Boerhaave Syndrome: an unusual complication in a case of Retropharyngeal Abscess.

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ABSTRACT

Introduction: Boerhaave's syndrome is a rare entity and it results from increased intraluminal esophageal pressure following sudden forceful vomiting. The Most common presentation of BS is sudden onset Retrosternal and/or epigastric pain following vomiting, and CT scan is the investigation of choice. The most common site of perforation is in the posterolateral aspect of the distal esophagus, and surgical intervention is the gold standard approach for treating BS. Case detail: A 27-years old male presented with dysphagia and throat pain. 6 hours after the admission, the patient had multiple episodes

of vomiting. Videolaryngoscopy revealed posterior pharyngeal wall ulcer. CT scan of neck shows features suggestive of rupture retropharyngeal abscess. After 24 hours patient developed respiratory distress and chest X-ray revealed right side pleural effusion. CT scan confirmed Boerhaave's syndrome and the patient was managed conservatively. Discussion: if diagnosed within 24hours surgery carries a 90% success rate but the main concern is the diagnosis and management of late presented cases. Conservative management and adequate drainage of the pleural cavity is the mainstay of treatment in such cases. Conclusion: Due to its non-specific presentation BS is difficult to diagnose. In our case, there was a little delay and due to the patient's poor general condition, we managed with conservative treatment.

Keywords: Boerhaave syndrome, Retropharyngeal Abscess, primary repair, Endoscopic therapy, conservative treatment.

1. INTRODUCTION

Spontaneous rupture of the esophagus or Boerhaave syndrome (BS) is a rare and fatal entity [1]. Sudden forceful vomiting leads to an increase in intraluminal esophageal pressure resulting in transmural tear [2]. BS classically presents with sudden onset of severe retrosternal and/or epigastric pain following vomiting [3]. The most common site of tear is in the posterolateral aspect of the distal esophagus [4]. Surgical intervention is the golden standard treatment irrespective of the duration of the perforation [5].

Here we present a case of Boerhaave syndrome developed in a patient with a retropharyngeal abscess.

2. CASE REPORT:

A 27-year-old male presented with complaints of throat pain, dysphagia for 1 week. It was associated with a productive cough and fever. The severity of Pain and dysphagia increased persistently. On the initial assessment, the patient had tachycardia, a temperature of 101˚F, poor general condition with oral candidiasis. After admission, the patient had a spontaneous onset of several episodes of vomiting. There was no history of overindulgence of food and alcohol consumption. The patient was kept nil per oral along with other conservative measures. Videolaryngoscopy (VL) reveals a large ulcer on the posterior aspect of the pharyngeal wall with slough (figure 1 a). Computer Tomography (CT) scan Neck was advised which showed features suggestive of a ruptured retropharyngeal abscess (figure 2). After 24 hours the patient developed chest discomfort and breathing difficulty. On examination, air entry was reduced on the right basal area along with bilateral crepitus on auscultation. Chest X-Ray showed right pleural effusion. Pleural fluid was turbid, and the analysis was negative for Acid Fast Bacilli (AFB). A chest tube was placed accordingly and after stabilization CT scan of the thorax was done based on the nature of draining fluid from the intercostals drainage tube and a high level of suspicion. The fluid was turbid with thick sediments. CT scan showed a contrast leak and the diagnosis of Boerhaave syndrome was established. Due to delayed diagnosis and poor general condition we followed conservative management. Repeat VL shows a large ulcer in the posterior pharyngeal wall with exposed paravertebral muscles. Feeding jejunostomy (FJ) was done under local anesthesia and subsequently feeding was started along with oral care and other conservative management. The patient was discharged with proper explanation of chest tube care and chest

physiotherapy and advised to review after 1 month. The patient came for review after one and half month and repeat CT scan shows no evidence of contrast leak and healing pharyngeal wall ulcer with vocal cord paresis on VL. The oral trial feed was started and the chest tube was removed after confirming no leak. Posterior pharyngeal wall ulcer was healed and vocal cord paresis was improving on subsequent follow up after 3 months (Figure 1 b). 6 months follow up shows no vocal cord paresis and healed pharyngeal wall. The patient was tolerating orally both solid and liquid diet and his general condition also improved with no voice change.

3. DISCUSSION

Dutch admiral BJV Wassenaer died after self-induced vomiting during a feast. According to the autopsy, there was a full thickness rent in the lower esophagus with an odor of the roast duck he had during the feast. Spontaneous rupture of the esophagus is termed as Boerhaave syndrome after Dutch Physician Hermann Boerhaave who first described this condition in 1724 [2].

BS is a rare condition and accounts for 10% of all oesophageal perforation. It is commonly seen in males between 50 – 70 years [3]. The most common site of perforation is in the left posterolateral aspect of the distal esophagus because of the anatomical weakness due to different muscle fiber orientation at the gastro-oesophageal junction region [2].

BS most often develops during or after intense vomiting caused by excessive eating or drinking alcohol. Sudden forceful vomiting leads to an increase in the intraluminal oesophageal pressure resulting in the tear [2]. Rarely BS can occur in the absence of vomiting and seen in a condition where a muscular layer is absent, during sleep, or in patients with Gastroesophageal reflux disease, Barrett's esophagus, peptic stricture of the esophagus, esophagus dysmotility, paraesophageal hernia and bleeding duodenal ulcer [6]. In our case repeated episodes of vomiting due to unknown reasons along with proximal

blockage from the retropharyngeal abscess must have increased the intraoesophageal pressure and subsequently resulted in an esophageal tear.

BS has a variable and non-specific presentation [2, 3]. Most common presentation seen in BS are sudden onset Retrosternal and/or epigastric pain following vomiting whereas Mackler's triad (chest pain, vomiting, subcutaneous emphysema) or Anderson triad (subcutaneous emphysema, tachypnoea, abdominal rigidity) and other symptoms and sign associated with BS are seldom seen [3]. Non-specific symptoms in BS mimic clinical conditions like myocardial infarction, pulmonary embolism, dissecting aorta, perforated peptic ulcer, pancreatitis pneumonia etc resulting in a delay in diagnosis [4].

A high index of suspicion is crucial for the timely diagnosis of BS. Chest X-ray (CXR) should be the scout investigation for a suspected case of BS. In the case of early presentation, CXR can be normal (15%). CXR findings of BS are subcutaneous and/or mediastinal emphysema, mediastinal widening, pleural effusion (specifically rapidly developing or evolving effusion), pneumothorax, hydrothorax, intrathoracic air-fluid levels or masses or V-sign (radiolucent streak of air dissecting the retrocardiac facial planes [4]. Few authors recommend upper endoscopy (sensitivity and specificity of 100 % and 83 % respectively) to confirm BS but as it can increase the size of the tear and deteriorate the pneumomediastinum its use is limited [7, 8]. Though oesophagography is considered as the gold standard for the diagnosis of BS it still has a false-negative rate of up to 10-25% % which can be attributed due to tissue edema or muscular spasm and because of this reason, it is now replaced by CT scan [3, 7, 8]. CT scan can detect the site of the perforation and the surrounding inflammatory process (e. g., mediastinitis) [6]. Additional features in CT scans are extraluminal air, periesophageal fluid, oesophageal thickening and extraluminal contrast [2].

The current management of BS includes.

1. Conservative, and
2. Surgical treatments.

3. Endoscopic therapy.

The survival rates for each treatment are 75%, 81% and 100%, respectively [9].

1. The conservative approach is suitable in patients with well-contained perforation without the signs of sepsis. It includes cessation of the oral intake, administrations of intravenous fluids and parenteral nutrition or feeding on a nasogastric tube or feeding jejunostomy, broad-spectrum antibiotics [4, 5].

2. Surgical intervention retains a dominant role in the management of BS. Surgical management can be performed by either open or by a minimally invasive approach. The minimally invasive approach is suitable in early presentation with stable hemodynamics and in the absence of septic complications. Surgical options are ranging from debridement and drainage of the mediastinal and pleural cavity to resection of the esophagus. The minimally invasive approach is always considered better especially in critically ill patients however open repair and drainage are still considered as the gold standard treatment [4, 5].

After reviewing the literature we can divide the treatment approach in two ways, based on presentation:

1. BS diagnosed more than 24 hours: The primary repair of an esophageal perforation remains the gold standard of therapy, with a 94.7 % survival rate, provided the treatment is performed within 24 hours in the absence of esophageal diseases [7].
2. BS diagnosed late (less than 24 hours): The dispute regarding treatment (conservative of surgery) comes when we encounter patients with BS presented or diagnosed late. In a case series of 21 patients, Han D et al have reported a leak in all patients (with delayed diagnosis or presentation after 72 hours) who underwent surgical procedures [8]. The factors contributing to postoperative leakage are friable stiff and oedematous edges [5, 10]. In late diagnosed cases due to severe inflammation, friable tissues, necrosis, and infection it is advisable

to use possible alternative modalities such as conservative treatment, stenting, and drainage instead of repairing the rent [11].

Han D et al suggested some technical points for the better surgical outcome:

1. Extend the rupture of the muscle layer to expose the entire length of the mucosal rupture and remove the necrotic muscle tissue,
2. Adequate needle pitch and margin and not to tie the knots very tightly to avoid cut through in the tissue, and
3. Vascular tissue flaps (Omentum) reinforcement [8].

In 1995, the stent was used for the first time in the management of spontaneous rupture of the esophagus [7]. Stents bridges over the rent and helps to seal the oesophageal leaks and prevent fistula formations in patients with delayed diagnosis and in patients who are diagnosed early and without widespread contamination [9, 10]. In Endoscopic therapy, the rent can be seal by placing the self-expanding metallic stents (SEMS) over the tear or over the scope clipping using clips or sutures [9, 12].

Endoscopic therapy can be useful in selected patients without evidence of systemic sepsis. According to Dickinson KJ et, al endoscopic therapy can be subjected in selected patients with a minimal leak and without any systemic sepsis. They suggested:

a) a) Closure through the scope or over the scope: in early diagnosed cases with a small defect of ≤1 cm.
b) b) Primary endoluminal closure with stent placement and drainage: in cases with defect ≥ 1 cm.
c) c) Esophageal diversion or exclusion in late diagnosed cases, defect involves more than 50% of the esophagus or inadequate drainage, and

Video-assisted thoracotomy (VAT) or open thoracotomy adjunct drainage if there is a massive or persistent leak [12].

The main drawbacks and adverse effects of stent placement are the migration of the stent, pressure-induced ischemia, ulceration, and perforation; development of new reactive stenosis at the ends of the endoprosthesis; bleeding or injury upon removal; an unsuccessful retrieval of the device at a later date [7]. Though it is a promising newer modality Shen G et all reported that it offers no advantage over surgical management and is associated with frequent treatment failure that eventually requires surgical intervention [5]. On the other hand, another report stated that stents placement avoid surgery in 60% of patients and displayed 100% clinical success [9].

Most Pharyngeal perforation can be managed conservatively if it is confined and uncomplicated [13]. In our case also we managed the patient conservatively which consist of adequate nutrition, oral and wound care, proper antibiotic, Proton Pump Inhibitor (PPI) and chest physiotherapy.

4. CONCLUSION

Conclusion: Diagnosis of Boerhaave syndrome is very difficult if there is no high index of suspicion. There are no issues with the management if diagnosed early but the problem arises when it is diagnosed late. In our case, we don't know what triggered the vomiting, but it was the abscess collection which might have obstructed and increased the intra-oesophageal pressure leading to Boerhaave syndrome. A high level of suspicion leads us to the diagnosis followed by successful conservative management without any complications.

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COMPETING INTERESTS

NIL

AUTHORS' CONTRIBUTIONS

The Author is responsible for the protocol writing, literature searches and drafting and final approval of the manuscript.

CONSENT (WHERE EVER APPLICABLE)

N/A

ETHICAL APPROVAL (WHERE EVER APPLICABLE)

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APPENDIX

FIGURES:

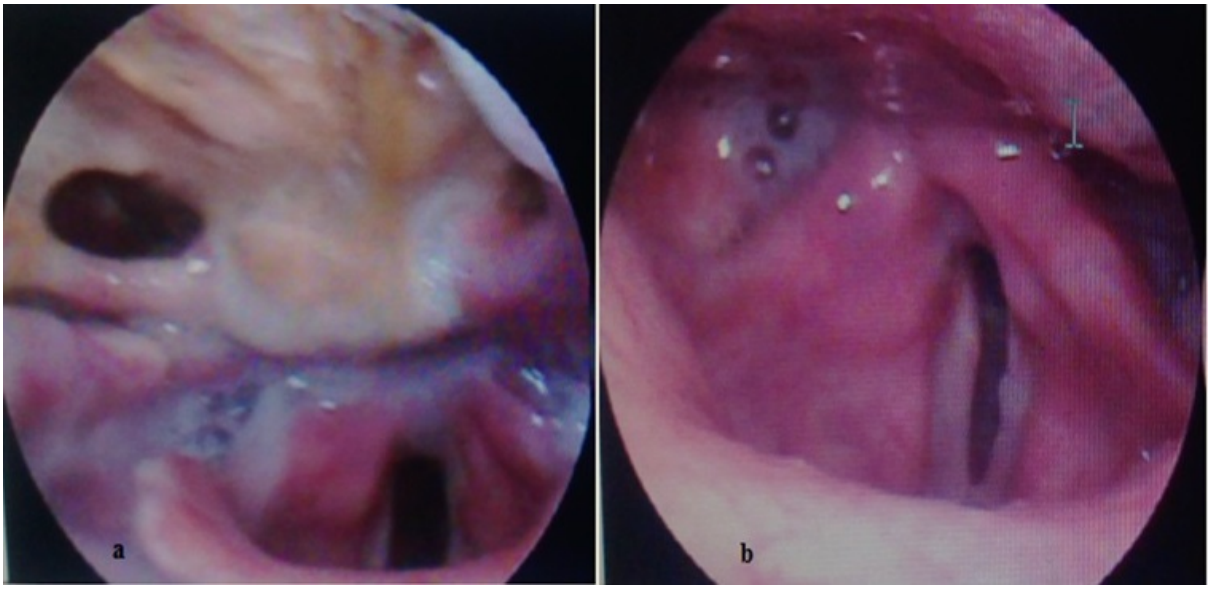

Fig 1 Video laryngoscopy (VL) picture showing rent in the posterior aspect of the pharynx (a) and repeat VL after 3 months (b) showing a healed posterior pharyngeal wall.

Fig 2 CT scan of neck showing rent in the right posterior aspect of the pharynx (white arrow).

Fig 3 CT scan thorax showing contrast leak (Black arrow)

LEGEND:

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