Intracranial pial arteriovenous fistulas (AVFs) are rare vascular lesions of the brain. These lesions differ from arteriovenous malformation (AVMs) as they are direct artery to vein connections, have no nidus, and are composed of one or more direct arterial feeders with a single draining vein. Many of these have associated venous varix or giant venous aneurysms. Pial fistulas can be treated by endovascular occlusion or by microneurosurgery and endovascular treatment offers a simple and safe option. We report a case of pial fistulas treated with embolization using n-butyl cyanoacrylate (NBCA) and Lipiodol mixture in association with a balloon in the afferent artery to slow down the flow within the fistula.

**Key Words**: Pial arteriovenous fistula; Embolization; Glue; Balloon

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**CASE REPORT**

A 29-year-old man presented with tremor of head and both hands since childhood was referred to our hospital. On neurological exam he had no weakness or paresthesia of his extremities. Non-enhanced CT scan revealed hyperdense tortuous vascular structures at left temporal lobe. MRI revealed tortuous and dilated vascular structures at superficial area of the left temporal lobe (Fig. 1A). Cerebral angiography of the left internal carotid artery with 3D rotational angiogram revealed a single-channel pial AVF fed by the inferior division of left middle cerebral artery (MCA) (Fig. 1B). Large aneurismal venous dilation was seen distal to the fistula with continuous varicose dilation of the vein of Labbe. Delayed drainage of the dilated normal cortical veins of left temporal and occipital lobes was noted.

We planned endovascular embolization of fistula and proximal draining vein with balloon assisted flow control. Under the general anesthesia, the Prowler microcatheter (Cordis, FL, U.S.A.) was navigated just
proximal to the fistula site and then a nondetachable Hyperform occlusion balloon (ev3, CA, USA) was navigated to the inferior division of the MCA, proximal to the prior Prowler microcatheter tip. Before starting glue embolization, stagnation of the contrast material within aneurismal venous dilation was confirmed under the temporary balloon occlusion of MCA. Under flow control by temporary balloon occlusion, embolization with NBCA (50%) mixed with lipiodol and tungsten powder was performed at the fistula site including both the feeding artery and the proximal aspect of the aneurismal venous dilation (Fig. 1C). Immediately after satisfactory embolization, the microcatheter was swiftly pulled back with simultaneous deflation of the balloon, however due to friction, the balloon bursted.

Postembolization angiogram revealed complete occlusion of the AVF (Fig. 1D). Postprocedural MRI after 1 day presented no arterial flow within the aneurismal venous sac with contrast stagnation within draining vein. MRI also showed mild brain swelling adjacent to the fistula site with no acute ischemic lesion on diffusion-weighted images. The patient was discharged 3 days later without any remarkable neurological symptoms except mild headache.

DISCUSSION

Intracranial pial arteriovenous fistulae are rare lesions that have been described in 90 cases since 1970 and account for only 1.6% of a series of 320 AVMs (3). A communication between an arterial feeder directly into a solitary draining vein without an intervening tangle of vessels creates conditions for high, turbulent flow that producing venous varices (3). Pial AVFs are usually congenital but can be acquired as in traumatic or iatrogenic cases. Pial AVFs come to clinical attention

![Fig. 1. T2-weighted image shows the dilated venous varices at the left temporal area (A). 3D rotational angiogram reveals the single-channel fistula from the inferior division of the left MCA with dilated venous structures (B). Flow control was achieved by temporary balloon inflation proximal to the fistula. High concentration glue was infused into the nidus and the proximal aspect of the venous sac through another microcatheter (C). Final angiogram shows complete occlusion of the fistula (D).]
with intracerebral hemorrhage, seizures, headache, focal neurological deficit and raised intracranial pressure. In neonates and infants they may present with high output cardiac failure, increased head circumference and bruit (1).

Owing to the absence of a nidus, closure of the shunt by either endovascular or surgical technique represents a satisfactory therapeutic procedure. Conventional flow disconnection involves clipping or cauterization of the fistula vessels, which is effective but carry a high surgical risk for neurological morbidity if the lesions are deep-seated or located in eloquent areas(4). Abrupt disconnection may result in hyperemia upon evocation of the normal perfusion pressure break-through phenomenon (5).

Endovascular treatments are less invasive, allow straight forward localization of the lesion, and allow relatively safe treatment of lesions in deep and/or critical areas (6). Recent endovascular applications have been reported as successful means of disconnecting AV fistulae with a variety of different agents. However, endovascular attempts are not always successful or safe. For a variety of reasons, it can be technically difficult to deliver embolic materials precisely at the AV communication (3). Furthermore, embolization of the proximal feeder may give rise to new arterial connections and to fistula recurrence. And endovascular treatment carries the risk of migration of embolic materials into the draining vein, lung, or elsewhere in the cerebral vasculature (7, 8). Embolization of a normal vein draining into the venous channel of the pial AVF may result in venous infarction (4). In a meta-analysis, treatment failed in 40% who had undergone endovascular treatment, or had their fistulas recanalized and required further treatment (6).

A variety of agents may be used in endovascular applications including balloons, coils, glue, silk suture, and polyvinyl alcohol. NBCA was the embolic agent of choice in 5 cases of Limaye (1). It gives fluidity to fill the vascular spaces, control during injection and provides a permanent occlusion. With glue injection, it is desirable that the feeding artery is occluded close to the fistula along with glue penetration into the proximal part of the vein (1). However, because of rapid flow at the fistula, significant risk exists for embolization of normal distal draining vein and possible pulmonary embolization. Glue injection in association with balloon occlusion at the afferent artery or with partial coil embolization in the proximal vein can be used to slow down the flow within the fistula and prevent passage into the veins (4).

In our case, cerebral angiography revealed a single-channel pial AVF with turbulent, high flow. Because it had risk of migration of glue into the distal normal vein or pulmonary artery, we planned endovascular embolization with glue injection in association with temporary balloon occlusion at the proximal feeding artery. After confirming the stasis of flow across the fistula under the balloon occlusion, we injected concentrated glue carefully and occluded the fistula, proximal vein, and feeding artery just proximal to fistula. Because the microcatheter used in glue injection must be removed immediately after finishing injection, retrieval was performed without complete deflation of the balloon. Though the retrieval was resistant, it was successfully performed with simultaneous burst of the balloon. This may carry a risk of vascular injury. Although such complication has not been reported, careful attention should be given to the withdrawal process to minimize such risk.

CONCLUSION

Owing to the absence of a nidus in the pial AV fistula, closure of the shunt by either endovascular or surgical technique represents a satisfactory therapeutic procedure. Endovascular embolization is successful means of disconnecting AV fistulae with a variety of different agents. Glue injection in association with flow control by temporary balloon occlusion at the proximal feeding artery can be an effective and safe therapeutic option.

References

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