

## Near fatal recurrent laryngospasm: Unusual presentation of gastric volvulus

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### Abstract

A 47 years old male patient presented repeatedly to emergency department with attacks of severe episodic cough and breathing difficulty. He was seen by different specialities including ENT and a diagnosis of severe reflux laryngitis with laryngospasm was made. Worsening of condition with apnoeic collapse resulted in admission to hospital and investigations. Radiological investigation revealed gastric volvulus and severe esophageal reflux. His condition worsened rapidly resulting in prolonged severe laryngospasm and apnea. This required endotracheal intubation, transfer to ICU and urgent corrective surgery. Here we describe this unique case of severe laryngospasm secondary to and as a presenting symptom of chronic gastric volvulus. Such association has not been reported previously in the literature.

### Keywords

laryngeal spasm; esophageal reflux; reflux laryngitis; gastric volvulus

### Abbreviations

ENT: Ear nose throat; GERD: Gastro esophageal reflux disease; LPR: Laryngopharyngeal reflux; ICU: Intensive care unit; GP: General practitioner; ER: Emergency room

### Introduction

Otolaryngologists commonly come across patients with gastro esophageal reflux disease (GERD). However another large group of patients seen in ENT clinics complain of symptoms related to the reflux of gastric contents into the upper aero digestive tract. They do not complain of heartburn [1], on the contrary they complain of hoarseness, Globus pharyngeus, dysphagia, aspiration, chronic cough, and throat clearing [1]. These presenting symptoms of this group of patients appear to be different from those diagnosed with typical GERD. To better describe these patients and differentiate them from patients suffering from GERD, the term gastro-pharyngeal or laryngo-pharyngeal reflux (LPR) has been described. Laryngospasm

is another symptom which may occur in LPR but is uncommon. When it occurs it is a highly significant and distressing symptom [2-4].

Association of laryngospasm with gastric volvulus, especially as its presenting symptom, has not been described before in the literature. Here we describe a unique case with this association.

## Case Presentation

A 47 years old obese gentleman presented to the GP clinic with complaints of severe episodic coughing and associated choking for about one week. He was diagnosed as having respiratory tract infection and started on antibiotics, antihistamine and cough syrup. Two days later he presented in ER of the hospital with worsening of his symptoms. He was afebrile with stable vital signs, there was no clinical evidence of respiratory tract infection, hematological investigations and X-ray chest was normal. During his stay in ER he was asymptomatic and was referred to ENT for consultation. Further questioning did not reveal any precipitating factors for the cough or any swallowing disorder; however there was history of dyspepsia and heartburn. Complete ENT examination was performed including indirect laryngoscopy (IDL) and flexible naso-laryngoscopy, which showed normal vocal cords movement but with significant posterior laryngitis (congested and edematous mucosa over the arytenoids). These finding indicated laryngo-pharyngeal reflux disease (LPR). At the end of the examination, the patient experienced a severe episode of coughing and choking. It was associated with severe difficulty in breathing and brief periods of apnoea, the whole episode lasted more than a minute. A clinical diagnosis of severe laryngo-pharyngeal reflux (LPR) with episodic laryngeal spasm was made. The patient was prescribed a proton pump inhibitor, dietary and lifestyle advice was provided along with a referral to gastroenterology out-patient department.

There was only brief improvement and after two days he presented in ER again with same symptoms. He was seen by pulmonologist on-call, hematological investigations and chest X-rays were normal. Some wheeze was noted on chest examination, adult onset asthma was suspected for which bronchodilator and steroid anti-inflammatory inhalers were prescribed. Four days later he was brought to ER by ambulance; his condition had worsened with history of collapse and unconsciousness at home. He was referred to and seen by ENT again; naso-laryngoscopic examination findings and conclusion were same as before. The general physical examination did not reveal any positive finding, hematological investigations were within normal range and X-ray chest was clear. In view of worsening condition it was decided to admit the patient and investigate. A Gastrograffin swallow was performed, which showed severe gastro-esophageal reflux and in addition there was incidental finding of gastric volvulus (Figure 1). The stomach was twisted in the middle dividing it into two compartments. The distal part was emptying into the duodenum but the proximal part was refluxing into esophagus. General Surgical team was approached for the management of the patient. It was decided to keep the patient NPO (nil per oral), naso-gastric tube was passed to keep the proximal stomach decompressed and he was nursed at 45 degrees to reduce reflux. Unfortunately the attacks of cough, laryngeal spasm and apnea worsened. During the night, patient collapsed with respiratory arrest, endotracheal tube was inserted and patient was shifted to Intensive Care Unit after resuscitation. After preparation, the patient was taken to operating theatre, Nissen fundoplication and gastropexy was

performed. Patient was extubated post operatively but naso-gastric tube was maintained for feeding and decompression of stomach for 48 hours. Patient was transferred to the ward and was recovering well until the fourth day when the episodic cough with laryngospasm recurred. The naso-gastric tube was reinserted and other conservative measure mentioned before were restarted. Repeat gastrograffin swallow showed unresolved gastric volvulus and persistent gastro-esophageal reflux, indicating surgical failure. The situation was further complicated by abdominal wound dehiscence, due to recurrent severe coughing. Revision surgery was performed and the patient had an extended postoperative stay in ICU due to chest infection and required temporary surgical tracheostomy. He made slow recovery and was shifted to the ward after about ten days. There was no recurrence of his symptoms and he was finally discharged home.

He was followed-up in the ENT and General Surgical clinics with no further episodes of cough, apnoea or laryngospasm. Repeat gastrograffin done three months postoperatively showed no gastric volvulus and good fundoplication with no reflux (Figure 2). He has since been discharged with no further follow-up.

## Discussion

Here we have described a unique case of chronic gastric volvulus presenting with laryngospasm, a symptom of LPR. Although neither laryngospasm nor chronic gastric volvulus is a rare disorder, however there combination in presentation makes the case interesting and rare.

The patient was seen in GP clinic and then many times in ER by different doctors. He always complained of attacks of severe cough and difficulty in breathing. Acute paroxysmal cough in adults with breathing difficulty may be caused by a variety of conditions like common cold, influenza, pertussis, asthma, bronchiectasis, bronchitis, GERD, LPR, pneumonia, tuberculosis, ACE inhibitors and many more. The patient had no other complaints, was always afebrile, with no history of ACE inhibitor intake, normal hematological investigations and clear chest X-rays. This, without going into details, eliminates most of these conditions from the list of differential diagnosis. The patient was treated for respiratory tract infection, LPR and adult onset asthma but with limited and temporary improvement only. On the other hand, chronic gastric volvulus was never considered in the differential diagnosis as there was nothing in the history or examination to suggest it. Here we will not go into details about GERD or LPR, rather we will discuss laryngospasm, chronic gastric volvulus and their relationship in our reported case.

Laryngospasm is a sudden, prolonged, forceful apposition of the vocal cords; it is a protective laryngeal reflex in response to noxious stimuli. Chodosh was the first to describe reflux induced laryngospasm in humans [3], since then much work has focused on this [1-10] including animal model studies [11-13]. These studies implicated acid contact with the endolaryngeal mucosa as a cause of both laryngospasm and reflex central apnea [11-13]. The superior laryngeal nerve forms the sensory arm and recurrent laryngeal nerve forms the motor arm of the reflex laryngospasm. The reflex central apnoea results from inhibition of the respiratory centre preventing further downward movement of the gastric acid secretions [2,5,11-13]. Both actions, laryngospasm by forceful apposition of vocal cords and reflex central apnea by stopping respiration, together provide effective protection against aspiration into respiratory tract. Because of these actions, LPR has also been named as a possible cause of sudden infant death syndrome (SIDS) [4,11-15].

Exact incidence of laryngospasm in LPR is uncertain, but it is an important symptom causing severe distress and may endanger life of the sufferer. It is part of the symptom complex occurring in LPR, which is usually diagnosed by endoscopic laryngeal examination and double probe pH monitoring. Although LPR associated laryngospasm has been reported [2,6] but laryngospasm in association with gastric volvulus and especially as the presenting symptom, has not been described in the literature before.

Gastric volvulus is as an abnormal rotation of the stomach of more than 180°, creating a closed loop obstruction that can result in incarceration and strangulation. Depending on the degree of twisting and the rapidity of onset, the volvulus may be acute, presenting as an abdominal emergency or it may be chronic and intermittent. Many cases of chronic volvulus are not diagnosed, as patients typically present with intermittent epigastric pain and abdominal fullness following meals. Hence patients are often investigated for other common disease entities such as cholelithiasis or peptic ulcer disease and chronic gastric volvulus is discovered incidentally. The chronic gastric volvulus may be diagnosed by gastric endoscopy and barium swallow, but barium swallow may be preferred as it is non-invasive and more accurate in demonstrating the degree of rotation and obstruction [16].

Our patient had chronic gastric volvulus, without any abdominal complaints. Instead he presented with cough and breathing difficulty secondary to reflux of gastric acid contents in the larynx. Barium swallow was performed to assess the presence and extent of gastric reflux, discovery of gastric volvulus was incidental. The barium swallow showed the stomach to have volvulated into two compartments. The distal part was emptying its contents effectively into the duodenum; however the proximal compartment was unable to empty distally due to the volvulus; instead it was emptying into the esophagus resulting in severe gastric reflux. Probably initially the volvulus was intermittent and or incomplete hence the symptoms in the beginning were occasional and less severe. But with time the volvulus probably progressed, becoming more complete and persistent, resulting in symptoms becoming worse. Probably, the proximal segment of the volvulated stomach would gradually distend due to food and secretions. Eventually forceful contraction of this distended proximal stomach would forcefully empty into the esophagus. This sudden reflux of large amount of gastric contents would rush into the pharynx and larynx. This resulted in episodic attacks of coughing, choking, gasping and apnoea. The episodes of laryngospasm and apnoea became more severe and frequent as the amount of refluxate increased. Usually coughing is able to clear small amount of acid secretions reaching the larynx as in LPR. But in our patient, the acidic gastric contents refluxed into the larynx were sudden and in large amount. These could not be cleared by coughing and resulted in laryngospasm and also central apnoea leading to patient collapse. This was also the reason for the failure of conservative medical treatment. However in LPR, acid contents are refluxed slowly and in small amounts, hence proton pump inhibitors in higher doses are quite effective. This created a life threatening situation requiring surgery to save the life of the patient.

Such a presentation of chronic gastric volvulus has not been described before in literature.

## Figures



**Figure 1:** Showing Pre-operative Gastrograffin study consistent with Volvulus



**Figure 2:** Postoperative Gastrograffin study showing normal study

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