## **Orolingual Dyskinesia in Central Pontine Myelinolysis**

Nai-Shin Chu



Figure. MRI T2-weighted images revealed a hypertense pontine lesion occupying mainly the basis pontis.

A 64-year-old housewife presented with involuntary movements of the lips and tongue for one month. Recently her mood became more labile, tended to be worry and had frequent nightmares. She was in her usual health until one year ago when she had diabetes diagnosed and noticed mild jaw tremor after a traumatic experience involving a minor motorcycle accident. There were no family history of movement disorders and no history of taking psychiatric drugs, liver disease, severe head trauma, and neurological disorders such as stroke. General examination was unremarkable. Neurological examination revealed an intact mental status, but a decreased facial expression. There were continued irregularly rhythmic movements of the month and protruding of the tongue. There was mild jaw tremor. Speech was dysarthric. Bradykinesia and rigidity were absent. Gait was normal. Deep tendon reflexes were moderately increased. Plantar response was flexor. Sensations were intact.

Laboratory tests including VDRL, thyroid, liver and renal functions, ceruloplasmin, copper, electrolytes and

From the Department of Neurology, Chang Gung Memorial Hospital, Linkou Medical Center, Taoyuan, Taiwan. Received July 9, 2004. Revised and Accepted July 26, 2004. Reprint requests and correspondence to: Nai-Shin Chu, MD. Department of Neurology, Chang Gung Memorial Hospital, No. 199, Tung-Hwa N Road, Taipei, Taiwan. E-mail: chu060@cgmh.org.tw acanthocytes were normal. However, serum glucose was 304 mg/dl (normal: 90-120). SEP, BAEP and blink reflex were normal. Brain MRI revealed a hyperintense pontine lesion on T2 and FLAIR images suggesting central pontine myelinolysis (CPM) (Fig.). Extrapontine myelinolysis (EPM) was not detected. There was mild cortical atrophy.

Haldol 2mg a day improved orolingual dyskinesia to such extent that the patient was satisfied with eating and speaking. Diabetes was treated and under control. Six months later follow-up MRI revealed no interium change of the pontine lesion.

CPM may be asymptomatic or presents as movement disorders<sup>(1)</sup>. Over dozen cases of CPM associated with movement disorders have been reported<sup>(2-4)</sup>. The movement disorders include dystonia, parkinsonism and choreoathetosis, and occurred as sequela of CPM usually weeks or months after onset. In most cases, these extrapyramidal symptoms are thought due to EPM, but there are cases without EPM<sup>(3)</sup>. Dystonia may also occur in other demyelinating diseases such as multiple sclerosis<sup>(5)</sup>.

CPM may be an unusual complication of diabetes<sup>(6)</sup>. Two patients with diabetes developed CPM after recurrent vomiting due to diabetic gastroparesis in the absence of documented electrolyte changes.

Dystonia in CPM may be generalized, focal or segmental. Salerno et al. reported a 44-year-old woman with a history of hypertension, depression and schizophrenia treated with perphenazine developed upper extremity and orolingual dystonia several weeks after onset of CPM without evidence of EPM<sup>(3)</sup>. In this case, orolingual dystonia might be the complication of anti-psychotic drug therapy. On the other hand, our patient presented with severe orolingual dyskinesia and mild pseudobulbar palsy without a history of anti-psychotic drug use. The present case demonstrates that rarely orolingual dystonia may develop in CPM, although the mechanism still remains unclear.

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