

Lateral Medullary Syndrome Presenting With Recurrent Hiccups: A Case Report

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Lateral medullary infarction (LMI) or Wallenberg's syndrome is one of the best known vascular syndromes of the posterior circulation. . The posterolateral part of the medulla oblongata of the brain stem and cerebellum receiving arterial blood supply from the posterior inferior cerebellar artery are the areas commonly affected. The usual symptoms of LMI include vertigo, dizziness, nystagmus, ataxia, nausea and vomiting, dysphagia, hoarseness, impaired sensation over half the face, impairment of pain and thermal sensation over the contralateral hemibody and limbs and the ipsilateral face, and Horner's syndrome.(1,2,3) A hiccup is a rare symptom in lateral medullary syndrome (Wallenberg's Syndrome). The aetiology of hiccups includes failure of the digestive organs, irritation of the diaphragm, ingestion of alcohol, excessive smoking, and any disease of the central nervous system that involves the brain stem. We present a case of a 75 years old gentleman referred to our hospital for persistent intractable hiccups as presenting symptom of LMS. He presented to our emergency room with persistent hiccups and generalised weakness. The patient had tobacco (smoking) dependence since 30 years apart from being a hypertensive.

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I. Case Report

A 75 years old gentleman with a history of nicotine (smoker) dependence since last 30 years was referred to our hospital with persistent hiccups and generalized weakness since 7 days. There was no history of recent fever, ear discharge or significant surgical interventions. Patient was known hypertensive on treatment, but with no history of diabetes. The initial computed tomography (CT) scan of brain from the referral hospital revealed no abnormality. His symptoms persisted when he presented to our emergency room. His vital parameters were stable. Neurological examination revealed mild cerebellar signs on the left side.

Diagnostic tests failed to reveal the cause and the hiccups failed to respond to medications and other attempted treatments. Further investigation with abdominal ultrasonography and gastroscopy revealed no abnormalities. The patient developed hoarseness of voice and difficulty in swallowing from the second day onwards since admission. Neurological examination revealed that the uvula was deviated towards right side suggestive of left sided of 9th and 10th cranial nerve palsy. Patient had cerebellar signs on left side. Patient had grade –II central nystagmus with rotatory on left lateral gaze. Power, tone and deep tendon reflexes were normal. The plantar reflex was normal. The cardiovascular and abdominal examination was normal. Respiratory examination revealed coarse crepitation on right lower zone of lung suggestive of aspiration pneumonia and confirmed by chest radiogram. On investigations complete blood count was (total leucocyte count - 9800/um, Hb- 13 gm%, platelet count – 1.53 lakhs/um), blood sugar level was 110 mg/dl and renal functions was normal. Chest x-ray showed right lower zone haziness suggestive of right lower zone pneumonia secondary to aspiration. Because of neurological signs MRI brain was obtained which showed a hypo-intense on T-1 weighted image and hyper-intense lesion on T2 and FLAIR sequences in left lateral medullary region (figure no.1, 2). This lesion was hyperintense on diffusion weighted images with decline of apparent diffusion coefficient (ADC) on ADC map, compatible with an acute ischemic stroke. Electrocardiogram was suggestive of left ventricular hypertrophy. This patient was started on antihypertensives, antiplatelet agents, statins and multivitamin supplementations. Nutrition was maintained on nasogastric tube and feed. Additionally Baclofen was given for the hiccups. , the patient was given 5 mg of baclofen by mouth three times per day, and the hiccups abated within 48 hours. The baclofen was discontinued after one week of therapy, and the hiccups did not return.

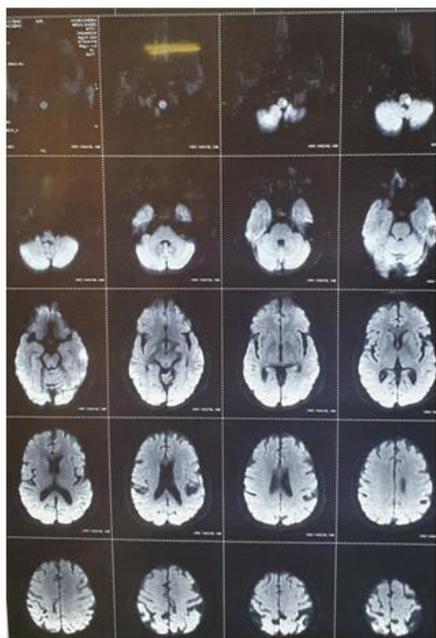


Fig.1 T2 images showing left medullary infarct

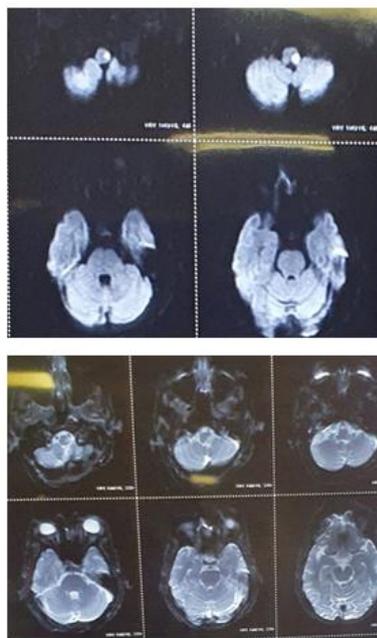


Fig 2. T1 images showing left medullary infarct

II. Discussion

Persistent hiccups are uncommon (lasting for more than 48 h) warrants complete medical investigations to uncover underlying pathology and needs immediate medical assistance.(3) Persistent hiccups can be very bothersome and disabling, affecting food intake, causing sleep deprivation, physically exhaustion and can lead to other potentially fatal consequences (aspiration pneumonia).

The neuroanatomical center for hiccups is not well-known, although the central connection is presumed to be a part of the brain stem which probably interacts with its respiratory centers, phrenic nerve nuclei, medullary reticular formation and hypothalamus. (4,5,6) The afferent pathway is made up of the sensory branches of the phrenic and vagus nerves and the dorsal sympathetic fibers, whereas the efferent pathway is formed by the motor fibers of the phrenic nerve. Hiccups have central and peripheral causes.(2,6) Central hiccups occur with lesions between the pathway from the central nervous system to the phrenic nerve, mainly in diseases of the brain stem such as ischemic stroke, dolichoectatic basilar artery, tumors, encephalitis and multiple sclerosis. Peripheral hiccups can be caused by diseases at phrenic nerve level such as gastric distension.(4,5)

Lateral medullary syndrome (LMS) remains an interesting clinical entity with a wide range of clinical presentations of cerebrovascular accidents. The area of the brain stem involved in LMS is the posterolateral part of the medulla oblongata, which is the portion receiving arterial blood supply from the posterior inferior cerebellar artery (PICA). (7,8)

Many different pathologies of the medullary region may cause hiccups. Hiccups seemed to be related to a more complete lesion in the middle part of the lateral medullary area and have been clinically correlated with symptoms of the 9th and 10th cranial nerves. A hiccup like reflex can be elicited by electrical stimulation of a limited area within the medullary reticular formation, the hiccup-evoking site (HES) and hiccups are rapidly suppressed after microinjection of baclofen, a gamma-aminobutyric acid (GABA) analogue, into the HES.(2,4)

Patients with LMIs had hiccups, mainly when the lesions occurred in the dorsolateral region of the middle medulla. There was a close correlation between hiccups and symptoms of cerebellar, vestibular and 5th, 9th and 10th cranial nerve involvement. Use of gabapentin in the treatment of persistent hiccups in cases of LMS has been reported.(9) Gabapentin may act by causing enhancement of GABA-mediated inhibition or by selectively diminishing calcium influx by the inhibition of voltage-operated calcium channels. In this way, it may interrupt the intermittent myoclonus of the diaphragm brought about by repeated activity of the solitary nucleus.(10)

This case attempts to highlights the importance of vigilant approach to seemingly trivial hiccups particularly when presented and resistant symptoms to preliminary treatment methods.

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