JOURNAL OF NEUROSURGERY:
Case Lessons

J Neurosurg Case Lessons 3(19): CASE2248, 2022
DOI: 10.3171/CASE2248

Pseudo-residual nidus after arteriovenous malformation surgery: illustrative case

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BACKGROUND Intraoperative indocyanine green video angiography (ICG-VA) is useful for determining the extent of lesion removal during cerebral arteriovenous malformation (AVM) surgery. The authors described a case of surgical removal of an AVM presenting with early venous filling mimicking a residual nidus on intraoperative ICG-VA.

OBSERVATIONS A 7-year-old girl experienced a sudden disturbance of consciousness. Computed tomography revealed right frontal intracerebral hemorrhage. Digital subtraction angiography showed a Spetzler-Martin grade 1 AVM in the right frontal lobe. The patient received surgical removal of the AVM after endovascular embolization. After removal of the nidus, the first intraoperative ICG-VA revealed early venous filling of the cortex around the excision cavity. Additional resection of the cortex around this area was performed. Histopathological examination of the lesion revealed a dilated normal vascular structure without an AVM.

LESSONS Early venous filling in the surrounding brain tissue after AVM removal does not necessarily indicate a residual nidus. The need for additional resection of the lesion depends on the eloquence of the area.

https://thejns.org/doi/abs/10.3171/CASE2248

KEYWORDS arteriovenous malformation; brain; perinidal microvasculature; residual nidus; neurosurgery

The residual nidus after cerebral arteriovenous malformation (AVM) surgery can cause postoperative bleeding; therefore, its accurate diagnosis is crucial for preventing this serious complication.[1] Indocyanine green video angiography (ICG-VA) is a routine procedure during AVM surgery to confirm the complete removal of AVM.[2–4] We report the first case of AVM with early venous filling mimicking residual nidus, as observed on ICG-VA during surgery and review of the pathophysiology.

Illustrative Case

A 7-year-old girl presented to our hospital's emergency department with a report of disturbed consciousness. Neurological examination revealed 6 points (E1V1M4) on the Glasgow Coma Scale (GCS) and left hemiparesis. Brain computed tomography revealed a right frontal intracerebral hemorrhage (Fig. 1A). She received emergency craniotomy and hematoma evacuation. The day after surgery, her consciousness level recovered to 15 points on the GCS without hemiparesis. Right carotid angiography showed a right frontal Spetzler-Martin grade 1 AVM (Fig. 1B). The patient received presurgical endovascular embolization of the AVM using the Onyx liquid embolic system. After embolization, right carotid angiography revealed an 80% reduction in the nidus (Fig. 1C). The patient received surgical removal of the AVM 5 days after endovascular embolization.

As in the standard procedure, we performed the following steps during surgery: nidus confirmation, feeder extirpation, nidus dissection, drainer occlusion, and nidus removal. After the procedure, a part of the cerebral cortex around the excision cavity was slightly hyperemic (Fig. 2A). The first ICG-VA revealed early venous filling in the cortex (Fig. 2B). Because residual nidus was suspected, we additionally resected the hyperemic brain area and vein. The second ICG-VA showed no abnormalities (Fig. 2C). Intraoperative digital

ABBREVIATIONS AVM = arteriovenous malformation; DSA = digital subtraction angiography; GCS = Glasgow Coma Scale; ICG-VA = indocyanine green video angiography; PDCN = perinidal dilated capillary network.

INCLUDE WHEN CITING Published May 9, 2022; DOI: 10.3171/CASE2248.
SUBMITTED January 28, 2022. **ACCEPTED** February 23, 2022.

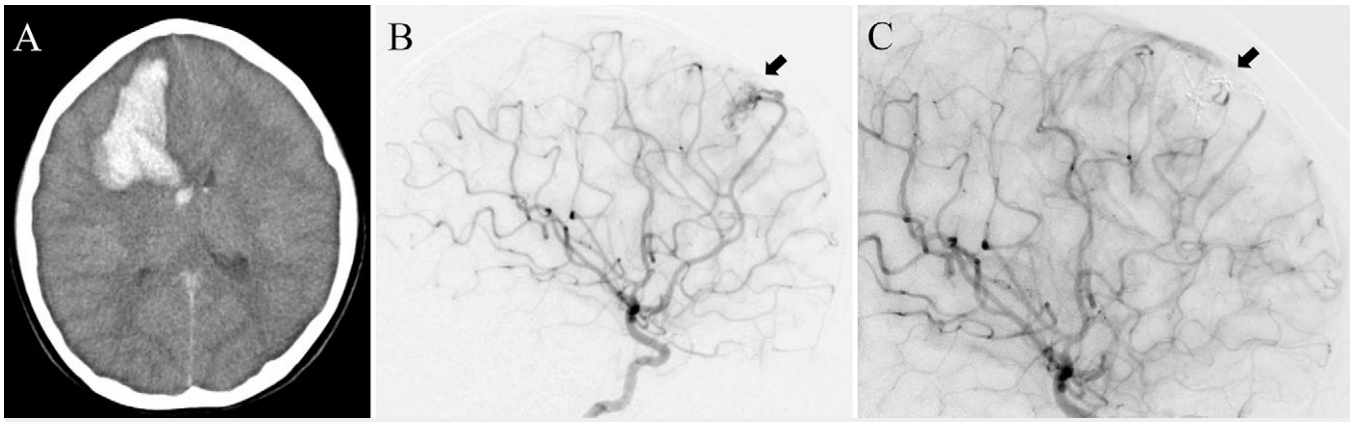

FIG. 1. Computed tomography (CT) on admission and DSA. **A:** CT shows a right frontal intracerebral hemorrhage. **B:** Right internal carotid artery angiogram, lateral view, shows right frontal arteriovenous malformation (*arrow*). **C:** Post–endovascular embolization right internal carotid artery angiogram shows partial occlusion of the nidus (*arrow*).

subtraction angiography (DSA) revealed total removal of the nidus (Fig. 2D). Histopathological examination of the additionally removed specimen revealed no lesions suggestive of an AVM, but arterioles and capillaries with abnormal dilation were observed (Fig. 3A and B). Follow-up DSA performed 1 year after surgery showed no recurrence of brain AVMs.

Discussion

Observations

During surgery for brain AVMs, residual nidus can cause postoperative intracerebral hemorrhages. Solomon et al.[1] reported a surgical case of posterior temporal AVM with a small residual nidus detected using postoperative DSA. The patient experienced postoperative

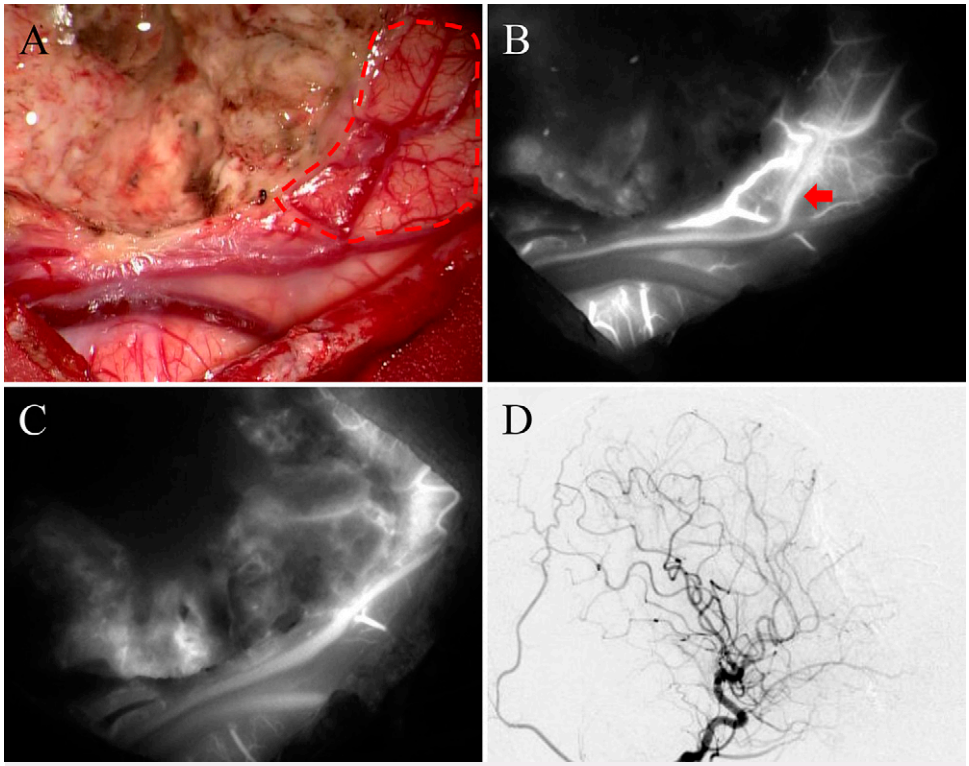

FIG. 2. Intraoperative findings. **A:** Surgical microscopic view shows hyperemic cortex around the excision cavity after complete removal of the nidus (*red dotted line*). **B:** The first ICG-VA shows early venous filling from the cortex (*red arrow*). **C:** The second ICG-VA after additional resection of the cortex shows no abnormal flow pattern. **D:** Intraoperative right carotid artery angiogram shows complete removal of the arteriovenous malformation.

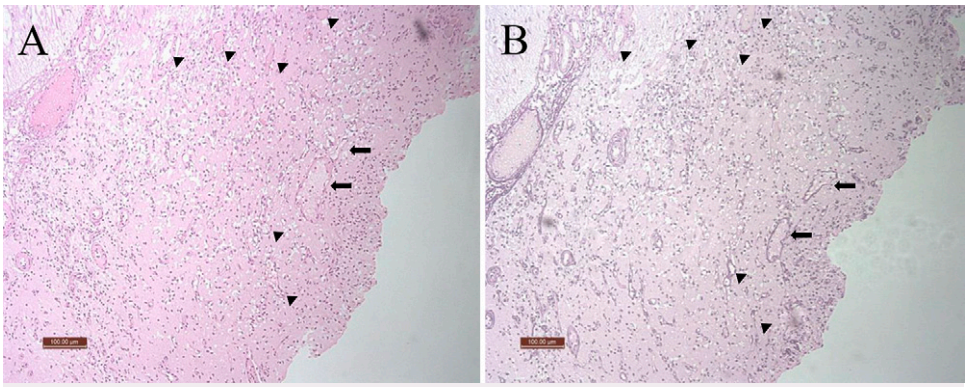

FIG. 3. Histopathology of the additionally resected specimen. Hematoxylin and eosin (**A**) and Elastica van Gieson (**B**) staining show dilated capillaries (*arrowheads*) and venules (*arrows*) with no evidence of arteriovenous malformation. Bar = 100 μm.

bleeding 48 hours after resection.[1] Furthermore, postoperative bleeding that led to new neurological deficits was reported in some patients.[5] Therefore, it is crucial to identify the residual nidus and achieve complete removal of the nidus using intraoperative imaging studies to perform safe and reliable AVM surgery.

Many reports have been published on the usefulness of ICG-VA in surgery for cerebrovascular diseases, and it is becoming a routine procedure in AVM surgery.[2,3,6] The procedure not only contributes to understanding the hemodynamics of the nidus but is also useful in confirming the extent of removal in real-time.[2] Thus, ICG-VA is necessary for performing safe AVM surgery.

On the other hand, false-negative results have been reported that need to be considered to ensure total removal of the nidus using ICG-VA. Bilbao et al. reported 37 patients with brain AVMs who received resection using ICG-VA and intraoperative cerebral angiography.[7] They noted that in two cases, in which ICG-VA confirmed total nidus removal, residual nidus was observed with subsequent intraoperative cerebral angiography. Therefore, it is dangerous to rely solely on ICG-VA to confirm the total removal of the AVM nidus, and it is necessary to ensure accuracy by including other modalities of imaging, such as intraoperative DSA.[4]

In the present case, ICG-VA for confirmation after removal of the nidus showed early venous filling of the cerebral cortex around the resected cavity, suggesting a residual nidus. However, after additional resection of the cortex, pathological examination showed no AVM in the tissue. Thus, a false-positive ICG-VA finding, which has not been reported previously, was observed in this case. We call this phenomenon the pseudo-residual nidus sign.

We attribute this phenomenon to the abnormal structure and local hemodynamic changes in the perinidal microvasculature of AVMs. Sato et al. reported perinidal dilated capillary networks (PDCNs) around cerebral AVMs based on pathological examination.[8] The PDCN is a dilated vascular network whose origin is in the capillary network adjacent to the nidal border of AVMs. The PDCN is connected to the nidus, feeding arteries, draining veins, and normal capillaries. Pennings et al. analyzed microcirculatory hemodynamic changes of the human brain during AVM surgery using intraoperative orthogonal polarization spectral imaging.[9] They reported a dramatic increase in microvascular flow in the perinidal brain tissue after resection of the nidus of AVM. Such structural abnormalities of the perinidal microvasculature and significant changes in local hemodynamics after AVM resection can contribute to early venous filling, as observed on ICG-VA despite complete removal of the AVM, leading to the pseudo-residual nidus sign.

The necessity of performing additional resection of the brain tissue containing perinidal microvasculature showing the pseudo-residual nidus on ICG-VA is controversial. Some reports suggest that the tissue may contribute to postoperative bleeding or AVM recurrence even after complete resection of the nidus.[10,11] Therefore, Sato et al. recommended meticulous treatment of the perinidal microvasculature.[8] On the contrary, Solomon et al. reported that the residual dysplastic vessels imaged on postoperative angiography after AVM resection would proceed to complete spontaneous resolution.[1] Consequently, they recommend managing the patient with the lesion conservatively without additional resection. If the pseudo-residual nidus is a lesion in a noneloquent area, additional aggressive resection of the lesion should be considered to prevent postoperative bleeding and AVM recurrence. On the other hand, strict follow-up with repeated postoperative imaging without additional resection should be performed if the lesion is in an eloquent area to prevent postoperative neurological sequelae.

Lessons

Early venous filling in the surrounding brain tissue after AVM removal, which we call “pseudo-residual nidus sign,” does not necessarily indicate a residual nidus. Confirmation of complete removal of the nidus using intraoperative DSA may be useful if this phenomenon is suspected. The need for additional resection of the lesion depends on the eloquence of the area.

Acknowledgments

We would like to thank Editage for English language editing.

Funding provided to Tsuyoshi Izumo by Grants-in-Aid Scientific Research (C) (grants 18K08973 and 21K09180), and to Takayuki Matsuo by Grants-in-Aid Scientific Research (C) (grant 21K09129).

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Izumo, Matsuo. Acquisition of data: Izumo, Okamura, Takahira, Sadakata, Morofuji. Analysis and interpretation of data: Izumo. Drafting the article: Izumo, Takahira, Matsuo. Critically revising the article: Izumo, Yoshida, Okamura, Takahira, Sadakata, Baba, Hiu, Anda, Matsuo. Reviewed submitted version of manuscript: Yoshida, Okamura, Takahira, Sadakata, Yamaguchi, Baba, Morofuji, Anda, Matsuo. Approved the final version of the manuscript on behalf of all authors: Izumo. Administrative/technical/material support: Yoshida.

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