Intractable Hiccup Due to Multiple Sclerosis: MR Imaging of Medullary Plaque

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Abstract: We describe a 31-year-old man with clinically definite multiple sclerosis who presented with intractable hiccup for one month prior to admission. The responsible lesion was confirmed by magnetic resonance imaging to be a plaque in the ventral paramedian portion of the medulla oblongata.

Résumé: Hoquet incoercible dû à une sclérose en plaques: visualisation d’une plaque médullaire par RMN. Nous décrivons le cas d’un homme de 31 ans, atteint d’une sclérose en plaques cliniquement certaine, qui a présenté un hoquet incoercible un mois avant son admission. L’imagerie par résonance magnétique a confirmé que la lésion en cause était une plaque située dans la portion paramédiane ventrale du bulbe rachidien.

Although multiple sclerosis (MS) is characterized by protean manifestations, intractable hiccup (IH) is rare and has been described only in isolated case reports.1–3 We describe a patient with clinically definite MS presenting with IH who responded well to corticosteroid therapy.

CASE REPORT

A 31-year-old businessman experienced IH, vertigo and vomiting for two weeks in December 1987. Six weeks later, the patient developed transient episodes of double vision on left gaze and paroxysmal attacks of left facial numbness and ipsilateral trigeminal hypesthesia with brisk tendon reflexes, right dysmetria and symmetrical sensory disturbance below the T10 level bilaterally.

Laboratory examinations were unremarkable, except that cerebrospinal fluid (CSF) study showed a slight mononuclear pleocytosis as well as mild protein elevation with 18% IgG. Pattern-shift visual-evoked potentials (EP) revealed prolonged latencies bilaterally. Studies of short-latency somatosensory EP and brainstem auditory EP were compatible with intrinsic brainstem lesions. However, a cranial computed tomographic (CT) scan was unremarkable. In the following 4 years, he had one relapse and remission of the right hemiparesis and brachial monoparesthesia.

He was readmitted to our hospital in September 1992 because of IH for about one month. The patient had 10 to 15 episodes of hiccup per day and each episode lasted minutes to hours. The hiccups could not be suppressed by Valsalva maneuver, vagal stimulation, or irritation of the uvula. Therapeutic trials with metoclopramide, chlorpromazine and diphenylhydantoin were all in vain. The patient also complained of episodic vomiting, easy choking, double vision, unsteady gait and a symmetrical ascending numbness and tight sensation up to the occipital region.

On neurologic examination, slow saccadic movement on right gaze was present with concomitant horizontal nystagmus. Dysarthria, bilateral trigeminal hypesthesia, cerebellar dysmetria and diminished proprioception of all extremities were also noted. There was no motor weakness, but a generalized hyperreflexia was identified. A cranial CT scan showed no discernible abnormality. Magnetic resonance imaging (MRI) with T1-weighted sagittal images demonstrated hypo-intense paramedian lesions along the ventral medulla. Axial views showed several plaques in the rostral and medial portion of the medulla oblongata. The neighboring paramedian plaques straddled the midline portion to form a large bilateral plaque (Figure 1A, B). The plaque involved bilateral pyramidal tracts, medial lemnisci, olives and paramedian reticular formations. On prednisone (60 mg/day) and metoclopramide, the hiccups resolved within one week, and his speech, sensory and cerebellar dysfunction all improved. His condition has been stable since then.

DISCUSSION

Newsom-Davis described hiccup as being controlled by a suprasegmental mechanism entirely independent of the respiratory system.4 IH caused by brainstem dysfunction has been attributed to the lesions in the hiccup reflex arc entailing the following structures: the dorsal portion of the nucleus tractus solitarius (inspiratory center), the ventral group of nucleus ambiguus (expiratory center), pneumotaxic center, and hypothalamic apneustic center.5–8 The MS plaques in the ventral medulla of our patient suggest that they may potentially perturb the neighboring hiccup reflex arc or interfere with the descending inhibitory cortico-medullary pathways.9

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The concept of IH as a dysfunction of hiccup reflex arc indicates that we should search cautiously for underlying hazardous and/or treatable condition. Howard et al. reported two MS patients with insidious onset of hiccups led to respiratory failure. We stress that an early diagnosis may prevent a potential catastrophe.

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REFERENCES