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Allodynia in the flank after thalamic stroke

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Abstract

Lesions responsible for thalamic pain are often thought to involve the ventral posteromedial nucleus and ventral posterolateral nucleus of the thalamus. We describe two patients with allodynia and hyperpathia in the contralateral flank caused by a small lesion in the posteroventral part of the thalamus. When considered with the literature, involvement of the ventral posteroinferior nucleus may be responsible for this unique post-stroke pain syndrome.

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1. Introduction

Sensory abnormalities after a minor stroke are occasionally restricted to a small portion of the body. This restricted sensory syndrome most often presents in the pattern of a cheiro-oral syndrome, a combined sensory abnormality over the contralateral hand or fingers and perioral region. Since the first report by Fisher,¹ patients with restricted sensory changes in the palm and foot (cheiro-pedal syndrome) or in the palm, foot, and perioral area (cheiro-oral-pedal syndrome) have been reported. However, despite numerous case reports describing restricted sensory syndrome following minor cerebral infarction,² to our knowledge there have been no reports in which the sensory disturbance included the flank. We describe two patients with allodynia and hyperpathia in the contralateral flank region in combination with paraesthesia in the palm, perioral area, or foot.

2. Case reports

2.1. Patient 1

A 75-year-old man with a history of chronic hepatitis developed sudden onset numbness of the right forearm. Within a few minutes, he also noticed numbness of the right lower leg and developed a slight right hemiparesis. There was no diplopia, nausea, or vomiting. On presentation to the emergency department, his blood pressure was 188/112 mmHg. His consciousness level was normal. Eye movements were normal with no nystagmus. There was no ton-

gue deviation on protrusion and the other cranial nerves were normal. There was no limb weakness, however deep tendon reflexes were slightly exaggerated on the right. He had paraesthesia over the forearm and lower leg on the right. Superficial sensation was normal in these areas, but deep sensation was slightly decreased. One week later, he developed allodynia and hyperpathia in the right flank (Fig. 1), which gradually increased in intensity over the next month. His liver transaminases were slightly elevated due to chronic hepatitis but other routine blood and urine examinations were normal. Brain CT scan after the onset of the stroke was normal. A subsequent MRI scan revealed a lesion restricted to the ventrocaudal nuclei of the thalamus (Fig. 2A). The MR angiogram was normal. Somatosensory evoked potentials (SEPs) by median and ulnar nerve stimulation showed normal latencies of N9, N11, N13, N20 and N32 bilaterally, but the amplitude of N20 and N32 elicited by right median and ulnar nerve stimulation was decreased. The chest X-ray was normal. Abdominal CT showed chronic hepatitis with gall bladder stones. A repeat brain MRI after the appearance of allodynia in the right flank showed no new changes. The paraesthesia in the right thumb, index finger and right foot and the allodynia and hyperpathia in the right flank persisted long-term.

2.2. Patient 2

A 46-year-old man came to the emergency room for evaluation of left hemi-paraesthesia. He had a 10-year history of hypertension. He suddenly developed numbness on the left side of the body, most notably in the forearm and perioral region, along with a mild loss of power in the left hand and leg. His blood pressure was 170/100 mm Hg. Conscious level was normal. He had mild dysarthria. Cranial nerves were normal. Subjective numbness of the left cheek, around the mouth and in the left hand and foot were reported. Sensation in response to pinprick, light touch and temperature on the left side of the mouth, left hand and foot were decreased. Vibratory sensation of the left hand was also decreased. Mild weakness in the left arm and leg was noted. Deep tendon reflexes were slightly brisk on the left, although the Babinski response was normal. The symptoms gradually improved and the dysarthria and weakness disappeared within one week. One month later, the paraesthesia of the left foot resolved completely. However, the paraesthesia of the left thumb and index finger and the left side of the mouth was persistent. Two months after the onset of stroke, allodynia and hyperpathia of the left flank commenced and gradually worsened (Fig. 1). Routine blood and urine examination and electroencephalogram were normal. Brain CT scan at onset of stroke showed a small hemorrhage in the right lateral thalamus. Subsequent MRI showed hyperintensity on T1- and T2-weighted images in the ventrocaudal nuclei of the thalamus (Fig. 2B). MR angiography and cerebral angiography showed mild vascular sclerosis. Follow up MRI after the onset of the sensory disturbance in the left flank showed no new changes.

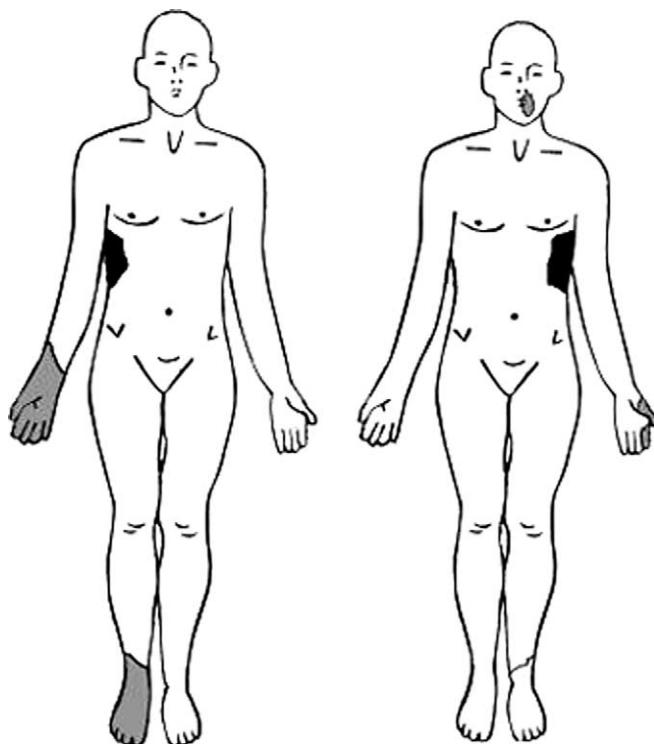


Fig. 1. Diagrammatic representation of the distribution of the sensory deficits in patient 1 (left) and patient 2 (right).

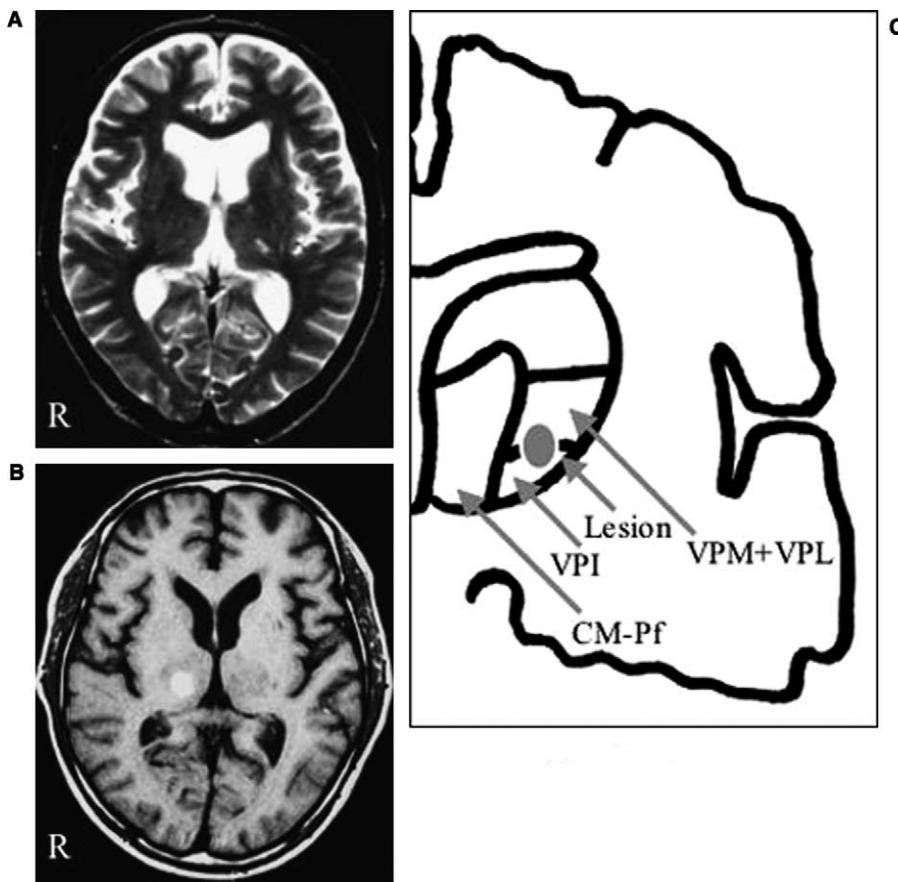


Fig. 2. A. Axial T2-weighted MRI showing the left thalamic infarct in patient 1. B. Axial T1-weighted MRI showing the right thalamic hemorrhage in patient 2. C. Diagrammatic representation of the location of the lesions in A and B. VPL: ventral posterolateral nucleus, VPM: ventral posteromedial nucleus, VPI: ventralis posteroinferior nucleus, CM-Pf: centrum medianum-parafascicularis nuclei.

3. Discussion

To our knowledge, there have been no previous reports of thalamic stroke causing allodynia in the flank in combination with sensory disturbance of the hand and foot or perioral area without other neurological symptoms.

In the two cases presented, the characteristics and onset of the sensory disturbance in the flank differed from that in other regions. Allodynia and hyperpathia in the flank occurred one or several weeks after the onset of the sensory disturbance of the hand and foot or of the mouth. Leijon et al. reported that 85% of patients who developed a central pain syndrome after stroke, did so several weeks after the stroke.³ In many patients, the pain began months later and was gradual in onset. The original description, by Dejérine and Roussy, also highlighted the delayed onset of central pain following thalamic infarction.⁴ It is generally assumed that the delayed onset of post-thalamic stroke pain is due to the reorganization of sensory modalities in the involved thalamus and its surroundings⁵ or sensitization of the somatosensory cortex.⁶ Thus, the lesion or mechanism responsible for sensory disturbance in the flank may differ from that in other regions occurring earlier in the course of the stroke.

According to previous studies using macaque monkeys,⁷ sensory projection fibers from the face, hand and foot are arranged from the ventral posteromedialis nucleus (VPM) to the ventral posterolateralis nucleus (VPL) where the tips of individual digits occupy large representation areas. In the hand representation area, the area for the thumb is located most medially, and that for the little finger most laterally. The projection areas for the proximal limbs and the trunk are relatively small and located in the dorsal area. A similar topography is suggested in human studies.⁸ Therefore, the sensory pattern seen in cheiro-oral-pedal or cheiro-pedal syndrome is possible without involvement of the trunk or the proximal limbs, however, simultaneous involvement of the flank seems unlikely as the flank representation area is probably not in close proximity and, further, is disproportionately small compared to those of the hand, mouth, and foot.

Lenz et al. reported that a region of the thalamus which has lost normal somatosensory input contains neurons that exhibit abnormal spontaneous and evoked activity, that electrical stimulation of these regions can produce a sensation of burning dysesthesia and that microstimulation of the right ventralis posteroinferior nucleus (VPI) of the thalamus evoked an painful sensation in the left flank region.⁹

As has been pointed out by several authors, spinothalamic tract fibers project not only to the VPM and the VPL but also to the VPI.¹⁰ The two cases presented here had involvement of the ventrocaudal nuclei of the thalamus including the VPI, demonstrated by MRI (Fig. 2). Therefore, we consider that a lesion in the VPI was responsible for delayed onset allodynia and hyperpathia in the flank. This post-stroke central pain syndrome may be misdiagnosed as intercostal neuralgia or referred pain.

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Cerebral amyloid angiopathy presenting with vasculitic pathology

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Abstract

We present an elderly patient with an unusual extensive multifocal central nervous system mass lesion, with dramatic imaging changes but only minor disturbance of cerebral function. Cerebral biopsy revealed an unexpected finding of severe cerebral amyloid angiopathy with secondary florid vasculitic appearances, which is a very rare but recognised association. Immunosuppression has produced significant sustained clinical and radiological remission.

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Keywords: Cerebral amyloid angiopathy; Vasculitis; Cerebral mass

1. Introduction

We present an elderly patient with an unusual widespread extensive multifocal central nervous system (CNS) mass lesion, with dramatic imaging changes on CT and MRI but only minor symptoms. Cerebral biopsy revealed the unexpected finding of severe cerebral amyloid angiopathy (CAA), with secondary vascular inflammatory changes.

CAA typically presents clinically as part of a dementing process, and is also a common cause of lobar haemorrhage in the elderly. Our patient did not display symptoms of headache, significant neurological signs or prominent cognitive dysfunction, which often point toward a diagnosis of CNS vasculitis; nor was there any evidence of a dementing process to suggest CAA. Prominent vasculitis-like changes are a distinctly uncommon but potentially treatable finding in CAA. This case underlines the value of cerebral biopsy

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