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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 at a postcommunication site. One branch was an accessory anterior cerebral artery (ACA) situated in the epicallous 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-

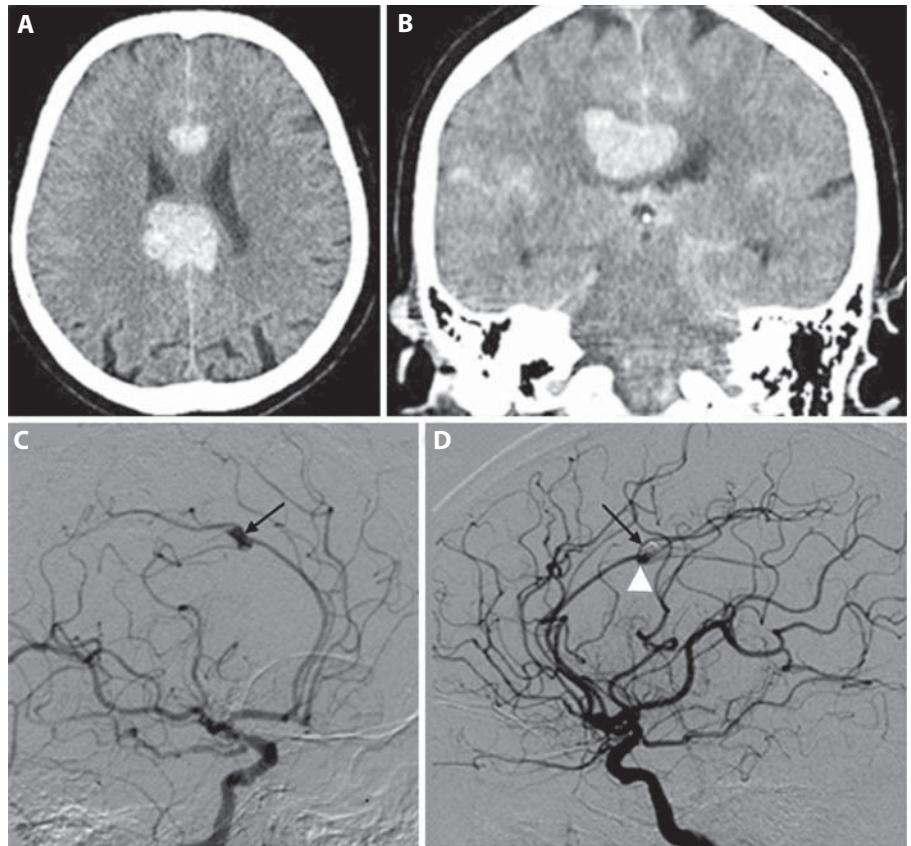


Fig. 1. **A, B** CT scan showing diffuse subarachnoid hemorrhage and an interhemispheric hematoma above the isthmus of the corpus callosum. **C, D** Right carotid angiogram showing triplication of the ACA and a twin-domed aneurysm at the distal accessory ACA. **C** The arrow identifies the aneurysm on the right lateral oblique view. **D** The embolized aneurysm (arrow) and the residual aneurysm (arrowhead) are shown on the left lateral view.

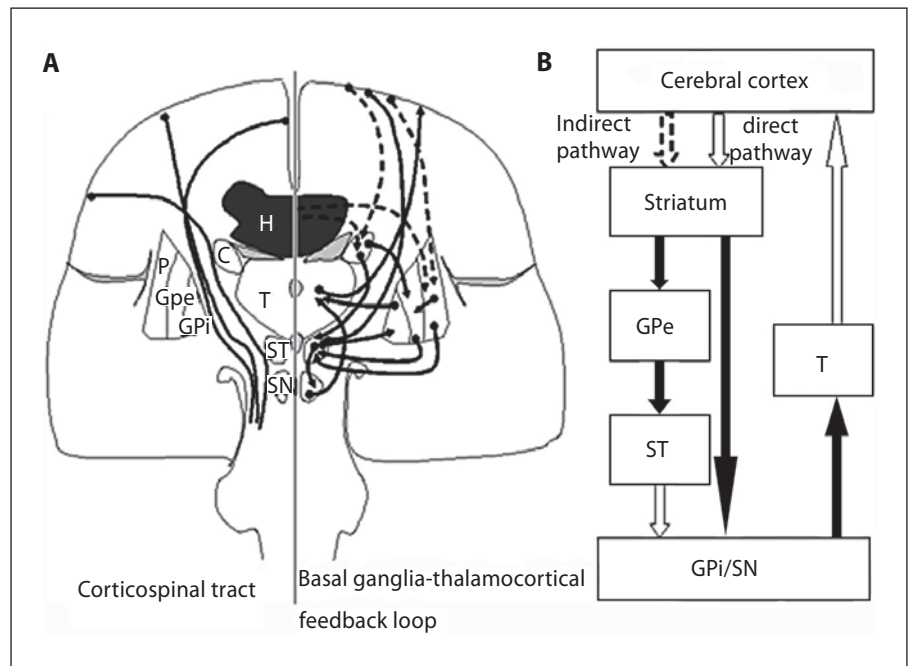
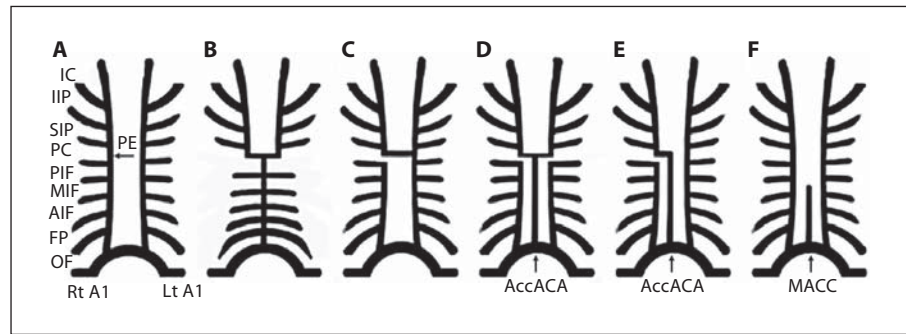


Fig. 2. Fiber tracts (**A**) and simplified cortico-striato-pallido-thalamo-cortical feedback loop (**B**). Inhibitory neurons are shown as filled symbols and excitatory neurons as open symbols. Dotted line indicates corticostriatal fibers that are supposed to be involved in the mechanisms of choreoathetosis. P = Putamen; C = caudate nucleus; T = thalamus; H = hematoma; Gpe = external segment of globus pallidus; Gpi = internal segment of globus pallidus; ST = subthalamic nucleus; SN = substantia nigra pars reticulata.

Fig. 3. ACA types. **A** Normal. **B** Unpaired (azygous). **C** Bihemispheric lateral. **D** Bi-hemispheric accessory. **E** Unihemispheric accessory. **F** ACA with median artery of the corpus callosum remnant. The pericallosal (PE), orbitofrontal (OF), frontopolar (FP), anterior internal frontal (AIF), middle internal frontal (MIF), posterior internal frontal (PIF), paracentral (PC), superior internal parietal (SIP), inferior internal parietal (IIP) and inferior callosal (IC) arteries are depicted.



rysms including our case were located at the A5 segment at the junction from which the first cortical branches of the postcommunical segment diverge [9–11]. This suggests a direct contribution by hemodynamic stress to aneurysm formation and growth.

With respect to the etiology of our patient's choreoathetosis we postulate that the presence of an ACA anomaly may have led to aneurysm formation at the A5 portion and subsequent rupture due to hemodynamic stress. The interhemispheric hematoma above the corpus callosum that derived from rupture of the distal ACA aneurysm may have led to selective disinhibition of indirect corticostriatal fibers of the feedback loop at the corona radiata and resulted in the manifestation of choreoathetosis.

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