

Idiopathic Intractable Hiccups : A Case Report**D Prabath Kumar¹, P Thejeswini², D Neeharika², Alladi Mohan³****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”¹. 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 at least 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 month².

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

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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 0.8mg/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 (25mg 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)³. 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 hiccups⁴.

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 problems⁵. 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 treatment⁶. In addition to pharmacological therapy, there are few advances in treatment of intractable hiccups, such as,

Table 1⁵ : Causes of persistent and intractable hiccups

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

Source : reference 5

phrenic nerve, cervical epidural and stellate ganglionic block⁷.

In the present case, the hiccups were of more than 1-month duration, and there was no obviously evident identifiable cause of the hiccups. As the patient did not respond to treatment with domperidone, rabeprazole, chlorpromazine, baclofen was started, which provided rapid symptom relief. Baclofen is a gamma-aminobutyric acid (GABA) analogue that results in depression of reflex hiccup activity. It has been found to be effective in controlling the intractable hiccups⁸. Baclofen was useful in controlling intractable hiccups in our patient as well. Other GABA agonists such as pregabalin have also been found to be useful in the treatment of intractable hiccups⁸. Some precautions to be kept in mind while prescribing baclofen include ensuring normal kidney functions (as it is mainly excreted by kidneys) and tapering the dose gradually to prevent withdrawal symptoms.

Patients with chronic intractable hiccups run a risk of dehydration, insomnia, depression, gastrointestinal disorders, such as gastroesophageal reflux and even death⁵. The present case report

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 :

1. Sharma RC. Successful treatment of idiopathic intractable hiccup with baclofen and supportive treatment : a case report. *J Neuropsychiatry Clin Neurosci* 2015;27:62-3.
2. Moretto EN, Wee B, Wiffen PJ, Murchison AG. Interventions for treating persistent and intractable hiccups in adults. *Cochrane Database Syst Rev* 2013;CD008768.
3. Hansen BJ, Rosenberg J. Persistent postoperative hiccups : a review. *Acta Anaesthesiol Scand* 1993;37:643-6.
4. Amirjamshidi A, Abbassioun K, Parsa K. Hiccup and neurosurgeons: a report of 4 rare dorsal medullary compressive pathologies and review of the literature. *Surg Neurol* 2007;67:395-402.
5. Chang F-Y, Lu C-L. Hiccup: Mystery, Nature and Treatment. *J Neurogastroenterol Motil* 2012;18:123-30.
6. Homer JR, Davies JM, Amundsen LB. Persistent hiccups after attempted interscalene brachial plexus block. *Reg Anesth Pain Med* 2005;30:574-6.
7. Kuusniemi K, Pyylampi V. Phrenic nerve block with ultrasound-guidance for treatment of hiccups: a case report. *J Med Case Reports* 2011;5:493.
8. Guelaud C, Similowski T, Bizec JL, Cabane J, Whitelaw WA, Derenne JP. Baclofen therapy for chronic hiccup. *Eur Respir J* 1995;8:235-7.