

CASE REPORT



Mallory-Weiss tear after violent hiccups: a rare association

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ABSTRACT

We present an unusual case of a 44-year-old male who developed violent hiccups soon after a ureteroscopy for nephrolithiasis; later, the forceful hiccups were followed by hematemesis. Upper esophagogastroduodenoscopy revealed Mallory-Weiss tears and esophageal erosions in the lower esophagus. Esophageal biopsy was unremarkable. The patient did not have a prior history of the gastrointestinal disorder. Although extracorporeal shock wave lithotripsy has on rare occasion been implicated in the development of gastrointestinal erosions, no such correlation exists for ureteroscopy and upper gastrointestinal bleeding in the management of nephrolithiasis. It was the development of violent hiccups for several hours before the onset of hematemesis that likely led to the upper gastrointestinal bleed.

ARTICLE HISTORY

Received 9 September 2016
Accepted 12 December 2016

KEYWORDS

Hiccups; esophageal laceration; hematemesis; Mallory-Weiss

1. Introduction

Hiccups are mostly considered to be benign and self-limited. They are triggered by the activation of the hiccup-reflex-pathway leading to the irritation of the diaphragm. Hiccups, although distressing, have rarely been attributed to gastrointestinal bleeding. This case report documents a yet another rare association between hiccups and upper gastrointestinal bleed due to hiccup-induced Mallory-Weiss tears. Typically, Mallory-Weiss tears are preceded by vomiting and retching. This case highlights that Mallory-Weiss tears could also be triggered by hiccups by causing forceful movements to the diaphragm, and the classical prodromal phase of vomiting and retching may not be necessarily present. It also emphasizes the need for the early institution of therapy in cases of strenuous hiccups, as in rare instances, it may lead to esophageal tears. Recommendations for clinical practice and subsequent practice are discussed.

2. Case report

The patient was a 44-year-old male with past medical history of nephrolithiasis presenting with the chief complaint of hematemesis and epigastric pain. The patient reported that due to his recurrent kidney stones he underwent ureteroscopic procedure four days before presentation. Soon after the ureteroscopic procedure, the patient developed violent hiccups that worsened over the course of time. His severe forceful hiccups were followed by hematemesis which prompted him to visit the emergency department.

The patient denied ingestion of alcohol or non-steroidal anti-inflammatory drugs. His vitals were reported to be stable. His labs revealed elevated white cell count at 19,900. His complete blood count, liver enzymes, and lipase were found to be within the reference range. Abdominal X-ray showed a moderate amount of stools in his colon. His hiccups were controlled with intravenous metoclopramide. Due to his history of hematemesis, and epigastric pain, an esophagogastroduodenoscopy was performed. Esophagogastroduodenoscopy revealed severe mucosal bleeding characterized by erythema and friability. Esophageal erosions and Mallory-Weiss lacerations were found in the lower one-third of the esophagus (Figures 1 and 2). No active bleeding was identified. The patient was discharged on pantoprazole and laxatives with close follow up advised. The patient's symptoms improved over the course of time.

3. Discussion

The pathophysiology of singultus also commonly known as hiccup is complex. It is attributed to irritation of diaphragm that causes it to shift inwards which result in air being pulled against the closed larynx, producing the characteristic 'hic' sound. There are many probable causes (some of which mentioned in Table 1), whereby the diaphragm becoming irritated triggers the hiccup reflex pathway. This hiccup reflex pathway consists of three components:

- (1) The afferent limb of the phrenic nerve, vagus and sympathetic nerves carrying the sensory fibers to the midbrain

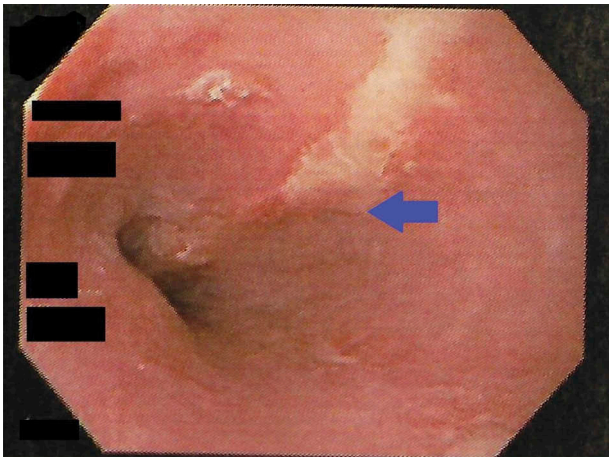


Figure 1. Longitudinal tear consistent with Mallory Weiss (blue arrow).

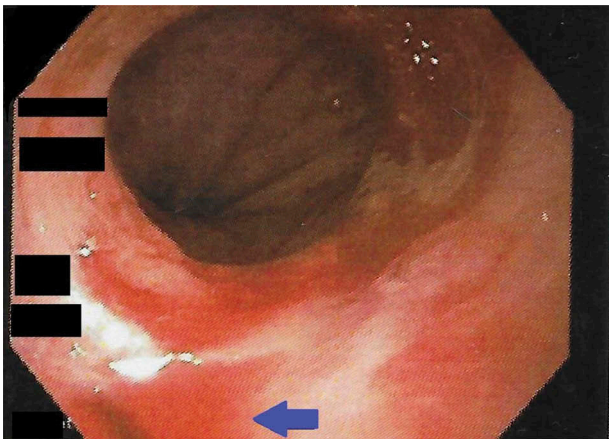


Figure 2. Esophageal erosions as depicted by the blue arrow.

Table 1. Causes of hiccups

Gastrointestinal causes of hiccups

1. Gastroesophageal reflux disease
3. Rapid eating
4. Aerophagia
5. Abdominal surgery
6. Gall bladder pathology

Non-gastrointestinal causes of hiccups

1. Stroke
2. Central nervous system infections
3. Central nervous system lesions (trauma, multiple sclerosis)
4. Drugs (e.g., barbiturates, dexamethasone, alcoholism, anesthetics, etc.)
5. Uremia
6. Electrolyte disorders
7. Pneumonia

- (2) The central processes in the midbrain.
- (3) And the efferent limb from the central processes consisting of the motor fibers of phrenic nerve to the diaphragm and subcostal muscles resulting in the contraction of the diaphragm [1,2].

Hiccups are relatively common, usually self-limited and typically not related to adverse effects; however, in prolonged cases, hiccups have been attributed to sleep disturbances, gastroesophageal reflux disease, cardiac arrhythmias and wound dehiscence in post-operative patients. Hematemesis due to Mallory-

Weiss tear or esophageal erosions after a bout of strenuous hiccups has rarely been documented in the literature.

First described in 1929, Mallory-Weiss is an esophageal tear or laceration of the esophageal mucosa at the gastroesophageal junction, commonly due to vomiting or retching [3]. Although the pathophysiology is poorly understood, it has been postulated that vomiting or retching is anti-peristaltic that raises the intra-abdominal pressure causing the acid content of the stomach to move towards the gastroesophageal junction. Also, the increased intraabdominal pressure due to vomiting causes extension of the of the gastroesophageal junction resulting in the laceration [4]. Overall, the most common cause of upper gastrointestinal bleed is peptic ulcer disease comprising of about 50% of the cases, while Mallory-Weiss tears are associated with less than 25 % of the cases [5,6]. There are several risk factors for the development of Mallory-Weiss tears, the most common risk factor of these tears is chronic or binge alcohol usage [5]. Hiatal hernia is seen in 40% cases which is also a predisposing factor for the development of the Mallory-Weiss tear [4,7]. Hiccups, however, are rarely associated with the gastroesophageal laceration. Our review of the literature revealed only five cases reporting upper gastrointestinal hemorrhage due to Mallory-Weiss laceration after hiccups [7]. Of these, four cases were associated with adults and one an infant [7–9]. In our patient, while a small hiatal hernia was found during the upper esophagogastro-duodenoscopy, it appears that hiccups were the principle factor contributing to the Mallory-Weiss tear.

This documents another rare clinical event of upper gastrointestinal bleeding due to esophageal laceration that was triggered by a bout of violent and strenuous hiccups that persisted several hours after the operative procedure. The hiccups were likely triggered due to general anesthesia and surgery. Surgery induced gastroparesis and anesthetic agents such as propofol have been well described in the literature as the potential causes that could lead to hiccups [1,2].

Extracorporeal shock wave lithotripsy (ESWL) for the management of kidney stones could potentially cause gastric and duodenal lesions [10,11]. However, the patient in question underwent a ureteroscopic procedure and not ESWL for his renal stone. Unlike ESWL, ureteroscopy is not directly linked to upper gastrointestinal hemorrhage. His symptoms appear to have been triggered by the prolonged bout of hiccups due to the anesthesia and procedure related gastroparesis.

The treatment of Mallory-Weiss is supportive if no active bleeding is noticed. The patient was

discharged on acid suppression therapy which is the standard of care for non-bleeding Mallory-Weiss tear [12].

4. Conclusion

This is a rare case in which the patient with no prior esophageal symptoms developed esophageal laceration and upper gastrointestinal hemorrhage after a prolonged bout of strenuous hiccups for several hours. This case documents a rare clinical association between hiccups and esophageal laceration as a cause of upper gastrointestinal bleeding. It also highlights the importance of early recognition and treatment of prolonged and strenuous hiccups, which have the propensity to cause esophageal tears.

Authors' contributions

All authors have contributed to the design of the manuscript, literature review, drafting and revision of the manuscript.

Acknowledgements

Technical, editorial, and writing assistance was provided by Cameron G. Shultz, PhD, MSW.

Compilation of images and documents was provided by Ms. Elizabeth Chavez

Disclosure statement

No potential conflict of interest was reported by the authors.

Funding

None.

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