

# Case Report

## Persistent Hiccups (Singultus) as the Presenting Symptom of Lateral Medullary Syndrome

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### ABSTRACT

Lateral medullary syndrome (LMS) is a constellation of varied neurologic manifestations seen in cerebrovascular accidents. 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. We present a case of a middle aged gentleman referred to our hospital for persistent intractable hiccups as presenting symptom of LMS. He presented to our emergency room with persistent hiccups and left sided cerebellar signs. The patient had significant past history of alcohol and tobacco (smoking) dependence since 30 years apart from being a hypertensive.

**Key words:** Cerebellar signs, lateral medullary syndrome, persistent hiccups, posterior inferior cerebellar artery

### REVIEW OF LITERATURE

Hiccups are repeated involuntary, spasmodic and temporary contractions of the diaphragm accompanied by sudden closure of the glottis, producing a distinguishing “hic” sound.<sup>[1]</sup>

Hiccups, or Singulata (hiccup is singultus), are commonly experienced by most people at 1 time or another and are usually brief and self-limiting. The physiological purpose of hiccups is uncertain.<sup>[1,2]</sup> Because fetuses and premature infants hiccup often, it is suggested that hiccups are a programmed isometric inspiratory muscle exercise, which is useless after the neonatal period but may be restimulated by irritation along the reflex arc.<sup>[1,3]</sup>

Persistent hiccups are uncommon (lasting for more than 48 h) warrants complete medical investigations to uncover underlying pathology and needs immediate medical assistance.<sup>[2]</sup> Persistent hiccups can be very bothersome and disabling, affecting food intake, causing sleep deprivation, physical 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.<sup>[4-6]</sup> 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.<sup>[2,4]</sup> 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.<sup>[2,4,7]</sup>

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Lateral medullary syndrome (LMS) remains an interesting clinical entity with a wide range of clinical presentations of cerebrovascular accidents.<sup>[8]</sup> The area of the brain stem involved in LMS is the posterolateral part of the medulla oblongata,<sup>[4]</sup> which is the portion receiving arterial blood supply from the posterior inferior cerebellar artery (PICA).<sup>[7,9]</sup> The usual symptoms of lateral medullary infarction (LMI) include vertigo, dizziness, nystagmus, ataxia, nausea and vomiting, dysphagia, hoarseness, hiccups, impaired sensation over half the face, impairment of pain and thermal sensation over the contralateral side of the trunk, limbs and the ipsilateral face and Horner's syndrome. Among the symptoms, hiccups can easily be overlooked, though among other effects they may cause aspiration pneumonia, respiratory depression and esophagitis.<sup>[4,10,11]</sup>

The lateral medullary or Wallenberg's syndrome is the one most commonly described, indeed Keane's analysis found 56% of his experience with central hiccups were secondary to LMIs.<sup>[4,10]</sup>

The mechanism of hiccup in patients with LMI is not very clear, though few hypothesis have been proposed.

The region of the dorsolateral medulla, where the nucleus ambiguus is located — including the vagal motor neurons projecting to the larynx and the premotor neurons that control the diaphragm are implicated in this regard. Thus lesions in this region induce hiccups, as they affect the laryngeal motor neurons which control the glottis and the premotor neurons which control the inspiratory muscles.<sup>[1]</sup>

## CASE REPORT

A middle aged gentleman with a history of alcohol and nicotine (smoker) dependence since last 30 years was referred to our hospital with persistent hiccups, difficulty in walking and giddiness 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. He continued have above said symptoms when he presented to our emergency room. His vital parameters were stable. Neurological examination revealed mild cerebellar signs on the left side.

Patient received treatment for gastritis and alcohol detoxification. Initial treatment included oral xylocaine viscous, proton pump inhibitors, vitamin supplements and baclofen, but no improvement was achieved. Initial diagnostics involved the following procedures:

- Liver function test
- Blood count
- Chest X-ray
- Blood glucose levels.

Diagnostic tests failed to reveal the cause and the hiccups failed to respond to medications and other attempted treatments. Following consultation with the on-site specialized neurological unit, chlorpromazine was administered, but did not lead to improvement. Further investigation with abdominal ultrasonography and gastroscopy revealed no abnormalities.

With worsening persistent hiccups, difficulty in walking, giddiness and cerebellar dysfunction signs on the left side not responding to treatment. The psychiatry team with the opinion of neurologist and physician advised second CT scan as a diagnostic measure. The review CT scan revealed PICA infarct. Patient was immediately started on low molecular weight heparin, clopidogrel with aspirin and gabapentin along with initial treatment.

Following 3-4 days, patient showed signs of improvement. Hiccups decreased in frequency and intensity, general condition improved and there was marked recovery from cerebellar dysfunction.

## DISCUSSION

Persistent hiccups often imply a serious underlying disease process that requires an extensive evaluation. Among the more than 100 causes of hiccups, the most common are located in the gastrointestinal tract. Recently, greater attention has been focused on central causes, particularly lesions located in the brain stem.<sup>[12]</sup>

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 9<sup>th</sup> and 10<sup>th</sup> 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.<sup>[2,4]</sup>

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 5<sup>th</sup>, 9<sup>th</sup> and 10<sup>th</sup> cranial nerve involvement. Use of gabapentin in the treatment of persistent hiccups in cases of LMS has been reported.<sup>[13]</sup> 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.<sup>[12]</sup>

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

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