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Case Report

Transarterial embolization for convexity dural arteriovenous fistula with or without pial arterial supply: A report of four patients

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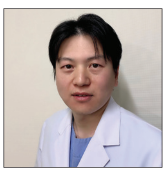

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ABSTRACT

Background: Convexity dural arteriovenous fistulae (dAVF) usually reflux into cortical veins without involving the venous sinuses. Although direct drainage ligation is curative, transarterial embolization (TAE) may be an alternative treatment.

Case Description: Between September 2018 and January 2021, we encountered four patients with convexity dAVFs. They were three males and one female; their age ranged from 36 to 73 years. The initial symptom was headache ($n = 1$) or seizure ($n = 2$); one patient was asymptomatic. In all patients, the feeders were external carotid arteries with drainage into the cortical veins; in two patients, there was pial arterial supply from the middle cerebral artery. All patients were successfully treated by TAE alone using either Onyx or N-butyl cyanoacrylate embolization. Two patients required two sessions. All dAVFs were completely occluded and follow-up MRI or angiograms confirmed no recurrence.

Conclusion: Our small series suggests that TAE with a liquid embolic material is an appropriate first-line treatment in patients with convexity dAVFs with or without pial arterial supply.

Keywords: Convexity, Dural arteriovenous fistula, Pial artery, Transarterial embolization

INTRODUCTION

Dural arteriovenous fistulae (dAVFs) are the result of anomalous connections between the meningeal arteries and dural veins.[7] Convexity dAVFs comprise 2.4% of all dAVFs.[6] They usually drain retrogradely into cortical veins and are categorized as Borden Type III.[1] As dAVFs with cortical venous reflux (CVR) risk the development of intracranial hemorrhage,[14] they must be addressed.

Direct surgery is a radical treatment for convexity dAVFs; however, endovascular treatment is another option. It is difficult to access dAVFs through their venous drainage route to the shunt point and there is also a risk of venous rupture. Therefore, transarterial embolization (TAE) can be selected instead of transvenous embolization. Our literature search found no reports of convexity dAVFs with pial arterial supply that were successfully treated by TAE alone.

Using Onyx (Medtronic, MN, USA) or N-butyl cyanoacrylate (NBCA) for TAE, we successfully treated four patients with convexity dAVFs; two patients presented with pial arterial supply. Here, we report their clinical features and their successful treatment with TAE alone.

CASE REPORTS

Representative case 1

A 63-year-old man scheduled for heart valve surgery underwent magnetic resonance imaging. A flow void in the left occipital lobe was detected incidentally [Figure 1a]. Angiography confirmed a dAVF on the left occipital convexity adjacent to the superior sagittal sinus (SSS). Drainage was into the cortical vein of the left occipital lobe leading to the SSS. The bilateral occipital arteries and the bilateral middle meningeal arteries (MMAs) were the feeders [Figures 1b-f]. Neither left carotid nor vertebral angiography revealed the pial arterial supply. We chose endovascular embolization because the presence of CVR raised the risk for intracranial hemorrhage.

With the patient under general anesthesia, we advanced a 6-Fr guiding catheter into the left external carotid artery through the femoral artery. A balloon catheter (Scepter XC; Microvention, CA, USA) was navigated to the distal portion of the posterior branch of the left MMA [Figure 1g] and under flow control using balloon inflation, Onyx 34 was injected; it traveled into the OA through artery-to-artery anastomosis and reached the shunt site. After the injection of a small additional amount of Onyx 18, the cortical draining vein and the feeders were sufficiently filled with the embolization material [Figure 1h] and the shunt disappeared [Figures 1i and j]. Postoperative MRI showed no brain edema and there was no dAVF recurrence in the 2 years following our treatment.

Representative case 2

A 42-year-old woman developed headache and vertigo. MRI identified flow voids and dilated cortical vessels in

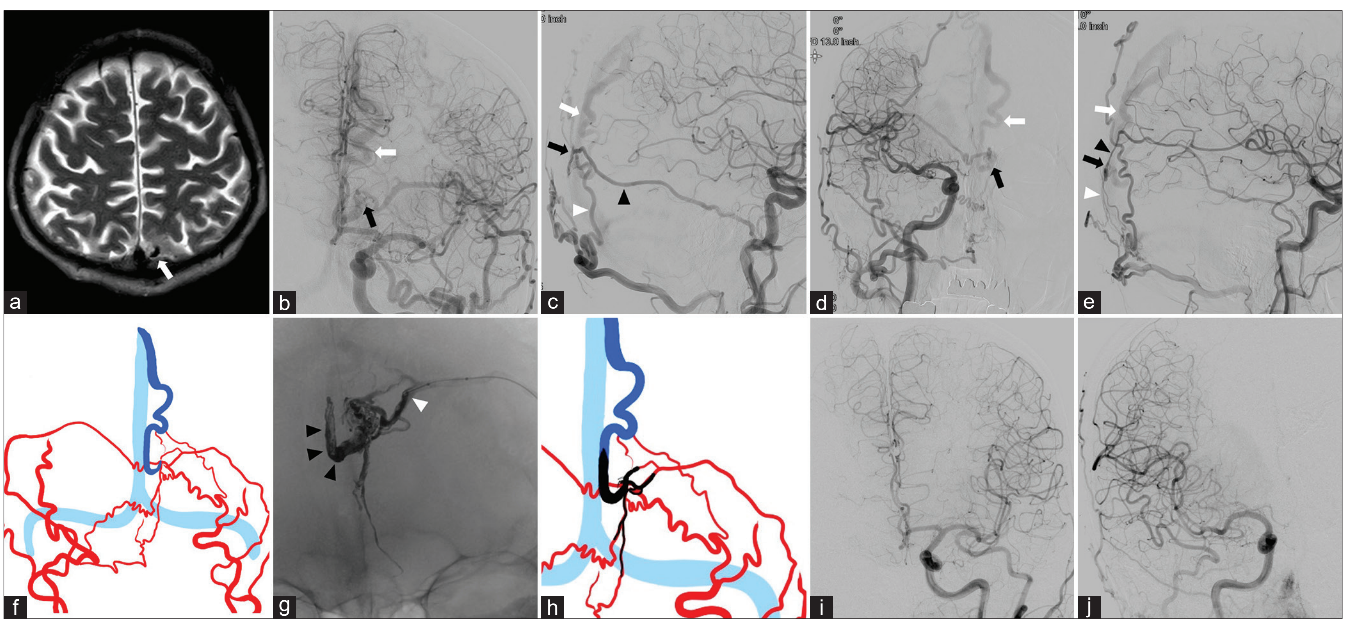

Figure 1: Representative Case 1 (a) T2-weighted image shows flow void in the medial aspect of the left occipital lobe (white arrow). (b and c) The left common carotid angiograms (b: frontal view and c: lateral view) confirm a dural arteriovenous fistula (dAVF) fed by the mastoid branch of the left occipital artery (OA) (white arrowhead) and middle meningeal artery (MMA) (black arrowhead) on the left occipital convexity with cortical venous reflux (CVR, white arrow). The black arrow indicates the shunt site. (d and e) The right common carotid angiograms (d: frontal view and e: lateral view) reveal the dAVF (white arrow) supplied by the mastoid branch of the right OA (white arrowhead) and MMA (black arrowhead) with the drainage into the occipital cortical vein. The black arrows indicate the shunt site. (f) Schema of the angioarchitecture delineating the feeding arteries (red), CVR (blue), and sinus (light blue). (g) Postoperative magnified X-ray (frontal view) demonstrates that the CVR, the mastoid branch of the left OA, and the posterior branch of the MMA were filled with Onyx (black arrowhead). The white arrowhead indicates the injection point on the MMA (tip of the balloon catheter). (h) Schematic drawing of the Onyx embolization material (black), feedeing arteries (red), CVR (blue), and sinus (light blue). (i and j) Postoperative left (i) and right (j) common carotid injections (frontal view) identify complete disappearance of the dAVF.

and around the left central sulcus [Figure 2a]. Angiography revealed a dAVF on the left parietal convexity arising from the anterior and posterior branches of the bilateral MMAs, pial arterial supply from the left middle cerebral artery (MCA) [Figures 2b-g], and cortical venous drainage into the SSS. Under general anesthesia and systemic heparinization, she underwent two TAE sessions with NBCA. In the first session, a 6-Fr guiding catheter was placed in the left and right external carotid artery through the bilateral femoral arteries. Then, a microcatheter (DeFrictor nanocatheter; Sumitomo Bakelite, Akita, Japan) was advanced to the distal part of the anterior branch of the right MMA [Figures 2h, i] using a TENROU S10 guidewire (Kaneka Medix, Osaka, Japan) supported by a TACTICS catheter (Technocrat Corp., Aichi, Japan), the distal access catheter that was placed in the foramen spinosum. Another DeFrictor microcatheter was retained at the shunt site [Figure 2i] through the anterior branch of the contralateral MMA as described above. NBCA (concentration 25%) was injected from that DeFrictor microcatheter. The injected NBCA penetrated the shunt site and filled the cortical venous drainer. Then, another DeFrictor microcatheter was advanced to near the shunt site [Figure 2i] through the posterior branch of the left MMA, and a 25% concentration of NBCA was injected. Finally, NBCA (concentration 17%) was injected through the DeFrictor microcatheter placed in the anterior branch of the

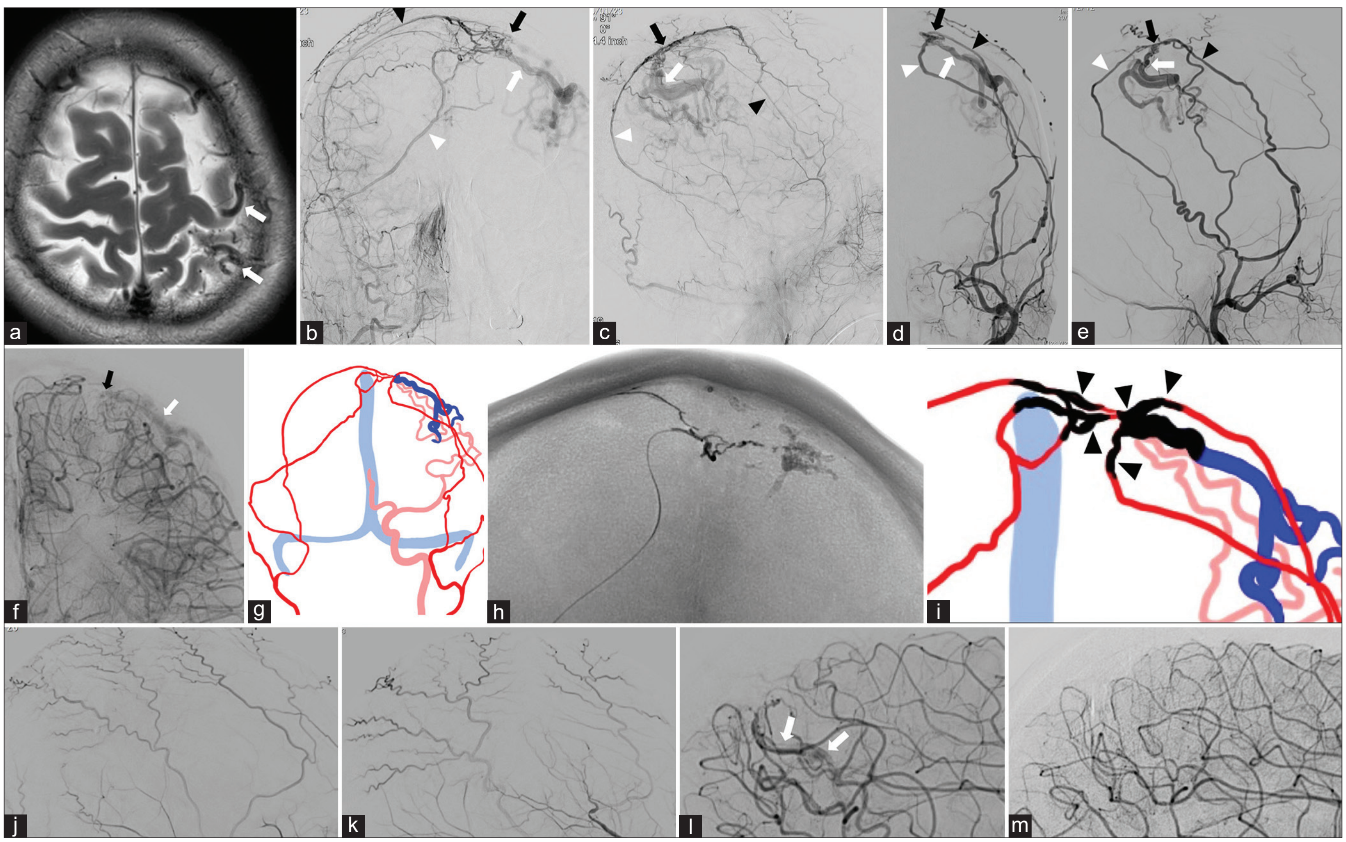

Figure 2: Representative Case 2: (a) T2-weighted image shows dilated cortical vessels (white arrows) in and around the left central sulcus. (b and c) Selective right external carotid angiograms (b: frontal view and c: lateral view) identified a dural arteriovenous fistula (dAVF) on the left parietal convexity. It was fed from the right anterior (black arrowhead) and posterior (white arrowhead) branch of the middle meningeal artery (MMA). Drainage was into a dilated Rolandic vein (white arrow). Black arrows indicates the shunt site. (d and e) Selective left external angiograms (D: frontal view and E: lateral view) also reveal the arteriovenous shunt arising from the left anterior (black arrowhead) and posterior (white arrowhead) branch of the MMA with cortical venous reflux (CVR) (white arrows). The black arrows indicate the shunt site. (f) Selective left internal carotid angiogram (frontal view) demonstrates pial arterial supply from the left middle cerebral artery (MCA). Black and white arrows indicate the shunt site and CVR, respectively. (g) Schema of the angioarchitecture delineating the feeding arteries from the external carotid arteries (red), the pial arterial supply from the left MCA (pink), CVR (dark blue), and the sinus (light blue). (h) X-ray (frontal view) demonstrates CVR and the bilateral MMAs filled with NBCA. (i) Schematic drawing of NBCA distribution (black). Black arrowheads indicate the 5 NBCA injection sites. Red, pink, dark blue and light blue also indicate feeding arteries, the pial arterial supply, CVR and the sinus respectively. (j and k) Selective right (j: lateral view) and left external (k: lateral view) carotid angiograms confirming complete obliteration of the dAVF immediately after the second TAE session. (l and m) Selective left internal carotid angiograms (lateral views) demonstrate a slight residual shunt flow (white arrows) from the left MCA just after the second session (l), and complete disappearance of the shunt 6 months after the procedures (m).

right MMA. Although some shunt flow from the posterior branch of the right MMA and the anterior branch of the left MMA persisted, her symptoms improved immediately after the first session.

At a second TAE session, performed 1 month later, we injected NBCA (25% concentration) through the posterior branch of the right and the anterior branch of the left MMA [Figure 2i]. Shunt flow from the bilateral external carotid arteries disappeared [Figures 2j and k], but a slight residual shunt flow from the pial supply delivered by the left MCA remained [Figure 2l]. All shunt flow and CVR had completely and spontaneously disappeared 6 months after the second session [Figure 2m].

RESULTS

The etiology of their dAVFs was unknown in all four patients. They had no history of sinus or venous thrombosis, no blood coagulation abnormalities, no head trauma, and none had undergone craniotomy. As shown in Table 1, the initial symptoms were headache (Case 2) and convulsion (Cases 3 and 4), one patient (Case 1) was asymptomatic. In all but Case 3, the fistulous site was on the medial side. In Cases 2 and 4, angiograms revealed pial arterial supply from the MCA, direct cortical venous drainage was observed in all four patients. We injected Onyx in Case 1; in Cases 2 and 3, we used NBCA, and in Case 4, we embolized with coils, NBCA and Onyx (first session), and with Onyx in the second session. Angiograms confirmed obliteration and there were no complications. In the course of follow-up (mean 22.8 months), no patient suffered recurrence.

DISCUSSION

Since convexity dAVFs are non-sinusal, they often exhibit CVR. Intracranial hemorrhage was elicited by 43% of convexity dAVFs reported by Kobayashi *et al.*[5] Although direct ligation of the draining vein near the shunt site is an acceptable treatment, TAE with a liquid embolic material is a minimally invasive option.

During TAE for convexity dAVFs, the fistula site must be penetrated, therefore, the microcatheter must be advanced as close as possible to that site. In Cases 2 and 3, the microcatheter was a DeFrictor; the outer diameter of the tip was 1.3-Fr. The DeFrictor microcatheter features a flow-direct and a guidewire catheter. It provides excellent flexibility, trackability, and extremely high distal reachability. Because we were able to advance the DeFrictor very close to the fistula site in Cases 2 and 3, we were able to deliver NCBA to the cortical vein.

Others[2,3,11] reported that 11.3–23.8% of cranial dAVFs received pial arterial supply. Hetts *et al.* suggested that dAVF patients with pial arterial supply who are treated endovascularly are at greater risk for ischemic stroke than patients without pial arterial supply. Our literature search found six earlier patients whose convexity dAVFs with pial arterial supply required direct surgery for cure [Table 2].[4,8,-10,12,13]

Although both injected Onyx and NBCA may flow into the pial artery, we think that Onyx is more likely to result in complete obliteration because the liquid material tends to move toward the venous side that is under lower pressure before filling the pial arterial feeders. Consequently, before embolization, it is important to identify the appropriate distance from the fistula to the Onyx injection site and the delivery of Onyx must be stopped when the liquid material reaches the predefined position. In cases, where the microcatheter can be advanced to be close to the fistulous site, NBCA may be an alternative embolization material. When the dAVF with pial arterial supply is adjacent to the eloquent area such as the motor cortex, direct surgery may be indicated to avoid ischemic complications.

Table 1: Clinical features of four patients of convexity dAVF treated by TAE alone.

Case No.	Age/Sex	Symptom	Side	Location/ Deviation	Pial supply	Feeder	Drainer	Embolic material	Result	Complication
1	63/M	None	Left	Occipital/Medial	No	Bil. MMA Bil. OA	Cortical vein	Onyx	Cure	None
2	42/F	Headache	Left	Parietal/Medial	Yes	Lt. MCA Bil. MMA	Cortical vein	NBCA	Cure	None
3	36/M	Convulsion	Left	Temporal/Lateral	No	Lt. MMA Lt. OA	Cortical vein	NBCA	Cure	None
4	73/M	Convulsion	Left	Occipital/Medial	Yes	Lt. MCA Rt. APA Bil. MMA Lt. OA	Cortical vein	Onyx (Coil) (NBCA)	Cure	None

APA: Ascending pharyngeal artery, Bil.: Bilateral, dAVF: Dural arteriovenous fistula, Lt.: Left, MCA: Middle cerebral artery, MMA: Middle meningeal artery, NBCA: n-butyl-2-cyanoacrylate, OA: Occipital artery, Rt.: Right, TAE: Transarterial embolization

Table 2: Reported cases of convexity dAVF with pial supply.

Author (Year)	Age/Sex	Presentation	Etiology	Side	Location/ Deviation	Feeders Dural	Feeders Pial	Drainer	Treatment	Result	Complication
Miyasaka *et al.* (1996)[9]	41/F	Epilepsy	Past Craniotomy	Right	Parietal/ Medial	MMA STA	MCA	Cortical vein	Direct surgery	Cure	None
Ozawa *et al.* (1998)[12]	39/M	Brain edema	SSS thrombosis	Right	Parietal/ Medial	MMA STA	MCA	Cortical vein	Embolization +Direct surgery	Cure	None
Saito *et al.* (2008)[13]	40/F	Hemorrhage	SSS thrombosis	Bilateral	Frontal/ Medial	MMA	MCA	Cortical vein SSS	Direct surgery	Flow-reduction	None
Kato *et al.* (2012)[4]	41/M	Hemorrhage	N/A	Left	Parietal/ Medial	MMA OA, STA	MCA	Cortical vein (Vein of Trolard)	Direct surgery	Cure	None
Matsubara *et al.* (2014)[8]	38/M	Hemorrhage	SSS thrombosis Protein S- deficiency	Right	Parietal/ Medial	MMA STA	ACA MCA	Cortical vein	Direct surgery	Cure	None
Okamoto *et al.* (2020)[10]	58/M	Brain edema	N/A	N/A	Frontal/ N/A	AEthA MMA	ACA MCA	Cortical vein	Embolization × 2 + Direct surgery	Cure	None

ACA: Anterior cerebral artery, AEthA: Anterior ethmoidal artery, APA: Ascending pharyngeal artery, AVF: Arteriovenous fistula, MCA: Middle cerebral artery, MMA: Middle meningeal artery, OA: Occipital artery, SSS: Superior sagittal sinus

CONCLUSION

Open surgery can be chosen as a radical therapy to treat dAVFs. However, our small case series suggests that TAE can be a less invasive and curative treatment in patients with convexity dAVFs, even in the presence of pial arterial supply. Embolization with Onyx may be more advantageous in terms of the cure rate, although the delivery of NBCA is possible in cases where a very thin microcatheter can be advanced very close to the shunt site.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Borden JA, Wu JK, Shucart WA. A proposed classification for spinal and cranial dural arteriovenous fistulous malformation and implications of treatment. J Neurosurg 1995;82:166-79.
2. Brinjikji W, Cloft HJ, Lanzino G. Clinical, angiographic, and treatment characteristics of cranial dural arteriovenous fistulas with pial arterial supply. J Neurointerv Surg 2021;13:331-5.
3. Hetts SW, Yen A, Cooke DL, Nelson J, Jolivalt P, Banaga J, *et al.* Pial artery supply as an anatomic risk factor for ischemic stroke in the treatment of intracranial dural arteriovenous fistulas. AJNR Am J Neuroradiol 2017;38:2315-20.
4. Kato N, Tanaka T, Suzuki Y, Sakamoto H, Arai T, Hasegawa Y, *et al.* Multistage indocyanine green videoangiography for the convexity dural arteriovenous fistula with angiographically occult pial fistula. J Stroke Cerebrovasc Dis. 2012;21:918. e1-e5.
5. Kobayashi E, Wakamatsu S, Tominaga S. A case of dural arteriovenous malformation on the convexity adjacent to the superior sagittal sinus. No Shinkei Geka 1994;22:643-8.
6. Kuwayama N, Kubo M, Endo S, Sakai N. Present status in the treatment of dural arteriovenous fistulas in Japan. Jpn J Neurosurg (Tokyo) 2011;20:12-9.
7. Lasjaunias P, Chui M, ter Brugge K, Tolia A, Hurth M, Bernstein M. Neurological manifestations of intracranial dural arteriovenous malformations. J Neurosurg 1986;64:724-30.
8. Matsubara S, Satoh K, Satomi J, Shigekiyo T, Kinouchi T, Miyake H, *et al.* Acquired pial and dural arteriovenous fistulae following superior sagittal sinus thrombosis in patients with protein S deficiency: A report of two cases. Neurol Med Chir

(Tokyo) 2014;54:245-52.
9. Miyasaka Y, Kurata A, Saegusa H, Yuzawa I, Utsuki S, Ohwada T. Dural-pial arteriovenous malformation with unusual venous drainage. Neurol Med Chir (Tokyo) 1996;36:91-5.
10. Okamoto M, Sugiyama T, Nakayama N, Ushikoshi S, Kazumata K, Osanai T, *et al*. Microsurgical findings of pial arterial feeders in intracranial dural arteriovenous fistulae: A case series. Oper Neurosurg (Hagerstown) 2020;19:691-700.
11. Osada T, Krings T. Intracranial dural arteriovenous fistulas with pial arterial supply. Neurosurgery 2019;84:104-15.
12. Ozawa T, Miyasaka Y, Tanaka R, Kurata A, Fujii K. Dural-pial arteriovenous malformation after sinus thrombosis. Stroke 1998;29:1721-4.
13. Saito A, Sugawara T, Mikawa S, Akamatsu Y, Saito H, Seki H. A case of multiple pial arteriovenous fistulas associated with dural arteriovenous fistula. J Neurosurg 2008;109:1103-7.
14. van Dijk JM, ter Brugge KG, Willinsky RA, Wallace MC. Clinical course of cranial dural arteriovenous fistulas with long-term persistent cortical venous reflux. Stroke 2002;33:1233-6.

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