Case Report

Pediatric Neurosurgery

Pediatr Neurosurg 1998;28:198–203

Received: April 21, 1998
Accepted: May 6, 1998

Saleh S. Baeesa[a]
Tommy Dang[c]
Daniel L. Keene[b]
Enrique C.G. Ventureyra[a]

Divisions of
[a] Neurosurgery and
[b] Neurology of the Children's Hospital of Eastern Ontario, and
[c] University of Ottawa, Ont., Canada

Unusual Association of Intractable Temporal Lobe Seizures and Intracranial Aneurysms in an Adolescent: Is It a Coincidence?

Key Words
Epilepsy
Complex partial seizures
Aneurysms

Abstract
Intracranial aneurysms in the pediatric age group are rare occurrences. They usually present with subarachnoid hemorrhage or mass effect. Their association with epilepsy has rarely been reported; such concurrence may not be a coincidence. We present a 16-year-old girl with a 5-year history of medically intractable complex partial seizures. Preoperative electrophysiological and neuroimaging studies demonstrated an epileptogenic focus and atrophy in the right mesial temporal lobe, and ipsilateral incidental aneurysm at the carotid artery bifurcation. The patient underwent a complete right anterior temporal lobectomy, followed by clipping of the aneurysm. We concluded that the epilepsy management in association with cerebral aneurysms is controversial, but when surgery is indicated, clipping of the aneurysm and resection of the epileptogenic focus may provide the optimal outcome. The relevant literature is reviewed and the possible mechanisms of production of epilepsy by intracranial aneurysms are discussed.

Introduction

Seizures as the first presenting symptom of intracranial aneurysms are uncommon [1], and rarely become medically intractable [2–6]. Dandy [7], in 1944, was the first to describe such unusual association in a patient with a 15-year history of seizures. At surgery, he found a giant thrombosed middle cerebral artery aneurysm replacing most of the temporal lobe tip [7]. In general, the exact incidence of aneurysmal epilepsy remains unknown. Only 4% of the 2,621 patients in the cooperative study of intracranial aneurysms and subarachnoid hemorrhage reported by Locksley had seizures prior to bleeding [1]. Currie et al. [8] reported structural abnormalities in 169 out of 666 patients (25.4%) with complex partial seizures (CPS), and only 1 (0.15%) had a middle cerebral artery aneurysm [8]. However, less than 17% of the patients had an angiogram, and CT scan was not available. On the other hand, Falconer et al. [9] reported 100 patients operated on for CPS, of whom none had aneurysms.

We report here a 16-year-old girl with a 5-year history of medically intractable complex partial seizures who was diagnosed incidentally as having a right internal carotid artery bifurcation aneurysm. The possible mechanisms are discussed, and the relevant literature on epilepsy caused by aneurysms is reviewed.

KARGER

Fax + 41 61 306 12 34
E-Mail karger@karger.ch
www.karger.com

1016–2291/98/0284–0198$15.00/0

Accessible online at:
http://BioMedNet.com/karger

Enrique C.G. Ventureyra, MD
Division of Neurosurgery
Children's Hospital of Eastern Ontario, 401 Smyth Road
Ottawa, ON K1H 8L1 (Canada)
Tel. +1 613 737 2316, Fax +1 613 738 4228

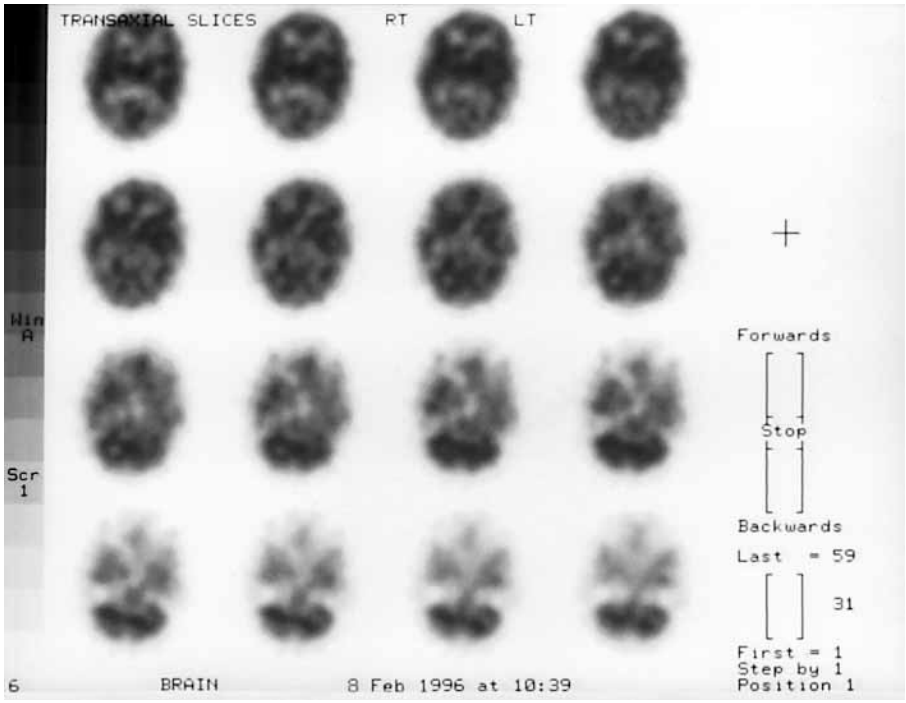


Fig. 1. Ictal ^{99m}Tc-HMPAO SPECT scans demonstrating an increase tracer uptake in the right temporal lobe.

Case Report

This 16-year-old female presented with initial onset of complex partial seizures at the age of 11. She was the product of an uneventful pregnancy, and was born at 34 weeks of gestation by cesarean section due to breech presentation. Her initial seizure onset consisted of sudden eye blinking followed by staring to the left side for 1 min with impairment of consciousness. These episodes were on most occasions preceded by shortness of breath. There was no history of febrile convulsions in early life or family history of epilepsy or intracranial aneurysms.

General and neurological examinations were normal, and preoperative routine laboratory tests were within normal limits. Her initial computed tomography scan was normal, and electroencephalogram (EEG) demonstrated slow-wave activity in the region of the right temporal lobe.

For a period of 5 years, her seizures had increased (7–10 per day) in frequency and severity despite adequate management with several anticonvulsant medications, including Dilantin, Depakene, Tegretol, Mysoline, Sabril, Rivotril, and Mogadon. Her seizures were considered refractory, and surgery was then indicated. She was on four antiepileptic medications prior to surgery.

The preoperative workup included EEG, MRI, SPECT scans, and cerebral angiogram with intracarotid injection of sodium amobarbital (Wada test). Video-telemetered interictal EEG recording showed bursts of theta activity mixed with sharp discharges over the right temporal region, and during the usual seizure the EEG showed rhythmic 4- to 6-Hz sharp-wave activity in the same area. This finding was confirmed with ^{99m}Tc-HMPAO SPECT scans, which demonstrated increased radiotracer localization in the right temporal lobe during seizure episodes (fig. 1). MRI scans demonstrated a mild atrophy of the right hippocampal gyrus, and a flow void signal was seen adjacent to it (fig. 2). This finding was shown on angiogram to be an 8 × 13 mm incidental aneurysm of the right carotid bifurcation origin (fig. 3). Wada test demonstrated left language dominance and bilateral memory representation.

At surgery, through a right frontotemporal craniotomy, electrocorticographic monitoring demonstrated neocortical as well as allocortical interictal discharges. A tailored temporal lobectomy (5 cm) was then performed removing the neocortex first. The aneurysm was found adherent to the right hippocampal gyrus, identified and clipped, then amygdalohippocampectomy (AHC) was performed. There was no evidence of remote hemorrhage.

Pathological examination revealed marked neuronal loss and gliosis, most prominent in the hippocampal cortex adjacent to the aneurysm (fig. 4). There was no evidence of hemosiderosis.

The patient had an unremarkable postoperative course apart from four motor focal seizures in the first 72-hour postoperative period; each lasted less than 30 s. Postoperative EEG demonstrated that they originated from the right frontal lobe. Postoperative MRI demonstrated adequate resection of the temporal lobe, and no filling of the aneurysm was seen on angiography (fig. 5). She has been followed for 2 years and has no CPS, but developed frontal lobe seizures refractory to antiepileptic medications that required surgical intervention and resection of a well-defined epileptogenic focus by electrocorticography. Pathological examination of this new epileptogenic focus revealed focal areas of cortical microdysgenesis. She has been seizure-free since then on 300 mg of Dilantin daily.

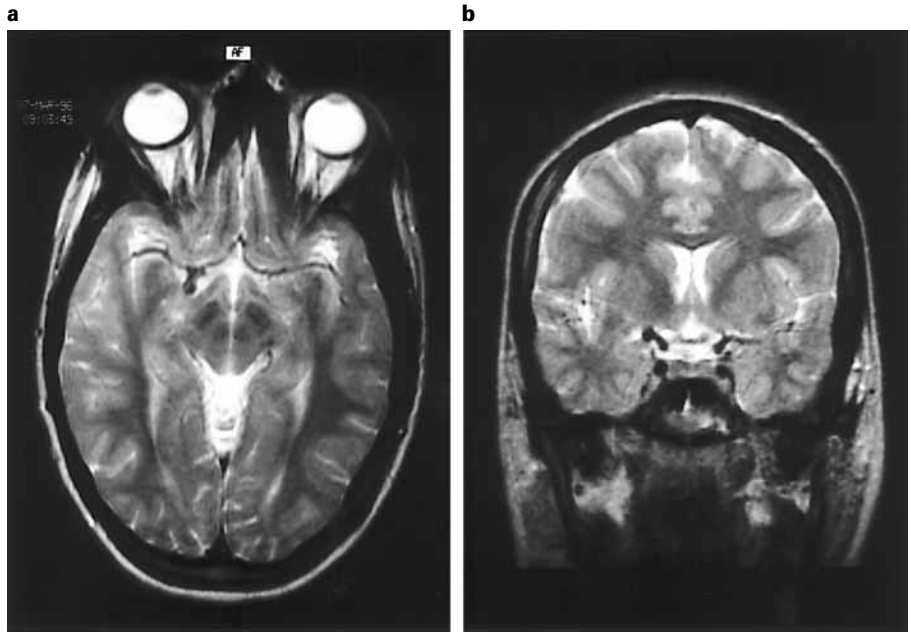


Fig. 2. Axial (**a**) and coronal (**b**) T2-weighted MRI demonstrating a flow void signal adjacent to the right hippocampus. The right hippocampal gyrus appears atrophied.

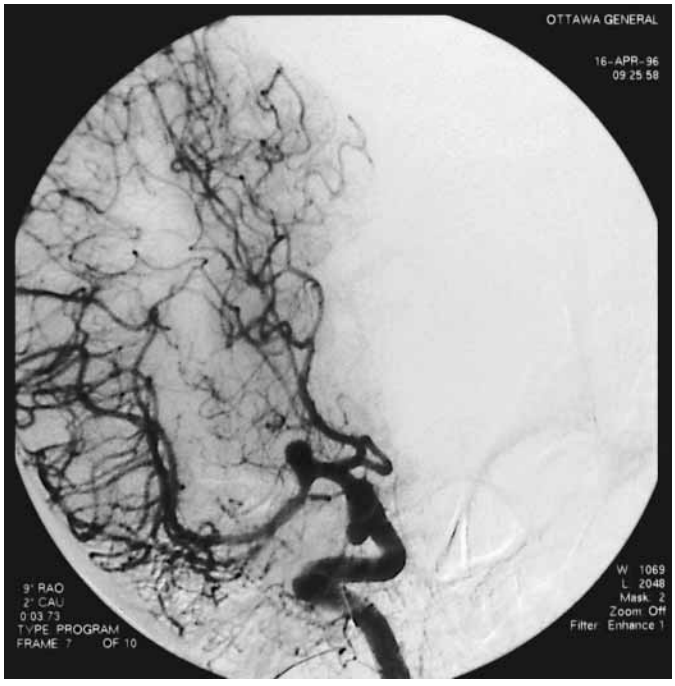


Fig. 3. Digital subtraction angiogram demonstrating an 8 × 13 mm aneurysm of the right carotid artery bifurcation.

Discussion

Intracranial aneurysms in children account for 0.5–4.6% of all aneurysms in this age group [10, 11]. Their etiology remains controversial, and several speculations have been made. Birth trauma has been reported in several cases as a contributing factor in aneurysm formation [12, 13]. Piatt et al. [12] have reported a case of an aneurysm in a cerebral artery that may have been injured by the free edge of the tentorium secondary to frontooccipital compression during the passage through the birth canal [12]. In addition to birth trauma, infection, moyamoya disease, arterial hypertension, arteriovenous malformation, fibromuscular hyperplasia, polycystic kidney disease, coarctation of the aorta, hydromyelia, tuberous sclerosis, generalized arterial dysplasia with myxoid medial degeneration, and agenesis of the corpus callosum all have been associated with aneurysms in this age group [10, 14]. Some authors have suggested that pediatric aneurysms are congenital [15, 16].

The most common presentation of intracranial aneurysms in children is subarachnoid hemorrhage [10, 15, 17]. Presentation secondary to mass effect (i.e. hydrocephalus or cranial nerve deficits) are less common [10, 15, 18, 19]. There is a higher incidence of giant aneurysms (>2.5 cm) in children compared to adults [10, 15, 19]. In addition, aneurysms in children are more likely to occur at the internal carotid bifurcation [17, 20, 21] or the posterior circulation [15, 19, 21]. However, some authors have found these aneurysms to be more peripherally located [10, 22].

The mechanism of intracranial aneurysms causing epilepsy is unknown. Several hypotheses have been suggested in the literature. Liang-fu and Da-jie [23] reported intra-

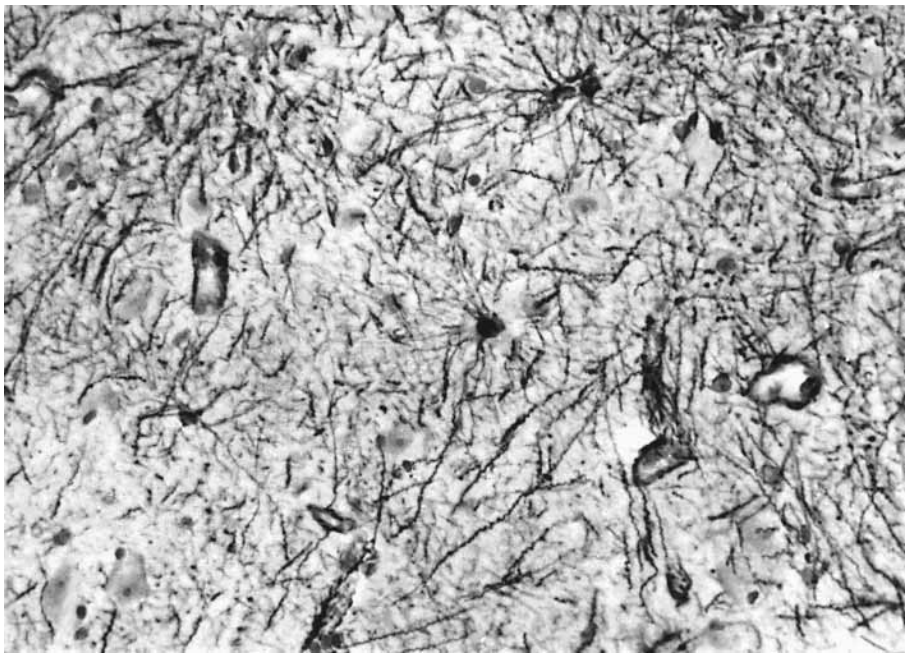
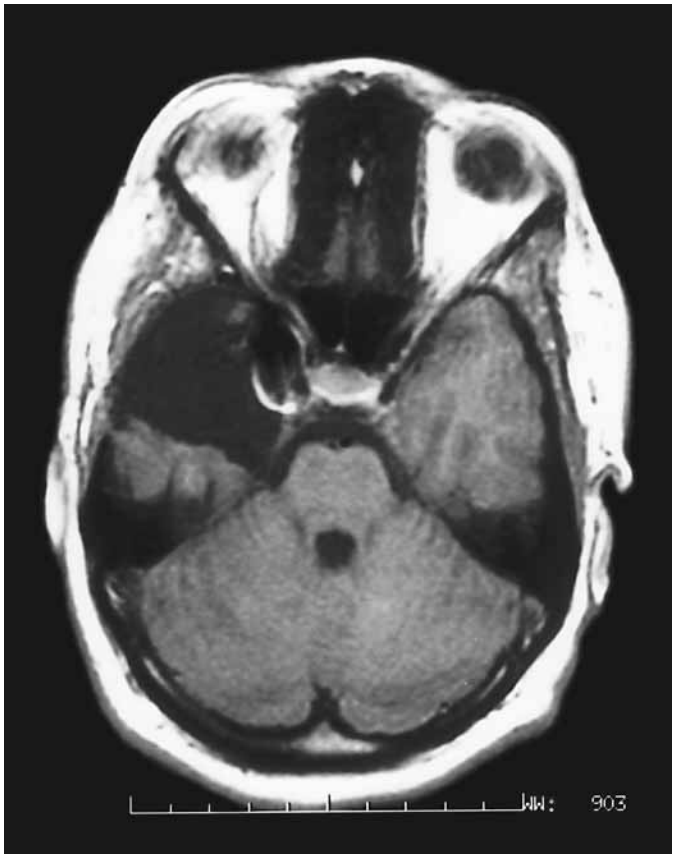
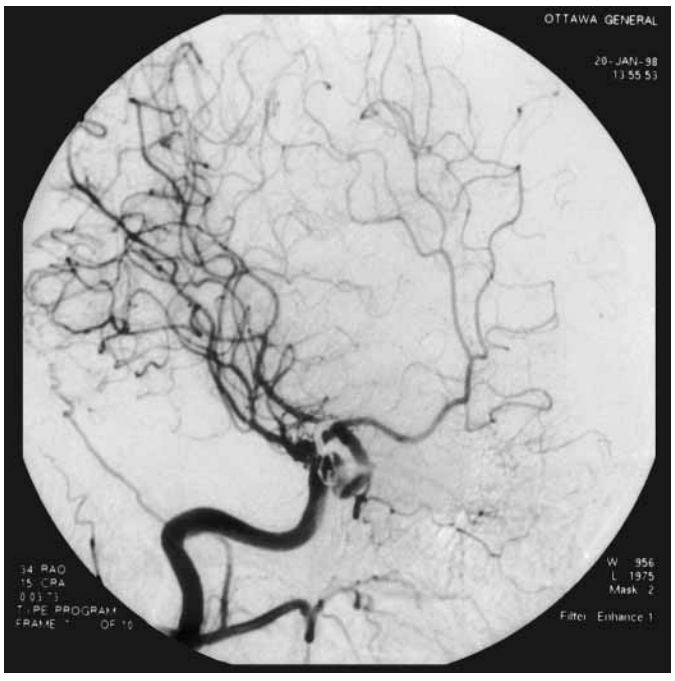


Fig. 4. Photomicrograph of the surgical specimen taken from the hippocampal gyrus adjacent to the aneurysm demonstrating residual neurons surrounded by marked gliosis. Glial fibrillary acidic protein. Original magnification × 400.



a



b

Fig. 5. Postoperative MRI (**a**) and angiogram (**b**) demonstrating adequate resection of the temporal lobe and no filling of the aneurysm.

Table 1. Summary of the surgical cases reported in the literature

Author	Ref.	Age years	Sex	Presentation	Aneurysm location	Treatment	Seizure outcome/ follow-up
Kamrin 1966	2	33	M	Visual and olfactory illusions with headache	MCA	Surgical clipping	Seizure-free/18 mo.
		45	M	Grand mal seizures	MCA	Aneurysmal excised & temporal lobectomy	Improved/10 mo.
		48	F	Grand mal seizures	MCA	Surgical clipping	Improved/4 yr.
		53	F	Left focal seizures	MCA	Surgical clipping	Improved/1 yr.
Sengupta et al. 1978	3	52	M	Temporal lobe seizures	MCA	Surgical clipping	Seizure-free/3 yr.
		56	F	Temporal lobe seizures	MCA	Surgical clipping	Improved/5 yr.
		57	F	Right-sided seizures	MCA	Surgical clipping	Seizure-free/3 yr.
		15	M	Right-sided seizures	MCA	Surgical clipping	Seizure-free/11 mo.
		42	M	Right-sided seizures	MCA	Surgical ligation	Lost to follow-up
Whittle et al. 1985	5	50	F	Complex partial seizures	MCA (2)	Surgical clipping (1) & excision (1)	Seizure-free/2 yr.
		52	M	Complex partial seizures	MCA	Surgical clipping	Seizure-free/4 yr.
Putty et al. 1990	13	7[1]	F	Left-sided focal seizures	PCA	Surgical clipping	Seizure-free/1 yr.
Yacubian et al. 1994	6	30	M	Complex partial seizures	PCA	Proximal occlusion of PCA & AHC	Seizure-free/15 mo.
Present case		16	F	Complex partial seizures	Carotid artery bifurcation	Surgical clipping, temporal lobectomy & AHC	Seizure-free/2 yr.

[1] Weeks.

cranial aneurysms in 2 of their patients and speculated that the calcified walls of the aneurysms acted as hamartomas and may have caused epilepsy. Another possibility is that minor leakage or recurrent minor bleeding from these aneurysms, without producing clinical manifestations of subarachnoid hemorrhage, may produce localized brain damage with glial scarring acting as an epileptogenic focus [3, 5, 24]. Yet another mechanism is compression of a giant aneurysm against the hippocampal gyrus causing epilepsy [25]. Lastly, there may be transient ischemia secondary to embolization by thrombus from an unruptured thrombosed aneurysm [4]. In our case, there was evidence of gliosis of the right hippocampal gyrus.

Only a few cases of epilepsy with intracranial aneurysms have been reported [2–6, 13, 23]. Table 1 summarizes the surgical outcomes of 15 relevant cases (including our case) that have been reported in the literature. The most common location of the aneurysm is the middle cerebral artery (MCA) bifurcation. EEG studies revealed a temporal lobe epileptogenic focus in all. Seven were females, and 3 were below the age of 18. All but 2 were treated with surgical clipping and remained seizure-free or improved in a follow-up varying from 1 to 5 years [2, 3, 5, 6, 13]. One case, a 30-year-old male with posterior cerebral artery (PCA) aneurysm, was treated with proximal PCA occlusion and selective AHC and remains seizure-free [6]. In another case, temporal lobectomy and excision of a calcified thrombosed MCA aneurysm was performed in a 48-year-old woman with significant improvement of her seizures, but she died 10 months later from myocardial infarction [2].

The preoperative evaluation of the child with epilepsy is of critical importance. This evaluation not only may reveal the cause of the epilepsy in the majority of cases, but can also help in the appropriate management decisions, medical or surgical. In addition to EEG, CT and SPECT scans, other imaging modalities and tests such as MRI, cerebral angiography, and Wada test are also important. MRI has increased the sensitivity for the detection of focal cerebral pathology and vascular abnormalities [26]. Thus, it has replaced cranial CT scan as the imaging modality of choice in epilepsy. Vascular lesions will be

shown on MRI as flow-void signals [13]. Putty et al. [13] reported the case of a cerebral aneurysm in an infant diagnosed by MRI. However, cerebral angiography is still necessary to confirm the lesion and identify the vascular anatomy for appropriate treatment. The Wada test is difficult to perform in young children because it must be done with the patient awake and cooperative. It is rarely done in children less than 10 years of age [27]. The Wada test defines how each hemisphere functions, regarding memory and language support, following inactivation of the opposite hemisphere. Recent memory is a function of bilateral mesial temporal structures. For temporal lobe seizures involving mesial structures, the Wada test demonstrating bilateral memory representation is a good indicator of minimal effect on recent memory loss with unilateral hippocampectomy.

Conclusions

Epilepsy in association with intracranial aneurysms is rare, and usually treated with anticonvulsant medication. Surgical or endovascular obliteration of the aneurysm is usually considered in patients at significant risk of aneurysmal rupture. When the seizures become intractable, and there is imaging evidence of mesiotemporal lobe atrophy, both aneurysmal clipping and resection of the epileptogenic focus may provide the best option for seizure control.

References

1 Lodksley HB: Report on the cooperative study of intracranial aneurysms and subarachnoid hemorrhage; natural history of subarachnoid hemorrhage, intracranial aneurysms, and arteriovenous malformations. J Neurosurg 1966; 25:219–239.
2 Kamrin R: Temporal lobe epilepsy caused by unruptured middle cerebral artery aneurysm. Arch Neurol 1966;14:421–427.
3 Sengupta RP, Saunders M, Clarke PRR: Unruptured intracranial aneurysms; an unusual source of epilepsy. Acta Neurochir 1978;40: 45–53.
4 Tanaka K, Hirayama K, Hattori H, Matsuoka O, Sakamoto H, Hakuba A, Murata R: Case report: A case of cerebral aneurysm associated with complex partial seizures. Brain Dev 1994; 16:233–237.
5 Whittle IR, Allsop JL, Halmagyi GM: Focal seizures: an unusual presentation of giant intracranial aneurysms. Surg Neurol 1985;24:533–540.
6 Yacubian EMT, Rosemberg S, da Silva HCA, Jorge CL, de Oliveira E, de Assis LM: Intractable complex partial seizures associated with posterior cerebral artery giant aneurysm: A case report. Epilepsia 1994;35:1317–1320.
7 Dandy WE: Intracranial Arterial Aneurysms. New York, Comstock, 1944.
8 Currie S, Heathfield KWG, Henson RA, Scott DF: Clinical course and prognosis of temporal lobe epilepsy. A survey of 666 patients. Brain 1971;94:173–190.
9 Falconer MA, Serafetimides EA, Corsellis JAN: Etiology and pathogenesis of temporal lobe epilepsy. Arch Neurol 1964;10:233–248.
10 Ferrante L, Fortuna A, Celli P, Santoro A, Fraioli B: Intracranial arterial aneurysms in early childhood. Surg Neurol 1988;29:39–56.
11 Frank E, Zusman E: Aneurysms of the distal anterior cerebral artery in infants. Pediatr Neurosurg 1990/91;16:179–182.
12 Piatt JH, Clunie DA: Intracranial arterial aneurysm due to birth trauma. J Neurosurg 1992; 77:799–803.
13 Putty TK, Luerssen TG, Campbell RL, Boaz JC, Edwards MK: Magnetic resonance imaging diagnosis of a cerebral aneurysm in an infant. Case report and review of the literature. Pediatr Neurosurg 1990/91;16:48–51.
14 Tekkok IH, Ventureyra ECG: Spontaneous intracranial hemorrhage of structural origin during the first year of life. Childs Nerv Syst 1997; 13:154–165.
15 Amacher AL, Drake CG: Posterior circulation aneurysms in young people. Neurosurgery 1981;8:315–320.
16 Lipper S, Morgan D, Krigman MR, Staab EV: Congenital saccular aneurysm in a 19-day-old neonate: Case report and review of the literature. Surg Neurol 1978;10:161–165.
17 Almeida GM, Pindaro J, Plese P, Bianco E, Shibata MK: Intracranial arterial aneurysms in infancy and childhood. Childs Brain 1977;3: 193–199.
18 Amacher AL, Drake CG: The results of operating upon cerebral aneurysms and angiomas in children and adolescents. Childs Brain 1979;5: 151–165.
19 Ventureyra ECG, Choo SH, Benoit BG: Super giant globoid intracranial aneurysm in an infant. A case report. J Neurosurg 1980;53:411–416.
20 Sedzimir CB, Robinson J: Intracranial hemorrhage in children and adolescents. J Neurosurg 1973;38:269–281.
21 Storrs BB, Humphreys RP, Hendrick EB, Hoffman HJ: Intracranial aneurysms in the pediatric age-group. Childs Brain 1982;9:358–361.
22 Amacher AL, Drake CG: Cerebral artery aneurysms in infancy, childhood and adolescence. Childs Brain 1975;1:72–80.
23 Liang-fu Z, Da-jie J: Large and giant intracranial aneurysms – diagnosis and surgical treatment. Chin Med J 1987;100:392–397.
24 Fried I, Jung KH, Spencer DD: Hippocampal pathology in patients with intractable seizures and temporal lobe masses. J Neurosurg 1992; 76:735–740.
25 Stewart RM, Samson D, Diehl J, Hinton R, Ditmore QM: Unruptured cerebral aneurysms presenting as recurrent transient neurologic deficits. Neurology 1980;30:47–51.
26 Bergen D, Bleck T, Ramsey R, Clasen R, Ristanovic R, Smith M, Whisler WW: Magnetic resonance imaging as a sensitive and specific predictor of neoplasms removed for intractable epilepsy. Epilepsia 1989;30:318–321.
27 Jones-Gotman M: Presurgical psychological assessment in children: Special tests. J Epilepsy 1990;3(suppl):93–102.