Letter to the Editor

Embolization of the esophageal branch of intercostal artery for treatment of spontaneous intramural hematoma of the esophagus: a case description

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Introduction

Intramural hematoma of the esophagus (IHE), a rare clinical entity, was described for the first time by Williams in 1957 (1). It is one of the acute esophageal injuries that includes the more common Mallory-Weiss tear and Boerhaave syndrome (2,3). It is characterized by a concentric or eccentric intramural hematoma related to dissection of the esophageal wall (2). The disease is associated with factors such as a rapid increase in intraesophageal pressure, trauma, and emetogenic and aortic disease. It can also occur spontaneously without any evident inducement, a phenomenon known as spontaneous intramural hematoma of the esophagus (SIHE), which was first reported by Marks and Keet in 1968 (4-6). The most common clinical symptoms are retrosternal pain, dysphagia, and minor hematemesis (2). Differential diagnosis is crucial for correct and appropriate clinical treatment. Nevertheless, due to the lack of specificity in clinical manifestation of this disease, an accurate diagnosis of this disease is difficult, and misdiagnosis can occur (7-9). In this paper, we describe a case of spontaneous giant submucosal hematoma of the esophagus treated with transarterial embolization. To our knowledge, only a few cases of SIHE treated with transarterial embolization have been reported in the literature (10,11).

Case presentation

A 66-year-old male, with renal failure and a history of hypertension and trauma surgery (the patient underwent surgical treatment for a left lower limb tibial fracture caused by a car accident in 2014), was sent to the emergency department of Jiangyin Hospital affiliated to Nantong University with sudden persistent retrosternal pain, dysphagia, and hematemesis. After examination, lung computed tomography (CT) showed the presence of a soft tissue-like density shadow in the esophagus (*Figure 1*), and gastroscopy showed esophageal mucosal protrusion. A massive lump was observed at a distance of 22 cm from the incisor teeth, with a blue and smooth surface that occupied the entire esophageal cavity. There was resistance when the endoscope was inserted (*Figure 2*). The patient was admitted to the hospital after being diagnosed with esophageal hematoma in the emergency department. Emergency blood routine on the day of admission showed a white blood cell count of 6.28×10^9/L, neutrophil percentage of 81.30%, a lymphocyte count of 0.64×10^9/L, and a hemoglobin level of 108.00 g/L. On the morning of the second day after admission, the blood routine hemoglobin level was 82 g/L. Troponin levels and electrocardiography, myocardial zymogram, and other physical examination results were normal. The possible cause of the patient's esophageal

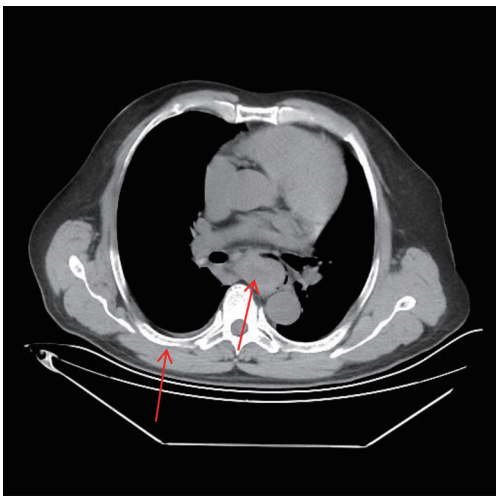

Figure 1 Lung CT showed soft tissue–like density in the esophagus (the arrow on the left indicates a small amount of pleural effusion; the arrow in the middle indicates soft tissue like-density in the esophagus). CT, computed tomography.

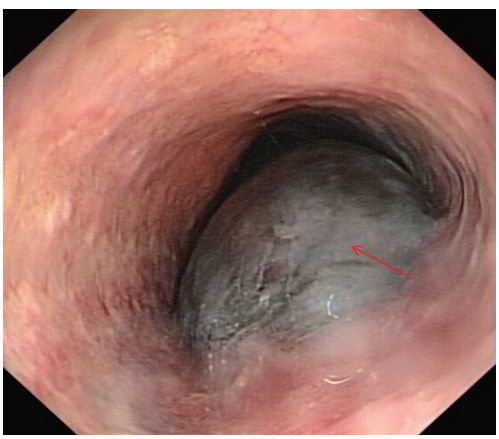

Figure 2 Gastroscopy showed esophageal mucosal protrusion (the arrow indicates the protruding area of the esophageal mucosa on gastroscopy).

hematoma was poor control of hypertension, prolonged consumption of rough and hard food, repeated nausea and vomiting, and sudden changes in esophageal pressure. After symptomatic treatments such as fasting, hypotension, acid suppression, hemostasis, nutritional support, etc., the related symptoms were not relieved. After 2 days of symptomatic treatment, chest enhanced CT showed increased pleural effusion on the right side (*Figure 3*). Moreover, noncoagulant fluid was extracted via right chest

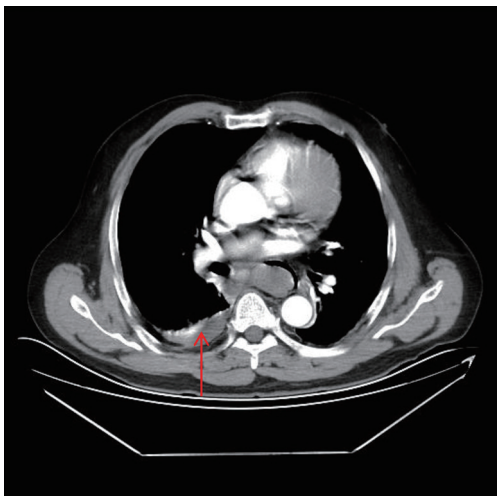

Figure 3 Chest enhanced CT showed increased pleural effusion on the right side (the arrow indicates the location of increased pleural effusion on the right side on chest enhanced CT). CT, computed tomography.

diagnostic puncture, and active bleeding was considered.

Given the continuing chest pain and dysphagia, after multidisciplinary consultation, the patient was taken to the interventional radiology room. Intra-arterial access was performed via a puncture of the right femoral artery with the modified Seldinger method, and a 5F catheter sheath was placed into the femoral artery. Subsequently, a 5F MIK catheter (Terumo Corp., Tokyo, Japan) was passed through the sheath to the aortic arch and extended to the thoracic aorta to provide angiography of the right intercostal artery and bronchial artery one by one. The opening of the esophageal artery branch of the right eighth intercostal artery was found to be abnormal on angiography. A Cobra catheter (Cook Medical, Bloomington, IN, USA) catheter was substituted into the opening for angiography to confirm the esophageal artery abnormality and the bleeding point. A microcatheter (Terumo Corp.) was used to superselect to the distal end of the eighth intercostal artery on the right side. Two microcoils (Cook Medical) 4.0 mm × 4.0 cm and a 2.0 mm × 2.0 cm in size, respectively, were released at the rear end of the esophageal artery opening. Subsequently, 10 mg of 350 to 560 μm particles of gelatin sponge (Hangzhou Ai Li Kang Pharmaceutical Technology Co., Ltd., Hangzhou, China) were administered for embolization, and then a 6.0 mm × 6.0 cm spring coil was released at the opening (*Figure 4*). Postoperative angiography showed that the artery was successfully embolized with particles

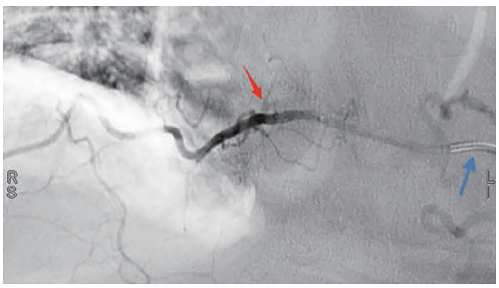

Figure 4 Results of arteriography (the red arrow indicates signs of contrast agent overflow; the blue arrow indicates that the catheter is superselected to the eighth intercostal artery).

and microcoils. Completion angiography was satisfactory, and a subsequent chest CT plain scan and enhanced scan showed that the pleural effusion had decreased and that the esophageal hematoma had basically been absorbed (*Figure 5*). Postoperative pain was relieved. On follow-up CT 3 days later, the hematoma appeared smaller. On the fourth day after surgery, the patient was instructed to take liquid food, and on the seventh day after operation, the patient took semiliquid food. The patient was discharged without complications after 1 week.

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

Discussion

SIHE is an uncommon cause of acute thoracic pain and is usually accompanied by additional digestive symptom such as dysphagia and hematemesis (7,8). SIHE can occur at any age, with the lower segment of the esophagus being the more common site of emergence. This can be explained by the absence of striated muscles and the lack of supporting structures at this level (10), which makes this area is the weakest part of the esophagus. Moreover, the submucosa of the esophageal wall is formed by loose connective tissue. There are abundant blood vessels and lymphatic vessels in it, which connect the mucosa and muscle layer. Therefore, SIHE usually occurs in the mucosa and submucosa.

Although there is no definite conclusion regarding the etiology of SIHE, it can be classified into the following categories (3,12,13): (I) esophageal injury; (II) a rapid rise of intraesophageal pressure; (III) iatrogenic injury, involving improper gastroscopy, retention of the gastric tube and small intestinal nutrition tube, etc; (IV) abnormal coagulation function; (V) related diseases, such as aortoesophageal fistula, esophageal cancer, esophageal hiatal hernia, etc.; (VI) special treatment, such as myocardial infarction thrombolysis, atrial fibrillation ablation, hemodialysis, etc; (VII) idiopathic disease without identifiable cause; and (VIII) spontaneous emergence (i.e., occurring naturally). The etiology type in this case was spontaneous.

The clinical manifestations of SIHE are not specific; the most common symptom is acute thoracic pain, present in 66–84% of cases, which is followed by hematemesis and dysphagia/odynophagia, present in 70% and 26% of cases, respectively (2,14-17). Additionally, 35% of SIHE patients can show all 3 of the abovementioned symptoms, while at least 50% of can have at least 2 of these symptoms (18). Moreover, 26% of patients with SIHE have dysphagia as the only clinical manifestation. If dysphagia occurs, it indicates that the disease is progressing rapidly, and the pressure of intramural hematoma is high and the volume is large. Some patients experience respiratory and circulatory disorders due to the compression of the trachea and adjacent aorta by hematoma. Therefore, it is important to differentiate SIHE from angina pectoris, acute myocardial infarction, arterial dissection, or spontaneous pneumothorax. In the process of clinical diagnosis and treatment, if the symptom of chest pain appears, especially in older adult patients, a diagnostic strategy should be further developed, in which a hematoma in the esophageal wall should be considered to avoid misdiagnosis and mistreatment.

With the improvement in medical practice and the application of advanced instruments, the methods for achieving the differential diagnosis methods of SIHE have become increasingly rich and mainly include the following (2,14,19): (I) upper gastrointestinal radiography, with the characteristic manifestation being “double tube sign”; (II) gastroscopy, in which the mucosa is longitudinally raised, the surface is obviously congested, and the base is blue; (III) chest wall CT, which can directly display the hematoma of the esophageal wall, showing concentric or eccentric thickening of the esophageal wall, which is helpful for differential diagnosis; (IV) endoscopic ultrasound, which is helpful for distinguishing solid tumor, cyst, or hematoma in the blood vessel wall and can detect the depth and scope of hematoma; (V) digital subtraction angiography (DSA),

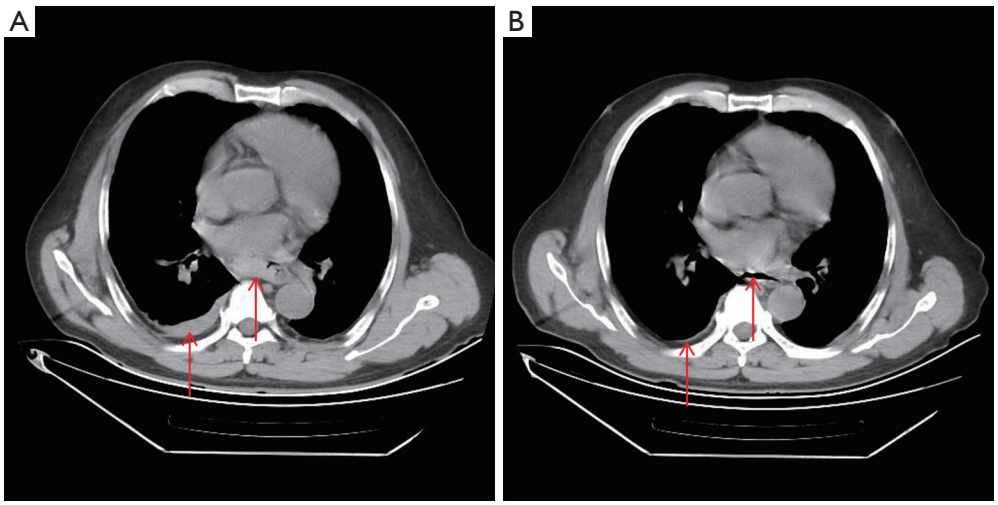

Figure 5 CT results of postoperative follow-up. (A) At 3 days (the arrow on the left indicates a decrease of pleural effusion; the arrow in the middle indicates reduction of esophageal hematoma. (B) At 1 week (the arrow on the left indicates the site where pleural effusion had basically disappeared; the arrow in the middle indicates the site where the esophageal hematoma had been basically absorbed). CT, computed tomography.

which can be used to identify patients with hematoma rupture and massive hemorrhage; and (VI) magnetic resonance imaging (MRI), which can be a valuable tool for differentiating IHE from aortic dissection.

After an accurate diagnosis is made, corresponding treatment is needed. The treatment of SIHE depends on syndrome and conditions. Treatment methods include (14,20,21) the following: (I) conservative treatment in internal medicine (involving fasting, acid suppression, hemostasis, protection of esophagus, gastric mucosa, etc.) which is applicable to a condition in which the muscle layer is not involved and with hematoma being absorbed in 1–3 weeks; (II) interventional embolization, which is applicable to mediastinal hematoma, hemothorax, etc.; and (III) surgery, which should be applied when esophageal rupture occurs.

Generally speaking, SIHE is usually managed conservatively with hemostasis, parenteral nutrition, and supportive treatment. However, Brown *et al.* reported that for the embolization treatment of the 8th to 10th intercostal arteries, there may be a risk of Adamkiewicz artery ectopic embolism leading to spinal cord ischemia and paraplegia during and after intervention (22). To reduce this risk, intraoperative angiography to confirm that there is no spinal artery, superselective avoidance of spinal artery embolization, and the use of spring coils combined with gelatin sponge particles can be applied. Transcatheter arterial embolization should be taken into account as a useful treatment when the symptoms are not relieved after conservative treatment and endoscopic bleeding is uncontrolled. It has been reported that arterial embolization is safe and effective for SIHE as compared to surgery (10,11).

In conclusion, SIHE should be considered in patients presenting with acute chest pain, dysphagia, and minor hematemesis. Although most symptoms of pain and bleeding in SIHE can be managed medically, more appropriate treatment should be attempted for those symptoms that are not relieved and for unmanageable bleeding after conservative treatment. In such cases, transarterial embolization may be a safe and useful treatment alternative to surgery.

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Footnote

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://qims.amegroups.com/article/view/10.21037/qims-23-564/coif).

The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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