

Akinetic mutism and utilization behavior after bilateral thalamo-polar artery stroke

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Reports of bilateral thalamo-polar artery stroke are rare and data of long-term outcome are exceptional.

A 46-year-old healthy and professionally active male worker was admitted in the emergency room with fever, meningism, without neurological deficits. The diagnosis, as supported by neuroimaging and LCR analysis, were pneumococcal meningitis and mastoiditis. Intravenous antibiotics were started and mastoidectomy was performed. 48 h later the patient showed a sudden deterioration of consciousness without lateralized signs. The clinical picture, as suggested by MRI-DWI images (Fig. 1), was due to an acute bilateral ischemic stroke in the ventral anterior thalamus and anterior thalamic peduncles. Brain MRI showed neither focal cortical lesion nor dural or internal venous thrombosis. An extensive workup excluded, besides meningeal vessels inflammation, other causes of stroke (i.e., endocarditis, atrial thrombi, and patent foramen ovale).

A normal sleep–waking cycle appeared within 4 weeks, but the patient did not show further signs of recovery for the following 48 months until the last follow-up (of which the first 9 were spent in a dedicated neurorehabilitation center and then in a nursing home). The patient did not show any emotional expression. He spent all the time mute

with his eyes open, but neither was he able to comprehend and/or to follow simple instructions nor did he show any verbal initiative, and had no finalistic movements, even for feeding and sphincteric functions. However, he showed an unceasing grasping (e.g., clothes, urinary and enteral tubes, chair) with unrestrainable manipulation of whatever object the examiner placed in the personal space. A gastric feeding tube was used for long-term nutrition. The intrinsic and extrinsic ocular motility was spared. There were no signs of corticospinal involvement, there were no focal neurological signs or motor deficits, and the patient had pain withdrawal reflexes. In the upright posture (with the help of two persons) the patient remained with the head and four limbs flexed.

The patient's condition did not respond to any pharmacological intervention (dopamine, amantadine, acetylcholine-esterase inhibitors). We supposed that the persistent akinetic mutism and utilization behavior was related to

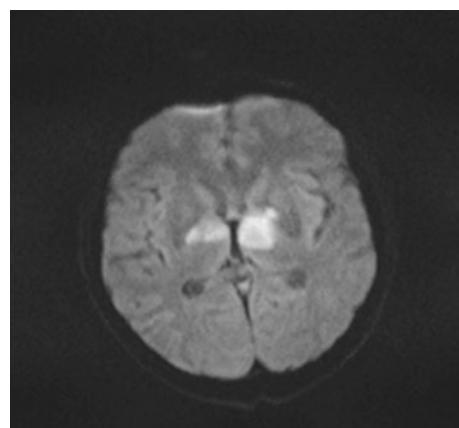


Fig. 1 IRM DWI images showing isolated bilateral ischemic acute stroke in the thalamo-polar artery territory distribution

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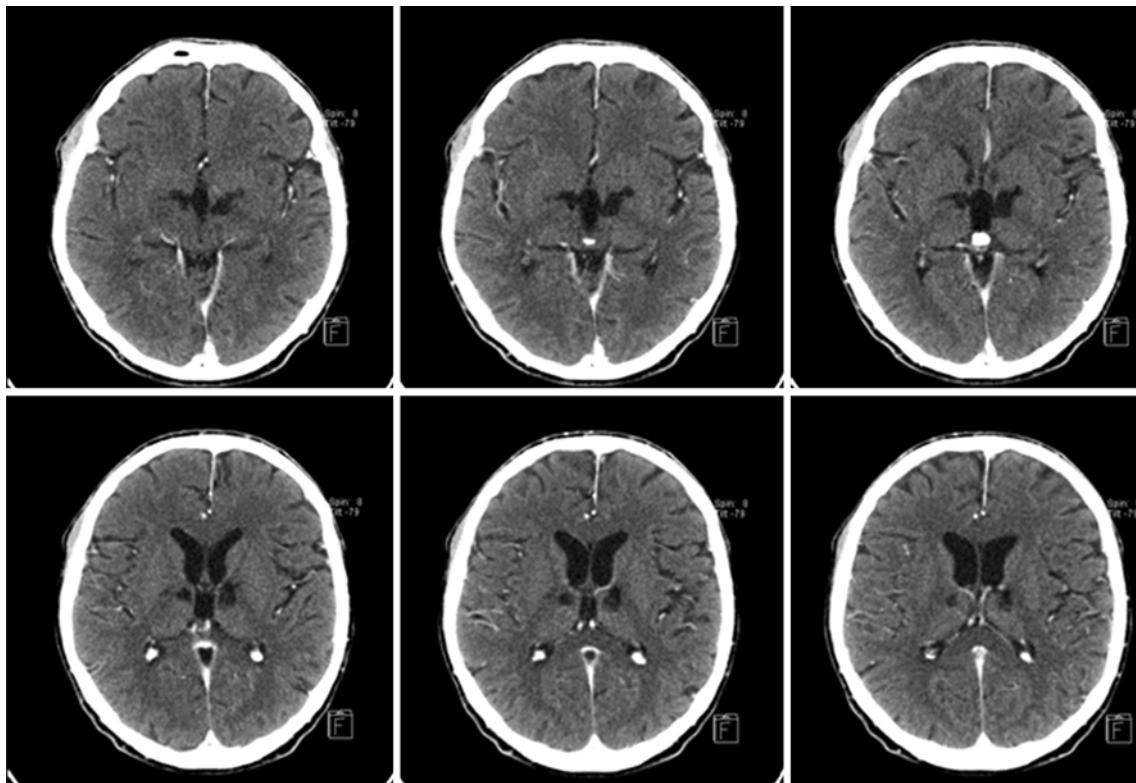


Fig. 2 Transverse brain CT-scan sections showing residual bilateral tuberothalamic artery territory stroke involving mainly the anterior thalamic nuclei and also thalamic peduncles

the ischemic strokes in the vascular territory of the thalamo-polar arteries (with the involvement of the anterior thalamic nuclei, anterior thalamic peduncles and the mammillo-thalamic tract) and hypothalamic perforant vessels (Fig. 2).

A vast majority of patients with unilateral thalamo-polar stroke (independently from the hemisphere) manifest prominent executive dysfunction (especially superposition of unrelated information and perseveration), apathy, word finding difficulties and severe memory deficits [1, 2]. Although data from literature are scanty, the reported outcome is mainly negative (often resulting in “strategic-infarct-dementia”) [2].

Reports of thalamic stroke associated with akinetic mutism [3] and utilization behavior [4] are rare and generally the dorsomedial nuclei are involved. However, we were not able to find similar cases with bilateral ischemic lesions limited to the anterior thalamic nuclei. The anterior thalamic nuclei receive projections from the mammillo-thalamic tract (MTT), monoaminergic and cholinergic nuclei, and have dense reciprocal connections with the extended limbic system (including the cingulate gyrus, hippocampus, parahippocampal formation, and orbitofrontal cortex) and with the medial and lateral prefrontal cortex [5].

Thus, the clinical picture of our patient could be related to the bilateral disconnection of the anterior nuclei from limbic and frontal cortices throughout the damage of the thalamic anterior peduncles. While, at least, some functions of a brain area affected by a unilateral thalamic lesion can, in principle, be compensated by the contralateral side, bilateral lesions lead to a loss of function in both hemispheres.

This single case report suggests that thalamic anterior nuclei and their related cortical loops are fundamental relays that subserve motivation and purposeful behaviors. Their interruption might result in a persistent apathetic and extremely severe dysexecutive syndrome (akinetic mutism with unceasing grasping and manipulation behavior).

Conflict of interest The authors declare that they have no conflicts of interests and that they have no grants to disclose for this study.

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