

## Persistent Singultus: Addressing Complexity With Simplicity

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### Abstract

The hiccup reflex arc involving the brainstem, phrenic and vagus nerves, and the sympathetic chain is associated with singultus. There are many possible organic, psychogenic, idiopathic, and iatrogenic instigators. We describe a case of singultus in a 69-year-old man secondary to his CPAP mask, with resolution after he stopped using the mask. Our case establishes that CPAP may be a cause of iatrogenic aerophagia leading to gastric distention, singultus, and emesis and highlights the importance of a complete history.

### Introduction

Singultus, or hiccups, is derived from the Latin word *singult*, meaning “the act of attempting to catch one’s breath while sobbing.”<sup>1</sup> Hiccups are thought to be secondary to a reflex arc, which involves an afferent tract, a brain center, and an efferent tract. The afferent tract is composed of the phrenic nerve, sympathetics from T6-T12, and cranial nerves V-VII. The brain center includes the brainstem that receives feedback from the hypothalamus, the temporal lobe, and the reticular activating system. Finally, the efferent portion consists of the phrenic nerve, accessory nerves to the external intercostal muscles, and the recurrent laryngeal nerve. When there is stimulation of a portion of the afferent component, the hiccup reflex arc is activated, resulting in hiccups.<sup>2</sup> There are many possible types of instigators: organic, psychogenic, idiopathic, or, in our case, iatrogenic.<sup>3</sup>

### Case Report

A 69-year-old man presented with a 3-month history of intermittent hiccups and vomiting. Hiccups occurred weekly and often in the morning after breakfast, and were accompanied by non-bilious and non-bloody vomiting. His history was significant for hypertension, hyperlipidemia, osteoarthritis, and obstructive sleep apnea. He had undergone an esophagogastroduodenoscopy (EGD), colonoscopy, and an abdominal/pelvic computed tomography (CT), all of which were unremarkable. He reported trying baclofen, esomeprazole, and chlorpromazine without relief, although he had some relief with metoclopramide. Evaluation revealed a well-developed man with an unremarkable head, neck, and neurological exam, normal gastric emptying study, normal cranial CT for central nervous system causes, and a morning cortisol of 6.5 µg/dL (normal: 7-28 µg/dL). However, about 2 weeks prior to a follow-up visit, the patient stopped using his continuous positive airway pressure (CPAP) mask, and his symptoms stopped and did not recur. His CPAP mask was recently changed and the new mask did not fit him properly. We deduced that iatrogenic aerophagia secondary to the CPAP machine led to gastric distention, singultus, and emesis.

### Discussion

Fass et al demonstrated that distention of the proximal esophagus precipitated hiccups, and showed how a large food bolus or carbonated beverage in the proximal esophagus may cause stimulation of the vagus nerve causing hiccups.<sup>4</sup> A similar scenario may have occurred in our patient. The improperly-fitted CPAP mask likely resulted

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in aerophagia, which caused hiccups. It is unclear whether gastric distention had a role in the pathophysiology, although the patient did complain of bloating. No studies have evaluated the role of gastric distention in hiccups, and could be a promising subject for future research.

Conversely, another case report has shown CPAP to be an effective treatment for hiccups. It demonstrated that a CPAP mask will alter esophageal function by increasing the basal lower esophageal sphincter (LES) pressure and decreasing the duration of the LES relaxation during swallowing.<sup>5</sup> A CPAP mask also increases intrathoracic pressure, which prevents the drop in pressure and diaphragmatic contraction that is necessary to cause a hiccup.<sup>5</sup> However, this study did not examine the effects of a poorly fitted mask.

Although CPAP is not known to be a common cause of hiccups, gastric distention may be, and this connection may have been made earlier if it was known that the patient had recently replaced his mask. Singultus may seem a trivial problem due to its regular occurrence, but its presence may indicate more problematic underlying causes. When physicians encounter this complaint, it is important to thoroughly investigate and rule out the possible serious causes of singultus while taking a thorough history to identify simple and easily reversible causes.

## Disclosures

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