

Obstructive Hydrocephalus Induced Tremor in Patient with Mesencephalic Lacunae

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We report a case of hydrocephalus in a 8-year-old boy who presented bilateral hand tremor. The hydrocephalus was caused by the aqueductal stenosis due to expanding lacunae in the mesencephalothalamic area on MR findings. The tremor was improved after CSF drainage by spinal tap and ventriculoperitoneal shunt. The authors present the possible mechanism of hydrocephalus induced tremor

KEY WORDS : Hydrocephalus · Tremor · Ventriculoperitoneal shunt · Lacunae .

Introduction

Hydrocephalus is one of the most common and treatable conditions encountered in neurosurgical practice⁶⁾. Hydrocephalus typically produces headache, nausea and vomiting, and progressive obtundation⁵⁾. Papilledema, gait difficulties, and hyperactive tendon reflexes may occur, but tremor is a rare and little-known sign of obstructive hydrocephalus⁵⁾. In our case, hydrocephalus was induced by aqueductal obstruction. The tremor was improved as hydrocephalus was resolved by cerebrospinal fluid(CSF) drainage by spinal tap or ventriculoperitoneal shunt. The cause is unclear. The tremor may be caused by lacunae at the pretectile lesion or pure hydrocephalus. In the current report we describe a patient in whom mesencephalic lacunae was observed on magnetic resonance (MR) imaging and in whom the referring complaints were related to obstructive hydrocephalus induced tremor.

Case Report

Clinical history

An 8-year-old man was admitted to our hospital because of tremor of the both arms. The symptoms appeared 2 years earlier. The past history was nonspecific. The patient's neurological examination showed no gait disturbance, bradykinesia or rigidity. An intentional tremor of the both hands

appeared when the patient sustained posture and tried to write, draw and pour. The tremor had a frequency of 2 to 4Hz. The patient's score on the Mini-Mental State Examination(MMSE) was 23.5/30, which is at the lower limit of the normal range. The patient's score on the Fahn's tremor scale¹⁾ was 16. All findings of routine blood laboratory examinations were normal.

Radiological findings

Cranial MR imaging revealed triventricular hydrocephalus and effaced cerebral cortical sulci and demonstrated, in the mesencephalothalamic region, a 3mm -sized, round well-defined areas, that exhibited a signal intensity identical to CSF. No enhancement was seen after intravenous administration of contrast agent within or near the lacunae (Fig. 1). The lesion was located in the mesencephalic tegmentum, creating an obstruction of the cerebral aqueduct and displacing the quadrigeminal plate posteriorly. A portion of this lesion occupied the posterior portion of the left red nucleus. No focal lesion was seen on chest x-ray films.

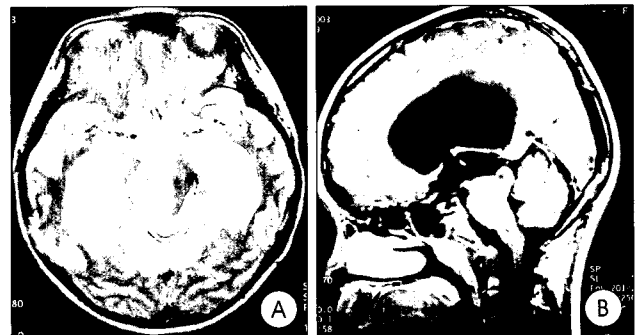


Fig. 1. Magnetic resonance images (A : T2WI axial, B : T1WI sagittal) show lateral and third ventricular enlargement and high signal intensity in mesencephalic region. The signal intensity of mesencephalic lesions is identical to that of CSF.

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Preoperative treatment and shunt placement

CSF drainage of 30cc was done by lumbar puncture. The patient's CSF pressure in lumbar puncture was 38mmHg (normal 15mmHg). The CSF analysis showed 11.4g/L proteins (normal 0.1~0.45g/L), 73mg/dl glucose (normal 50~80mg/dl), and no leukocyte (normal 10); no tumor cells or microorganisms were observed during the microscopic examination. After CSF drainage, the patient's score on the tremor scale was improved 16 to 7 and aggravated to 20 after 3 days. A CSF ventriculoperitoneal shunt with a programmable valve was inserted via a left Kocher's point.

Postoperative course

Neurological examination showed improvement of the both hand tremor. The patient denied having any gait disturbance, urinary urgency, or sudden falls. His score on the tremor scale was 8 and his score on the MMSE 30/30. The shunt pressure was changed from 100mmH₂O to 110mmH₂O. Postoperative MR imaging demonstrated resolution of the triventricular hydrocephalus and no interval change in the size of the mesencephalic lacunae, which failed to enhance after administration of contrast agent (Fig. 2).

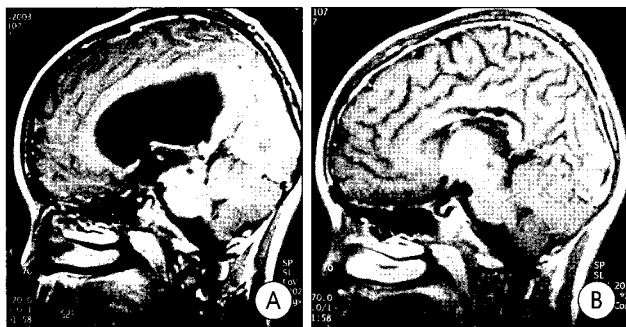


Fig. 2. Preoperative (A : T1W sagittal) and postoperative (B : T1W sagittal) magnetic resonance image show resolution of ventricular enlargement and no interval change of mesencephalic lacunae.

Discussion

The lacuna is an uncommon cavity of the brain that is filled with cerebrospinal fluid⁹. The cause is unclear, although they are possibly a result of abnormally dilated perivascular (Virchow-Robin) spaces that exert a mass effect on adjacent parenchyma and typically bulge into the ventricular cavities⁹. Based on pathological features, Poirier and Deroesne⁹ classified brain lacunae into three fundamental types : Type I lacunae correspond to lacunar infarction; Type II lacunae correspond to small intraparenchymal hematomas; and Type III (expanding) lacunae correspond to dilated pe-

rivasular (Virchow-Robin) spaces. The tiny perivascular spaces (expanding lacunae) had no clinical correlate⁷.

The expanding lacunae in the mesencephalic tegmentum can be regarded as an additional rare cause of benign aqueductal obstruction and noncommunicating normal-pressure hydrocephalus^{4,8}. In our patient, mesencephalothalamic lacunae caused aqueductal stenosis, therefore triventricular hydrocephalus developed. Hydrocephalus is a well-known clinical entity comprising disorder of stance and gait, urinary urgency or incontinence, and dementia associated with ventricular enlargement^{3,10}. In addition to the impairment of gait, many patients with NPH complain of imbalance of stance, difficulties with fine finger movements, tremor of the hands, and impairment of handwriting². The impairment of motor function in the upper extremities and hand tremor were found in some patients, indicating that hydrocephalus may cause a more generalized motor disorder¹⁰. But like our case, only tremor is a rare and little-known sign of hydrocephalus.

The mechanism by which the ventricular enlargement produces motor disturbances is not fully elucidated. The gait disturbances have been ascribed to stretching of fibers from the precentral motor cortex as they course around the enlarged lateral ventricles². Although exact pathogenesis of hydrocephalic tremor are uncertain, probably a fine, rapid hand tremor may indicate involvement of frontal premotor areas due to ventricular enlargement¹⁰. Therefore, the tremor was successfully treated by CSF ventriculoperitoneal shunt placement if expanding lacunae do not cause symptoms per se. The persistence of dorsal mesencephalic syndrome or rubral tremor after CSF shunt placement indicate that expanding lacunae may cause focal neurological dysfunction⁷. In our case, tremor improved after CSF diversion.

Conclusion

Rare cases of tremors that have resulted from hydrocephalus was reported. The tremor may be a result of hydrocephalus or lacunae at the mesencephalothalamic area on MR findings. When located in the mesencephalothalamic region, expanding lacunae can become symptomatic, producing either triventricular hydrocephalus due to aqueductal obstruction or focal symptoms and signs due to the compressive effect on adjacent brain parenchyma. Placement of a CSF shunt or ventriculocisternostomy can treat the hydrocephalus syndrome but does not modify the focal symptoms and signs due to expanding lacunae in mesencephalic region. In our case, the expanding lacunae cause hydrocephalic tremor, and do not produce neurological symptom per se.

References

1. Fahn S, Tolosa E, Marin C : Clinical rating scale for tremor in Jankovic J, Tolosa E, (eds) : **Parkinson's disease and movement disorders**, Baltimore : Williams and Wilkins, 1993, pp271-280
2. Fisher CM : Hydrocephalus as a cause of disturbances of gait in the elderly. **Neurology** **24** : 224-231, 1974
3. Hakim S, Adams RD : The special clinical problem of symptomatic hydrocephalus with normal cerebrospinal fluid pressure : observations on cerebrospinal fluid hydrodynamics. **J Neurol Sci** **2** : 307-327, 1965
4. Homeyer P, Cornu P, Lacomblez L, Chiras J, Derouesne C : A special form of cerebral lacunae : expanding lacunae. **J Neurol Neurosurg Psychiatry** **61** : 200-202, 1996
5. Keane JR : Tremor as the result of shunt obstruction : four patients with cysticercosis and secondary parkinsonism : report of four cases. **Neurosurgery** **37** : 520-522, 1995
6. Kim YD, Hwang SK, Hwang JH, Sung JK, Hamm IS, Park YM : The contributory factors of CSF shunt failure. **J Korean Neurosurg Soc** **30** : 79-84, 2001
7. Mascalchi M, Salvi F, Godano U, Nistri M, Taiuti R, Tosetti M, et al : Expanding lacunae causing triventricular hydrocephalus. **J Neurosurg** **91** : 669-674, 1999
8. Poirier J, Barbizet J, Gaston A, Meyrignac C : Thalamic dementia, expansive lacunae of the thalamo-paramedian mesencephalic area, hydrocephalus caused by stenosis of the aqueduct of sylvius. **Rev Neurol(Paris)** **139** : 349-358, 1983
9. Poirier J, Derouesne C : The concept of cerebral lacunae from 1838 to the present. **Rev Neurol(Paris)** **141** : 3-17, 1985
10. Soelberg Sorensen P, Jansen EC, Gjerris F : Motor disturbances in Normal-pressure Hydrocephalus. Special reference to stance and gait. **Arch Neurol** **43** : 34-38, 1986