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Case Report

Hemichorea-hemiballismus caused by postoperative hyperperfusion after clipping of a giant unruptured middle cerebral artery aneurysm

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Abstract

Background: Movement disorders after the clipping for an unruptured giant aneurysm are rare. The information on the pathogenesis and treatment options for this condition is largely unknown.

Case Description: An 82-year-old female with no neurological deficits underwent a clipping for a giant middle cerebral artery (MCA) aneurysm. Immediately after surgery, she presented with hemichorea–hemiballismus (HC–HB) on the left side. Postoperative angiograms and single-photon emission computed tomography demonstrated the hyperperfusion in the right frontal cortex and the decreased perfusion in the basal ganglia, indicating that the abrupt hemodynamic changes due to the obliteration of the giant aneurysm caused the dysfunction of the frontal cortical and subcortical pathway and the basal ganglia. Administration of tiapride hydrochloride was dramatically effective in controlling the HC–HB until the hyperperfusion resolved. Single-photon emission computed tomography obtained 8 weeks after surgery revealed that the cerebral blood flow had been normalized in the right frontal cortex. The relative hypoperfusion of the right basal ganglia was also resolved. Then tiapride hydrochloride was discontinued without a relapse of HC–HB.

Conclusion: This case appears consistent with the theory that the connecting fibers responsible for the development of HC–HB are also located in the frontal lobe. The treatment of giant aneurysms involving the M1 portion can cause abrupt hemodynamic changes in both frontal cortex and the basal ganglia, which can potentially induce postoperative movement disorders.

Key Words: Aneurysm, chorea, clipping, giant aneurysm, hyperperfusion, single-photon emission computed tomography

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INTRODUCTION

Movement disorders can occur as secondary to a variety of neurological, metabolic, infectious, traumatic, and other systemic diseases.[20,24] Cerebrovascular events account for approximately 22% of these secondary movement disorders.[24] A large series study reported that chorea was the most common movement disorder (35.7%).[2]

Hemichorea–hemiballismus (HC–HB) has been observed in patients with hemodynamic insufficiency caused by moyamoya disease,[10,13,14,16,21,25,29,30,32,33,38,40,41] intracranial arterial stenosis,[15,17] and extracranial carotid artery stenosis.[8,22,27] The basal ganglia, particularly the lentiform nucleus and the thalamus, has traditionally been deemed responsible for HC–HB.[2,3,5,9,20] However, recent studies investigating the distribution of hemodynamic insufficiency using single-photon computed tomography (SPECT) demonstrated that the frontal cortical subcortical motor pathway also plays a significant role in the development of HC–HB.[14,22]

Here we report a case of HC–HB on the left side of the body that started immediately after clipping of a right unruptured giant middle cerebral artery (MCA) aneurysm at the M1 portion. This patient had no hemorrhage or infarct appreciable on postoperative magnetic resonance (MR) images. However, SPECT showed some degree of postoperative hyperperfusion in the right frontal lobe as well as the hypoperfusion of the right basal ganglia. These hemodynamic changes resolved 8 weeks after surgery with remission of HC–HB. Our findings indicated that hyperperfusion as well as hypoperfusion can induce contralateral HC–HB and substantiated the previous theory that dysfunction of the frontal cortical subcortical motor pathway is one of the possible mechanisms of HC–HB. The treatment of giant aneurysms involving the M1 portion might be at an increased risk of postoperative movement disorder because it can potentially cause abrupt hemodynamic changes in both frontal cortex and the basal ganglia.

CASE REPORT

An 82-year-old female was referred to our hospital with a 1-month history of pulsatile headache. A head computed tomography (CT) scan taken at the previous hospital showed a 2.5-cm sized mass in the right temporal region. MR imaging demonstrated a round and partially thrombosed aneurysm of the MCA [Figure 1a]. After thoroughly discussing treatment options and risks with the patient and her family, they decided to continue to watch the aneurysm. We scheduled a follow-up visit in 6 months, but this was not accomplished because the patient felt she was stable. When she revisited our hospital for worsening headache 2 years after her last visit, the aneurysm had grown in size from 2.5 to 4.0 cm, with the development of a significant perifocal edema [Figure 1b]. She had no neurological deficits. We recommended that she undergo clipping of the aneurysm, and she agreed to our proposal. We performed a right frontotemporal craniotomy through the orbitozygomatic approach, with intraoperative monitoring of the motor-evoked potential. The aneurysm was buried in the temporal lobe [Figure 1c]. Dissecting the sylvian fissure,

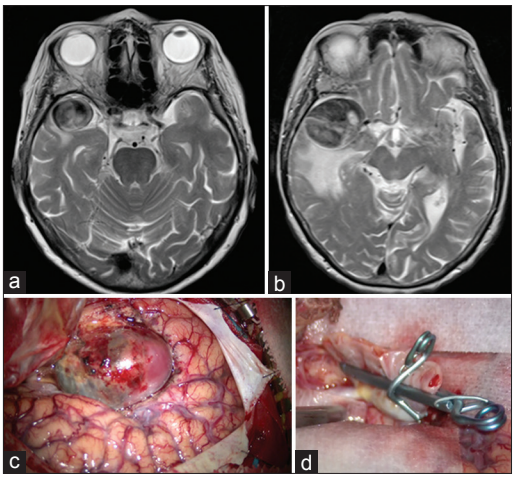

Figure 1: (a and b) Axial T2-weighted MR images showing an increase of the right middle cerebral artery aneurysm diameter from 2.5 to 4 cm in 2 years. (c) Intraoperative photograph of the aneurysm. (d) The aneurysm was clipped using two clips

we found the aneurysm had arisen from the nonbranching portion of the M1, not M1–M2 bifurcation. First, we anastomosed the right superficial temporal artery (STA) to the temporal cortical branch of the MCA to secure distal blood flow during the temporary occlusion of M1. After confirming patency of the anastomosis and the distribution of bypass flow, we trapped the M1 using temporary clips. Next, we sectioned the aneurysm and completely removed the clots. Two clips were then applied in an angioplastic fashion [Figure 1d]. A good patency was confirmed using Doppler flowmetry and indocyanine green fluorescence angiography. The patient showed a good arousal from anesthesia, but presented a few hours after surgery with an irregular, purposeless, and hyperkinetic movement in the left arm and leg, which she was still able to move voluntarily (see Supplementary Video). Postoperative CT scan obtained immediately after surgery revealed no abnormal findings. MR images obtained 1 day after surgery did not reveal any new hemorrhage or infarction [Figure 2a-c]. We diagnosed her abnormal movement as HC–HB. Angiography delineated a complete obliteration of the aneurysm with no other occluded arteries. However, the arterial flow in the peripheral MCA territory obviously increased, compared with preoperative findings [Figure 2d and e]. Although STA–MCA bypass flow was also confirmed, the flow via the bypass was only toward the distal portion of the cortical temporal artery [Figure 2f]. The 99mTc-ECD SPECT obtained 3 days after surgery demonstrated hyperperfusion of the MCA territory [Figure 3a]. There was slight hypoperfusion in the right basal ganglia including the subthalamic regions. Cerebral blood flow (CBF) was decreased in the lateral insular cortex, but the area of hypoperfusion corresponded to that of the perifocal edema existing before surgery. We suspected that the patient's HC–HB may be related to

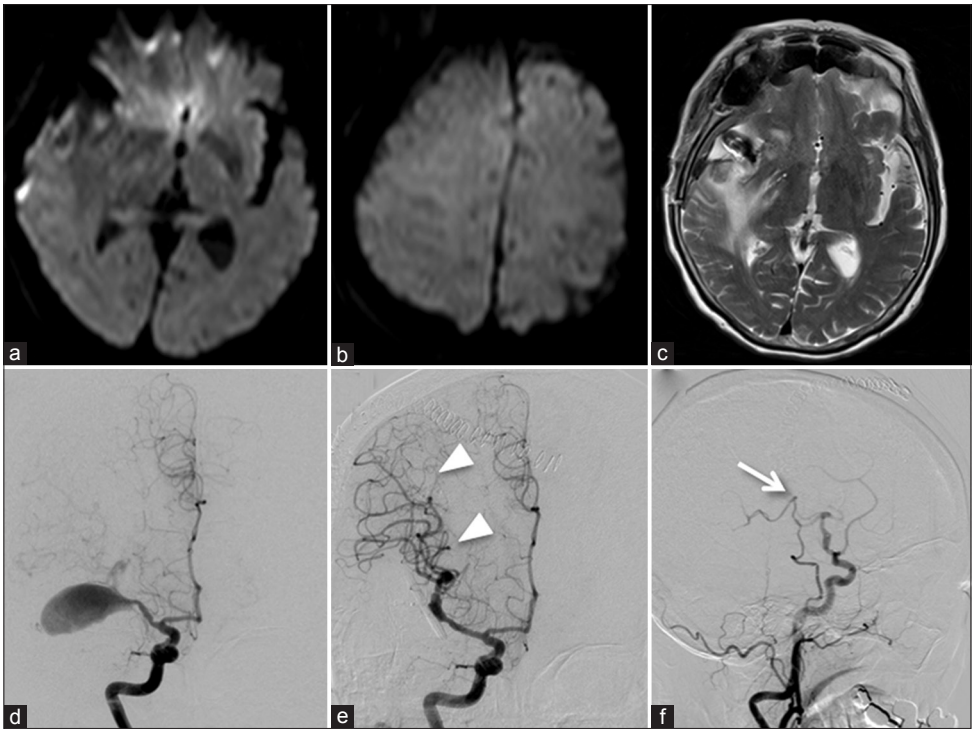

Figure 2: (a and b) Axial diffusion-weighted MR images obtained 1 day after surgery revealing no acute ischemia. (c) Axial T2-weighted MR image performed 1 day after surgery showing no new changes other than the preexisting edema around the aneurysm. (d) Preoperative angiogram demonstrating a giant MCA aneurysm. (e) Postoperative angiogram showing the complete obliteration of the aneurysm with preservation of the parent artery. Note the remarkable increase of the arterial flow in the MCA territory (arrowheads). (f) Right lateral carotid angiogram demonstrating that the bypass flow covered only a small area of the frontal lobe distal to the site of anastomosis (arrow)

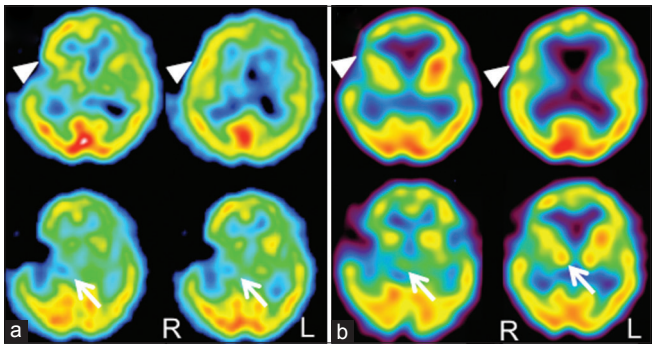

Figure 3: (a) 99mTc-ECD SPECT performed 3 days after surgery revealing hyperperfusion in the frontal cortex (arrowheads). There was also slight hypoperfusion in the right basal ganglia including the subthalamic nucleus (arrows). (b) 99mTc-ECD SPECT obtained 8 weeks after surgery showed the resolution of hyperperfusion in the right frontal cortex (arrowheads) with the resolved laterality of the perfusion in the subthalamic regions (arrows)

the hyperperfusion in the right frontal cortex in addition to the decreased perfusion in the right thalamic region. Although her neurological findings were otherwise normal, HC–HB persisted without any improvement for the following week, irrespective of our attempt to maintain her systolic blood pressure under 130 mmHg. We next tried tiapride hydrochloride, 75 mg twice a day, which dramatically relieved her symptoms. The patient was discharged with no neurological deficit. SPECT obtained 8 weeks after surgery revealed that CBF had been normalized in the right frontal cortex with the resolved laterality of the perfusion in the thalamic regions [Figure 3b]. The semiquantitative data calculated by the Patlak plot method demonstrated the normalization of regional CBF in the right frontal lobe and the resolution of the disparity in the bilateral subthalamic nuclei [Table 1]. At that point, the tiapride hydrochloride was discontinued without a relapse of HC–HB.

DISCUSSION

HC–HB can occur as a sequela to a variety of strokes.[4,20,24] Stroke types associated with HC–HB include cerebral infarction,[11,26,28,36] moyamoya disease,[10,13,14,16,21,25,29,30,32,33,38,40,41] intracranial arterial stenosis due to atherosclerotic change,[14,15] delayed vasospasm after subarachnoid hemorrhage,[31] extracranial carotid artery stenosis,[8,22,27] and cerebral hemorrhage.[2,18] Traditionally, the mechanism underlying poststroke HC–HB is believed to be ischemia of the basal ganglia, particularly the lentiform nucleus and the thalamus.[2,3,5,9,20] However, previous literature also shows that HC–HB can be caused by subcortical ischemia, without involvement of the basal ganglia.[1,7] In addition, recent studies examining CBF show that the frontal cortical and subcortical motor pathway may also play a significant role in the development of

Table 1: Cerebral blood flow calculated by the Patlak plot method (ml/100 g/min)

	Immediate post-op		8 weeks after surgery	
	Right	Left	Right	Left
Frontal operculum	48.4	36.0	37.0	38.8
Subthalamic nucleus	35.8	39.1	30.2	29.5

HC–HB.[14] Morigaki *et al*. also reported two cases of HC–HB that subsided after revascularization.[22] Their study proposes that the disrupted hemodynamics in the watershed areas of the frontal lobe account for the onset of hemichorea associated with carotid artery occlusive disease. In our case, the findings from postoperative angiography and SPECT demonstrated hyperperfusion, not hypoperfusion, in the frontal lobe immediately following surgery. It is well known that hyperperfusion can cause a variety of neurological deficits following STA–MCA anastomosis,[6,12,39] carotid endarterectomy (CEA),[37] obliteration of arteriovenous malformation,[34] and clipping of giant aneurysms.[19,23,35] We presume that abrupt elimination of the giant aneurysm from the M1 segment, which had long served as a buffering reservoir, may have led to a massively increased blood flow in the cortical MCA territory, resulting in dysfunction of the frontal cortical and subcortical motor pathways. It is unlikely that the STA–MCA bypass caused the hyperperfusion in our case because bypass flow covered only a small area of the frontal lobe distal to the site of anastomosis. We also speculate that the slight decreased perfusion in the right basal ganglia, although it was less prominent than the change in the frontal cortex, might have exacerbated the dysfunction of the frontal cortical and subcortical motor pathways. This hypoperfusion in the basal ganglia might be due to the flow reduction of the perforators caused by clipping of the giant M1 aneurysm with a wide neck. The development of HC-HB in this case could have a bearing on the morphological and anatomical characteristics of a giant M1 aneurysm mentioned above.

In summary, we encountered a case of HC–HB that developed immediately after clipping of a giant unruptured MCA aneurysm. Postoperative angiography and SPECT indicated that hyperperfusion in the frontal cortex and hypoperfusion in the basal ganglia appeared to provoke this hyperkinetic movement disorder. Administration of tiapride hydrochloride was effective in controlling the HC–HB until the hyperperfusion resolved. This study also supports the recent theory that the connecting fibers in the frontal cortical and subcortical motor pathway play a significant role in the development of HC–HB.

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