Case Report

Intractable Hiccup as the Presenting Symptom of Cavernous Hemangioma in the Medulla Oblongata: A Case Report and Literature Review

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A case of intractable hiccup developed by cavernous hemangioma in the medulla oblongata is reported. There have been only five previously reported cases of medullary cavernoma that triggered intractable hiccup. The patient was a 28-year-old man who was presented with intractable hiccup for 15 days. It developed suddenly, then aggravated progressively and did not respond to any types of medication. On magnetic resonance images, a well-demarcated and non-enhancing mass with hemorrhagic changes was noted in the left medulla oblongata. Intraoperative findings showed that the lesion was fully embedded within the brain stem and pathology confirmed the diagnosis of cavernous hemangioma. The hiccup resolved completely after the operation. Based on the presumption that the medullary cavernoma may trigger intractable hiccup by displacing or compression the hiccup arc of the dorsolateral medulla, surgical excision can eliminate the symptoms, even in the case totally buried in brainstem.

Key Words: Brainstem · Cavernous hemangioma · Hiccup · Medulla oblongata · Surgery.

INTRODUCTION

Hiccup is a repeated involuntary, spasmodic, and temporary contraction of the diaphragm accompanied by a sudden closure of the glottis, producing the characteristic inspiratory sound “hic” and discomfort}. It can be considered persistent or intractable when it lasts more than 24 hours}. The exact etiology of hiccup remains unclear in most cases. Regarding various central causes, the medulla oblongata has been investigated as one of the most important centers in the hiccup circuit. In addition to neurological disorders, including medullary infarction and hemorrhage, tumors and tuberculoma reportedly generate intractable hiccup. Although few reports demonstrated surgically treated and pathologically confirmed cavernous hemangioma (CH) in the medulla oblongata, most of them were superficially located in the dorsal part of the medulla oblongata. In the current case, the authors demonstrate the fully embedded medullary cavernoma with intractable hiccup surgically treated without morbidity and discuss possible pathogenesis of this condition with a review of the reported cases and related literature.

CASE REPORT

History

A 28-year-old man presenting with persistent hiccup for 15 days was admitted to our hospital. The symptom developed suddenly and aggravated progressively in its frequency and intensity. The patient noticed motor weakness and sensory changes in the left side of his body three days prior to admission. Hiccup did not respond to any types of medication, but occurred only occasionally while asleep. There were no abnormal findings in endoscopy and computed tomography scans for the chest and abdominal organs.

Presentation and examination

On admission, neurological examination revealed no impair-
medulla oblongata, surgery was performed via the midline suboccipital approach under prone position. The lesion was covered by normal parenchymal tissue and made a bulging contour of the medulla oblongata with superficial abnormal draining veins (Fig. 2A). The shortest trajectory to the lesion was confirmed by neuronavigation (StealthStation S7®️, Medtronic, Minneapolis, MN, USA) and a 2 cm longitudinal pial incision was made caudally from the obex. After dissecting a few millimeters deep, hemosiderin staining of the neural tissue was encountered (Fig. 2B). The plane of the dissection between the cavernoma and the parenchyma was well-distinguished (Fig. 2C). The lesion was removed en bloc. Histopathologically, the brain specimen revealed irregularly dilated vascular spaces without intervening neural tissue, which are typical features of cavernous hemangioma (Fig. 3). Additionally, there was reactive piloid gliosis with numerous Rosenthal fibers in the periphery of the lesion and occasional hemosiderin-laden macrophages. Hiccup resolved immediately after surgery. The patient had slight hemiparesis (motor grade IV+/IV+) and hemisensory changes that cleared entirely at the time of discharge.

**DISCUSSION**

The clinical manifestations of the brainstem CHs closely correlated with the anatomical location of the lesion. The common signs and symptoms include various types of cranial neuropathy, sensory/motor deficits, headache, diplopia, ataxia, vertigo, nausea/vomiting, dysarthria, dysphagia, and dysmetria, but...
The hiccup has rarely been reported as the presenting symptom in three cases. Contrary to this, Ward et al. reported that intractable hiccup was not an infrequent clinical presentation in medullary cavernoma (5/18 cases, 27.8%). However, these case series did not reveal the detailed description on the exact location, and clinico-radiological characteristics. For this reason, medullary cavernoma presenting as intractable hiccup has rarely been reported as a single case report, with the explanation of unique clinical course. Table 1 illustrates surgically resected medullary cavernomas presenting as intractable hiccup.

Table 1. Surgically resected medullary cavernomas presenting as intractable hiccup

<table>
<thead>
<tr>
<th>Authors</th>
<th>Age/sex</th>
<th>Hiccup characteristics</th>
<th>Lesion characteristics</th>
<th>Operation related</th>
<th>Complication</th>
</tr>
</thead>
<tbody>
<tr>
<td>Eisenächer and Spiske</td>
<td>26/Male</td>
<td>Sudden</td>
<td>Not defined</td>
<td>Midline SOC</td>
<td>Immediately after operation</td>
</tr>
<tr>
<td>Mattana et al.</td>
<td>40/Male</td>
<td>Sudden</td>
<td>Not defined</td>
<td>Midline SOC</td>
<td>Immediately after operation</td>
</tr>
<tr>
<td>Musumeci et al.</td>
<td>46/Male</td>
<td>Sudden</td>
<td>Not defined</td>
<td>Midline SOC</td>
<td>Immediately after operation</td>
</tr>
<tr>
<td>Pechlivanis et al.</td>
<td>33/Male</td>
<td>Sudden</td>
<td>Not defined</td>
<td>Midline SOC</td>
<td>Immediately after operation</td>
</tr>
<tr>
<td>Thaci et al.</td>
<td>36/Female</td>
<td>Sudden</td>
<td>Not defined</td>
<td>Midline SOC</td>
<td>Immediately after operation</td>
</tr>
<tr>
<td>Present case</td>
<td>28/Male</td>
<td>Sudden</td>
<td>Not defined</td>
<td>Midline SOC</td>
<td>Immediately after operation</td>
</tr>
</tbody>
</table>

*Presenting sudden deterioration in consciousness and respiration due to intrallesional hemorrhage, †May be deep seated lesion based on representative MR images, ‡Presumptive size of lesion based on representative MR images, §Recurrent and lasting up to 3 months, ¶Presenting sudden developed quadriaparesis, headache, unsteady gait, and facial paresis due to intrallesional hemorrhage, SOC : suboccipital craniotomy
excitatory function for hiccup genesis. On the other hand, the latter two cases, including the current one, were located in the deep portion of the dorsolateral medulla caudal to the obex. Intractable hiccup in the latter ones may be induced by inactivation of the inhibitory function of GABA-containing neurons suggested by Oshima et al. Considering the aforementioned hiccup pathogenesis and the case summary, we presume that the present medullary cavernoma may also have reduced the inhibitory function or induced a stimulatory signal on the hiccup reflex by displacing or compressing the hiccup arc of the dorsolateral medulla.

CONCLUSION

Cavernous hemangioma on the dorsolateral medulla can generate intractable hiccup by compressing or displacing the normal reflex arc. Even in totally buried cases like the current one, surgical excision could be performed for eliminating symptoms without a major neurological deficit.

References