CASE REPORT



Circumferential intramural esophageal dissection with large mucosal defect

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Abstract

Intramural esophageal dissection (IED) is an uncommon entity characterized by mucosal rupture creating a false lumen. It usually develops following endoscopy or in old patients with bleeding tendencies and spontaneous presentation possesses diagnostic dilemma. Managing partial IED is simpler than circumferential IED. Due to rarity of presentation, management is not standardized. We report circumferential IED in a young patient and challenges faced during diagnosis and management.

Keywords Intramural esophageal dissection · Esophagectomy · Stent placement

Introduction

Intramural Esophageal Dissection (IED) is an uncommon disorder characterized by a lengthy laceration deeper to the mucosal layer of the esophageal wall, without complete perforation. This condition is an intermediate form of esophageal injury between superficial Mallory–Weiss tear and full thickness Boerhaave's syndrome. While the latter conditions occur following violent bouts of vomiting, IED often occurs spontaneously in people with underlying bleeding diathesis or iatrogenically following endoscopy [1, 2].

Majority of cases respond well to conservative management [1]. Interventions include endoscopy and surgery and are rarely required in partial IED. However, management of circumferential IED is complicated requiring long-term endotherapy and at times surgery [3]. This is report of a young male with circumferential IED with large mucosal defect managed successfully with esophagectomy.

Short report of case

A 40-year-old diabetic male following a bout of vomiting developed sudden onset chest pain, dysphagia and nasal regurgitation of food for 4-week duration. Initial evaluation elsewhere at presentation with barium swallow and

Santhosh Anand kssa.5149@gmail.com endoscopy revealed tight stricture causing complete obstruction at Gastro Esophageal Junction (GEJ) level (Fig. 1a). He lost 7 kg during this period. He was managed with intravenous fluids and proton pump inhibitors during this period. Patient also had alcohol-induced chronic calcific pancreatitis. He had earlier undergone right hemicolectomy for ileocecal tuberculosis-induced intestinal obstruction 10 years back and open mesh hernioplasty for incisional hernia 2 years later.

Contrast-Enhanced Computed Tomography (CECT) showed normal GEJ and stomach. There was "double barrel" appearance [4] of esophagus suggesting IED and no pneumomediastinum or pleural effusion (Fig. 1b–d). Endoscopy was done which revealed a circumferential IED starting 18 cm from incisor with a long mucosal defect for 8 cm and a thin bridge of mucosa connecting them. The dissection is circumferential as even the mucosal bridge connecting proximal and distal tubes was not adherent to surrounding muscular tube. The distal mucosal tube below remained collapsed and endoscope was predominantly entering the cul de sac ending blindly near GEJ (Fig. 2).

In view of very long mucosal defect and dissection starting from cervical esophagus, endoscopic management would have been difficult and outcome was uncertain. He underwent Trans Hiatal Esophagectomy (THE) and gastric pull through. He had transient left vocal cord palsy with complete recovery of voice in 3 weeks. He started tolerating oral feed from day 7 following surgery. Histopathology of esophagus showed nonspecific inflammation and possibilities of described inciting factors like eosinophilic esophagitis and

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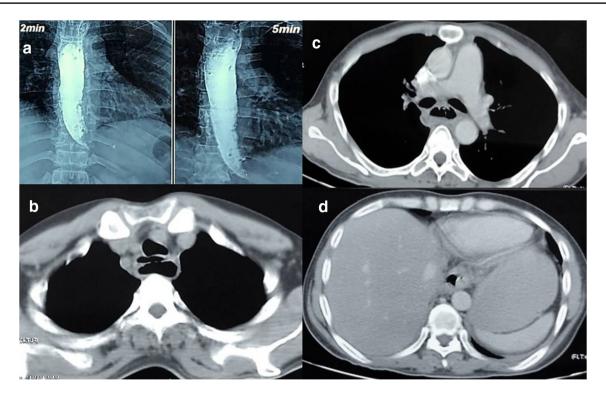


Fig. 1 (a Barium swallow picture showing entry of contrast into cul de sac and retention at GEJ. b, c, d) Contrast-enhanced computed tomogram from upper esophagus to GEJ level showing double barrel appearance consistent with IED



Fig.2 Endoscopic picture at mid esophagus level. Thin mucosal bridge (*) connecting proximal and distal mucosal tube (solid arrow). Note the large mucosal defect with cul de sac surrounding the mucosal tube

mycosis were ruled out. Patient had gained 5 kg of weight 6 months following surgery.

Discussion

IED was initially reported as intra-mural esophageal hematoma with submucosal bleed dissecting the esophagus and mucosal break preceding or following it. This occurred spontaneously in elderly patients on anti-coagulation. This can also occur due to pressure changes in esophagus like retching, vomiting or uncoordinated swallowing, as in our case report. Blunt trauma caused by endoscopic interventions, nasogastric tube insertion and ingestion of harsh food also has been reported to cause esophageal dissection [5].

Majority of case reports have described partial IED, which respond to conservative or simple endoscopic management. Very few cases of circumferential IED are available in literature. Conservative management is usually not successful in latter. Our patient had circumferential IED with long mucosal defect, hence necessitating endotherapy or surgery [6, 7].

Endoscopic interventions like adhesiolysis, distal mucosal incision, bouginage and stent placement have adjunctive roles during resolution of symptoms. Fischer et al. reported a case of eosinophilic esophagitis developing circumferential IED starting 25 cm from incisor managed successfully with early deployment of partially covered self-expandable metal stent. Two weeks later, stent was removed and patient was symptom free on long-term budesonide therapy [8]. Endotherapy success is not uniform and at times has protracted and complicated course. Kim et al. treated a case of circumferential IED for a period of 16 months before resolution of dysphagia with multiple endoscopic interventions including mucosal incision, endoscopic dilatation, permanent stent placement and repeated dilatations. Three Endoscopic interventions may at times fail or result in complications like perforation or esophagopleural fistula. In such cases, an additional esophagectomy may be required [7].

Our patient had an option of endoscopic stent placement. However, the dissection started at 18 cm from incisor with a 7-cm-long mucosal defect and long cul de sac till GEJ. He would have required very long stent with proximal end near cricopharynx and had extensive scarring at mucosal defect level, making its removal difficult. In such situation, a chance of endotherapy failure was high. Patient was young and was not having any bleeding tendencies. Hence, esophagectomy was done, providing a definitive treatment with early resolution of oral feeding.

Conclusion

This was a report of extensive circumferential IED in a young patient. Surgery in such cases would provide a single time prompt resolution of symptoms outweighing perioperative morbidity and expected outcome of endotherapy.

Compliance with ethical standards

Conflict of interest Dr. Santhosh Anand declares that he has no conflict of interest.

Human rights All procedures followed have been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments.

Informed consent Informed consent was obtained from all patients for being included in the study.

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