

## CASE REPORT

# SPONTANEOUS THROMBOSIS IN CEREBRAL ARTERIOVENOUS MALFORMATION AFTER MINOR SURGICAL MANIPULATION: A CASE REPORT

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The complete or partial spontaneous disappearance of a cerebral arteriovenous malformation is rare. Spontaneous thrombosis in large arteriovenous malformations has been reported in only two cases. We report the case of a large arteriovenous malformation in a 17-year-old man, that thrombosed spontaneously after minor surgical manipulation and review the literature and some possible mechanisms for spontaneous thrombosis of arteriovenous malformation.

**Keywords** arteriovenous malformation mechanism spontaneous disappearance thrombosis

### Introduction

Arteriovenous malformation (AVM) of the brain may manifest various degrees of spontaneous obliteration when examined angiographically after a time lag; however, complete or partial spontaneous disappearance of a cerebral AVM is rare.<sup>1-6</sup>

To our knowledge, 30 cases of complete spontaneous regression of AVM, documented by angiography, prior to and after thrombosis, have been reported so far. Here we report the case of a young man with AVM, that thrombosed spontaneously after minor surgical manipulation. Follow-up data (angiography, computerized tomography [CT] and magnetic resonance imaging [MRI]) confirmed thrombosis of the lesion. Only two previous reported cases had large AVMs, more than 6 cm in size;<sup>7-9</sup> our case is the third. The two earlier reported cases were in patients older than 20 years.

### Case Report

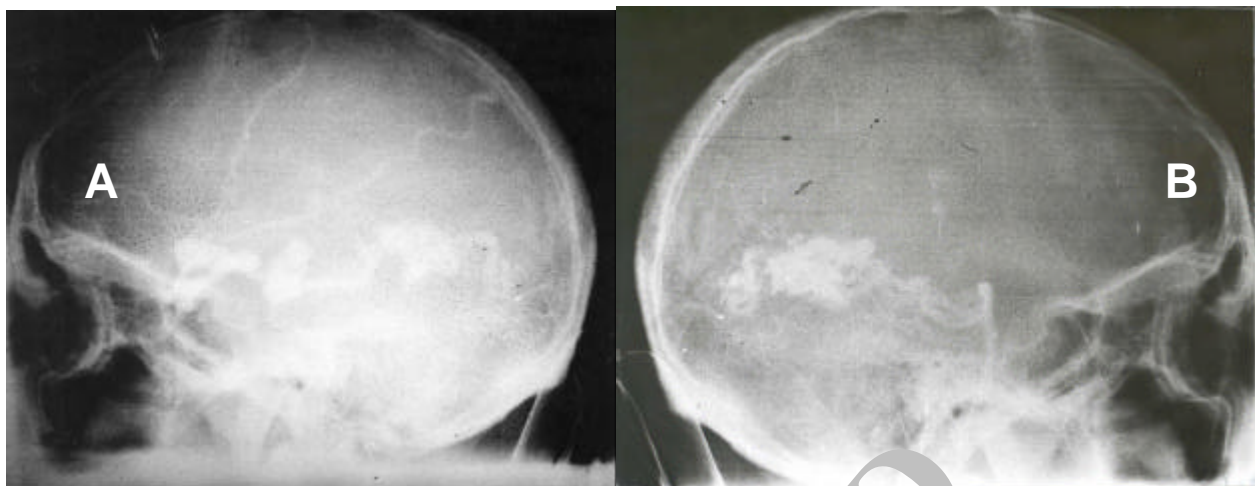
A 17-year-old man was admitted to our hospital

for loss of consciousness following a seizure in March 1998. There was no history of seizure and his family history was also not helpful. Two hours after the seizure, he was conscious and spoke coherently. He complained of headache. There was no evidence of motor weakness. Neurological examination and plain skull films were all normal. Hematological data including those for coagulation did not indicate any abnormality. CT scan revealed a local subarachnoid hemorrhage on the left side of the basal cistern and a cystic image in the deep frontotemporal area. Angiography demonstrated a huge AVM with two vascular tufts, one in the deep left frontotemporal region that was supplied by the carotid system and the other in the left paratrigenal area supplied by the basilar system; blood was drained by the vein of Galen (Figure 1).

Contrast-enhanced CT scan and MRI showed abnormal dilated tortuous vessels on the left from the deep frontotemporal to the paratrigenal area and also a cystic image in the deep frontotemporal area without enhancement that compressed the left frontal horn and caused mild dilation of the left lateral ventricle, probably due to narrowing of the foramen of Monro (Figure 2).

Four months after the diagnosis, the patient underwent surgery via a left parietooccipital

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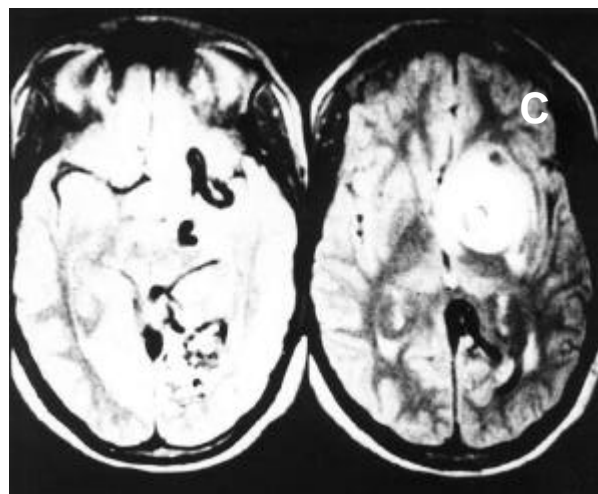
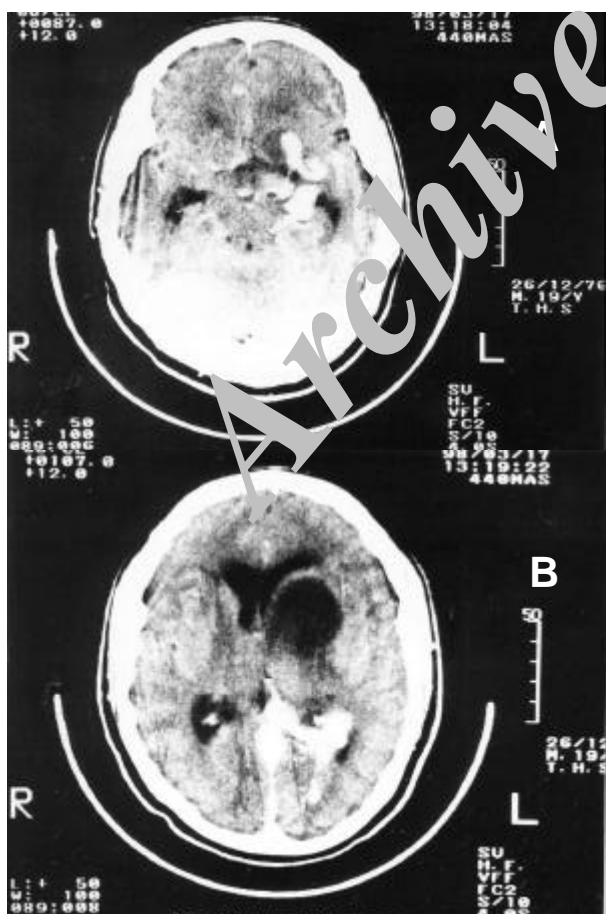


**Figure 1.** Conventional angiography demonstrates a huge AVM from the deep left frontotemporal area to the left paratrigenal area (A, carotid and B, vertebral angiography)

craniotomy using the transcortical approach, through the posterior parietal area with the patient in the right lateral position. The transcortical dissection proceeded through a relatively non-eloquent area until the trigonal area was reached. Here we encountered a large abnormal vasculature and, fearing that surgical intervention would cause a neurological deficit, a piece of surgical was placed on the surface of the AVM and the

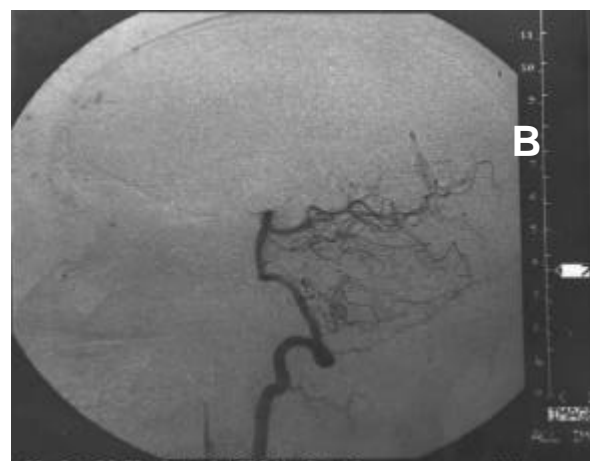
operation was finished. The patient was scheduled for arterial embolization. We approached the cystic lesion via a left frontal craniotomy through the 12 area to evacuate the cyst. There was no abnormal tumor-like lesion but a fine biopsy was taken from the wall of the cyst. The cyst fluid was xanthochrom and no abnormal cells were found in cytological study. Biopsy of the wall of the cyst was also normal. Repeat angiography, performed 5 months later to plan embolization, showed complete absence of the lesion (Figure 3).

Follow-up MRI one year after the operation showed no evidence of flow phenomenon and abnormal tortuous vessels in the area that were previously seen (Figure 4). In fact, there was evidence of thrombosis at the site where the AVM



**Figure 2.** Abnormal dilated tortuous vessels on the left side from the deep frontotemporal to the paratrigenal area in contrast-enhanced computerized tomography (A and B) and MRI (C). A cystic image without enhancement is seen in the deep frontotemporal area (B).

## Spontaneous Thrombosis of AVM



**Figure 3.** Complete absence of AVM is seen on angiography 8 months after the initial diagnosis (A left carotid angiography; B and C vertebral angiography).

lesions at sites not directly related to the AVM have been reported as one cause of this phenomenon.<sup>14</sup> Mendelow et al reported that cerebral AVMs in younger patients tend to increase in size.<sup>15</sup> The same results were described by Minakawa et al<sup>10</sup> and Kraysenbuhl.<sup>16</sup> Spontaneous regression of cerebral AVMs has been mainly observed in older patients, except for some cases in infancy.<sup>12, 17, 18</sup> Only a few authors have documented this phenomenon before the age of 20 years.<sup>5, 6, 19, 20</sup> Our case fell within this age range. Waltimo observed changes in the size of AVMs angiographically over a median period of 44 months, and reported that small AVMs showed a rapid increase in size, while large malformations apparently tended to decrease in size.<sup>21</sup> On literature review we found only two cases of complete spontaneous thrombosis of large (>6 cm) AVMs;<sup>7-9</sup> our case is the third. Banker cited congenital heart disease and systemic and focal infection as the causes of thrombosis in AVM.<sup>22</sup> Subarachnoid or intracerebral hemorrhage with subsequent mass effect was the most frequent cause of spontaneous thrombosis of AVMs, because mass effect, vasospasm and edema decrease blood flow leading to thrombosis.<sup>1, 9, 18, 20, 23, 24</sup> Any iatrogenic manipulation may be a factor in thrombosis of AVM; the range of manipulation may be mild as in our case or extensive as documented in some other cases.<sup>7, 19</sup> Hemodynamic changes due to any cause and turbulent blood flow in AVMs have been suggested as possible mechanisms of thrombosis in AVMs.<sup>12, 13, 17, 28</sup> Arteriosclerosis may be a contributing factor in spontaneous thrombosis of AVMs.<sup>26, 27</sup> An

had been. The cystic mass lesion had decreased in size with evidence of deposition of hemosiderin around it.

### Discussion

Little is known, however about changes in the size of these lesions, in the natural course or after treatment.<sup>10</sup> Abdulrauf et al reported the incidence of spontaneous thrombosis of cerebral AVM to be less than %1.<sup>7</sup> It has been reported that a single draining vein is seen in 86% of AVM and 100% of AVM that disappeared spontaneously. The time-lapse from diagnosis to disappearance of AVM has varied from 2 months to 26 years, according to the literature.<sup>5, 7, 9-12</sup> In our case this period was 8 months. We summarized the etiology of spontaneous disappearance of intracranial AVM (Table 1). According to Minakawa et al, regression is most probable in small AVMs that are fed by a single vessel.<sup>10</sup> Superficial AVMs have a greater tendency toward regression than deep ones. AVM in our case was in the deep left hemisphere. Abnormal vessels and tortuous blood vessels might have a greater propensity for thrombosis than adjacent normal vessels.<sup>6, 13</sup> Associated vascular



**Figure 4.** Follow-up MRI shows no evidence of flow phenomenon and abnormal tortuous vessels. The arrow shows the surgical manipulating site

embolus could initiate thrombosis,<sup>2</sup> and clot embolism from the feeding artery has been suggested as a possible cause of partial regression of AVM.<sup>5</sup> Radiation therapy and changes in cerebral circulation may also influence the regression of AVM.<sup>6,14</sup> Other possible mechanisms suggested for thrombosis of AVM are hypercoagulable state in hormonally active females and drugs such as oral contraceptives.<sup>7,11,28</sup>

Hansen and Sogaard reported spontaneous thrombosis of an extra and intracranial AVM in the posterior fossa, which disappeared within 15 months after delivery (cesarean section) and 11 months after consumption of oral contraceptives.<sup>11</sup> In our patient, there was no intracerebral hematoma, therefore mass effect could not have been the cause of spontaneous thrombosis. On the other hand, the patient was too young to have arteriosclerosis and therefore this could not be a significant factor for the thrombosis. We suggest that minor surgical manipulation, protection of AVM by surgical and small hemorrhage in a venule or vein resulting in reduced blood flow in other vasculature of the AVM could have progressed to thrombosis of the AVM. Omojola et al reported two cases of spontaneous regression of intracranial AVMs; both were associated with cerebral mass lesions.<sup>5</sup> One patient had a large low-density structure deep in the left hemisphere and enhancing mural nodule. Surgical exploration and biopsy showed a low-grade cystic astrocytoma. The other patient had a large contrast-

**Table 1.** The etiology of spontaneous disappearance of intracranial AVM

Anatomical and pathological properties	
Single draining and single feeding artery <sup>10</sup>	
Superficial location <sup>10</sup>	
Abnormal vessel of AVM <sup>6,13</sup>	
Tortuous blood vessel <sup>6,13</sup>	
Associated vascular disease <sup>14</sup>	
Size of AVM <sup>7,8,9</sup>	
Infection (systemic and local) <sup>15</sup>	
Mechanical	
Intracerebral hemorrhage (mass effect, edema, and vasospasm) <sup>1,9,16-19</sup>	
Iatrogenic manipulation <sup>7,20</sup>	
Hemodynamic change and turbulent blood flow <sup>12,13,21,22</sup>	
Aging process; arteriosclerosis <sup>23,24</sup>	
Congenital vascular disease and thromboembolism <sup>3,5,15</sup>	
Other predisposing factors: radiation, <sup>6,14</sup> coagulopathy, <sup>7,11,25</sup> and drugs <sup>11</sup>	

enhancing mass lesion surrounded by a low-density zone in the right occipital region. Association of AVMs with tumor-like mass lesions has also been reported by Welcker and Seidel,<sup>29</sup> Crowell et al<sup>30</sup> and Heffner et al.<sup>31</sup> In our case, the cystic mass lesion in the deep left frontotemporal region was not enhanced in contrast study and cytological study of the fluid and biopsy from the wall of the lesion were normal. We thought that the cystic mass lesion in the deep frontotemporal area was due to extravasation of large vessels of the AVM and thrombosis of the AVM led to disappearance of the cyst.

## References

- Conforti P. Spontaneous disappearance of cerebral arteriovenous angioma. *J Neurosurg.* 1971; **34**: 432 – 4.
- Hatva E, Jaaskelainen J, Hirvonen H, et al. The endothelial cell-specific receptor tyrosine kinase is upregulated in the vasculature of arteriovenous malformation. *J Neuropathol Exp Neurol.* 1996; **55**: 1124 – 33.
- Kushner J, Alexander E Jr. Partial spontaneous regressive arteriovenous malformation; case report with angiographic evidence. *J Neurosurg.* 1970; **32**: 360 – 6.
- Magidson MA, Weinberg PE. Spontaneous closure of a dural arteriovenous malformation. *Surg Neurol.* 1976; **6**: 107 – 10.
- Omojola MF, Fox AJ, Vinuela FV, et al. Spontaneous regression of intracranial arteriovenous malformations. Report of three cases. *J Neurosurg.* 1982; **57**: 818 – 22.
- Svien HJ, Peserico L. Regression in size of arteriovenous anomaly. *J Neurosurg.* 1960; **17**: 493 – 6.
- Abdulrauf SI, Malik GM, Awad IA. Spontaneous angiographic obliteration of cerebral arteriovenous malformations. *Neurosurgery.* 1999; **44**: 280 – 8.
- Ezura M, Kagawa S. Spontaneous disappearance of a huge

## Spontaneous Thrombosis of AVM

- cerebral arteriovenous malformation: case report. *Neurosurgery*. 1992; **30**: 595 – 9.
- 9 Pasqualin A, Vivenza L, Rosta R, et al. Spontaneous disappearance of intracranial arteriovenous malformations. *Acta Neurochir (Wien)*. 1985; **76**: 50 – 7.
  - 10 Minakawa T, Tanaka R, Koike T, et al. Angiographic follow-up study of cerebral arteriovenous malformations with reference to their enlargement and regression. *Neurosurgery*. 1989; **24**: 68 – 74.
  - 11 Hansen JH, Sogaard I. Spontaneous regression of an extra- and intracranial arteriovenous malformation. Case report. *J Neurosurg*. 1976; **45**: 338 – 41.
  - 12 Mabe H, Furuse M. Spontaneous disappearance of a cerebral arteriovenous malformation in infancy. Case report. *J Neurosurg*. 1977; **46**: 811 – 5.
  - 13 Dyck P. Spontaneous thrombosis of an arteriovenous malformation. *Neurosurgery*. 1977; **1**: 287 – 90.
  - 14 Sukoff MH, Barth B, Moran T. Spontaneous occlusion of a massive arteriovenous malformation—case report. *Neuroradiology*. 1972; **4**: 121 – 3.
  - 15 Mendelow AD, Erfurth A, Grossart K, et al. Do cerebral arteriovenous malformations increase in size? *J Neurol Neurosurg Psychiatry*. 1987; **50**: 980 – 7.
  - 16 Krayenbuhl HA. Angiographic contribution to the problem of enlargement of cerebral arteriovenous malformations. *Acta Neurochir (Wien)*. 1977; **36**: 215 – 42.
  - 17 Nehls DG, Pittman HW. Spontaneous regression of arteriovenous malformations. *Neurosurgery*. 1982; **11**: 776 – 80.
  - 18 Takano S, Nose T, Maki Y, et al. Spontaneous occlusion of a cerebral arteriovenous malformation—report of a case [in Japanese]. *No Shinkei Geka*. 1987; **15**: 991 – 6.
  - 19 Hamada J, Yonekawa Y. Spontaneous disappearance of a cerebral arteriovenous malformation: case report. *Neurosurgery*. 1994; **34**: 171 – 3.
  - 20 Wakai S, Chen CH, Wu KY, et al. Spontaneous regression of a cerebral arteriovenous malformation. Report of a case and review of the literature. *Arch Neurol*. 1983; **40**: 377 – 80.
  - 21 Waltimo O. The change in size of intracranial arteriovenous malformations. *J Neurol Sci*. 1973; **19**: 21 – 7.
  - 22 Banker BQ. Cerebral vascular disease in infancy and childhood. Occlusive vascular diseases. *J Neuropathol Exp Neurol*. 1961; **20**: 127 – 40.
  - 23 Nukui H, Miyagi O, Tamada J, et al. Long-term follow-up study by cerebral angiography in cases with arteriovenous malformation of the brain. With special reference to spontaneous disappearance of arteriovenous malformation in cerebral angiography [in Japanese]. *Neurol Med Chir (Tokyo)*. 1982; **22**: 125 – 32.
  - 24 Sartor K. Spontaneous closure of cerebral arteriovenous malformations demonstrated by angiography and computed tomography. *Neuroradiology*. 1978; **15**: 95 – 8.
  - 25 Terao H, Hori T, Matsutani M, et al. Detection of cryptic vascular malformations by computerized tomography. Report of two cases. *J Neurosurg*. 1979; **51**: 546 – 51.
  - 26 Pool JL, Potts DJ. *Aneurysms and Arteriovenous Anomalies of the Brain. Diagnosis and Treatment*. New York: Harper; 1965; 326 – 73.
  - 27 Sasaki U, Itagaki T, Shimizu H, et al. Almost complete regression of cerebral arteriovenous malformation—report of a case [in Japanese]. *No Shinkei Geka*. 1976; **4**: 175 – 81.
  - 28 Shuey HM, Day AL, Quisling RG, et al. Angiographically cryptic cerebrovascular malformations. *Neurosurgery*. 1979; **5**: 476 – 9.
  - 29 Welcker ER, Seidel K. Combination of an arteriovenous aneurysmatic angioma with an astrocytoma. *Dtsch Z Nervenheilkd*. 1966; **189**: 231 – 9.
  - 30 Crowell RM, DeGirolami U, Sweet WH. Arteriovenous malformation and oligodendroglioma. Case report. *J Neurosurg*. 1975; **43**: 108 – 11.
  - 31 Heffner RR, Porro RS, Deck MD. Benign astrocytoma associated with arteriovenous malformation. Case report. *J Neurosurg*. 1971; **35**: 229 – 33.