

Recurrence of Completely Excised Arteriovenous Malformation : Review of the Literatures and Possible Explanation

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= Abstract = The authors report a case of recurrent arteriovenous malformation (AVM), which has been completely removed and disappeared on postoperative angiography four years ago. Initially, at the age of nine years, she presented intracerebral hematoma. The angiography demonstrated an aneurysm and AVM nidus located in the posterior frontal area, fed by the branches of the anterior and middle cerebral arteries. Eleven days after the ictus, rebleeding occurred, so left fronto-parietal craniotomy was done emergently. The hematoma and AVM nidus with aneurysm were removed. Her postoperative course was uneventful and postoperative angiography showed that the AVM had been completely excised. However, four years later, sudden focal motor seizure on right leg developed. Magnetic resonance(MR) images and angiography demonstrated that the AVM reappeared on the previously operated region. Extirpation of the recurrent AVM was carried out. We do emphasize a long term follow-up MR images or a repeated angiography is essential to confirm the complete absence after excision of the AVM.

Key words : *Arteriovenous malformation, Hematoma, Angiography, Magnetic resonance (MR) images, Reappear, Extirpation*

INTRODUCTION

Cerebral angiography has been considered the best method for diagnosing the AVM, and intra- or postoperative angiography was strongly recommended for confirming complete excision of an AVM (Lazar *et al.*, 1971 ; Smith, 1977).

After complete removal of the AVM which was proved by postoperative angiography, reported cases of the recurrence of AVM on previously operated region are very rare (Fuwa *et al.*, 1988 ; Higuchi *et al.*, 1991 ; Patil, 1982). We herein present such a case and call attention that a long term follow-up MR images or a repeated angiography is essential.

CASE REPORT

First admission

This 9-year-old girl suffered abrupt vomiting

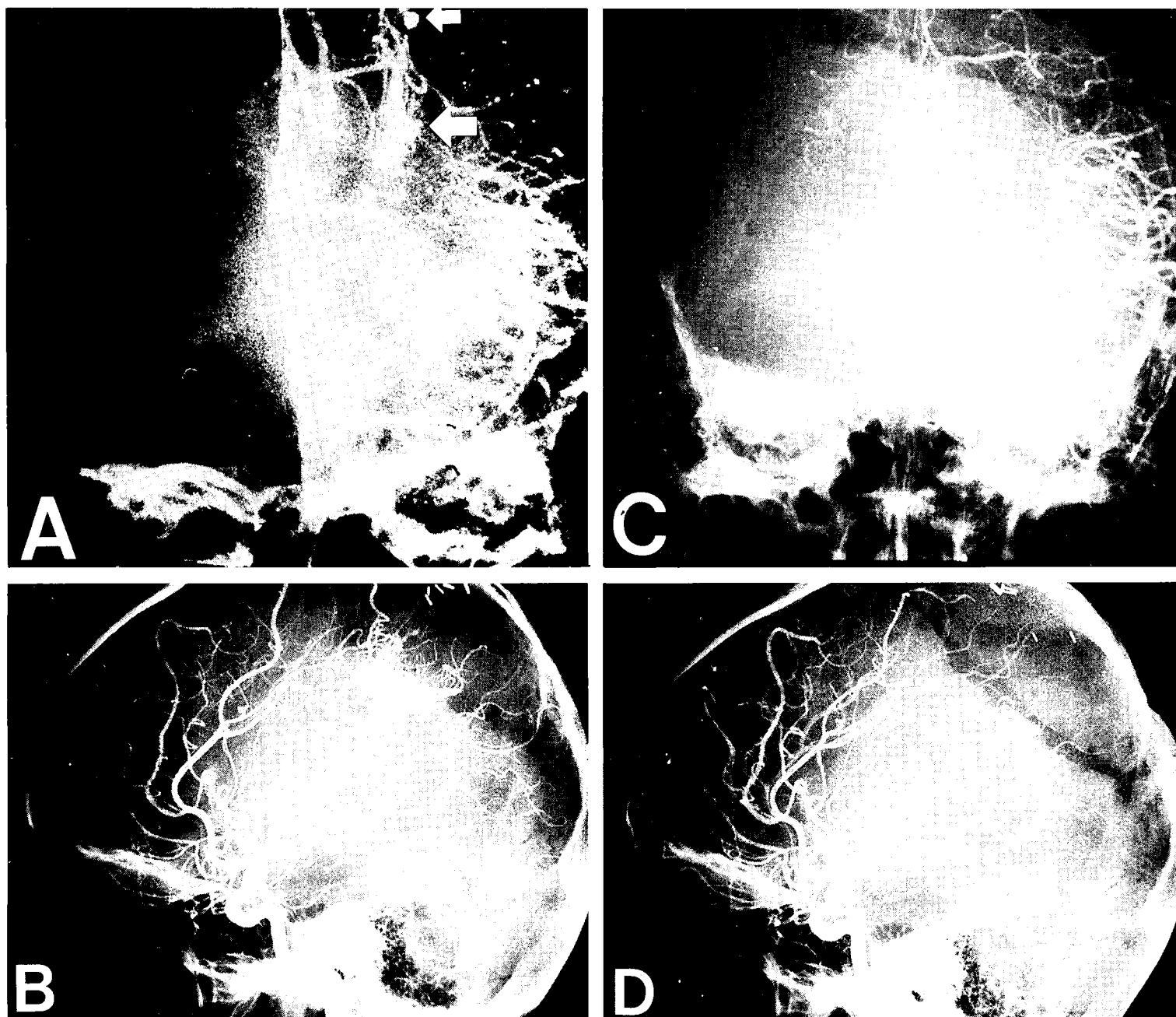


Fig. 1. A, B, C and D: Left carotid angiography after the initial hemorrhage demonstrates the aneurysm (small arrow) and the AVM (large arrow) fed by left pericallosal artery, left callosomarginal artery, and pre-rolandic branch of left middle cerebral artery (A, B). Sixteen days after operation, angiography demonstrates complete removal of the aneurysm and the AVM, and the presence of hemoclip (C, D).

and dysarthria. Several hours later, the level of consciousness deteriorated. On admission, she was slightly drowsy but properly oriented. Neurological examination revealed right hemiparesis (Grade II/V), pathologic reflex on right side and neck stiffness. The computerized tomography (CT) showed intracerebral hematoma in left posterior frontal area. Carotid angiography demonstrated an aneurysm and AVM nidus on posterior frontal

area. It was fed by widened left pericallosal artery, left callosomarginal artery, and pre-rolandic branch of left middle cerebral artery (Fig. 1. A,B) and drained to superior sagittal sinus.

Eleven days after the onset, sudden severe headache occurred and followed by deterioration of consciousness. The brain CT showed enlarged hematoma, so emergent left fronto-parietal craniotomy was done. The hematoma and the AVM ni-

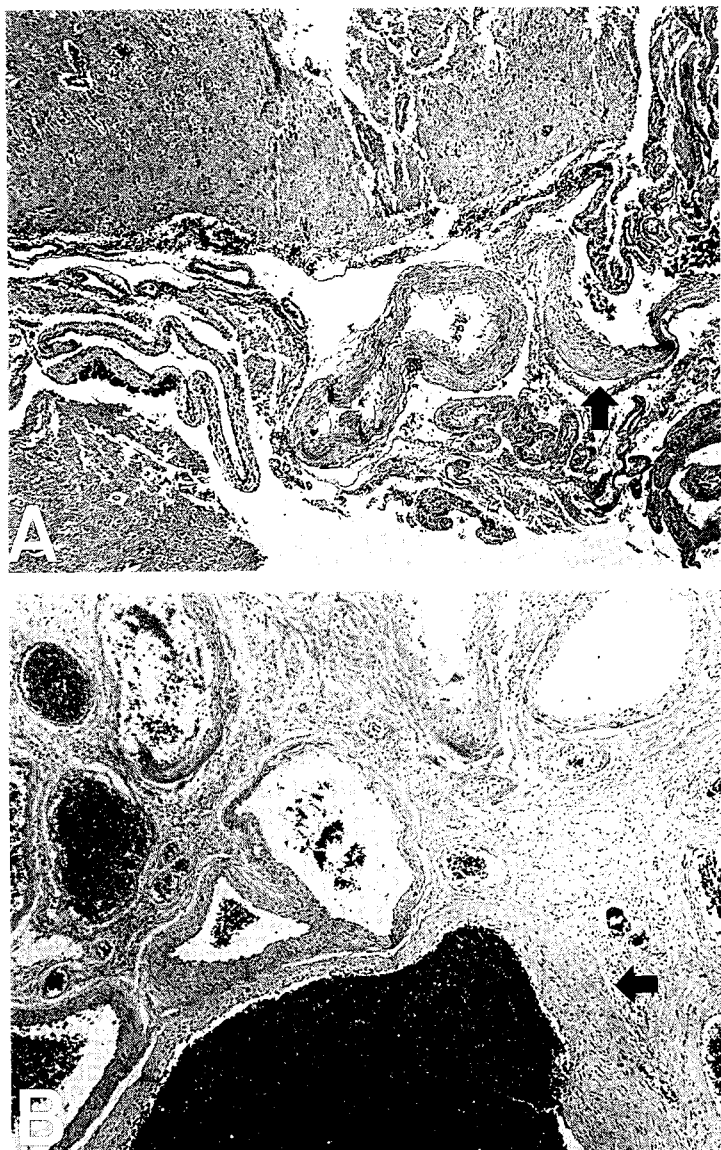


Fig. 2. A & B: Histologic findings. The biopsy revealed small artery and thickened vein (arrow).

cus with aneurysm were removed. Sixteen days after the operation, postoperative angiography demonstrated complete excision of the AVM and aneurysm (Fig. 1. C,D), and she was discharged with right hemiparesis (Grade III/V).

The histologic examination revealed small artery and thickened vein (Fig. 2. A).

Second Admission

During follow-up, her school performances was good and the right hemiparesis improved up to Grade IV/V. Four years after the excision of AVM, sudden focal motor seizure on right leg developed. MR images (2.0T; Goldstar, Korea) showed tortuous signal void mass with surrounding high

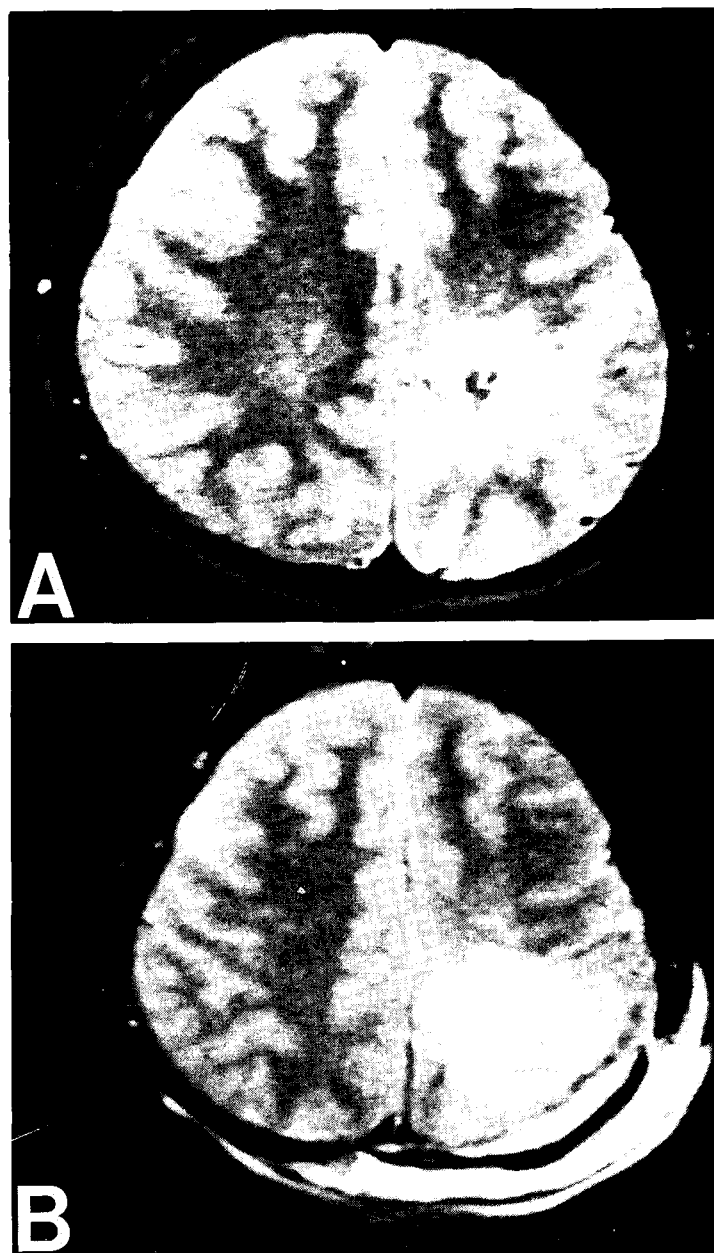


Fig. 3. A & B: Magnetic resonance images. T2-weighted magnetic resonance image shows signal void mass with surrounding high signal intensity lesion on previous operated region (A), and disappearance of signal void mass (B).

signal intensity lesion on previous operated region (Fig. 3. A). Angiography demonstrated that the AVM has reappeared. It was larger than former AVM and fed by left callosomarginal and pericallosal arteries and pre-rolandic branch of left middle cerebral artery (Fig. 4. A,B). Second operation was done, and postoperative angiography (Fig. 4. C,D) and MR images (Fig. 3. B) showed complete excision of the recurrent AVM.

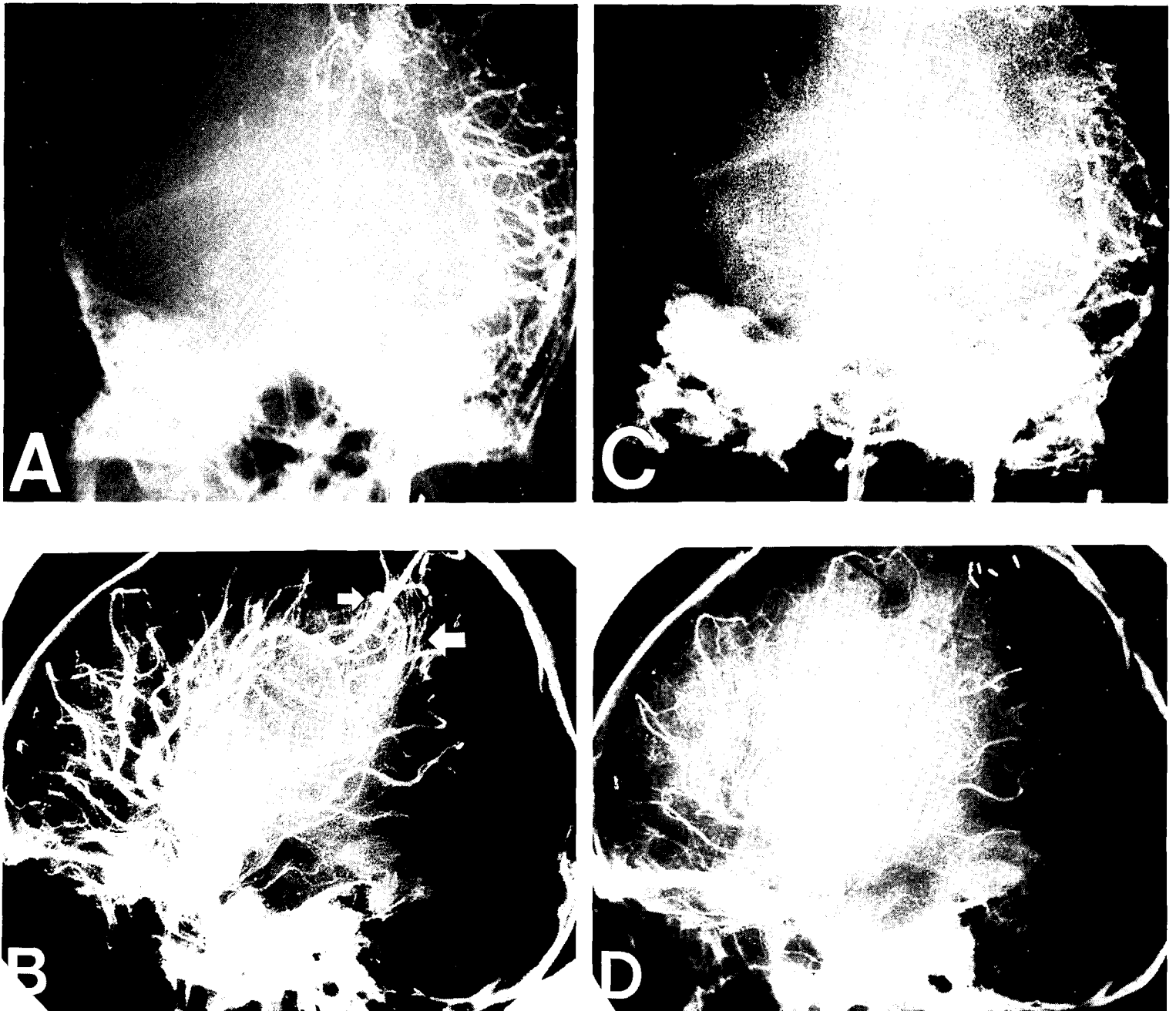


Fig. 4. A, B, C and D: Left carotid angiography. Angiogram after the focal motor seizure demonstrates reappearance of the AVM (A, B). Fourteen days after operation, angiography demonstrates complete removal of the recurrent AVM (C, D).

The histologic examination revealed same nature of previous examination (Fig. 2. B).

DISCUSSION

Cerebral AVM is not a true neoplasm, but a congenital development of vessels during an early embryonic stage. Therefore, theoretically there could not be true growth of AVM. But, many cases showing progressive enlargement of cerebral AVM have been reported (Hook and Johanson,

1958; Krayenbühl, 1977; Morioka *et al.*, 1988; Sano *et al.*, 1978; Spetzler and Wilson, 1975; Waitimo, 1973). Several possible mechanisms for growth or enlargement of AVM were summarized as followings (Morioka *et al.*, 1988): (1) the progressive vascular dilatation or pseudoaneurysmal formation occurring as a result of repeated occult hemorrhages that destroy the extravascular neural tissues (Krayenbühl, 1977; Waitimo, 1973). (2) the continuous hemodynamic stress causing progressive enlargement of the thin walled, undiffer-

entiated vessels forming the fistulous shunts of the AVM (Hook and Johanson, 1958), (3) the presence of more room for growth, for example, in the sylvian fissure (Hook and Johanson, 1958; Waltimo, 1973), (4) the autonomic growth (Krayenbühl, 1977; Spetzler and Wilson, 1975), and (5) the presence of a “reserve” nidus, which was not opacified initially and becomes visible angiographically after a change in hemodynamics (Sano *et al.*, 1978). But, there are a few reports of spontaneous disappearance of cerebral AVM (Golden and Kramer, 1978; Hook and Johanson, 1958; Levine *et al.*, 1973). It was explained by spontaneous thrombosis or emboli which occlude feeding vessels (Conforti, 1971), and atherosclerosis (Levine *et al.*, 1973).

An AVM that is verified operatively and/or pathologically but is not detected angiographically is usually called an angiographically occult AVM. Several mechanisms why angiography have failed to demonstrate a vascular malformation were speculated and could be summarized. There are (1) a small size (Becker *et al.*, 1979; Cohen *et al.*, 1982), (2) the partial or complete thrombosis caused by spontaneously or secondary to hemorrhage (Becker *et al.*, 1979; Chin and Harper, 1983; Cohen *et al.*, 1982; Davidoff, 1958; Golden and Kramer, 1978; Kramer and Wing, 1977; Patil, 1982), (3) the compression by adjacent hematoma and edema or destruction in the time of hemorrhage (Cohen *et al.*, 1982; Kramer and Wing, 1977), and (4) the spasm of the feeding vessels (Patil, 1982).

The recurrence of the AVM disappeared on postoperative angiography was very rarely reported (Fuwa *et al.*, 1988; Higuchi *et al.*, 1991; Patil, 1982). In the case of Higuchi *et al.* (Higuchi *et al.*, 1991), small abnormal vessels on postoperative angiography were noted retrospectively. But it was suggested that small abnormal vascular channels not demonstrated on postoperative angiography might have grown to nidus some years later (Fuwa *et al.*, 1988; Patil, 1982). Our case would be a well recognized phenomenon namely, failure of angiography to demonstrate an AVM following surgical excision with subsequent rediscovery of the AVM. So, we might consider that the small thrombosed vascular malforma-

tions, which located in the surrounding area of the AVM and was not visualized on postoperative angiography, could have grown to the recurrent AVM four years later. Therefore we suggest that a long term follow-up MR images or a repeated aggressive angiography is essential to confirm the complete removal after excision of the AVM, particularly in presentation with hemorrhage and in difficulty on hemostasis. Because in presentation with hemorrhage, the small vascular malformations could not be visualized on angiography due to compression and/or thrombosis.

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