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2 **Boerhaave Syndrome: an unusual complication**
3 **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.

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

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21 **1. INTRODUCTION**

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

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

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33 2. CASE REPORT:

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

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

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65 **3. DISCUSSION**

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

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

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

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

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

92 A high index of suspicion is crucial for the timely diagnosis of BS. Chest X-ray (CXR) should
93 be the scout investigation for a suspected case of BS. In the case of early presentation, CXR
94 can be normal (15%). CXR findings of BS are subcutaneous and/or mediastinal
95 emphysema, mediastinal widening, pleural effusion (specifically rapidly developing or
96 evolving effusion), pneumothorax, hydrothorax, intrathoracic air-fluid levels or masses or V-
97 sign (radiolucent streak of air dissecting the retrocardiac facial planes [4]. Few authors
98 recommend upper endoscopy (sensitivity and specificity of 100 % and 83 % respectively) to
99 confirm BS but as it can increase the size of the tear and deteriorate the
100 pneumomediastinum its use is limited [7, 8]. Though oesophagography is considered as the
101 gold standard for the diagnosis of BS it still has a false-negative rate of up to 10-25% %
102 which can be attributed due to tissue edema or muscular spasm and because of this reason,
103 it is now replaced by CT scan [3, 7, 8]. CT scan can detect the site of the perforation and the
104 surrounding inflammatory process (e. g., mediastinitis) [6]. Additional features in CT scans
105 are extraluminal air, periesophageal fluid, oesophageal thickening and extraluminal contrast
106 [2].

107 The current management of BS includes.

- 108 1. Conservative, and
- 109 2. Surgical treatments.

110 3. Endoscopic therapy.

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

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

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

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

125 1. BS diagnosed more than 24 hours: The primary repair of an esophageal
126 perforation remains the gold standard of therapy, with a 94.7 % survival rate,
127 provided the treatment is performed within 24 hours in the absence of esophageal
128 diseases [7].

129 2. BS diagnosed late (less than 24 hours): The dispute regarding treatment
130 (conservative or surgery) comes when we encounter patients with BS presented
131 or diagnosed late. In a case series of 21 patients, Han D et al have reported a
132 leak in all patients (with delayed diagnosis or presentation after 72 hours) who
133 underwent surgical procedures [8]. The factors contributing to postoperative
134 leakage are friable stiff and oedematous edges [5, 10]. In late diagnosed cases
135 due to severe inflammation, friable tissues, necrosis, and infection it is advisable

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

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

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

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

150 Endoscopic therapy can be useful in selected patients without evidence of systemic sepsis.

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

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

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

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

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

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175 **4. CONCLUSION**

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

183

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188

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190

191 NIL

192 **AUTHORS' CONTRIBUTIONS**

193

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

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197 **CONSENT (WHERE EVER APPLICABLE)**

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200 N/A

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202 **ETHICAL APPROVAL (WHERE EVER APPLICABLE)**

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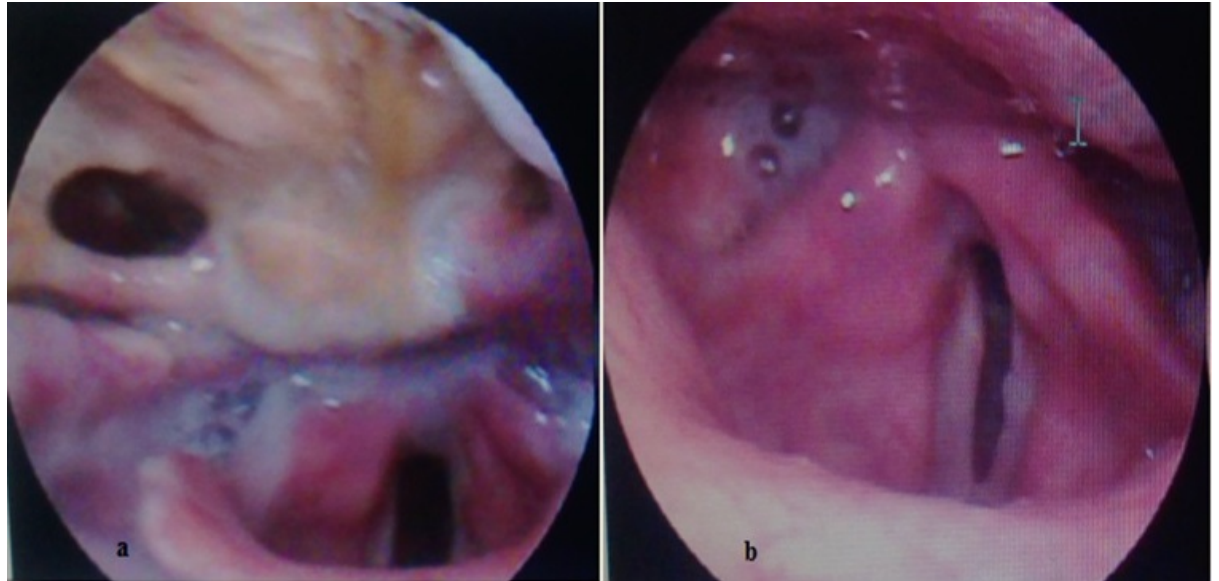
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263 **APPENDIX**

264

265 FIGURES:



266

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

270



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:

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).