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Case Report

Eventration of diaphragm with a rare association

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Abstract

Eventration of the diaphragm is a rare anomaly of unknown origin characterized by a permanent high position of one or rarely both the leaflets of the diaphragm, providing a potential space for the displacement of abdominal viscera on the affected side(s). The etiology, diagnosis, and management of this condition remains a controversial subject. We report a rare case of infiltrating squamous cell carcinoma of the esophagus in a patient with eventration of the diaphragm and gastric volvulus who presented to us with retrosternal discomfort. To the best of our knowledge, this rare association has not been reported in the literature, although one case of esophageal adenocarcinoma arising from Barrett's esophagus in association with eventration of the diaphragm has been reported previously. This case again emphasis the varied associations of an eventration of the diaphragm.

Key words

Eventration of diaphragm, gastric volvulus, infiltrating squamous cell carcinoma

Introduction

Eventration of diaphragm is a comparatively rare condition occurring in about 0.001-0.003% of live births. Incidence in adults is about 1 in 10,000. It is defined as a condition in which the left or the right leaf of the diaphragm has ascended abnormally high into the chest, and in rare cases both leaves of the diaphragm may be elevated.[1] The unbroken continuity differentiates eventration from diaphragmatic hernia. To the best of our knowledge, no case of primary esophageal squamous cell carcinoma and gastric volvulus associated with eventration of the diaphragm has been reported in the literature.

Case Report

A 53-year-old male, a daily wage worker, presented with complaints of postprandial retrosternal pain and discomfort associated with loss of appetite and loss of weight for one month. He also complained of cough with purulent sputum for the same duration. There was no history of fever, nausea, vomiting, hematemesis, or melena. His past medical and surgical history were noncontributory. Social history was significant in that he was an alcoholic and smoker for more than 20 years. On examination, his vital signs were stable. Examinations of heart and abdomen were normal. Chest revealed decreased breath sounds on the left base with fine basilar crackles. There was no palpable lymphadenopathy. Laboratory studies including complete hemogram and routine biochemistry were normal. Chest X-ray showed elevation of the left dome of diaphragm [Figure 1]. Ultrasonography abdomen and pelvis was normal.

Esophagogastroduodenoscopy revealed a growth in the distal one-third of the esophagus [Figure 2]. The endoscope could be negotiated beyond this esophageal growth, but it repeatedly coiled in the proximal stomach and could not be passed beyond. We decided to abandon the procedure and go ahead with barium series of upper gastrointestinal (GI) tract to visualize any pathology beyond the proximal stomach. On barium swallow, there was a void in the distal esophagus, and it also revealed an organo-axial type of gastric volvulus. Esophageal biopsies obtained at upper GI endoscopy revealed an infiltrating squamous cell carcinoma [Figure 3].

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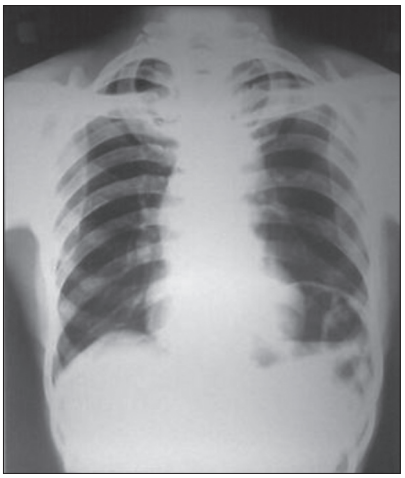

Figure 1: Chest X-ray showing eventration of diaphragm on the left side

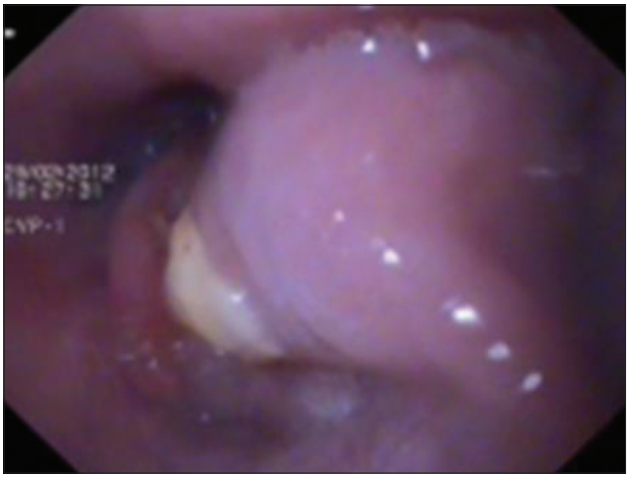

Figure 2: Upper endoscopy showing the mass in the distal esophagus

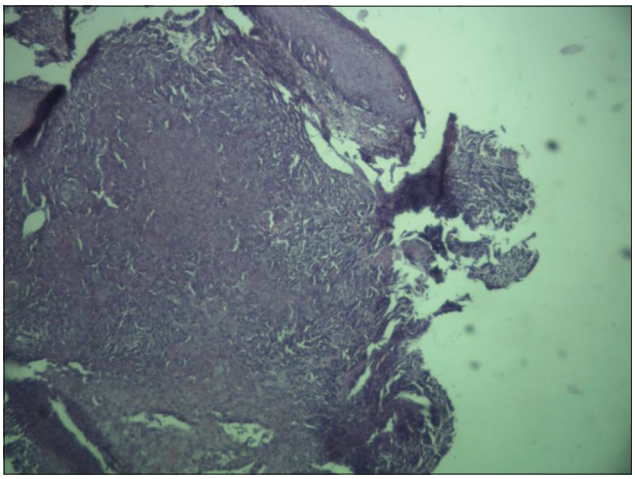

Figure 3: Histopathology of the distal esophageal mass revealing infiltrating squamous cells carcinoma

Discussion

Eventration of the diaphragm can be either congenital or nonparalytic and acquired or paralytic type. It may present at birth as a congenital condition due to a defect of the diaphragmatic development or in a later stage of life as an acquired condition ("acquired diaphragmatic paralysis" or "acquired diaphragmatic elevation"). It occurs almost exclusively on the left side, a point that may be of value in differentiating this from diaphragmatic paralysis.

Gastric volvulus is a condition where the stomach rotates more than 180 degrees creating a closed loop obstruction. It was first described by Berti in 1866 in a female autopsied patient, and the first operation was performed by Berg in 1897.[2,3] The reported frequency of gastric volvulus during barium X-ray studies is 0.15%. The stomach is relatively fixed at the esophageal hiatus and pylorus and is prevented from abnormal rotation by the gastric ligaments. Absence or weakness of these anatomic anchors results in abnormal mobility of the stomach within a wide subdiaphragmatic space created from eventration of the diaphragm and resulting in gastric volvulus. Most cases of gastric volvulus occur in association with eventration of left hemidiaphragm or a hiatus hernia. Organo-axial rotation is the most common type accounting for greater than 60% of the cases, followed by mesenteroaxial. A combined organo-axial and mesenteroaxial rotation is the least common type. Primary gastric volvulus, making up one-third of cases, occurs when the stabilizing ligaments are too lax as a result of congenital or acquired causes. Secondary gastric volvulus occurs in association with a paraesophageal hernia or other congenital or acquired diaphragmatic defects.[4]

Patients with diaphragmatic eventration are usually asymptomatic; however, some complain of dyspnea on exertion and orthopnea, due to a decrease in ventilation and oxygenation because of paradoxical motion of the affected diaphragm during inspiration and expiration. GI symptoms of diaphragmatic eventration may include nausea, heartburn, early satiety, postprandial vomiting, and epigastric discomfort. The classic triad, described by Borchardt in 1904, consisting of retching, severe and constant epigastric pain, and difficulty in passing a nasogastric tube is believed to be diagnostic for acute gastric volvulus.[5] Carter *et al*. suggested three additional findings that may be very suggestive of gastric volvulus: Minimal abdominal findings when the stomach is in the thorax; a gas-filled viscus in the lower chest or upper abdomen on chest radiograph, especially when associated with a paraesophageal hernia; and obstruction at the site of the volvulus shown by upper GI series.

In our patient, esophageal squamous cell carcinoma in addition to gastric volvulus with diaphragmatic eventration was present. To the best of our knowledge, no previous report of this association has been reported, although. Shimoji *et al*. have reported Barrett's carcinoma and eventration of diaphragm;[6] however, volvulus of the stomach was not present in their patient.

Conclusion

To the best of our knowledge, no case of primary esophaphageal adenocarcinoma associated with acquired eventration of diaphgram and volvulus of stomach has previously been reported in literature.

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