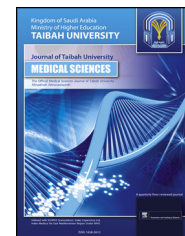




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Case Report

Psychogenic hiccups in an older adult: A case report and literature review



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المخلص

الفواق في الغالب حميدة وذاتية الحد. من ناحية أخرى، يمكن للفواق المستمر أو المستعصي أن يكون مؤلماً ويشير إلى وجود اضطراب نفسي أو فسيولوجي أساسي. قدم رجل يبلغ من العمر 63 عاماً إلى عيادة الطب السلوكي في مستشفى الرعاية الثالثية في مسقط، عمان، مع تاريخ 4 سنوات من الفواق المستعصي؛ بعد استبعاد جميع الأسباب العضوية للفواق المستعصي، تم تشخيص الفواق النفسي المنشأ. الفواق النفسي المنشأ نادر جداً، حيث تم نشر 7 سلسلة حالات وتقارير فقط حتى الآن على بوميد وقوقل سكولار. يتضمن هذا التقرير مراجعة شاملة للأدبيات المتعلقة بجميع حالات الفواق النفسي التي تم الإبلاغ عنها والتي نشرت حتى الآن على بوميد وقوقل سكولار للتحقيق في فعالية العلاجات المختلفة لهذه الحالة بدقة. الفواق النفسي المنشأ نادر جداً، ويجب أن يتم التشخيص بعد استبعاد الأسباب العضوية. يمكن أن تكون الإدارة صعبة بسبب نقص التدخلات القائمة على الأدلة.

الكلمات المفتاحية: زوبعة؛ فواق؛ نفسية المنشأ؛ مستعصية على الحل؛ طويلة الأمد

Abstract

Hiccups are typically benign and self-limiting. However, persistent or intractable hiccups can be debilitating, and may indicate the presence of an underlying physiological or psychological disorder. A 63-year-old man presented to the behavioral medicine clinic at a tertiary care hospital in Muscat, Oman, with a 4-year history of intractable hiccups. After exclusion of all organic causes of

intractable hiccups, a diagnosis of psychogenic hiccups was made. Psychogenic hiccups are very rare, and only seven case series and reports have been published to date, according to PubMed and Google Scholar. This report includes a comprehensive review of the literature on all reported cases of psychogenic hiccups published to date, according to these two databases, to thoroughly investigate the effectiveness of various therapies for this condition. Psychogenic hiccups are very rare, and diagnosis should be made after exclusion of organic causes. Management can be challenging because of the lack of evidence-based interventions.

Keywords: Hiccough; Hiccup; Intractable; Protracted; Psychogenic

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Introduction

A hiccup is a sudden repeated contraction of the diaphragm and inspiratory muscles that causes the glottis to close as a result of the abrupt rush of air into the lungs, thus causing a distinct “hic” sound.¹ Most hiccups are self-limiting and require treatment only in rare cases. However, various organic and psychogenic factors can result in chronic, intractable hiccups that can persist for years.² Intractable hiccups may lead to exhaustion, dehydration,

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malnutrition, weight loss, depression, insomnia, and low quality of life.²

Psychogenic hiccups are a rare condition described in adults and pediatric populations. Unfortunately, most evidence of treatments for psychogenic hiccups has come from case series or reports. In this case report, we describe the first reported case of psychogenic hiccups in an older adult patient. An older person is defined by the United Nations as a person who is over 60 years of age. In addition, we include a comprehensive review of the literature on all reported cases of psychogenic hiccups published to date, according to PubMed and Google Scholar, to thoroughly investigate the effectiveness of various therapies for this condition.

Case report

A 63-year-old retired male farmer presented to the behavioral medicine clinic at a tertiary care hospital, Muscat, Oman, with a complaint of intractable hiccups in the past 4 years. He had no known medical or psychiatric comorbidities. Four years earlier, the patient had experienced infrequent episodes of hiccups lasting for several minutes, which gradually worsened over time and became more persistent, and occurred throughout the day and night at equal frequency. The hiccups subsided only during sleep and eating of his regular meals. The hiccups were associated with breathing difficulty and abdominal pain. The patient denied having any stressors before the onset of the hiccups. He observed that the hiccups were triggered by speaking. Therefore, he avoided talking to his family members or friends, and spent most of his time alone in his bedroom. Sometimes, he induced vomiting by inserting his fingers down his throat and drinking excessive amounts of cold water to stop the hiccups, but these behaviors provided only temporary relief for approximately half an hour. The patient reported experiencing persistent low mood most days of the week for the prior 3 months, associated with undocumented weight loss, loss of energy, social withdrawal, insomnia, feelings of worthlessness, and recurrent thoughts of death. He had no symptoms of cognitive impairment, and he independently performed all activities of daily living. He denied any stressors before experiencing the depressive symptoms, alcohol consumption, smoking, and drug abuse.

Clinical examination revealed an overweight BMI of 29.7 m²/kg, a height of 163 cm, and a weight of 79 kg. The patient had normal vital signs, and his systemic examination was unremarkable. He was investigated thoroughly, and evaluated for possible gastroenterological and neurological disorders. His biochemical investigation findings were normal, except for dilutional hyponatremia attributed to the normal physiological response to excessive water consumption. His cardiac evaluation, including an electrocardiogram,

was normal. An extensive gastrointestinal workup was unremarkable, including chest X-ray, abdominal X-ray, abdominopelvic computed tomography, and gastroendoscopy. Neurologically, he was evaluated with electroencephalography, a computed tomography scan of the brain, magnetic resonance imaging of the brain, and phrenic nerve conduction study, all of which indicated normal findings. The patient was not taking any medication known to cause hiccups.

The patient denied any preceding or ongoing stressors or conflicts (e.g., financial, marital, family, or occupational stressors). After exclusion of all possible medical conditions, a diagnosis of psychogenic hiccups with co-morbid major depressive disorder was made. No other comorbid psychiatric diagnosis was made. The patient was administered various medications, including mirtazapine, at a dose as high as 45 mg at bedtime (HS); some improvement was observed in his depressive symptoms. However, he continued to experience intractable hiccups. Haloperidol, risperidone, and chlorpromazine were commenced at different times but did not reportedly decrease the frequency of the intractable hiccups. Later, the patient commenced pregabalin 75 mg in the morning and 150 mg in the evening along with mirtazapine 45 mg HS, in addition to relaxation therapy. He reported significant improvements, evidenced by fewer episodes of hiccups. In follow-up appointments, he reported that the hiccups were lower in intensity and frequency. He also maintained significant amelioration of his depressive symptoms.

Materials and Methods

A literature search of reported cases of psychogenic hiccups was conducted in PubMed and Google Scholar (Table 1). The keywords used were hiccup, intractable hiccup, persistent hiccup, protracted hiccup, and psychogenic hiccup. The results were filtered to include only case reports. On the basis of review of the abstracts, unrelated publications were excluded, and only cases with a diagnosis of psychogenic hiccups were chosen. Finally, a total of seven publications were reviewed (Table 2).

All case reports found to report psychogenic hiccups were reported in children or adults, whereas no cases were reported among older adults. The hiccup duration ranged between 4 days and 6 years, with varying episodes in each case. All patients were investigated thoroughly from a medical perspective, and organic causes were excluded. Four of seven cases^{3–6} were treated with a combination of pharmacological and psychological management. Pharmacological treatments included benzodiazepines, antipsychotics, antidepressants, and dopamine antagonists. Psychological management included psychoeducation, cognitive behavioral therapy,

Table 1: Research methods.

Initial Results	After Filter Application	Title and Abstract Evaluation	
PubMed (N = 84)	Case reports, any time (N = 41)	Articles were evaluated according to relevant titles and abstracts (N = 21)	Subsequently, only cases reporting psychogenic hiccups were included (N = 7)
Google Scholar (N = 1508)	Date 2006–2021 (N = 817)		

Table 2: Literature review results.

Author	Article Name	Sociodemographic	Nature of Hiccups	Psychiatric History	Duration	Intervention	Outcome
Theohar et al. ⁷	Hiccups of psychogenic origin: a case report and review of the literature	41-year-old, white, married mother of four children	Sudden onset, hiccups when awake; stopped during sleep	Conversion reaction to psychosocial problems	2 weeks	Hypnosis	10-month follow up: patient completely hiccup free for up to 5 months
Singh et al. ³	Conversion disorder presenting as intractable hiccups in middle-aged male	50-year-old man	Reoccurring at regular intervals every 5–10 s after quitting alcohol; relieved by drinking alcohol		5–6 years	(Lorazepam 1 mg BID, chlorpromazine 50 mg HS, escitalopram 10 mg BID) + behavioral therapy session	Good patient response to treatment
Mehra et al. ⁸	Psychogenic hiccup in children and adolescents: case series	13-year-old girl	Continuous; absent during sleep; presented as a manifestation of psychological distress		4–5 months	Counseling and psychoeducation for the family to reduce secondary gain.	Symptom resolution in 4 days
		11-year-old boy	Episodic; absent during sleep, talking, and eating; presented as a manifestation of psychological distress	Temper tantrum	12–14 months	Counseling and psychoeducation for the family to reduce secondary gain.	Improvement in 7 days; no recurrence of symptoms
		13-year-old boy	Episodic; absent during sleep, talking, and eating; presented as a manifestation of psychological distress	Social phobia	3–4 months	Counseling and psychoeducation for the family to reduce secondary gain.	Improvement on the day of detailed assessment when double bind used
		14-year-old boy	Episodic; absent during sleep, talking, and eating; presented as a manifestation of psychological distress		3–4 months	Low-dose benzodiazepines, counseling, and psychoeducation of family to reduce secondary gain.	Complete improvement on the day of detailed assessment; maintained well at follow up
Jambulkar et al. ⁴	Psychogenic hiccups – a case report	20-year-old single female, college student	Three episodes of hiccups, first and last episode continuous, stopping only during sleep		First episode: 4 days, second episode: 1 day, third episode: 10 days	Episodes 1 and 2 were treated with some medications (not specified in the article); last episode treated with haloperidol 0.25 mg TID and clonazepam 0.25 mg TID along with relaxation	First treatment relieved hiccups for 7 weeks; hiccups reoccurred. Episode 3: Patient was on medications for 8 weeks then gradually tapered off over the next 4 weeks; no further episodes reported in follow up

Vaidya ⁹	Sertraline in the treatment of hiccups	42-year-old divorced African American man	Intractable hiccups after a Mallory Weiss lesion of esophagus	History of depressive episodes and anxiety symptoms on and off for 15 years; diagnosed with adjustment disorder; history of alcohol abuse but quit 3 years prior	3 years	therapy + psychoeducation for the patient and relatives for adherence to medications and regular follow up Started on sertraline 50 mg OD and gradually increased to 150 mg OD	Reported improvement in depressive symptoms and decrease in hiccups; after gradual increase in sertraline to 150 mg OD for 2 weeks; brief hiccup episodes lasted 1–2 h and were usually associated with meals In 6 month follow up, reported improvement of depressive symptoms and relief of hiccups; after sertraline dose reduction, hiccups returned
Nishi and Rajput ⁵	Intractable hiccups (singultus) in case of anxiety neurosis	40-year-old woman	3–4 episodes per day	Anxiety, neurosis; on anti-anxiety and anti-depressants + frequent panic attacks	More than 2 months	Metoclopramide for 15 days TID + counseling + cognitive behavioral therapy + lifestyle modifications	Decrease in intensity of hiccups
Siddiqui et al. ⁶	Intractable hiccups (singultus) of psychogenic origin	16-year-old girl, student in grade 11, socially active	Hiccup-like breaths; rate 4–6 times per minutes with regular intervals		Initially persistent; lasted for up to 48 h, then slowly became intractable, lasting for more than 1 month	Initially escitalopram 10 mg OD, chlorpromazine 50 mg HS for 2 weeks Subsequently haloperidol 1.5 mg OD, then slowly increased to 1.5 mg BID along with escitalopram 10 mg OD and supportive therapy, such as reassurance, support, psychoeducation, counseling, and relaxation therapy	No relief reported with escitalopram and chlorpromazine Decreased frequency of hiccups after 1 week; completely subsided after 2 weeks; haloperidol was tapered down and stopped; escitalopram was continued and then stopped after 6 months; psychiatric symptoms and hiccups resolved

counseling, and relaxation therapy. In one case, hypnosis was used to manage psychogenic hiccups⁷ and one case was managed with only psychoeducation and counseling.⁸ In one case⁹ the effectiveness of sertraline as a treatment for hiccups of psychogenic origin was discussed. Table 2 illustrates the results of the seven articles reviewed.

Discussion

We reported a case of intractable hiccups in an older adult man with no known psychiatric history or ongoing stressors at the time of hiccup occurrence. A diagnosis of psychogenic hiccups associated with the co-morbid major depressive disorder was made after exclusion of all possible organic causes.

Among cases reported in children, psychological interventions significantly affect management of psychogenic hiccups. According to Mehra et al.,⁸ both counseling and family psychoeducation directly improve patients' hiccups. Furthermore, a combination of psychological and pharmacological intervention has been reported to have strong effects in causing hiccups to subside in all cases reported in both adults and children.³⁻⁶ In combination with psychoeducation, a low dose of benzodiazepines has been used to manage intractable hiccups in a 14-year-old boy⁸ and in a 20-year-old woman⁴ who was administered an antipsychotic agent. The hiccups were entirely resolved in both patients, and no further hiccups were reported in the follow-up period.^{4,8} Psychological intervention was not an option in our case because of the patient's age and level of education, which would have affected adherence to such an intervention. The use of antipsychotics such as haloperidol⁶ has also been reported to ameliorate psychogenic hiccups. However, chlorpromazine did not show an effect when used with an antidepressant in the same patient. In contrast, a 50-year-old patient³ has been reported to respond well to a combination of chlorpromazine, escitalopram, and lorazepam. In this case report, all antipsychotics combined with antidepressants did not relieve the patient's intractable hiccups. In a study investigating the effect of sertraline in treating psychogenic hiccups,⁹ improvements were observed in terms of a decrease in both the frequency of hiccups and the patient's depressive symptoms. In the case reported herein, mirtazapine was initially used to treat the patient's symptoms; however, although it improved his depressive symptoms, it did not decrease the frequency or intensity of his hiccups. Pregabalin was then started and significantly decreased his persistent hiccups. In one case of idiopathic intractable hiccups, significant improvements in stopping the patient's hiccups have been reported after treatment with pregabalin after unsuccessful trials of cervical epidural block and phrenic nerve block.¹⁰ Moreover, Jatzko et al.¹¹ have reported three cases wherein alpha-2-delta ligands effectively treated chronic idiopathic hiccups in older adult people. Pregabalin was used as an add-on medication with other hiccup medications in two cases, and improvements were observed in both patients. The pathophysiology of hiccups is associated with three components: the afferent limb, including the phrenic, vagus, and sympathetic nerves;

the central processing unit in the midbrain; and the efferent limb, which carries the motor fibers to the diaphragm and intercostal muscles.¹² The literature has indicated strong evidence that gamma-amino-butyric-acid (GABA) is a neurotransmitter involved in hiccup reflex in the central nervous system.¹² Pregabalin, a GABA analog, has a role in treating intractable hiccups, because it affects the presynaptic terminals of respiratory muscles by decreasing calcium influx, inhibiting voltage-operated calcium channels, and increasing serotonin levels.¹⁰

Finally, hypnosis is another intervention that has been reported in the treatment of a 41-year-old woman with intractable hiccups with a history of conversion disorder.⁷ The patient remained fully hiccup-free in the 10-month follow-up period.

Conclusion

Combined psychological and pharmacological intervention is crucial in treating intractable hiccups of psychogenic origin in children and adults. The use of pregabalin in treating psychogenic hiccups, along with other antidepressant medications, can help decrease hiccup intensity, as shown in this case report, or even cause hiccups to subside, as demonstrated in previously reported cases. However, more cases must be reported to compare and explore the effectiveness of different psychiatric medications and their value in treating these hiccups. In contrast, psychological interventions must be investigated thoroughly in older adult populations to observe the effects in those patients. Unfortunately, psychoeducation and counseling were not an option for treating the hiccups in our patient; therefore, psychoeducation of family and patients should also be included and investigated in further studies.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms for the patient to report clinical information for publication.

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Ethical Approval

The medical research ethics committee at Sultan Qaboos University does not require ethical approval as long as the patient has given informed consent to the publication of his clinical data and no personally identifiable information that could be used to identify the patient has been disclosed in the article.

Conflict of interest

The authors have no conflicts of interest to declare.

Authors contributions

TM, FS, and HS conceived and designed the study, conducted a literature review, and collected patient data and performed follow-up. TM, FS, and NB analyzed and interpreted data. TM, FS, AH, and HS wrote the initial and final drafts of the article. All authors have critically reviewed and approved the final draft and are responsible for the content and similarity index of the manuscript.

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