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Intractable hiccups as a sole manifestation of medullary cavernous hemangioma

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Abstract

Intractable hiccups lasting over 48 hours can indicate serious medical conditions, including neurological disorders. Typically, such disorders present with other neurological symptoms, but hiccups may, in some cases, occur as the sole manifestation, delaying the underlying diagnosis. We report the case of a 37-year-old previously healthy man who experienced intractable hiccups for more than 50 days. Initial investigations, including chest and abdominal imaging and endoscopy, were unremarkable except for gastroesophageal reflux disease (GERD), which did not improve with standard treatment. A neurological examination revealed no abnormalities, but brain MRI showed a small cavernous hemangioma with subacute hemorrhage near the solitary nucleus in the medulla, which was thought to be responsible for the intractable hiccups. This case highlights the importance of considering small medullary lesions in the differential diagnosis of persistent hiccups, even in the absence of other neurological symptoms, and underscores the role of the solitary nucleus in the pathophysiology of intractable hiccups.

Keywords: Intractable hiccups, medullary cavernous hemangioma, solitary nucleus

INTRODUCTION

Hiccups are a common phenomenon that usually resolve spontaneously within a few days. However, hiccups persisting for more than 48 hours may indicate a serious underlying condition.[1] Any lesion affecting the complex hiccup reflex arc, which includes the vagus nerve, phrenic nerve, sympathetic chain, recurrent laryngeal nerve, intercostal nerves, and structures spanning from the medulla to the midbrain, can trigger hiccups.[2] When caused by neurological disorders, patients often exhibit other neurological symptoms such as headaches, dizziness, blurred vision, tremors, nausea, or vomiting.[3] Neurological examination may reveal diplopia, nystagmus, weakness, sensory disturbances, or ataxia.[4,5] Nevertheless, rare cases have been reported where hiccups were the sole manifestation, usually due to extra-axial lesions such as hemangioblastomas compressing the dorsal part of the medulla.[6,7]

We report a case of a healthy 37-year-old man who experienced intractable hiccups for more than 50 days. Despite the absence of other neurological symptoms or deficits, imaging revealed a small cavernous hemangioma near the solitary nucleus in the medulla with associated subacute hemorrhage. This case suggests that intractable hiccups persisting beyond 48 hours, even without other neurological symptoms, may be due to intra-axial central nervous system disorders. It also highlights the significant role of the solitary nucleus in the pathophysiology of such hiccups.

CASE REPORT

A 37-year-old man presented to the neurology clinic with persistent hiccups lasting 50 days. The hiccups began following an episode of overeating and occurred daily, often disturbing his sleep and causing significant discomfort. Initial Valsalva maneuvers temporarily alleviated the hiccups for about an hour, but after two weeks, they became ineffective. The patient experienced shortness of breath and throat tightness, prompting a visit to a local emergency department, where ECG and chest and abdominal CT scans showed normal results. Despite taking medications such as metoclopramide, cisapride, baclofen, and gabapentin prescribed by various clinics, the hiccups persisted. Accompanied by symptoms of acid reflux, the patient underwent endoscopy at our gastroenterology department two weeks prior and was diagnosed with gastroesophageal reflux

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disease (GERD). Rabeprazole was prescribed for two weeks without improvement.

The patient had no prior medical history, surgeries, or regular medications. He was a non-smoker and consumed alcohol moderately, but ceased drinking after the onset of hiccups. He denied headaches, dizziness, or other significant discomfort besides hiccups and acid reflux.

Physical examination revealed a temperature of 36.5°C, blood pressure of 130/75 mmHg, heart rate of 71 beats per minute, and respiratory rate of 19 breaths per minute. Other physical examinations were normal. Red glass testing showed no diplopia, extraocular movements were normal, and no nystagmus was observed. Rapid alternating movements, finger-to-nose test, heel-to-shin test, tandem gait, and Romberg test were all normal. Comprehensive neurological examination showed no abnormalities.

Chlorpromazine was initiated, and brain MRI was performed. Imaging revealed a 0.5x0.5 cm hypointense lesion on gradient echo sequences located dorsomedially in the left medulla (Figure 1A). The lesion showed two small hyperintense areas on T2-weighted images, corresponding to different stages of hemorrhage, and hyperintense areas on T1-weighted images suggestive of methemoglobin, indicating subacute hemorrhage (Figure 1B to 1D). These findings were consistent with a cavernous hemangioma. The lesion was situated near the solitary nucleus, which is associated with intractable hiccups, and was believed to be the cause.

Following chlorpromazine treatment, the hiccups improved, occurring only once or twice a week for about 10 minutes each time. After three months, the hiccups ceased completely, allowing discontinuation of chlorpromazine. We thought that the intractable hiccups might have subsided as the subacute hemorrhage resolved. Follow-up brain MRI showed no change in the size of the medullary cavernous hemangioma, and no hiccups or other neurological symptoms were reported for one year.

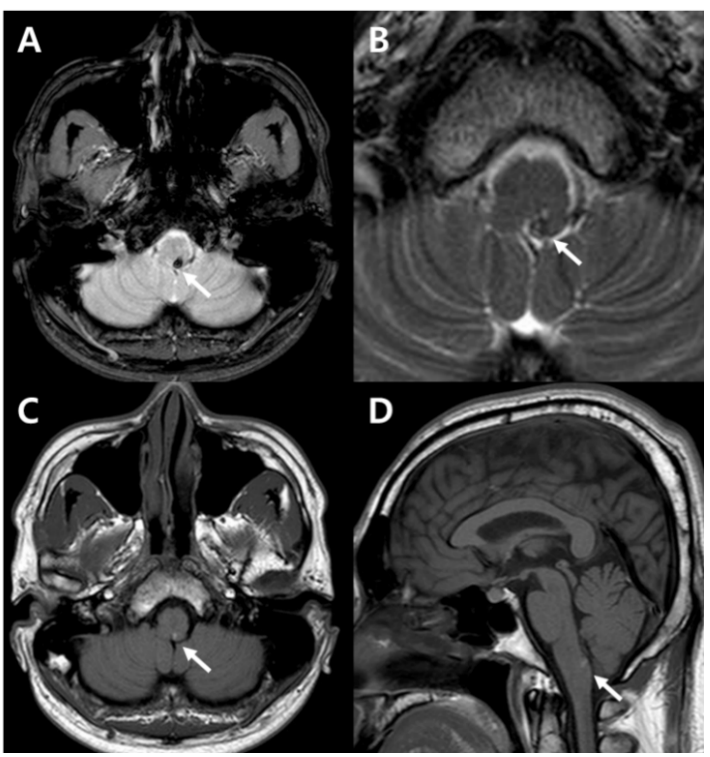

Figure 1. (A) A gradient-echo image shows a 0.5x0.5cm sized low signal intensity in the left dorsomedial medulla. (B) T2-weighted image reveals two small high signal intensities within the lesion, suggesting areas of hemorrhage at different stages. The locations of these high-signal areas differ from the high signal intensity observed on the T1-weighted images. (C) T1-weighted axial and (D) coronal images show a tiny high signal intensity within the lesion, indicating the presence of methemoglobin from a subacute hemorrhage. These findings are consistent with a small cavernous angioma in the left dorsomedial medulla. Arrows indicate the lesion.

DISCUSSION

A 37-year-old healthy man experienced intractable hiccups lasting more than 50 days. Imaging studies including chest and abdominal CT scans were normal, and esophagogastroduodenoscopy revealed GERD; however, the hiccups did not improve with rabeprazole treatment. Despite the absence of neurological symptoms or findings on examination, brain MRI identified a small cavernous hemangioma with subacute hemorrhage near the solitary nucleus in the medulla.

Hiccups are involuntary contractions of the diaphragm and intercostal muscles, followed by sudden closure of the glottis. The hiccup reflex arc is complex, involving afferent pathways (vagus nerve, phrenic nerve, sympathetic chain), central processing units (medulla to midbrain), and efferent pathways (phrenic nerve, recurrent laryngeal nerve, intercostal nerves).[1,2] Any disruption in these pathways can trigger hiccups.

In cases of hiccups persisting beyond 48 hours, a thorough evaluation is warranted. This includes reviewing general symptoms, surgical history, and medication use, as well as conducting comprehensive physical and neurological examinations. Imaging studies such as chest and abdominal CT scans and endoscopy may be necessary. If neurological symptoms or deficits are present, brain MRI is recommended.

Persistent hiccups can result from various neurological conditions including stroke (especially lateral medullary infarction), multiple sclerosis, tumors, infections, aneurysms, and cavernous hemangiomas.[2] Intra-axial lesions typically present with additional neurological symptoms or deficits.[3-5] However, rare cases have reported hiccups as the sole symptom without neurological deficits, usually due to extra-axial lesions such as hemangioblastomas compressing the dorsal medulla.[6,7] Although previous reports have suggested that hiccups may persist due to excessive activity of the solitary nucleus causing diaphragm spasms, these cases typically involved large intra-axial medullary lesions affecting multiple structures and accompanied by other neurological symptoms, making it difficult to isolate the solitary nucleus as the sole cause.[4,8]

In conclusion, this case is unique in that the lesion was very small and located near the solitary nucleus without affecting other medullary structures, resulting in intractable hiccups as the sole symptom. This highlights the importance of considering small, localized lesions in the medulla as potential causes of intractable hiccups, even in the absence of other neurological symptoms. It underscores the significant role of the solitary nucleus in the pathophysiology of persistent and intractable hiccups, providing valuable insights into their management and diagnosis.

DISCLOSURE

Ethics: Written informed consent was obtained from the patient for publication of the case details and accompanying images.

Conflicts of interest: None

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