Idiopathic Intractable Hiccups : A Case Report
D Prabath Kumar, P Thejeswini, D Neeharika, Alladi Mohan

ABSTRACT

We report the case of 18-year-old female, who presented with hiccups of 2 months duration. Hiccups were sudden in onset, progressive in nature, present throughout the day and were absent during sleep. There were no aggravating or relieving factors. There was no history of headache, seizures, ear discharge, neck pain, shortness of breath, cough, palpitations, pain abdomen, drug-intake or toxin exposure or trauma. There was no history of stress or anxiety related symptoms, loss of weight or appetite. Physical examination was unremarkable. She was started on oral rabeprazole, domperidone and investigated further. Laboratory investigations including complete haemogram, serum biochemistry, chest radiograph, electrocardiogram, ultrasonography of the abdomen were normal. Computed tomography of the brain did not reveal any intracranial lesions. Upper gastrointestinal endoscopy revealed no abnormality. She was diagnosed to have idiopathic intractable hiccups. As she had no relief with treatment with proton pump inhibitor and prokinetic agent over 3 weeks, she was started on oral chlorpromazine (25mg twice-a-day). As there was no improvement, started on baclofen (10 mg twice-a-day) and her symptoms gradually subsided over the next one week.

Introduction :

The term hiccup (hiccough) referred to as “singultus,” is derived from the Latin word singult which means “the act of catching one’s breath while sobbing”. Hiccups occur due to repeated involuntary spasmodic contraction of the diaphragm which followed by a sudden closure of the glottis.

Hiccups are usually of acute onset, self-limiting and benign; affecting almost all people atleast once in their lifetime. The self-limiting hiccup is thought to be induced by the rapid distension of stomach due to overeating, ingesting spicy food, eating too fast, drinking carbonated drinks, aerophagia and sudden change in the temperature of the ingested food. However, hiccups can sometimes be a manifestation of an underlying serious disorder as well. Hiccups are classified as acute hiccups that lasts for less than 24 hours, persistent hiccups that last for more than 48 hours and intractable hiccups that last for more than a month.

Intractable hiccups often impose a diagnostic challenge to physicians, especially when no identifiable organic cause is identified. Such cases are often referred for psychiatric evaluation and treatment. We report the case of a patient presenting with intractable hiccups who responded to baclofen treatment.

Case Report :

An 18-year-old female presented to the medical out-patient service with a history of hiccups of two months duration which were sudden in onset, progressive in nature, present throughout the day and were absent during sleep. There were no aggravating or relieving factors. There was no history of headache, seizures, loss of consciousness, ear discharge, neck pain, shortness of breath, cough, palpitations, pain abdomen, drug-intake or toxin exposure or trauma. There was no history of stress or anxiety related symptoms, loss of weight or appetite. No history of epilepsy, asthma, seizures, jaundice, tuberculosis, acute or chronic kidney injury, trauma in the past. No past blood transfusions, no past surgical history.

At the time of examination, pulse was 82 beats/min, blood pressure was 120/70 mm Hg, respirations 22/min and oxygen saturation (by pulse oximetry) was 98% while breathing room air at sea level. Detailed general physical examination and systemic
examination did not reveal any abnormality. Otorhinolaryngology consultation was unremarkable. She was started on oral proton pump inhibitor rabeprazole 20 mg once-daily and prokinetic domperidone 10 mg, thrice-daily.

Laboratory investigations revealed haemoglobin 11.6 g/dl, total leucocyte count 8,400/mm³, with a differential count of polymorphs 72%, lymphocytes 22%, and monocytes 6%; erythrocyte sedimentation rate 15 mm at the end of one hour (Westergren method); random blood sugar 89 mg/dl, serum creatinine 0.63 mg/dl, serum urea 27 mg/dl, serum sodium 132 mEq/l, serum potassium 3.7 mEq/l, serum total bilirubin 0.8 mg/dl, serum amylase 28 IU/L, serum lipase 52 IU/L. Chest radiograph, electrocardiogram, abdominal ultrasonography were normal. Computed tomography of the brain did not reveal any intracranial lesion. Upper gastrointestinal endoscopy was also normal.

As her symptoms persisted even after 3 weeks treatment with proton pump inhibitor and prokinetic, she was started on oral chlorpromazine (25 mg twice-daily). As the hiccups did not improve even after another day, she was started on oral baclofen 10 mg twice-a-day. The frequency of hiccups decreased after the initiation of baclofen and the hiccups completely subsided over a period of next one week. She was diagnosed to have idiopathic, intractable hiccups.

**Discussion:**

The hiccup is an involuntary medullary reflex of the chest wall. The reflex mechanism has an afferent pathway (the phrenic nerve, vagus nerve, and sympathetic chain arising from T6 to T12), the efferent pathway (phrenic nerves to the diaphragm, the intercostal nerves to the intercostal muscles, the nerve to the scalene muscles, and the recurrent laryngeal nerve to the glottis), and the central component (brainstem connections between C3 and C5 linking the respiratory center, phrenic nerve nuclei, medullary reticular formation, and hypothalamus). Disturbances at any level may result in intractable hiccups. Lesions in medulla such as stroke, multiple sclerosis and brain tumour are also known to cause intractable hiccups.

The aetiological causes of intractable hiccups (Table-1) include structural or functional disturbances of the medulla, metabolic and endocrine disorders, drugs, general anaesthesia, and psychological problems. Various non-pharmacological methods of treatment such as breath holding, drinking cold water, and inserting a nasogastric tube have been found to be ineffective in treating persistent hiccups. Several pharmacological agents have been used to treat intractable hiccups. Chlorpromazine has been the most frequently used drug in the treatment of hiccups. If hiccups are not controlled with chlorpromazine, other drugs such as haloperidol, clonazepam, olanzapine, amitriptyline, methylphenidate, metoclopramide, phenytoin, valproic acid, carbamazepine, nifedipine, lidocaine, midazolam and baclofen, have all been reported to be useful in treatment. In addition to pharmacological therapy, there are few advances in treatment of intractable hiccups, such as,

**Table 1**: Causes of persistent and intractable hiccups

<table>
<thead>
<tr>
<th>Central nervous system</th>
<th>Stroke, Brain stem tumours, Neuromyelitis optica Multiple sclerosis, Brain injury, Seizures, Parkinsonism</th>
</tr>
</thead>
<tbody>
<tr>
<td>Chest cavity</td>
<td>Mediastinal diseases, Lymphadenopathy, Diaphragmatic tumours</td>
</tr>
<tr>
<td>Cardiac</td>
<td>Myocardial ischemia</td>
</tr>
<tr>
<td>Gastrointestinal system</td>
<td>Esophageal tumours, GERD, Stomach volvulus H. pylori infection</td>
</tr>
<tr>
<td>Procedure related</td>
<td>Catheter ablation, Central venous catheterization, Oesophageal stent placement, Bronchoscopy, Tracheostomy, atrial pacing</td>
</tr>
<tr>
<td>Miscellaneous</td>
<td>Electrolyte imbalance, Ethanol users, Tuberculosis Chronic kidney disease</td>
</tr>
<tr>
<td>Drugs</td>
<td>Benzodiazepines, Levodopa, Nicotine, Ondansetron</td>
</tr>
</tbody>
</table>

*Source: reference 5*
suggests that early institution of baclofen in intractable hiccup cases of idiopathic origin is useful to prevent unnecessary morbidity, disability, and risk of mortality.

References: