vasoconstriction in brain haemorrhage is unclear. A possible mechanism is that when vasospasm subsides and perfusion is restored, artery rupture occurs. A drug-induced shift of upper limit of cerebrovascular autoregulation towards lower blood pressure levels together with a cannabis-induced transient arterial hypertension may have played a role in the pathogenesis of brain haemorrhage in our patient. A delayed and possible synergic effect of both drugs on cerebral autoregulation can be suspected.

A cervico-cephalic artery dissection is defined as hematoma within the artery wall. Spontaneous dissecting aneurysms of the middle cerebral artery (MCA) have been implicated in stroke. The giant serpentine aneurysm (GSA) is a partially thrombosed giant aneurysm (>2.5 cm) with tortuous vascular channels and separate entrance and outflow pathways [1–3]. The GSA is often situated on the MCA or its branch vessels [4]. Its pathogenesis has not been elucidated. We report a case in which a right MCA dissection was diagnosed after a stroke. Eleven years later, a second stroke occurred in the same territory, leading to the discovery of a GSA.

**Case Report**

In 1992, a 37-year-old woman suffered a sudden attack of weakness on the left side of the body. She was free from vascular risk factors and took no prescribed drugs. The CT scan revealed a right striatocapsular ischemia. Laboratory investigation failed to explain the stroke. The selective bilateral carotid, vertebral and renal angiograms showed only a very irregular aspect of the MCA, with multiple zones of narrowing interspersed with zones of dilatation (fig. 1a). The diagnosis of dissection of the MCA was proposed. The course of the disease was positive. Arterial hypertension was discovered during hospitalization. Antihypertensive and antithrombotic drugs were prescribed. Eleven years later, the patient presented sudden, persistent anesthesia and weakness of the left arm. No other neurological or general anomalies were observed. MRI in the frontal T1-weighted sequence revealed a partially thrombosed aneurysm, suggestive of a GSA, on the insular segment of the MCA (fig. 1b, c).

**References**

**Discussion**

This observation is the first, to our knowledge, showing a possible continuum from a dissection of the MCA to a GSA, supporting the idea that the dissection may trigger a pathological process leading to a GSA [5]. It has been hypothesized that spontaneous fusiform MCA aneurysms may develop as a result of arterial dissection with intramural hemorrhage between the intima and the media [6]. Some authors have reported the appearance of a GSA after internal carotid ligation for an intracavernous aneurysm [2], whereas others have incriminated the Coanda or the boundary wall effect, in which hemodynamic perturbations may lead to the development of GSAs [7]. The development of similar aneurysms has been reported in the posterior circulation [8]. Therapeutic strategy is unclear. Some authors have recommended revascularization surgery [9, 10], but the number of reports so far is too small to allow a definite choice of treatment. In the case presented here, the antithrombotic medication failed to prevent the development of the dissection into a GSA. Nevertheless, we have to take into account the high risk of bleeding in the initial phase of the disease, especially if anticoagulants are being administered. If, after the initial phase of the disease, an aneurysm appears to be developing into a GSA the use of anticoagulant drugs might be envisaged if aggressive surgery and endovascular treatment appear to be inadvisable. We cannot be certain that anticoagulants will prevent the development of a fusiform aneurysm into a GSA but they might be useful since the propagation of thrombosis seems to play an important role in the development of the GSA.

**Fig. 1.**

*a* Angiogram. Right carotid. The MCA presents a highly irregular aspect, with multiple zones of narrowing interspersed with zones of dilatation. The left carotid, vertebral and renal angiograms were normal. *b* T₁-weighted coronal MRI view. *c* Native MRA view. *d* 3D reconstruction view. The *b*, *c* and *d* views, taken 11 years after the angiogram shown in *a*, show a partially thrombosed giant aneurysm on the insular segment of the MCA.

**References**

Choreoathetosis due to Rupture of a Distal Accessory Anterior Cerebral Artery Aneurysm

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We encountered a patient with choreoathetosis subsequent to aneurysmal rupture. Although 3 cases of hyperkinetic movement disorders, dystonia, tremor and chorea associated with aneurysmal rupture, have been reported [1, 2], ours is the first documentation of choreoathetosis due to aneurysmal rupture and a mass lesion at the aneurysm site. We present a review of the literature to clarify the etiology of this extremely rare symptom.

Case Report

This 72-year-old right-handed woman with no history of hypertension, convulsive disease, orthostatic hypotension, diabetes mellitus, parkinsonism, severe infection or head trauma experienced sudden vomiting and lost consciousness. She did not have a familial history of any movement disorders. On admission, she was confused and manifested very mild left facial paresis, mild dysarthria and tetraparesis. Her muscle tone and proprioceptive sensation were normal. She exhibited choreoathetoid movements mainly in the distal part of her right arm that also involved her right leg and left arm 2 days later; her left leg was spared. The movements were continuous while she was awake and disappeared when she slept. Computed tomography revealed diffuse subarachnoid hemorrhage and an interhemispheric hematoma compressing the isthmus of the corpus callosum downward (fig. 1A, B). Angiography demonstrated absence of the left A1 segment and triplication of a postcommunication site. One branch was an accessory anterior cerebral artery (ACA) situated in the epi-callosal sulcus, the other 2 branches were the right and left ACA irrigated primarily by the mesial portion of the bifrontal convolution. An aneurysm with twin domes and a broad neck was located at the distal trifurcation of the accessory ACA and Fisher’s A5 segment (fig. 1C). There were no cortical branches ahead of the trifurcation; no obstruction or vasospasm and no venous congestion was evident. Hunt and Kosnik grade 3 was recorded and the patient underwent coil embolization of the aneurysm on the day of admission (fig. 1D). Although no special acute phase treatment for choreoathetosis was performed, nimodipine and fasudil were administered transvenously to prevent vasospasm. The involuntary movements of her bilateral upper and right lower limbs became gradually exacerbated during the following 12 days; they were slowly alleviated and ceased by 17 days after onset. Postoperative MRI showed gradual absorption of the hematoma. Perfusion-weighted MRI revealed mild hypoperfusion in the bilateral corona radiata beside the hematoma; there were no distinct areas of perfusion defect in the basal ganglia and thalamus. One month later she was referred to another hospital for rehabilitation. She was readmitted 3 months later and underwent right parietal craniotomy and clipping of the residual aneurysmal neck. She suffered no recurrence of choreoathetosis during an 11-month follow-up.

Discussion

The previously reported chorea occurred 8 days after subarachnoid hemorrhage and was due to vasospasm and secondary hydrocephalus [2]. The involuntary movements in our patient began shortly after subarachnoid hemorrhage onset. As she had no acute hydrocephalus, we posit that her symptoms were attributable to the hematoma on the corpus callosum. It has been suggested that these hyperkinetic involuntary movements occur in the presence of interruption in the cortico-striato-pallido-thalamo-cortical feedback loop (fig. 2) [3, 4]. Studies in humans of connecting motor fibers using diffusion tensor imaging and fiber tracking algorithms revealed that corticostriatal projections of the feedback loop pass through the corona radiata just lateral to the body and isthmus of the corpus callosum together with adjacent or mixed corticospinal pyramidal fibers [5]. As our patient initially manifested motor paresis indicative of pyramidal tract involvement, we postulate that her choreoathetosis was attributable to disinhibition of these adjacent fiber tracts due to transient hypoperfusion or mass effect of the hematoma. Pascual et al. [6] postulated that the hypoperfusion around the hematoma was significant in the subacute stage and disappeared completely during the second week. This may explain our patient’s exacerbation and alleviation of choreoathetosis. Corticostriate fibers cross to the contralateral striatum, therefore, interruption of crossing fibers at the corpus callosum is an alternative explanation for her choreoathetosis [7]. However, as the hematoma on the corpus callosum persisted for a relatively long period after her total recovery from choreoathetosis, this explanation appears inadequate. Our patient did not exhibit choreic movement of her left leg. It has been suggested that motor paresis due to increased involvement of the motor cortex or pyramidal tract may result in the elimination of dyskinesia [3, 4] and this hypothesis may also apply to our case.

The reported incidence of accessory ACA ranges from 3.3 to 15% [8, 9]. Only 4 distal accessory ACA aneurysms have been reported to date [9–11]. Baptista [8] described 3 patterns of distal ACA anomalies, i.e. unpaired (azygous) arteries (fig. 3B), bihemispheric arteries giving rise to branches on the contralateral hemisphere (fig. 3C, D) and triplication of the postcommunicational segment (fig. 3D–F). In our case the accessory ACA was bihemispheric; it gave rise to bihemispheric branches after the trifurcation (fig. 3D). There are only 2 previously reported cases with a bihemispheric accessory ACA who presented with aneurysmal rupture [9, 11]. Interestingly, all 5 distal accessory ACA aneu-