

## References

1. Sloof JL, Kernohan JW, MacCarty CS. *Primary Intramedullary Tumors of the Spinal Cord and Filum Terminale*. Philadelphia: WB Saunders Co; 1964.
2. Tibbs Jr RE, Harkey HL, Raila FA. Hemangioblastoma of the filum terminale: case report. *Neurosurgery* 1999;**44**:221–3.
3. Wolbers JG, Ponssen H, Kamphorst W. Hemangioblastoma of the cauda equina. *Clin Neurol Neurosurg* 1985;**87**:55–9.
4. Wyburn-Mason R. *The Vascular Abnormalities and Tumors of the Spinal Cord and Its Membranes*. London: Henry Kimpton; 1943., pp. 1–196.
5. Browne TR, Adams RD, Roberson GH. Hemangioblastoma of the spinal cord: review and report of five cases. *Arch Neurol* 1976;**33**: 435–41.
6. Morii K, Tawaka R, Washiyama K, Kumanishi T, Kuwano R. Expression of VEGF in capillary hemangioblastoma. *Biochem Biophys Res Commun* 1993;**194**:749–55.
7. Brisman JL, Borges LF, Ogivly CS. Extramedullary hemangioblastoma of the conus medullaris. *Acta Neurochir (Wien)* 2000;**142**:1059–62.
8. Chazono M, Shiba R, Funasaki H, Soshi S, Hattori A, Fujii K. Hemangioblastoma of the L-5 nerve root: case illustration. *J Neurosurg (Spine)* 1999;**90**:160.
9. Chu BC, Terae S, Hida K, Furukawa M, Abe S, Miyasaka K. MR findings in spinal hemangioblastoma: correlation with symptoms and with angiographic and surgical findings. *AJNR Am J Neuroradiol* 2001;**22**:206–17.
10. da Costa Jr LB, de Andrade A, Braga BP, Ribeiro CA. Cauda equina hemangioblastoma: case report. *Arq Neuropsiquiatr* 2003;**61**: 456–8.
11. Djindjian M, Djindjian R, Houdart R, Hurth M. Subarachnoid hemorrhage due intraspinal tumors. *Surg Neurol* 1978;**9**:223–9.
12. Giannini C, Scheithauer BW, Hellbusch LC, et al. Peripheral nerve hemangioblastoma. *Mod Pathol* 1998;**11**:999–1004.
13. Kitanaka C, Kuwahara M, Teraoka A. Intradural, purely extramedullary hemangioblastoma of the spinal cord: case report. *Neurol Med Chir (Tokyo)* 1993;**33**:377–80.
14. Lonser RR, Wait SD, Butman JA, et al. Surgical management of lumbosacral nerve root hemangioblastomas in von Hippel–Lindau syndrome. *J Neurosurg Spine* 2003;**99**:64–9.
15. Murota T, Symon L. Surgical management of hemangioblastoma of the spinal cord: a report of 18 cases. *Neurosurgery* 1989;**25**:699–708.
16. Raghavan R, Krumerman J, Rushing EJ, et al. Recurrent (non-familial) hemangioblastomas involving spinal nerve roots: case report. *Neurosurgery* 2000;**47**:1443–52.
17. Sasaki A, Hayakawa I, Tsuchida T, et al. Hemangioblastoma of the cauda equina: a case report. *No Shinkei Geka* 1978;**6**:695–700.
18. Michaud LJ, Jaffe KM, Benjamin DR, Stuntz JT, Lemire RJ. Hemangioblastoma of the conus medullaris associated with cutaneous hemangioma. *Pediatr Neurol* 1988;**4**:309–12.
19. Rohde V, Voigt K, Grote EH. Intra-extradural hemangioblastoma of the cauda equina. *Zentralbl Neurochir* 1995;**56**:78–82.
20. Roonprapunt C, Silvera VM, Setton A, Freed D, Epstein FJ, Jallo GI. Surgical management of isolated hemangioblastomas of the spinal cord. *Neurosurgery* 2001;**49**:321–7.
21. Yasargil MG, Antic J, Laciga R, de Preux J, Fideler RW, Boone SC. The microsurgical removal of intramedullary spinal hemangioblastomas. Report of twelve cases and a review of literature. *Surg Neurol* 1976;**6**:141–8.
22. Conway JE, Chou D, Clatterbuck RE, Brem H, Long DM, Rigamonti D. Hemangioblastomas of the central nervous system in von Hippel–Lindau syndrome and sporadic disease. *Neurosurgery* 2001;**48**:55–63.
23. Malis LI. Atraumatic bloodless removal of intramedullary hemangioblastomas of the spinal cord. *J Neurosurg Spine* 2002;**97**: 1–6.
24. Lee DK, Choe WJ, Chung CK, Kim HJ. Spinal cord hemangioblastoma: surgical strategy and clinical outcome. *J Neurooncol* 2003;**61**:27–34.

doi:10.1016/j.jocn.2005.02.025

## Allodynia in the flank after thalamic stroke

Mitsuhiro Yoshita <sup>a,b,\*</sup>, Masahito Yamada <sup>b</sup>

<sup>a</sup> Department of Neurology & Neurobiology of Aging, Kanazawa University Graduate School of Medical Science, Takara-machi 13-1, Kanazawa 920-8641, Japan

<sup>b</sup> Department of Neurology and Center for Neuroscience, University of California at Davis, 1544 Newton Court, Davis, CA 95616, USA

Received 3 November 2004; accepted 8 February 2005

### 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.

© 2006 Elsevier Ltd. All rights reserved.

**Keywords:** Thalamus; Infarction; Allodynia; Flank lesion

\* Corresponding author. Tel.: +1 530 757 8585; fax: +1 530 757 8827.  
E-mail address: myoshita@ucdavis.edu (M. Yoshita).

## 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,<sup>1</sup> 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,<sup>2</sup> 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.

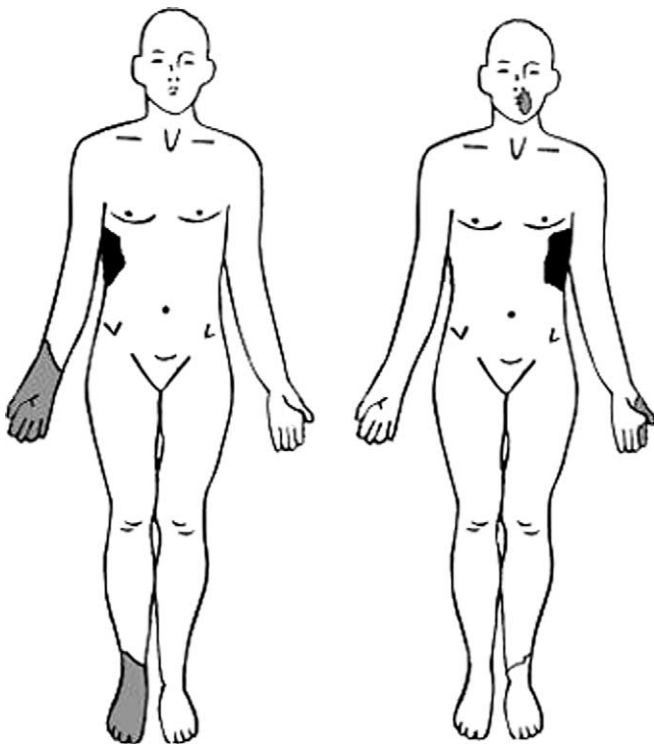


Fig. 1. Diagrammatic representation of the distribution of the sensory deficits in patient 1 (left) and patient 2 (right).

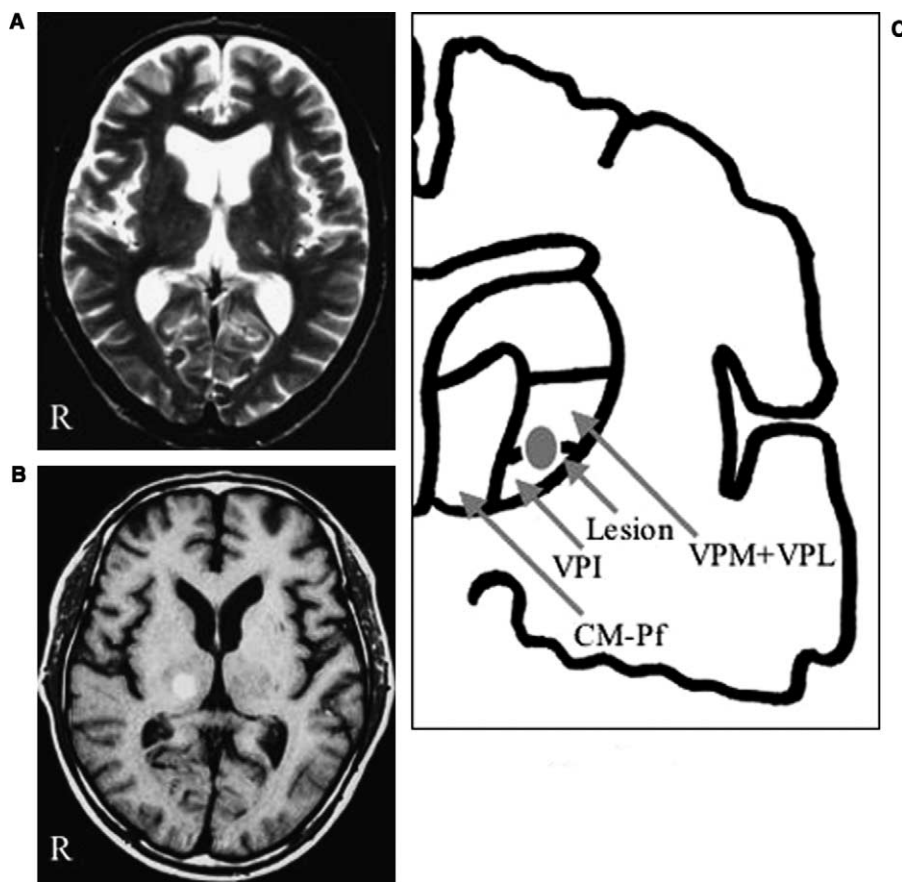


Fig. 2. A. Axial T2-weighted MRI showing the left thalamic infarct in patient 1. B. Axial T1-weighted MRI showing the right thalamic hemorrhage in patient 2. C. Diagrammatic representation of the location of the lesions in A and B. VPL: ventral posterolateral nucleus, VPM: ventral posteromedial nucleus, 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.<sup>3</sup> In many patients, the pain began months later and was gradual in onset. The original description, by Dejerine and Roussy, also highlighted the delayed onset of central pain following thalamic infarction.<sup>4</sup> 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 surroundings<sup>5</sup> or sensitization of the somatosensory cortex.<sup>6</sup> 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,<sup>7</sup> 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.<sup>8</sup> 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.<sup>9</sup>

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.<sup>10</sup> 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.

## References

1. Fisher CM. Pure sensory stroke involving face, arm and leg. *Neurology* 1965;**15**:76–80.
2. Kim JS. Restricted acral sensory syndrome following minor stroke. *Stroke* 1994;**25**:2497–502.
3. Leijon G, Boivie J, Johansson I. Central post-stroke pain – neurological symptoms and pain characteristics. *Pain* 1989;**36**:13–25.
4. Déjérine J, Roussy G. Le syndrome thalamique. *Rev Neurol* 1906;**14**: 521–32.
5. Lenz FA, Gracely RH, Hope EJ, Baker FH, Richardson RT, Dougherty PM. Reorganization of sensory modalities evoked by microstimulation in region of the thalamic principal sensory nucleus in patients with pain due to nervous system injury. *J Comp Neurol* 1998;**399**:125–38.
6. Baron R, Baron Y, Disbrow E, Roberts TP. Activation of the somatosensory cortex during Abeta-fiber mediated hyperalgesia. A MSI study. *Brain Res* 2000;**871**:75–82.
7. Nelson RJ, Kaas JH. Connections of the ventroposterior nucleus of the thalamus with the body surface representations in cortical area 3b and 1 of the *Cynomolgus* Macaque, *Macaca fascicularis*. *J Comp Neurol* 1981;**199**:29–64.
8. Hassler R. New aspects of brain functions revealed by brain disease. In: French JD, editor. *Frontiers in Brain Research*. New York: Columbia University Press; 1962. p. 242.
9. Lenz FA, Gracely RH, Hope EJ, et al. The sensation of angina can be evoked by stimulation of the human thalamus. *Pain* 1994;**59**: 119–25.
10. Berkley KJ. Spatial relationship between the termination of the somatic sensory and motor pathways in the rostral brainstem of cats and monkeys. I. Ascending somatic sensory inputs to lateral diencephalon. *J Comp Neurol* 1980;**193**:283–317.

doi:10.1016/j.jocn.2005.02.023

# Cerebral amyloid angiopathy presenting with vasculitic pathology

S.H. Wong <sup>a</sup>, P.D. Robbins <sup>b</sup>, N.W. Knuckey <sup>c</sup>, A.G. Kermode <sup>a,\*</sup>

<sup>a</sup> Department of Neurology and Clinical Neurophysiology, Sir Charles Gairdner Hospital, Hospital Avenue, Nedlands, Western Australia 6009, Australia

<sup>b</sup> Western Australian Centre of Pathology and Medical Research, Sir Charles Gairdner Hospital, Hospital Avenue, Nedlands, Western Australia, Australia

<sup>c</sup> Department of Neurosurgery, Sir Charles Gairdner Hospital, Hospital Avenue, Nedlands, Western Australia, Australia

Received 10 January 2005; accepted 22 March 2005

## 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.

© 2005 Elsevier Ltd. All rights reserved.

**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

\* Corresponding author. Tel.: +61 8 9346 3088; fax: +61 8 9346 2455.  
E-mail address: agkermode@bigpond.com (A.G. Kermode).