Early Intracerebral Hemorrhage Complicating the Successful Occlusion of a Carotid-Cavernous Fistula

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Summary

A 74-year-old woman with a direct carotid-cavernous fistula secondary to aneurysm rupture was treated by endovascular balloon occlusion of the fistula. Two days later, she suffered an intracerebral hemorrhage ipsilateral to the treated carotid cavernous fistula, perhaps due to normal perfusion pressure breakthrough.

Normal perfusion pressure breakthrough has been described as a cause of hemorrhage following treatment of arteriovenous malformations¹ and carotid stenosis^{2,3}. We report an unusual case of therapeutic endovascular occlusion of a direct carotid-cavernous fistula complicated by hemorrhage presumably caused by normal pressure perfusion breakthrough.

Case Report

A 74-year-old woman presented with a three-month history of left proptosis and diplopia. Physical examination revealed left proptosis and chemosis. She also had weakness in her left oculomotor, trochlear, and abducens nerves. Because her history and physical examination were consistent with a carotid-cavernous fistula, an angiogram was performed. Angiography revealed a direct, high-flow, left carotid-cavernous fistula with extensive cortical venous

drainage (figure 1A). The fistula was felt to be due to rupture of an aneurysm because there was no history of trauma and a 4 mm contralateral cavernous carotid aneurysm was present. A decision was made to perform endovascular occlusion of the fistula. The fistula was completely occluded with a single detachable silicone balloon (Target Therapeutics/Boston Scientific Corporation, Natick, MA) (figure 1B). There was immediate improvement in her proptosis but her oculomotor palsies progressed to complete ophthalmoplegia. Her post-operative course was uneventful until 40 hours after the balloon occlusion procedure, when she was found to have right hemiparesis. Her systolic blood pressure was noted to be 180 mm Hg. A CT scan of the brain was obtained immediately which showed a 2 cm hemorrhage centered in the left basal ganglia (figure 1C). She was transferred to the intensive care unit where her blood pressure was managed aggressively. Her condition deteriorated as the hemorrhage enlarged over the next 48 hours (figure 1D). The presence of hematocrit levels within the hemorrhage on the CT scans suggested a coagulopathy, though no coagulopathy was identified by a consulting hematologist. She died five days after the balloon occlusion procedure. An autopsy was performed, which revealed no intracerebral vascular abnormality.

A

Figure 1 74-year-old woman with left carotid-cavernous fistula caused by aneurysm rupture. A) Lateral projection angiogram demonstrates a high flow fistula with extensive cortical venous drainage.

B

Figure 1 B) Lateral projection angiogram following silicone balloon inflation and detachment demonstrates complete closure of the fistula and restoration of normal flow to the hemisphere.

Discussion

Normal perfusion pressure breakthrough as originally described by Spetzler et al occurs after the abrupt excision of a high flow intracerebral arteriovenous malformation¹. These investigators hypothesized that chronic cerebral ischemia due to a vascular “steal” through the arteriovenous malformation leads to chronic vasodilatation and eventual loss of autoregulation in the arteriolar resistance vessels. When the arteriovenous malformation is excised, local arterial blood pressure and cerebral perfusion pressure increase abruptly, and the inability of these arteries to maintain autoregulation leads to excessive perfusion of the capillary beds, which can result in malignant cerebral edema or hemorrhage¹. Experimentally, it has been demonstrated that there is an increase in capillary density and a loss of astrocytic foot processes surrounding capillaries in the brain subjected to chronic arteriovenous shunting⁴.

Normal perfusion pressure breakthrough has also been reported to occur following carotid

Figure 1 C) Unenhanced head CT demonstrates intracerebral hemorrhage 40 hours after occlusion of the fistula. D) Unenhanced head CT demonstrates enlargement of hemorrhage 72 hours after occlusion of the fistula.

endarterectomy. In eleven cases of postoperative ipsilateral intracerebral hemorrhage reported in a series of 1500 carotid endarterectomies, the mortality rate was 36%³. Postoperative systolic blood pressures were 200 to 240 mm Hg in six of eleven patients³. The time of occurrence of hemorrhage in these patients ranged from the immediate postoperative period to the tenth postoperative day, with a mean three day interval³. Solomon et al reported eight cases of postoperative intracerebral hemorrhage occurring in a series of 1930 carotid endarterectomies². Seven of these eight patients had high-grade (>90%) stenoses and the hemorrhages occurred between two days and three months following surgery. None of these patients had uncontrolled hypertension at the time of the hemorrhage.

The current standard treatment of direct carotid-cavernous fistulae is endovascular occlusion with detachable balloons^{5,6}. The complication rate of endovascular balloon occlusion of a carotid-cavernous fistula is low when performed by an experienced operator⁶. In a series

of 100 consecutive direct carotid-cavernous fistulas treated by endovascular techniques reported by Lewis et al, only one case was complicated by intracerebral hemorrhage⁶. No additional details were given about this case, so it is not clear if this was a result of normal perfusion pressure breakthrough, though it is quite possible that it was.

Halbach et al⁷ described a series of 165 carotid-cavernous fistulae, two of which were complicated by normal perfusion pressure breakthrough. In this report, they also included three cases of vertebral-venous fistulae that were complicated by normal perfusion pressure breakthrough. None of these cases were complicated by hemorrhage. All five of the patients had large, longstanding fistulae, ranging in duration from nine to thirty-two years. In four of these cases, the patients became symptomatic immediately on balloon inflation which completely occluded the fistula. The authors were able to occlude these fistulae gradually, so as to not precipitate symptoms of normal perfusion pressure breakthrough. Our patient did

not become symptomatic immediately, so we did not consider gradual occlusion to be indicated. A case of endovascular occlusion of a traumatic cavernous-carotid fistula complicated by headache and seizure with post-treatment hyperperfusion of the ipsilateral hemisphere documented by single photon emission tomography (SPECT) has been reported⁸. This case was not complicated by hemorrhage, however.

A direct carotid-cavernous fistula is a high-flow lesion, yet it can create a situation of cerebral hypoperfusion analogous to a severe carotid stenosis by shunting blood intended for the ipsilateral cerebral hemisphere into the cavernous sinus.

Abrupt occlusion of this shunt results in sudden restoration of normal perfusion pressure to the ipsilateral hemisphere. While normal perfusion pressure breakthrough is quite unusual following endovascular therapy of direct carotid-cavernous fistula, the original description of this entity included experimental sup-

port from a model of carotid-jugular fistula in cats¹.

Our patient's cerebral hypoperfusion ipsilateral to the fistula may have been exacerbated by venous hypertension as reflected by the presence of cortical venous drainage. A case of transverse and sigmoid sinus dural arteriovenous fistula with cortical venous drainage was reported to be complicated by post-operative hyperperfusion documented by SPECT⁹. It is also possible that venous thrombosis, which started in the cavernous sinus at the time of embolization, extended into the deep venous system, causing venous infarction and subsequent hemorrhage.

In retrospect, the chronic course of our patient's fistula may have been an indicator of higher risk for reperfusion injury. Aggressive blood pressure management aimed at minimizing hypertension is prudent following occlusion of direct cavernous-carotid fistulae, as it is following arteriovenous malformation resection and carotid endarterectomy.

References

- 1 Spetzler RF, Wilson CB, Weinstein P et Al: Normal perfusion pressure breakthrough theory. *Clin Neurosurg* 25: 651-672, 1978.
- 2 Solomon RA, Loftus CM et Al: Incidence and etiology of intracerebral hemorrhage following carotid endarterectomy. *J Neurosurg* 64: 29-34, 1986.
- 3 Pomposelli FB, Lamparello PJ et Al: Intracranial hemorrhage after carotid endarterectomy. *J Vasc Surg* 7: 248-255, 1988.
- 4 Sekhon LH, Morgan MK, Spence I: Normal perfusion pressure breakthrough: the role of capillaries. *J Neurosurg* 86: 519-524, 1997.
- 5 Debrun GM, Vinuela F et Al: Indications for treatment and classification of 132 carotid-cavernous fistulas. *Neurosurgery* 22: 285-289, 1988.
- 6 Lewis AI, Tomsick TA, Tew JM Jr: Management of 100 consecutive direct carotid-cavernous fistulas: results of treatment with detachable balloons. *Neurosurgery* 36: 239-244, 1995.
- 7 Halbach VV, Higashida RT et Al: Normal perfusion pressure breakthrough occurring during treatment of carotid and vertebral fistulas. *Am J Neuroradiol* 8: 751-756, 1987.
- 8 Nakashima H, Tomita T et Al: A case of atypical course after balloon occlusion for high flow traumatic carotid-cavernous fistula. *No To Shinkei* 47: 177-181, 1995.
- 9 Kuroda S, Ushikoshi S et Al: Postoperative hyperperfusion in dural arteriovenous fistula associated with venous ischemia: case report. *Surg Neurol* 49: 406-411, 1998.

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