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Case report

Life-threatening bleeding from dissecting Intramural Hematoma of Esophagus (IHE) treated by trans arterial embolization.

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ABSTRACT

Dissecting intramural hematoma of esophagus (DIHE) is an uncommon entity, characterized by accumulation of blood within the esophageal wall and usually managed conservatively. Only in rare circumstances, DIHE is associated with massive life-threatening hemorrhage requiring emergency treatment. We present a case of DIHE associated with cardiovascular collapse and treated by transcatheter arterial embolization. Transcatheter arterial embolization is a rare treatment option for DIHE associated with hemodynamic instability and only a handful of cases have been reported in the literature.

Introduction

Dissecting intramural hematoma of esophagus (DIHE) is considered intermediate form of the spectrum of esophageal wall injury. The most common clinical symptoms are retrosternal pain, dysphagia and minor hematemesis [1]. Therefore, it should be considered among differential diagnosis of acute chest pain and acute upper GIT bleeding in proper clinical scenario. Massive GIT bleeding secondary to DIHE is rare and may lead to critical consequences. In this case report we highlight the rule of transcatheter arterial embolization (TAE) as a treatment option in these cases. To the best of our knowledge, only a couple of TAE for DIHE were reported in the literature [2,3].

Case presentation

Our case is an elderly gentleman who developed a cardiovascular collapse following several episodes of hematemesis. His medical history was remarkable with diabetes mellitus, ischemic heart disease and peripheral vascular disease.

Cardiopulmonary resuscitation efforts were instituted immediately with defibrillation attempts and inotropic support. After 20 minutes of CPR, his hemodynamic circulation was re-established. He was admitted to the intensive care unit where he was put on the ventilator. Endoscopic examination revealed marked luminal narrowing along the esophagus caused by diffuse submucosal swelling with a bluish discol-

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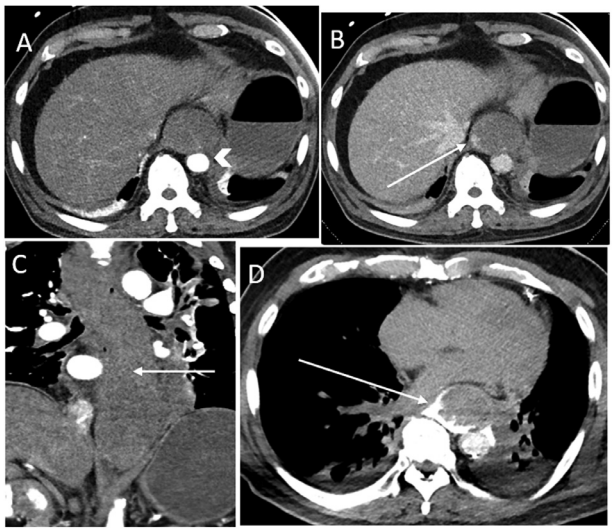

Fig. 1 – Axial (A) arterial and (B) portal venous (PV) phases of contrast enhanced CT reveals a linear streak of contrast in the distal esophageal wall on the arterial phase (arrow head) with subsequent pooling of contrast in the PV phase (arrow). Coronal reformatted image (C) shows a diffusely expanded esophagus by intramural hematoma and almost complete obliteration of the esophageal lumen (arrow).

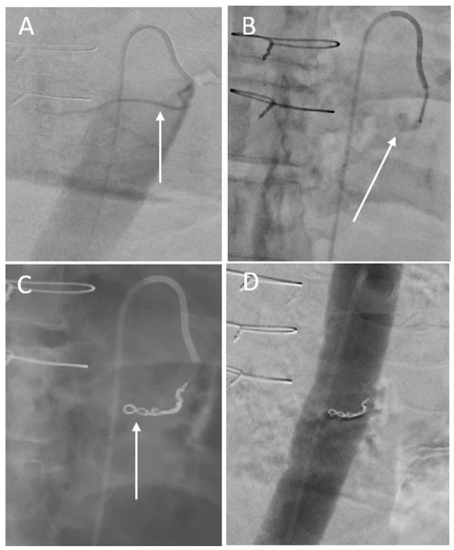

Fig. 2 – Trans-arterial access with superselective catherization (A) of the distal esophageal arterial branch arising from the thoracic aorta (arrow) using a 4-F Shepard Hook catheter followed by 2.2-F Progreat microcatheter. Injection of contrast (B) reveals contrast extravasation from the arterial branch indicating the site of active hemorrhage (arrow). (C) The artery was successfully embolized with PVA particles and several figure of 8 micro coils (arrow). Post-embolization completion angiogram (D) shows cessation of active hemorrhage.

oration of the mucosa without any evidence of perforation. The findings suggest a submucosal hematoma. Computed tomographic (CT) aortography revealed a diffusely swollen esophagus with a speck of active contrast extravasation in the distal esophageal wall on the arterial phase and further accumulation of contrast on the delayed phase. The findings are consistent with active hemorrhage secondary to AEF. Given the continuing hemodynamic instability, the patient was brought to the interventional radiology suite.

Intra-arterial access was performed by a puncture of the right common femoral artery. Superselective catherization of the middle to distal esophageal arterial branches were performed using a 4-F Shepard Hook catheter followed by 2.2-F Progreat microcatheter. Intra-arterial CT revealed active hemorrhage into the esophageal wall and gastric lumen from a distal esophageal branch arising from the thoracic aorta. The artery was successfully embolized with 355-500-micron PVA particles and several Figure of 8 micro coils. There was satisfactory completion angiogram, and subsequent Intra-arterial CT showed cessation of active hemorrhage.

Figure 1, Figure 2

After the procedure, the patient remained ventilated and he was unresponsive to arousal. Neurological examination revealed a comatose state without any signs of meningism. EEG showed continuous generalized slowing with periodic lateralized epileptiform discharges over the left cerebral hemisphere which are consistent with severe diffuse encephalopathy. Magnetic resonance imaging (MRI) revealed hypoxic ischemic encephalopathy with multiple brain infarctions. The patient succumbed to cardiac arrest subsequently in the ICU.

Discussion

IHE usually occurs at the distal third of esophagus; this can be explained by the absence of striated muscles and the lack of supporting structures at this level. Furthermore, the distal third of the esophagus is vulnerable to laceration due to sheering mechanisms from severe vomiting or coughing. On the other hand, traumatic injuries usually affect the upper third. In patients with abnormal hemostasis, IHE can occur more proximally or at multiple sites along the esophagus.

The etiologies of IHE can be classified into the following categories [4]; (1) Idiopathic without identifiable cause; (2) Abnormal hemostasis including use of anticoagulants and/or coagulopathic disorder; (3) Esophageal injury including laceration due to sudden change in intrathoracic/intraesophageal pressure, for example, forceful vomiting or cough; (4) Direct trauma by foreign body, food induced or iatrogenic (endoscopy, feeding tube or CVC); (5) Vascular malformation including arterio-venous malformation; (6) AEF.

AEF is a rare fatal condition, secondary to esophageal pathology (eg, malignancy, penetrating ulcer or corrosive esophagitis) or aortic cause (eg, rupture aortic aneurysm, complicated aortic or esophageal surgeries) [5]. The presenting symptoms are usually chest pain and sentinel bleed followed by massive hematemesis; also, known as the Chiari's triad [5].

CT aortography is the modality of choice to diagnose DIHE and potential causes. DIHE typically appears as eccentric or

concentric hyperattenuating submucosal mass like lesion that does not enhance [1]. In AEF, there is a communicating tract to the aorta with active contrast extravasation [1]. The presence of enhancing submucosal hematoma with underlying aortic aneurysm should suggest AEF. Other suspicious features of AEF on CT include abnormal air within the mediastinum, aortic lumen and persistent or expanding peri-graft fluid (in case of prior aortic repair) [6]. Given the close proximity of the esophagus and the thoracic aorta, CT is useful to distinguish between esophageal wall disintegrity and acute aortic conditions (eg, aortic dissection, intramural hematoma and penetrating aortic ulcer) [1].

Esophagogram with water-soluble iodinated contrast (eg, gastrografin) may demonstrate communication between the esophageal lumen and the hematoma cavity [5]. This appearance, known as "double barrel sign" is considered diagnostic for IHE [5].

Endoscopy should be avoided as it may inadvertently worsen the situation through gas insufflation with mechanical transmucosal perforation [7]. Endoscopic ultrasound carries similar invasive risks. Being superior to endoscopy, endoscopic ultrasound is able to demonstrate submucosal lesions besides evaluating adjacent mediastinal structures and exclude any fistulous communication [8]. MRI could be valuable to differentiate IHE from aortic dissection when CT is inconclusive or contraindicated. Additionally, MRI may show clear soft tissue planes around the aorta and esophagus as well as intramural hematoma of intermediate signal intensities on T1 and T2 weighted images [9].

DIHE should be considered in differential diagnosis of acute chest pain. It may be indistinguishable clinically from acute coronary or aortic conditions [10]. CT may help to detect DIHE or acute aortic syndrome spectrum. Needless to say, the more common critical conditions (acute coronary) should be excluded first by electrocardiogram and cardiac enzymes tests.

Emetogenic induced esophageal injuries spectrum includes Mallory-Weiss mucosal tear (mild), DIHE (intermediate) and transmural perforation (severe) which is known as Boerhaave syndrome (BS). The clinical picture may be identical in the whole spectrum [11] with chest pain, dysphagia and odynophagia that started after forceful vomiting or cough. Radiologically, presence of signs of pneumomediastinum or mediastinal contrast leak (in conventional or CT esophagogram) should suggest BS. Plain chest radiograph is abnormal in over 90% of BS cases [12], with signs of pneumomediastinum (eg, V sign of Naclerio), left side pneumothorax and left pleural effusion. CT has higher sensitivity to detect pneumomediastinum and perforation. Mallory-Weiss tear is usually radiographically unidentifiable. However, CT may show intraluminal haemorrhage or minimal extraluminal gas adjacent to the site of the tear [8].

On cross sectional imaging, all causes of symmetric and asymmetric esophageal wall thickening are among the differential diagnosis of DIHE. This includes esophagitis, diffuse esophageal spasm, foregut duplication cyst, leiomyoma and other esophageal tumours. Differentiation of DIHE can be made by its imaging characteristics. DIHE appears as hyper-attenuating & non enhancing well defined submucosal lesion.

IHE is usually managed conservatively with reversal of anticoagulation, parenteral nutrition and supportive treatment [7]. The only type of IHE that cannot be managed conservatively is cases resulting in hemodynamic collapse or those secondary to AEF [7]. If not recognized and treated promptly, AEF almost always results in death [6]. Treatment methods include endovascular aortic stent-graft with esophageal stent, sternotomy or left thoracotomy [6].

TAE is an extremely rare treatment option for DIHE associated with hemodynamic instability and only a handful of cases have been reported in the literature [2,3]. According to some authors, TAE can be considered as a treatment method for endoscopically unmanageable upper GI bleeding. In a meta-analysis, authors found that TAE is safe and effective for upper GI bleeding when compared to surgery [13].

Conclusion

- Intramural hematoma of the esophagus should be considered as a differential diagnosis of acute chest pain and upper GIT bleeding in proper clinical scenario.
- DIHE may be rarely associated with massive GIT bleeding which may lead to critical consequences and cardiovascular collapse.
- TAE can be considered as a treatment option in cases of DIHE associated with massive bleeding and cardiovascular collapse. Larger number of patients may be required to verify the safety and effectiveness of this treatment method.

Patient consent

Unfortunately, the patient is deceased and relatives are uncontactable.

We confirm that we have deleted all personal information and other patient specific data. All direct identifiers including age have been removed. The included radiological images are non-identifiable. The only indirect identifier in this manuscript is that the authors are from the same organization, otherwise the patient is anonymized.

Declaration of Competing Interest

The authors have declared that no competing interests exist.

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