Status epilepticus as clinical presentation of cerebral hyperperfusion syndrome after carotid endarterectomy: report of a case

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Abstract

Cerebral hyperperfusion syndrome (CHS) is an uncommon complication following carotid revascularization, that occurs within two weeks by carotid artery stenting (CAS) or carotid endarterectomy (CEA). CHS is clinically characterized by unilateral headache, seizures, focal neurologic defects. We describe a 74-year-old female patient who developed status epilepticus (SE) seven days after left CEA, with no response to benzodiazepines. Intravenous administration of phenytoin led to resolution of SE over a period of 48h. Neuroimaging findings showed reversible lesion in left frontal region, associated to acute evidence of hyperperfusion signs in left hemisphere suggestive for CHS.

KEY WORDS: status epilepticus, cerebral hyperperfusion syndrome, ictal hyperperfusion, periodic lateralized epileptiform discharges (PLEDs).

Introduction

Carotid endarterectomy (CEA) and, more recently, carotid artery stenting (CAS) are vascular reconstruction techniques for carotid artery stenosis. These treatment options are indicated when a patient has symptomatic stenosis >50% or when he/she presents asymptomatic stenosis ≥80% (1). Guidelines recommend carotid revascularization techniques within 14 days in patients suffering for transient ischemic attack (TIA) or stroke related to carotid artery stenosis (2, 3). Ischemic complications after CEA and CAS related to embolization or inadequate cerebral protection in patients with a poor collateral supply, represent a major cause of postoperative central neurological disorders. However, carotid reconstruction surgery may result in a rapid increase in ipsilateral cerebral blood flow (CBF) with consequent hyperperfusion, high above the metabolic demands of the chronic ischemic brain tissue. Cerebral hyperperfusion syndrome (CHS) represents therefore a dangerous complication following CAS and CEA, with increased risk of intracerebral hemorrhage (IH) (4); it may occur within few hours or up to 3 weeks (5) after CEA. Incidence of CHS after CEA and CAS ranges from 0.4 to 1.8% (6, 7) and from 0.4 to 3% (8-10), respectively. Risk factors include longstanding hypertension, highgrade stenosis, poor collateral blood flow, contralateral carotid artery occlusion (6) or recent contralateral CEA, diabetes mellitus, old age and history of administration of anticoagulants or antiplatelet agents. Clinical characteristics of CHS consist of unilateral headache, face and eye pain, seizures, and focal neurologic defects. The pathophysiological mechanisms underlying CHS after CEA and CAS are poorly understood but may be related to a deterioration in cerebrovascular autoregulation in ischemic brain regions with consequent abnormal increase of regional CBF (8). In most cases seizures and focal neurological symptoms are due to cerebral edema or intracranial hemorrhage. Electroencephalographic (EEG) examinations are important to better explain the pathophysiological mechanisms of CHS (11). We report a case of a 74-year-old female patient who developed CHS seven days after CEA, presenting with status epilepticus.

Case report

A 74-year-old woman with a history of diabetes mellitus type II, past evidence of intestinal metaplasia, hy-
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pertension and atrial fibrillation on oral anticoagulant therapy (TAO), was admitted to the Vascular Surgery Department due to the finding of a critical left internal carotid stenosis, with hemodynamic changes at eco-color Doppler examination. Computed Tomography (CT) of the brain showed multiple bilateral gliotic lesions, with particular involvement of the left internal border zone area. CT angiography (CTA) revealed a severe stenosis of the left internal carotid (>75%) and a mild stenosis (50%) at the right carotid bifurcation. The patient interrupted warfarin therapy which was replaced with low molecular weight heparin (LMWH) five days before performing left internal carotid endarterectomy (CEA). There were no reported complications during the periprocedural period. Neurological examination was normal before and after surgery approach. Seven days after CEA, she developed loss of consciousness with evidence of focal clonic contractions at the right face side and right arm. She was conducted to the Emergency Room (ER) for status epileptics with persistence of focal clonic movements and consciousness deterioration; she was administered benzodiazepines (Lorazepam 4 mg) with substantial reduction in clonic contractions. The patient underwent urgently CTA and brain CT scan that revealed hypervascularity on the left hemisphere (Fig. 1 a) and a regular course without significant stenosis of extra and intracranial left carotid artery; a left frontal cortical-subcortical hypodense lesion was detected in brain CT imaging. On admission in the Neurology Department her blood pressure was 180/90 mmHg; neurological examination showed deterioration in the level of consciousness (GCS score 8/15), unintelligible speech related to aphasic disorder, right hemiparesis. EEG-poligraphic recording, performed a few hours after admission, revealed a persistent focal status epilepticus with evidence of a rhythmic activity of slow spike waves from the fronto-centro-temporal regions of the left hemisphere without electromyographic activity (Fig. 2 a), an electro-clinical condition compatible

Figure 1 - a. Brain CT angiography performed at day 1, with hypervascularization of left hemisphere (inside the circular area); b. Brain MRI performed at day five that showed areas of T2 hyperintensity on left frontal region (below the arrows); c. Brain MRI performed at day 24; it can noted disappearances of these areas.
with non-convulsive status epilepticus (NCSE). Intravenous administration of levetiracetam (bolus injection of 40 mg/kg, then 1500 mg every 12 hours) did not modify the EEG pattern (Fig. 2 b). The patient was then started on intravenous phenytoin (at a dosage of 15 mg/kg in bolus followed by a maintenance dose of 400 mg/24 hours) with improvement in consciousness level and resolution of NCSE in 24 hours. Control EEG at 24 h and 48 h showed periodic lateralized epileptiform discharges (PLEDs) on the central and temporal regions of the left hemisphere (Fig. 3). On the second day of hospitalization the patient presented fever, which continued for a few days. Magnetic resonance imaging (MRI) of the brain with gadolinium, performed five days after admission, detected altered signal areas in the left frontal supero-lateral region, engaging the subcortical white matter and a probable focal cortical thickening in the anterior portion of the left inferior frontal gyrus (Fig. 1 b). Laboratory tests showed normal levels of leukocytes and a slight increase of the C-reactive protein level; urinalysis was normal. A lumbar puncture was also performed, with absence of LCR cells and normal values of LCR glucose and protein levels; the search for membrane neuronal auto-antibodies was normal. Clinical course improved in the following days, with resolution of fever, normalization of neurological examination, and disappearance of PLEDS on EEG recording with residual intercritical sporadic sharp waves in the left temporal region. In the 24th day the patient made brain MRI control with gadolinium, resulting in a substantial disappearance of altered signal areas in the left frontal region (Fig. 1 c).

Discussion

The increase of cerebral blood flow (CBF) above the metabolic requirements of brain tissue after CEA can
lead to a hyperperfusion with variable focal cerebral distress. Incidence of CHS after CEA ranges from 0.4 to 1.8% (6, 7); although acute symptomatic seizures are a common expression of brain distress, a few papers report EEG abnormalities or epileptic seizures in patients with CHS (11-13), and only one publication describes status epilepticus related to CHS post carotid revascularization without intracranial hemorrhage (IH) (5). In our comatose patient, a focal epileptic activity during NCSE has evolved in a PLEDs pattern with improvement of consciousness after intravenous phenytoin administration. Moreover, PLEDs represent a not uncommon EEG pattern in CHS (14) with a good prognostic value in patients who evolved from NCSE (15). In our patient, however, the occurrence of PLEDs associated with hyperpyrexia raised also the suspicion of a possible infectious etiology (i.e. herpetic encephalitis), but it was contradicted by negativity of CSF examination. Moreover, brain CT and MRI revealed altered signal areas in the left frontal supero-lateral region not related to a vascular territory and without hyperintensity on diffusion weighted imaging (DWI), excluding periprocedural thromboemolism. CTA images showed no re-occlusion of the left internal carotid, and mild stenosis (50%) of the right carotid bifurcation, with a minimal contribute to the cerebral hemodynamic reserve. The most important finding of angiography was evidence of a hypervascularization in the whole left hemisphere, expression of increased CBF. The loss of visualization of the altered signal areas in the left frontal lobe at control brain MRI (performed in the 24th day) suggests a transitory cortical dysfunction. In our reported case, there were multiple risk factors for CHS, such as contralateral stenosis, poor collateral circulation, old age, hypertension and administration of anticoagulant therapy. We suppose that the mechanism underlying the development of CHS in our patient was cortical hyperperfusion induced by an increase in regional CBF post-revascularization and ictal hyperperfusion. Recent case-studies demonstrated that the use of transcranial Doppler for the measurement of systolic mild cerebral artery velocity in conjunction with systematic blood pressure monitoring, performed at an early stage after CEA, can more accurately predict patients at risk of CHS (16, 17).

References


