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# **Bilateral Paramedian Thalamic Infarct in the Presence of an Unpaired Thalamic Perforating Artery**

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#### Summary

Bilateral paramedian thalamic infarction is rare. The suggested mechanism is occlusion of a central unpaired thalamic perforating artery – an anatomic variant. In the few existing reports of this condition, the diagnosis was based on computed tomography (CT) or magnetic resonance imaging (MRI) findings alone. Other causes of thalamic lesions were not ruled out, and there was no angiographic demonstration of the presumed variant artery.

We present a case of a 48-year-old man with a bilateral thalamic infarction seen on CT and MRI. Initial neurological examination revealed lethargy, severe combined motor and sensory aphasia, and a mild upward gaze limitation. The patient had no focal motor deficits. After 24 hours, the patient was more alert and his speech became more fluent, but Korsakoff-type amnesia with poor attention span became apparent. The patient improved slowly over 6 months of rehabilitation.

Bilateral thalamic lesions can be caused by several conditions. Among those are thiamine deficiency, cerebral lupus, toxoplasmosis, cysticercosis, cerebral syphilitic gumma, and even tumors and fungal infections. All these were ruled out in our case. Superselective digital subtraction angiography (DSA) demonstrated a single unpaired thalamic perforator. To our knowledge, this is the first time this anatomical variant has been demonstrated in vivo in association with bilateral thalamic infarction.

*Keywords:* Thalamic perforators; bilateral thalamic infarction; amnesia; cerebral angiography.

## Introduction

Bilateral paramedian thalamic infarction is rare. The suggested mechanism is occlusion of a single central thalamic perforating artery – an anatomic variant described by Percheron in 1977 [8]. This condition has been reported several times over the past 20 years [1, 4, 6]. However, these reports relied only on the CT or MRI findings of probable bilateral thalamic infarction. Other causes of thalamic lesions were not ruled out, and there was no angiographic demonstration of the presumed variant artery. Therefore, this hypothesis has never been supported by strong empirical evidence.

We present a case of bilateral thalamic infarction seen on CT and MRI, in which superselective DSA demonstrated the single thalamic perforator.

We describe the clinical syndrome seen in this patient and discuss the differential diagnosis of necrotic thalamic lesions.

#### Case Report

A 48-year-old right-handed Caucasian male was admitted to the emergency room at another hospital. He had been found unconscious in his home four days after failing to appear at a family function. On admission, he was found to have non-ketotic hyperosmolar hyperglycemia, with glucose levels of above 600 mg/dl. The patient also had hypothermia (core temperature 90 °F), coagulopathy, and hypothyroidism.

The patient was transferred to our hospital. On admission, physical examination was significant for dehydration; gangrene of toes bilaterally with clear demarcation; and multiple skin abrasions on the knees, abdomen, arms, and right shoulder. On neurological examination the patient was awake but oriented only to self, with a severe combined motor and sensory aphasia. He was able to follow only simple one-step commands. The patient had difficulty concentrating and participating in the neurological examination. His affect was flat and he did not initiate movement, indicating marked abulia. Cranial nerve examination was only significant for a mild upward gaze limitation. The patient had no focal motor deficit.

Further review of history and medications revealed that the patient had non-insulin dependent diabetes mellitus (NIDDM) that had been treated with Glucotrol, but several weeks before the hemorrhage, he had been instructed to take amoxicillin, stop oral agents, and control his diabetes with diet alone. CT scan and an MRI of the brain revealed bilateral small paramedian anterior thalamic lesions that enhanced minimally after injection of gadolinium, as well as a small amount of subarachnoid blood in the left posterior frontal and parietal area (Fig. 1).

Initial treatment included hydration and correction of metabolic abnormalities. Neurologically, the patient improved slowly. After 48

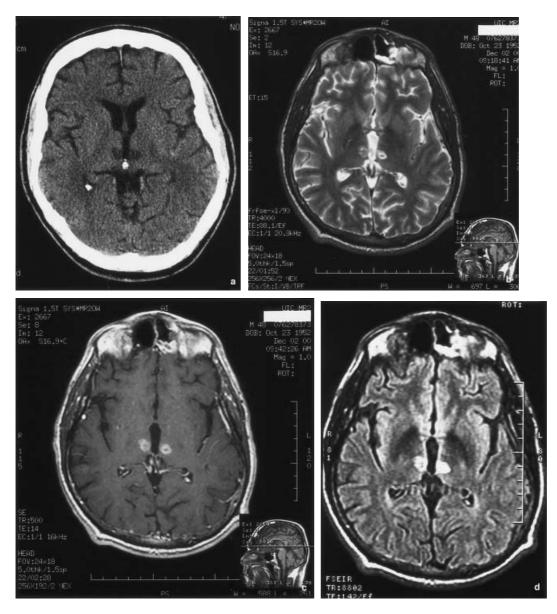


Fig. 1. Axial CT scan (a), T2 (b), T1 with gadolinium (c), and FLAIR (d) MRI images, demonstrating bilateral round thalamic lesions, suggestive of infarction

hours his speech became more fluent. He cooperated with a neuropsychological evaluation at that time, which demonstrated an alert and awake patient disoriented to place and time. Memory, both long- and short-term, and constructional abilities were markedly impaired, and he confabulated his answers. Comprehension and naming were also affected. Deficits were found in information processing, insight, and judgement. EEG revealed only a global slowwave abnormality.

Bilateral thalamic infarction was strongly suspected. Additional studies were done to clarify the diagnosis. Thiamine level was 2.0 ug/dl. Influenza A and B titer was <1:8. Anti double strand DNA antibody level was zero. Screening test for HIV AV was negative. Tests for *Toxoplasma* antibodies were negative. Echocardiogram was normal, showing no cardiac source of embolism and no

patent foramen ovale. A cerebrospinal fluid (CSF) sample obtained by lumbar puncture revealed no abnormal cells. Six weeks of fungal and acid fast bacilli culture of the CSF revealed no pathogens. Cerebrospinal fluid VDRL was non-reactive. Cerebral DSA revealed the presence of a variant central thalamic-subthalamic perforating artery. Based on these findings, we diagnosed a bilateral paramedian thalamic infarction secondary to embolism or hypoperfusion of a single unpaired perforating artery (Fig. 2).

After discharge to a rehabilitation facility, the patient and his parents reported slow improvement. After one month of rehabilitation, the patient continued to display multiple deficits in memory and higher language functions. Orientation accuracy was 10%, he identified 60% of common objects and demonstrated 65% comprehension on formal language testing. Expression was tangential with con-

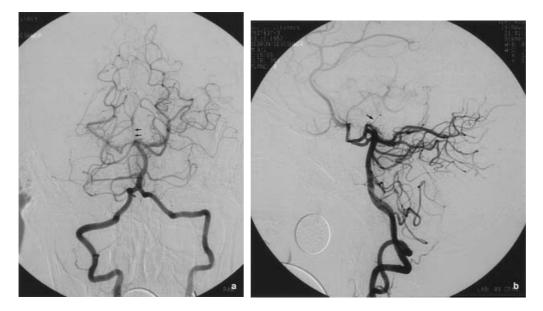


Fig. 2. Digital subtraction angiography – right vertebral injection. AP (a) and lateral (b) views demonstrate a single unpaired central thalamic perforating artery (arrows)

fabulations. He had difficulty reading and concentrating. His gait was wide-based. There were no focal motor or sensory deficits and no tremor or dyskinesia. At follow-up visits 3 and 6 months after discharge from the hospital, he continued to show gradual improvement.

## Discussion

We present a case of a patient who apparently suffered a prolonged coma, followed by a neurological syndrome of abulia, partial aphasia, amnesia with confabulation (Korsakoff type), and limitation of upward gaze. These neurological findings have been described in other cases of bilateral paramedian thalamic infarction [2, 4, 7]. Angiography and MRI in our case were consistent with a bilateral paramedian thalamic infarction which probably resulted from temporary embolic occlusion or hypoperfusion in the territory of a central thalamic perforating artery, an uncommon anatomical variant. The variant artery arose from the P1 segment of one posterior cerebral artery as an unpaired perforating artery, as described in a systematic review by Percheron [8]. The initial event could have been severe hyperglycemia, followed by a decreased level of consciousness and dehydration and probably exacerbated by a fall and head trauma. Hypotension, embolism, or the trauma itself could have caused an infarction in that territory [1].

Although bilateral thalamic infarction may be part of the top of the basilar syndrome [11], the isolated finding of bilateral paramedian thalamic infarction is rare. Two cases were reported by Guberman and Stuss in 1983 [4], who described a clinical syndrome including coma, Korsakoff type amnesia, and vertical gaze disturbance. The diagnosis was based on the clinical picture and CT findings only. Other reports by Lepore *et al.* [6] and Barontini and Maurri [1] demonstrated the lesions on autopsy and MRI, respectively. The presence of an unpaired thalamic perforator was presumed, but not proven.

Stroke was the working diagnosis in our case, but bilateral thalamic lesions may be caused by several other, less likely conditions. These include thiamine deficiency [13], cerebral lupus [12], abscesses caused by fungi or toxoplasmosis [3], cerebral syphilitic gumma [10], and tumors [5]; in children, influenza has been described with bilateral thalamic necrosis [9]. All these were ruled out in our case. The availability of highresolution digitally enhanced angiography allowed us to demonstrate the anatomical variant artery in vivo.

To the best of our knowledge, this is the first report of a case of bilateral thalamic infarction in which an unpaired thalamic perforator artery was demonstrated on superselective DSA. Our findings support the hypothesis that an unpaired thalamic perforator is present in cases of bilateral thalamic infarction.

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#### Comments

This is a highly interesting case report. Bilateral thalamic infarction has never, as far as I know, so beautifully and so completely been illustrated. Particularly the angiogram is unique. There can be little doubt that the artery which originates near the top of the basilar artery is an unpaired thalamic perforating artery. The combination of the bilateral lesions in the thalamus and the unpaired thalamic perforating artery strongly supports the conclusion of the authors that this is an infarction of the thalamus and no cerebral lupus, thiamine deficiency, toxoplasmosis, cysticercosis, syphylitic gummata or tumors and fungal infections.

C. Tulleken

In this well written case report, the authors describe the occurrence of bilateral paramedian thalamic infarct associated with the finding of a single posterior thalamic perforator, as revealed by digital substraction angiography.

Brassier *et al.* has explained this anatomical variation on the basis of the way that the embryonic P1 segments merge [1]. Symmetrical merging of the embryonic P1 segments into the superior basilar complex results in common trunks that originate on each P1 segment, supplying the mesencephalodiencephalic territory. When the merging is asymmetrical, a single mesencephalodiencephalic trunk supplies the whole area bilaterally.

This teaching file case, demonstrating clinical correlation with an anatomical variant, should remind readers of the existence of anatomical disparities and their potential clinical implications, especially in neurovascular pathology.

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