and Moosy [20] revealed agenesis of the corpus callosum. A 1-mm basilar artery aneurysm was discovered as an incidental autopsy finding in a 13-week-old boy who had polycystic kidney disease and died from respiratory arrest [40]. It is of interest that there was only one associated case of polycystic kidney

disease, which is known to have a fairly high association with cerebral aneurysms in the adult population.

Other Vascular Anomalies

Three of the cases had other vascular anomalies. Bolander et al. [22] reported an 11-month-old boy with a large anterior communicating artery aneurysm and renal artery fibromuscular hyperplasia (FMH). Several investigators have suggested a relationship between FMH and intracranial aneurysms. Palubinskao et al. [40] reported five cases of cerebral aneurysms in 70 patients with renal artery FMH. An additional four patients had cerebral aneurysms associated with carotid artery FMH. Arai et al. [36] reported a 20-month-old boy with a “huge” distal middle cerebral aneurysm and a “small” distal middle cerebral aneurysm with an associated AVM. Angiography also revealed dilatation of the internal carotid artery and dilatation and kinking of the middle cerebral artery. A curvilinear calcification was located in the large distal middle cerebral artery aneurysm. The autopsy on the 3-month-old boy reported by Ferry et al. [31] revealed widespread arterial aneurysms of the cerebral, iliac, and renal arteries.

Miscellaneous

Devadiga et al. [23] reported the “spontaneous” cure of a large intracranial aneurysm in a 14-month-old boy. A selective left carotid artery arteriogram demonstrated a large aneurysm in the intracavernous portion of the internal cerebral artery. The patient presented with proptosis and left-sided weakness. The parents refused surgery and the child was discharged. A repeat arteriogram 2 years later revealed complete obliteration of the aneurysm with better filling of the intracranial vessels and evidence of internal and external carotid anastomosis.

Treatment and Outcome

Of the 18 patients who underwent surgical treatment, 14 had a good surgical outcome. Two of the patients died after surgery and two were reported to have a fair surgical outcome.

Conclusions

Aneurysms in infancy are very rare but they do occur and are treatable. Their prognosis appears to be much more favorable than that of ruptured aneurysms in the adult. A good prognosis also depends on a prompt diagnosis. Our review clearly defines a childhood population in which aneurysms occur. Although the presentation and location of aneurysms in this age group differ from those in the adult population, the diagnostic workup, including CT and cerebral angiography, should be identical. A high index of suspicion must be entertained when a previously healthy infant presents with increasing irritability, vomiting, and/or lethargy.

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