Spontaneous Disappearance of a Cerebral Arteriovenous Malformation

—Case Report—

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Abstract

A 63-year-old male presented with a cerebral arteriovenous malformation in the dominant motor area which subsequently disappeared spontaneously. He had suffered from epileptic attacks in his right extremities for 2 years before he first presented aged 53 years. He was treated with anticonvulsants and was followed up. Bleeding from the cerebral arteriovenous malformation occurred when he was 61 years old. Two years later, angiography revealed the complete disappearance of the cerebral arteriovenous malformation. Such spontaneous disappearance is rare, and the patient should be followed up as the nidus may reappear later.

Key words: arteriovenous malformation, spontaneous disappearance

Introduction

Spontaneous disappearance of a cerebral arteriovenous malformation (AVM) is caused by internal embolization, and occurs in 2.2% of all AVMs.¹⁹⁾ The 28 cases described in the literature were mostly associated with hemorrhage from the AVM.

We describe a case demonstrating complete spontaneous disappearance of AVM 12 years after the epileptic onset.

Case Report

A 53-year-old male presented with a 2-year history of epileptic attacks in his left extremities. He was admitted to the Shizuoka Red Cross Hospital in April 1979. There was no neurological deficit or any abnormality in the laboratory data. Skull x-ray films showed no abnormality, but computed tomography (CT) detected an area of slight high density in the left frontoparietal lobe, which was clearly enhanced with contrast medium, accompanied by high density spots

suggesting abnormal vessels (Fig. 1). Angiography revealed an AVM of about $4 \times 3.5 \times 3$ cm in the left frontoparietal lobe, fed mainly by distal trunks of the left anterior cerebral artery (ACA) and the middle cerebral artery (MCA) (Fig. 2). The main draining vein was the superior sagittal sinus (SSS). The AVM involved the dominant motor area, so surgical intervention involved grave risk of neurological deficits. He was therefore given anticonvulsant drugs and followed up.

Eight years later, when he was 61 years old, he was admitted again with deteriorating right hemiparesis and motor aphasia. CT revealed a high density area surrounded by an edematous low density area in the left frontoparietal lobe, implying bleeding from the nidus. Postcontrast CT showed the nidus and abnormal vessels were enhanced (Fig. 3). Angiography showed the AVM to be the same size as at the first examination (Fig. 4). However, the two feeding arteries appeared slightly spastic and the arterial flow to the anterior portion of the nidus seemed to have decreased compared with before. Additionally, the

Received May 23, 1995; Accepted July 25, 1995

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Fig. 1 First admission. Precontrast CT scans demonstrating a slight high density area in the left frontal lobe (*upper row*). Postcontrast CT scans showing enhancement (*lower row*).

Fig. 3 Second admission. Precontrast CT scans demonstrating a high density area implying hemorrhage (*upper row*). Postcontrast CT scans showing enhanced surrounding abnormal vessels (*lower row*).

Fig. 2 Left internal carotid angiograms at the first admission, showing the branches of the ACA and MCA feeding the AVM, and the SSS as the primary draining vein.

Fig. 4 Left internal carotid angiograms at the second admission, demonstrating the AVM as the same size as at the first examination. The two feeding arteries are slightly spastic and the arterial flow to the anterior portion of the nidus seems to be decreased compared with before.

drainage flow to the SSS also appeared to be somewhat slow and decreased. He was treated conservatively for 1 month, and then discharged with very slight right hemiparesis and motor aphasia. He was

treated with anticonvulsants after the second discharge, but had no epileptic attacks.

Fig. 5 Left internal carotid angiograms 10 years after the first angiography, demonstrating the complete disappearance of the AVM.

Two years later, digital subtraction angiography showed the disappearance of the AVM. Conventional angiography revealed the complete disappearance of the AVM (Fig. 5). The AVM had thus spontaneously disappeared 12 years after the epileptic onset, and 2 years after the hemorrhagic episode.

Discussion

The complete disappearance of an AVM is rare, with only 28 previous cases in the literature (Table 1).^{2-9,11-19,21,23-25} Of the 29 patients (including ours), 18 were males and 11 were females. The mean age at the disappearance was 36.5 years. The AVM was small in 13 cases, medium in 10, and large in three, with three unknown. The most common site was the parietal lobe, and a branch of the ACA or MCA often fed the nidus. The SSS was frequently involved as a draining vein. Twenty-two cases had only one feeding artery and the other seven had two or more feeding arteries. Fifteen patients had hemorrhagic episodes at the onset, and seven during the course. The mean follow-up period was 7.7 years.

Histological examination often reveals that the disappearance of an AVM is associated with thrombotic changes of the nidus.^{1,10} Most of the 29 cases had hemorrhagic episodes during the course, suggesting that the spontaneous disappearance of an AVM is closely related to hemorrhage from the nidus.¹⁰ Hematoma accompanied by perifocal edema and vasospasm of the feeding artery is the only explanation for the acute shrinkage of an AVM occurring after hemorrhage from the nidus. However, this does not explain the disappearance of AVMs long after the hemorrhagic episode.^{14,23} Lakke¹⁰ found

that minor hemorrhages occurred frequently in his patients during their courses, which induced small clots in the nidus and surrounding gliosis and possibly caused kinking of the feeding arteries with stagnancy and finally complete closure of the nidus. This theory is supported by the fact that thrombosed AVMs, incidentally discovered by CT, provide histological proof of microhemorrhages.¹⁰ Other possible mechanisms include atherosclerotic changes of vessels, coagulation diseases, antipregnancy drugs, etc.^{15,20,23} Vessels associated with AVMs may be thrombosed more easily than normal vessels.²² The flow velocity and direction are not constant in AVMs, but easily change according to the pressure balance in the nidus. A partial decrease of blood flow and velocity in the nidus leads to stagnant blood flow and local thrombosis, and finally thrombosis of the entire nidus. Such a process could be accelerated by many incidents: cerebral hemorrhage, microhemorrhage, atherosclerotic change, antipregnancy drugs, etc.^{10,14,15,23}

Thrombosed AVMs carry no risk of bleeding, but sometimes can be the foci of uncontrollable epilepsy, in which case surgical removal should be considered.¹⁹ However, Nukui *et al.*¹⁷ reported an interesting case in which the nidus reappeared on angiograms after its complete disappearance. This suggests the necessity of careful follow-up even after the complete disappearance of the nidus.

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Table 1 Cases of complete spontaneous disappearance of AVM

Author (Year)	Age*	Sex	Location	Size**	Feeding artery—draining vein	Bleeding	Follow-up period***
Höök and Johansen (1958) ⁹⁾	54	M	lt FP	S	ACA, MCA—SSS, ICV	+	21 yrs
Conforti (1971) ³⁾	37	F	lt P	S	ACA—galenic vein	+	3 yrs
Kawafuchi and Nukui (1971) [#]	26	M	lt P	S	ACA—SSS	+	3 mos
Eisenman <i>et al.</i> (1972) ⁴⁾	34	F	lt P	?	MCA—deep vein	+	4 yrs
Levine <i>et al.</i> (1973) ¹²⁾	46	M	lt P	M	MCA—?	+	6 yrs
Hansen and Sogaard (1976) ⁸⁾	24	F	posterior fossa	?	OA, VA—torcular vein	—	1 yr
Yamazaki <i>et al.</i> (1977) ²⁵⁾	61	M	rt P	S	ACA—SSS	+	2 mos
Castroviejo <i>et al.</i> (1977) ²⁾	3	F	rt P	L	AChA, PCA—SS	+	3 yrs
Mabe and Furuse (1977) ¹³⁾	1	F	posterior fossa	?	PCA, SCA, PICA—SS, TS	—	7 mos
Sartor (1978) ²¹⁾	24	F	lt P	S	AChA—?	+	11 yrs
Omojola <i>et al.</i> (1982) ¹⁸⁾	35	F	rt P	M	ACA—SSS	+	5 yrs
	15	F	lt P	S	AChA—galenic vein	?	3 yrs
Nehls and Pittman (1982) ¹⁶⁾	59	M	rt P	S	MCA—SSS	+	13 yrs
Wakai <i>et al.</i> (1983) ²⁴⁾	18	M	rt F	M	MCA—sphenoparietal sinus	+	1.5 yrs
Pasqualin <i>et al.</i> (1985) ¹⁹⁾	41	M	lt P	L	ACA, MCA—SSS	?	7 yrs
	56	M	lt P	M	MCA—SSS	+	9 yrs
	15	M	rt P	S	ACA—SSS	+	1 yr
	40	F	rt P	S	MCA—SSS	—	15 yrs
Leramo (1987) ¹¹⁾	22	M	rt T	S	AChA—basal vein	+	6 mos
Takano <i>et al.</i> (1987) ²³⁾	65	M	rt P	S	MCA—Labbé vein	+	3 days
Megison <i>et al.</i> (1989) ¹⁴⁾	25	M	lt PT	S	MCA—Labbé vein	+	1 yr
Minakawa <i>et al.</i> (1989) ¹⁵⁾	30	M	lt BG	M	AChA—?	+	17 yrs
	35	M	lt P	M	ACA—?	+	18 yrs
	57	M	rt P	S	ACA—?	+	25 yrs
	58	M	lt P	M	ACA—?	+	26 yrs
Ezura and Kagawa (1992) ⁵⁾	53	F	lt O	L	PCA, MMA, OA—SSS, ISS, galenic vein	+	6 yrs
Hamada and Yonekawa (1994) ⁷⁾	14	F	lt Thal	M	PChA—sylvian vein	—	6 yrs
Ha <i>et al.</i> (1994) ⁶⁾	47	M	lt FP	M	ACA—?	—	9 yrs
Present case	63	M	lt P	M	ACA, MCA—SSS	+	12 yrs

*Age at complete disappearance of AVM. **S: small (< 2 cm), M: medium (2–6 cm), L: large (> 6 cm). ***Time from onset to the last angiography which revealed complete disappearance of AVM. #Cited by Nukui *et al.*, *Neurol Med Chir (Tokyo)* 22: 125–132, 1982.¹⁷⁾ AChA: anterior choroidal artery, BG: basal ganglia, F: frontal, FP: frontoparietal, ICV: internal cerebral vein, ISS: inferior sagittal sinus, MMA: middle meningeal artery, O: occipital, OA: occipital artery, P: parietal, PCA: posterior cerebral artery, PChA: posterior choroidal artery, PICA: posterior inferior cerebellar artery, PT: parietotemporal, SCA: superior cerebellar artery, SS: straight sinus, T: temporal, Thal: thalamus, TS: transverse sinus, VA: vertebral artery.

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