JOURNAL OF BEHAVIORAL HEALTH, 2019
VOL 8, NO. 1, PAGE 20–23
10.5455/jbh.20181212031940

CASE REPORT

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Intractable hiccups (Singultus) of psychogenic origin—A case report

Javed Ather Siddiqui[1,2], Shazia Farheen Qureshi[1,3], Adel Allaithy[1], Talal Abdullah Mahfouz[1]

[1]Department of Psychiatry, Mental Health Hospital, Taif, Saudi Arabia

[2]Seth GS Medical College & KEM Hospital, Mumbai, India

[3]Government Medical College, Aurangabad, Maharashtra, India

ABSTRACT

Hiccups are repeated involuntary, intermittent spasmodic contractions of the diaphragm and inspiratory intercostal muscles which results in sudden inspiration, followed by a sudden closure of the glottis. It is naturally benign, self-limited but maybe most of the time organic in nature and in some cases, chronic, debilitating, and psychogenic. Here, we report a case of a 16-year-old female student who developed hiccups of a psychogenic origin started by stress, which last up to 48 hours sometimes for a month and is successfully treated by antipsychotics haloperidol and psychological interventions. Physicians always face a diagnostic challenge, especially when no identifiable organic cause is elicited. However, such cases are invariably referred for psychiatric evaluation and treatment.

ARTICLE HISTORY

Received December 12, 2018
Accepted February 12, 2019
Published February 19, 2019

KEYWORDS

Persistent hiccups; singultus; psychogenic; antipsychotics

Introduction

The word singultus means hiccups and it comes from singult; a Latin word that means "gasp" or "sob." The hiccup is produced by the sudden repeated involuntary spasmodic contractions of the diaphragm and intercostal muscles, leading to sudden contraction of the glottis. The incoming air hits the closed glottis and produces a characteristic hic sound [1]. It is always benign and self-limiting. The physiological purpose of hiccups is not known [1,2]. There are different types of hiccups according to their duration: Acute hiccups last up to 48 hours [3,4], persistent hiccups are hiccups that last for more than 48 hours [3,4]. Intractable hiccups are hiccups that last more than 1 month [4] or 2 months [3].

Psychogenic hiccups are considered a subtype of intractable hiccups. The causes of hiccups can be idiopathic, psychogenic, organic, structural, or functional disturbances of the medulla. Hiccups result when afferent or efferent nerves to the respiratory muscles and the medullary center controlling these muscles are irritated. Other causes are metabolic and endocrine disorders, drugs, general anesthesia, and psychological problems [5]. Intractable hiccups may result in severe discomfort, decreased physical strength, and mental depression. If left untreated may cause death also. Drug-induced causes are antibiotics, benzodiazepines, barbiturates, corticosteroids, opioids, methyldopa, and cytotoxic agents [4]. There are many other causes for hiccups as gastric distention, alcohol abuse, carbonated beverages, very hot and cold drinks, cancer, lesions of the central nervous system, anxiety, and stress. Some case studies said that hypothesis on the neurotransmitters involved to cause hiccups. The central neurotransmitters like gamma amino butyric acid, dopamine, serotonin, and peripheral neurotransmitters like epinephrine, norepinephrine, acetylcholine, and histamine are enlaced in hiccups. The exact etiology is not clearly stated, as well as which kinds of neurotransmitters and receptors in this nervous system are associated in the pathophysiology of hiccups [6,7]. Hiccups occur for short period in healthy individuals after a large meal, intake of alcoholic beverages, or sudden excitement of any event. The intractable hiccups are more common in men around 82% than in women, and men suffering from hiccups are 50 years of age or older [8]. Psychogenic hiccups have been reported to occur more commonly in women.

Contact Javed Ather Siddiqui ✉ javedsiddiqui2000@gmail.com Mental Health Hospital, Taif, Saudi Arabia.

Regarding pharmacological treatment options for hiccups, chlorpromazine and metoclopramide are the most frequently used first-line treatments [6,7]. Chlorpromazine is the only approved medication for hiccups by the Food and Drug Administration but it is not effective for all patients, so other adjunct therapies should be used. Dopamine is associated with the pathophysiology of hiccups, so chlorpromazine and other antipsychotics get effective results in the treatment of hiccups. Drugs like dopamine-blocking agents, baclofen, clonazepam, and phenytoin have widely varying mechanisms of action, which is effective in the treatment of hiccups [9]. Various non-pharmacological treatment methods also play an important role such as breath holding, drinking cold water, and inserting a nasogastric tube, which are found ineffective in cases of persistent hiccups. The medications such as chlorpromazine, haloperidol, amitriptyline, olanzapine, phenytoin, valproic acid, carbamazepine, methylphenidate, metoclopramide, nifedipine, lidocaine, baclofen, and midazolam have been reported to be effective in such hiccups [3,10,11]. Interventional methods such as phrenic nerve block have been effective for the treatment of intractable hiccups [12].

Case Report

A 16-year-old, good body built, and a socially active female student was referred by otorhinolaryngologist for psychiatric evaluation. She had no prior past history of psychiatric and medical illness. She is a school student studying in 11th standard. She was apparently alright 6 months before when her first term examination was near and she had to appear in the examination. She had a lot of stress of completing her syllabus and she became very anxious. She started experiencing persistent and rhythmic hiccup-like breaths due to which her speech was disturbed, and she was unable to concentrate on her studies. Her symptom disappears at sleep. Her elders advised to perform non-pharmacological treatment methods such as breath holding, drinking cold water while pinching nose, and she was successful initially, but whenever she got stressed, her hiccups like breaths restarted. It is initially persistent and last up to 48 hours, then slowly sometime it become intractable, lasts for more than a month. Her rate was 4–6 times per minute with regular intervals. Then the patient was referred to otorhinolaryngologist and neurologist opinion. She was diagnosed with intractable hiccups and the organic cause was ruled out. They started antitussive, as well as treated gastritis with pantoprazole plus domperidone, but the patient did not improve and was advised to have a psychiatric referral.

On psychiatric examination, no significant findings were present, except that she has stress and anxiety features during the examination period. She had a similar episode before but always during the examination period. There was no family history of psychiatric illness, as well as hiccups. The routine laboratory findings including a complete blood count, all biochemical tests, and C-reactive protein were all within normal range. On local examination, her pharyngeal noises that heard like "gyu," "gyu," and it is heard almost every second. The noises present all day long but disappeared while the patient was sleeping. She was diagnosed with a case of intractable hiccup secondary to anxiety. She was prescribed with escitalopram 10 mg daily with chlorpromazine 50 mg at bedtime for 2 weeks, but she did not get relief. She was admitted for detailed workup. Her history revealed that the patient had repetitive hiccups lasting for a whole day and worsen by speaking and was absent during sleep. There was no associated complaint such as pain in the abdomen and difficulty in breathing. She is always preoccupied in thought and become anxious due to the stress of an examination. All relevant investigations were normal, as done before. Chlorpromazine was withheld and started haloperidol 1.5 mg daily, then slowly increased up to 1.5 mg twice per day, along with escitalopram 10 mg per day continued. The frequency of the hiccups started decreasing after 1 week. After 2 weeks, her hiccups completely subsided. We tapered haloperidol and stopped, and continued escitalopram to lift her stress and anxiety; later it was also stopped after 6 months. Her psychiatric symptoms and hiccups completely disappeared. We have also given supportive treatment in the form of assurance, support, psycho-education, and counseling. She was also taught relaxation therapy. No reports of further episodes have been reported.

Discussion

We have reported an unusual case of intractable hiccups due to psychosomatic causes, which was successfully treated by low doses of antipsychotic therapy along with psychological interventions. This case report is associated with stress and intense emotions such as fear of examination and anxiety are known cause for hiccups [13–15].

Table 1. Some case report review on hiccups of psychogenic origin.

Author	Article name	Treatment
Theohar and McKegney [14]	Hiccups of psychogenic origin: A case report and review of the literature	Hypnotic suggestion
Mehra et al. [16]	Psychogenic hiccups in children and adolescents: A case series.	In this case series, three cases were treated by counseling and psycho-education of a family member to cut down the secondary gain and the fourth case was treated by a low dose of benzodiazepine and psycho-education.
Elife [17]	Successful treatment of intractable hiccups due to psychosomatic disorder.	Treated by acupuncture treatment modality and Sulpiride 50 mg twice per day.
Jambulkar et al. [18]	Psychogenic hiccups—a case report.	Treated by Haloperidol 0.25 mg thrice a day and Clonazepam 0.25 mg thrice a day.
Nishikawa et al. [19]	Intractable hiccups (singultus) abolished by risperidone but not by haloperidol.	Syrup Risperidone 3 ml.

Patient got benefit from hiccups initially with non-pharmacological measures such as breath holding and cold water drinking while pinching nose, as it has been taught by her elders. Hiccups were not observed in sleep. This case seems psychogenic in origin. Though it is rare, it always encounters in clinical practice. There are many case reports [14,16–19] of hiccups treated by different medications, but as per our knowledge, there are no similar case reports treated with combinations of escitalopram and a low dose of haloperidol along with psychological interventions. We reviewed the existing literature and many case reports on hiccups of psychogenic origin, which has been treated with different medications and psychotherapy is depicted in Table 1. Initially, we ruled out of medical cause and later we focused on the comorbid psychiatric cause, and we diagnosed her as an intractable hiccup secondary to anxiety. Our patient improved with an antidepressant low dose of antipsychotic along with psychological interventions.

In the present case, the hiccups are episodic, lasts more than month duration and particularly during stressor of examination. There was no identifiable medical cause for the hiccups occurrence. The patient did not respond to pantoprazole with domperidone, antitussive, and chlorpromazine. Therefore, we started an antidepressant, with a low dose of antipsychotic-like haloperidol, which is effective. It is also important that the patient should be given a supportive treatment of psychological interventions in the form of assurance, encouragement, and support. A complete psycho-education and counseling of the patient and her relatives are also given.

Conclusion

Such type of hiccups raises the possibility of a diagnostic challenge to physician and otorhinolaryngologist, especially when no identifiable organic cause is elicited. It is also important to rule out the comorbid psychiatric cause and such cases are always referred for psychiatric evaluation and treatment, as well later follow-up at psychiatric outpatient department. This case showed significant improvement with antidepressant and a low dose of haloperidol. Psychosomatic causes such as anxiety and stress should be considered in the underlying cause of intractable hiccups.

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