CASE REPORT/*OPIS PRZYPADKU*

Bilateral paramedian thalamic infarction with hypothalamic dysfunction

Obustronny udar wzgórza z dysfunkcją podwzgórza

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Abstract

Unilateral thalamic lesions cause transient or permanent behavioral, sensory and oculomotor disturbances; bilateral lesions of thalamus result in more severe and longer lasting symptoms. We present an atypical case of bilateral paramedian thalamic infarct with concomitant hypothalamic dysfunction. The only risk factor of ischaemic stroke found in the patient was a short lasting episode of atrial fibrillation. Bilateral paramedian thalamic infarcts may result from occlusion of one paramedian thalamic artery, which arises from the posterior cerebral artery, either with separated or with a common trunk, thus supplying the thalamus bilaterally. Independently of anatomical variants of thalamus blood supply, the most probable cause of infarct in our patient was unilateral or bilateral occlusion of the posterior cerebral artery by cardioembolism, probably in the course of basilar artery occlusion. Hypothalamic dysfunction may accompany thalamic infarcts; thus hypothalamo-pituitary function should be routinely assessed in bithalamic infarcts.

Key words: ischaemic stroke, bilateral thalamic infarct, hypothalamic dysfunction.

Streszczenie

Jednostronne udary w obrębie wzgórza mogą być przyczyną przemijających lub trwałych zaburzeń zachowania, objawów czuciowych lub zaburzeń gałkoruchowych. Obustronne udary wzgórza skutkują zwykle bardziej nasilonymi i dłużej trwającymi objawami. W niniejszym artykule zaprezentowano przypadek obustronnego udaru wzgórza z jednoczesną dysfunkcją podwzgórza. Jedynym czynnikiem ryzyka udaru niedokrwiennego mózgu, jaki stwierdzono u pacjenta, był krótkotrwały epizod migotania przedsionków. Obustronne udary wzgórza mogą być wynikiem zamknięcia jednej tętnicy przyśrodkowej wzgórza, odchodzącej od tętnicy tylnej mózgu albo w postaci dwóch osobnych gałęzi, albo też jednego wspólnego pnia, zaopatrującego jednak wzgórze obustronnie. Niezależnie jednak od wariantów anatomicznych unaczynienia wzgórza najbardziej prawdopodobną przyczyną udaru niedokrwiennego u przedstawionego pacjenta było jednostronne lub też obustronne zamknięcie tętnicy tylnej mózgu przez materiał zatorowy, prawdopodobnie w przebiegu zamknięcia szczytu tętnicy podstawnej. Dysfunkcja podwzgórza może towarzyszyć udarom wzgórza, autorzy sugerują więc, aby funkcja układu podwzgórzowo-przysadkowego była rutynowo oceniana w przypadku obustronnych udarów wzgórza.

Słowa kluczowe: udar niedokrwienny, obustronny udar wzgórza, dysfunkcja podwzgórza.

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Introduction

Acute bilateral infarction of the thalamus is not a common condition as it represents 0.6% of first-ever acute ischemic strokes [1], but is usually associated with specific neurological and neuropsychological symptoms. As the thalamus is one of the strategic regions of the human brain, the knowledge of its functions and clinical consequences of its lesions is vital [1-3]. Vascular lesions destroy thalamic nuclei in different combinations producing sensorimotor and behavioral syndromes depending on the nuclei involved. Unilateral thalamic lesions cause transient or permanent behavioral, sensorial or ocular motor disturbances; bilateral lesions of the thalamus result in more severe and longer lasting symptoms and complete recovery in these cases is rare [4]. Bilateral paramedian thalamic infarctions account for one third of all paramedian infarcts [5]. Here, we discuss an atypical case of acute bilateral paramedian infarct with concomitant hypothalamic dysfunction.

Case report

A fifty-eight-year-old man with sudden onset of vigilance disturbances, without previous history of concomitant disorders, was found by his family in the morning in a comatose state with preserved breathing and circulation. The patient was referred to the department of toxicology, where toxic causes of coma were excluded. Before admission to the toxicology department, he was consulted by a neurologist, and brain computed tomography (CT) was performed a few hours after disease onset, which revealed no brain abnormality; the patient's blood pressure on first neurological examination was normal. The second brain CT performed on day 2 after disease onset (< 48 hours) revealed no pathology again. On the 3rd day he was referred to our Department of Neurology to continue diagnostics. Neurological examination performed on admission revealed a comatose patient (Glasgow Coma Scale – 3 points) with equal pinpoint pupils unreactive to light and discrete right hemiparesis, right-sided Babiński sign was also present. The patient remained comatose for 3 days, after which he recovered consciousness but presented pathological sleepiness. From the 4th day, the consciousness was fully recovered, but the patient presented hypersomnia, amnesia, transcortical aphasia, vertical gaze palsy and discrete right hemiparesis; his reflexes were decreased in the right limbs.

On the brain magnetic resonance imaging (MRI) performed on the 4th day after the disease onset, bi-thalamic ischemic lesions in the paramedian territories were found (hyperintense lesions in T2-weighted and FLAIR sequences located bilaterally in the paramedian thalamus) (Figs. 1A and 1B). Brain angio-CT scans were performed

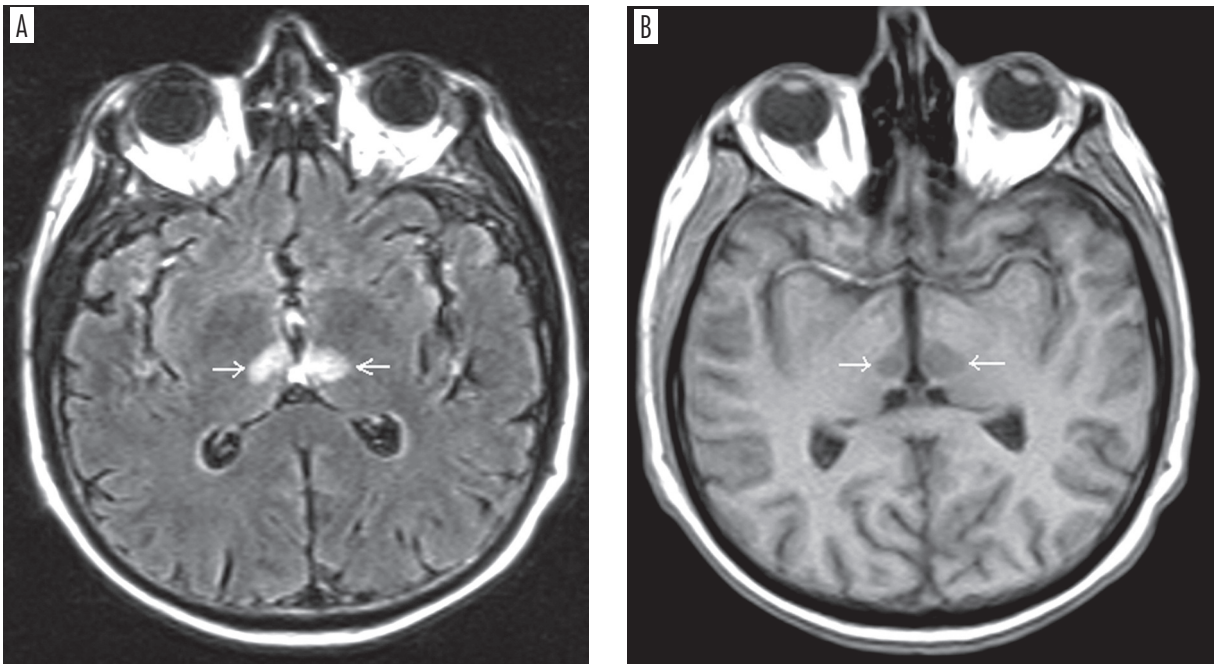

Fig. 1. Magnetic resonance imaging of the patient with bilateral thalamic infarct. Axial FLAIR images (A) and T1-weighted images (B) are shown. Sites of lesion are indicated by arrows

subsequently (to exclude cerebral deep venous thrombosis), revealing a filling defect of both P1 segments of posterior communicating arteries. Complete blood cell count, biochemical profile, and urinalysis were within normal limits. Thiamine deficiency, cerebral lupus and toxoplasmosis were excluded. Additionally, we performed transcranial Doppler and duplex sonography of the carotid and vertebral arteries, 12-lead electrocardiography (ECG), 24-hour ECG monitoring, and transthoracic and transesophageal echocardiography. Neither transthoracic nor transesophageal echocardiography revealed pathology. Only 24-hour ECG monitoring revealed one paroxysmal short-lasting episode of atrial fibrillation, which remained unnoticed by the patient. Blood coagulation (including mutation of factor V, antithrombin, and lupus anticoagulant) were normal.

Results of more detailed blood tests revealed hypothyroidism, secondary to hypothalamo-pituitary dysfunction; serum free T4 (fT4) and free T3 (fT3) levels were lowered, and the thyroid stimulating hormone (TSH) level was undetectable. The patient also presented ACTH, cortisol, gonadotropin, dehydroepiandrosterone (DHEA) and testosterone deficiency. The prolactin and growth hormone levels were within normal limits. After a week, the patient also presented symptoms of diabetes insipidus. Repetitive measurement of blood electrolytes revealed hypernatremia; urinalysis demonstrated a dilute urine with a low specific gravity.

In the follow-up period the patient required substitutional treatment with hydrocortisone, thyroid hormones and oral preparations of vasopressin analogues. On brain MRI, no structural damage of the pituitary gland were found, but the presence of multihormonal deficiency indicated loss of function of upper centers in the hypothalamus.

On discharge from hospital, the patient presented amnestic syndrome, reduced verbal fluency and vertical gaze paresis. Although the memory and concentration improved, 6 months after stroke onset the patient did not regain his premorbid levels. He was referred for intensive cognitive neurorehabilitation.

Discussion

The typical cause of bilateral paramedian thalamic infarcts is an occlusion of the paramedian thalamic artery resulting in loss of consciousness or somnolence at stroke onset, subsequent concentration and orientation deficits as well as memory deficits [6]. The paramedian artery arises from the P1 section of the posterior cerebral artery, but it may differ substantially [7]. Percheron distinguished three variants of this artery. The paramedian arteries can arise as a pair from each P1 section (type I), but they also may arise from one P1 portion of the posterior cerebral artery either with separated (type IIa) or with a common trunk (type IIb), thus supplying the thalamus bilaterally [8]. In type III, we observe an arcade of thalamic perforators branching from an artery which connects both P1 segments. The paramedian artery supplies a variable part of the thalamus, but usually the dorsomedial nucleus, internal medullary lamina and the intralaminar nuclei: central lateral, centromedian and parafascicular. Sometimes lateral dorsal, lateral posterior and ventral anterior nuclei may also be supplied. In the absence of tuberothalamic artery, the paramedian artery assumes that territory as well. Percheron's artery (tuberothalamic artery, TTA) originates from the middle third of the posterior communicating artery and may be visualized in vivo by superselective early angiography; we did not attempt to verify its presence because it could have carried a high risk because of the state of the patient.

Our patient was unusual, because he presented concomitant hypothalamic dysfunction. Normally, the TTA and the paramedian artery are two arteries responsible for the vascular supply of posterior parts of the hypothalamus where the sympathicoexcitatory tract has its origin [9]. For this reason, infarction in the hypothalamic region occurs rarely and when it happens the clinical and radiological damage is often limited [10]. In our case angio-CT scans of brain vessels revealed a bilateral filling defect in posterior communicating arteries; thus we hypothesize that in our patient the paramedian artery was responsible for blood supply to the posterior part of the hypothalamus, and its occlusion resulted in bilateral thalamic infarct with hypothalamic dysfunction. Usually paramedian artery occlusion results from occlusion of the P1 portion of the PCA as a variant of tip basilar artery (BA) occlusion, or is subsequent to tip BA occlusion. Cases of concurrent infarction in the paramedian and tuberothalamic territory are rare in the literature [11]. Nevertheless, independently of anatomical variants of thalamus blood supply, the most probable cause of infarct in our patient was occlusion of the P1 segment of the PCA, unilateral or bilateral, in the course of BA top occlusion, resolved by the 4th day, which was seen in angio-CT scans.

Bilateral infarction in the paramedian artery territory may result in disorientation, confusion, hypersomnolence, deep coma, 'coma vigil' or akinetic mutism and severe memory impairment often accompanied by eye movement abnormalities [12]. Anterograde and retrograde

memory deficits and apathy are typical, as well as impulsive, aggressive behaviors, emotional blunting, loss of initiative and absence of spontaneous mental activities. Nevertheless, complete recovery from bilateral paramedian thalamic infarction has also been reported [4]. It is worth noting that bilateral and left-sided infarcts have worse outcome than right-sided ones [13]. The main structures involved in patients with memory deficits are the mamillothalamic tract, internal medullary lamina and dorsomedial nucleus [4,6], although there are some data suggesting that the latter is not involved in memory deficits [14]. Loss of psychic activation and thalamic dementia are probably caused by interruption of the striatal-ventral pallidal-thalamic-frontomesial limbic loop [2,15].

Typical oculomotor signs resulting from lesions in the paramedian artery territory may cause complete or partial vertical gaze paresis, loss of convergence, bilateral intranuclear ophthalmoplegia, miosis and sometimes intolerance to bright light [6]. Lesions of the medial thalamus disrupt the corticofugal fibers that lead from motor and premotor cortices to the nuclei of the midbrain, which are responsible for vertical gaze (up and down); they also disrupt the fibers to the rostral nucleus of the medial longitudinal fasciculus responsible for downgaze [16]. In our patient, we observed at the beginning a complete vertical gaze paresis, which improved with time; after 8 weeks the patient presented a partial vertical gaze paresis. Nevertheless, the prognosis of full recovery of vertical eye movements is poor [3].

The most frequent causes of stroke in patients with bilateral thalamic involvement are firstly lacunar strokes (small vessel disease), and secondly embolic ones (artery to artery embolism, cardioembolism) [1,6]. In our case no history of concomitant disorders was present, but a single episode of atrial fibrillation confirmed in 24-hour ECG was found; therefore the most probable cause of bithalamic stroke seems to be embolus to the tip of the basilar artery with subsequent occlusion of the paramedian artery. However, it is worth noting that bilateral thalamic infarcts may rarely be symptomatic of cerebral deep venous thrombosis [17], which we excluded by angio-CT and brain MRI angiography. It is also worth noting that in paramedian infarcts a cardioembolic source is more frequent than in other thalamic infarcts [6]. Atrial fibrillation and atrial flutter remain the most frequent causes of cardioembolic stroke, but they usually escape from standard ECG or Holter monitoring [18].

To conclude, hypothalamic dysfunction may be a symptom of damage of the area adjacent to the thalamus, resulting from occlusion of the variants of the paramedian artery or tuberothalamic artery. Therefore hypothalamo-pituitary function should be routinely assessed in bithalamic infarcts.

Disclosure

Authors report no conflict of interest.

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