Aortoesophageal fistula with hemorrhagic shock successfully treated with resuscitative endovascular balloon occlusion of the aorta

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Case Report

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Abstract

Background

Aortoesophageal fistula (AEF) is a rare cause of upper gastrointestinal hemorrhage. Despite diagnostic and therapeutic advances, the mortality rate in AEF patients remains high because of its fulminant course, even with maximal intensive care. Resuscitative endovascular balloon occlusion of the aorta (REBOA) is a resuscitation technique to control life-threatening bleeding. It has become an important modality in the management of life-threatening, traumatic or non-traumatic, arterial bleeding. However, its use in hemorrhagic shock caused by cancer has rarely been reported.

Case presentation

: A 51-year-old female with a history of esophageal cancer presented to our emergency department with hematemesis. Computed tomography was performed because of a strong suspicion of hemorrhagic shock. With a diagnosis of AEF due to esophageal cancer, emergency thoracic endovascular aortic repair was performed while the bleeding was controlled using REBOA. Staged elective esophageal reconstruction was successfully performed.

Conclusions

Hemostasis is of paramount importance in individuals presenting with suspected hemorrhagic shock attributable to arterioenteric fistula (AEF). The timely implementation of Resuscitative Endovascular Balloon Occlusion of the Aorta (REBOA) has shown promise in these circumstances, showcasing potential effectiveness.

Background

Resuscitative endovascular balloon occlusion of the aorta (REBOA) was developed in 1953 by Edwards et al. (1). A balloon-bearing catheter is introduced through the femoral arterial sheath and inserted into the aorta for endovascular aortic occlusion. REBOA ensures blood flow to the vital organs through early proximal control of bleeding. It temporarily controls bleeding below the inflation site and increases the cerebral and coronary circulation, providing a window for definitive care, including surgery and embolization, and preventing death.

Case Presentation

A 51-year-old female was transported to our hospital by ambulance with a chief complaint of hematemesis. She was already under follow-up at our hospital for esophageal cancer. Upon arrival of the emergency services, she had difficulty moving. The emergency crew entered through a window and

performed the rescue operation. She was brought to the hospital 40 min after the initial report. Our university hospital includes advanced emergency, cardiovascular surgery, cardiology, and gastroenterology services and is capable of performing emergency surgeries.

She was diagnosed with oropharyngeal and esophageal cancer 2 years previously. She had undergone oropharyngeal resection for oropharyngeal cancer, and chemotherapy and photodynamic therapy for esophageal cancer 1 year previously. The patient also had a history of dyslipidemia and chronic kidney disease.

She was admitted to our hospital for hematemesis. Upon examination, she had facial pallor and her body temperature was 38.9°C. Her pulse rate and blood pressure were 146 bpm and 88/35 mmHg, respectively. Oxygen saturation was 100% without supplemental oxygen, and the respiratory rate was 20 breaths/min. She was diagnosed with hemorrhagic shock and a double lumen central venous catheter (CVC) and 4Fr arterial sheath were inserted from the right inguinal vein. A REBOA (Tokai Medical Products, Inc., Aichi, Japan) device was kept on standby. Blood tests showed a markedly decreased hemoglobin level of 6.8 g/dL, and high-dose fluid replacement therapy was started through the CVC. The patient responded to fluid replacement and the blood pressure improved slightly. Computed tomography (CT) was performed with the REBOA deflated, which showed an aortoesophageal perforation. Numerous blood clots were found in the gastrointestinal tract, indicating upper gastrointestinal bleeding (Fig. 1). During the CT scan, systolic blood pressure decreased to 47 mmHg, and the patient's consciousness level deteriorated. The aortic diameter in CT was 23 mm, and the REBOA was 21 mm. A balloon was immediately inserted through the right femoral artery and placed at the perforation site (Fig. 2). Following balloon inflation, the systolic blood pressure improved to 105 mmHg.

Transfusion therapy and tracheal intubation were performed simultaneously. The patient was taken to the operating room for an emergency thoracic endovascular aortic repair. A stent was inserted at the lower edge of REBOA and rapidly deployed to the area of contrast-medium leakage, seen in CT with REBOA deflation. The patient was admitted to the intensive care unit for further management. The patient developed sepsis due to stent infection. An elective esophagectomy, aortic vascular replacement, and gastrointestinal reconstruction were performed at a later date.

Discussion and Conclusions

Aortoesophageal fistula (AEF) is a rare cause of upper gastrointestinal bleeding, with an incidence of approximately 0.07% (2). Its etiology includes aneurysms (54.2%), foreign bodies (19.2%), and esophageal cancer (17%) (3). Conservative medical treatment has a poor prognosis and invariably results in a fatal outcome (4). To the best of our knowledge, no patient has survived emergency surgery following hemorrhagic shock. A literature search showed that there have been no cases of emergency surgery leading to hemorrhagic shock.

REBOA is a means for resuscitation in cases with intractable hemorrhagic shock, including gastrointestinal bleeding, ruptured aortic aneurysms, and critical traumatic or obstetric bleeding. Aortic

clamping through resuscitative thoracotomy is the classic technique for bleeding control, but it is an invasive procedure associated with chest wall bleeding, hypothermia, and infection. Aortic occlusion through REBOA may improve myocardial and cerebral blood flow by restricting the cardiac output to thoracic aortic vasculature (5, 6). In this case, renal artery occlusion due to REBOA placement led to blood flow disturbances and worsening renal function. The patient required dialysis therapy but was weaned after 27 days.

The cut-off time to the operating room was 60 min. In some cases, REBOA may cause bleeding and organ damage, and proper ballooning is important. The aorta is divided into three zones to determine the location of balloon occlusion based on the source of bleeding. Zone 1 extends from the left subclavian artery to the celiac artery, zone 2 extends from the celiac artery to the renal artery, and zone 3 extends from the renal artery to the aortic bifurcation (7). If the source of bleeding can be identified, zone 1 is chosen for intra-abdominal bleeding and zone 3 is chosen for pelvic fractures and gynecological organ bleeding (8). Relatively accurate positional information can be obtained through CT with contrast. The patient survived in this case, became alert, and recovered to the rehabilitative status.

In addition, early sheath placement and REBOA insertion were noteworthy in this case. Early hemorrhage is characterized by sympathetic system activation, resulting in compensatory vasoconstriction aimed at normalizing arterial blood pressure (9). After a certain amount of blood loss, sympathetic inhibition occurs, vascular resistance decreases, and bradycardia develops, which is rapidly followed by cardiocirculatory arrest (10). In this case, there was a high probability of hemorrhagic shock based on the chief complaint and the medical history. Therefore, appropriate measures were taken from the beginning. Furthermore, previous CT scans did not show any vascular anomalies, and we expected REBOA to be effective even before the patient reached the hospital. A less invasive primary therapeutic approach may be used in frail patients with high surgical risk, which may lead to lower intraoperative and perioperative mortality rates than those associated with thoracotomy.

Early insertion of the sheath into a collapsed vessel can cause vascular damage and promote bleeding. In addition, device insertion can cause deterioration of the patient’s condition if other conditions, such as septic shock, are present.

The present case suggests that, even in cases of potentially fatal hemorrhagic shock, appropriate hemostasis with REBOA can be effective. The wound was in a good condition postoperatively, but the patient remained paraplegic. After rehabilitation, she was able to move around in a wheelchair without assistance. She was transferred to another hospital on day 89 for long-term rehabilitation. She is regularly followed up at our hospital and was still alive 1.5 years after the event.

Hemostasis is important in AEF patients with suspected hemorrhagic shock. Early REBOA may be effective, as in this case, but further research is required to explore its use.

Abbreviations

AEF
Aortoesophageal fistula
REBOA
Resuscitative endovascular balloon occlusion of the aorta
CVC
central venous catheter
CT
Computed tomography

Declarations

Ethics approval and consent to participate:

Ethics approval is not applicable. Written informed consent was obtained from the patient for writing this case report and the use of accompanying images. The study was performed in accordance with the ethical standards as laid down in the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards.

Consent for publication:

Written informed consent was obtained from the patient for writing this case report and the use of accompanying images.

Availability of data and materials: Not applicable.

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Authors' contributions: TH, MO had taken responsibility for all aspects of the reliability and freedom from bias of the data presented and their discussed interpretation, and TH wrote this report. TN collected and interpreted the clinical data, and drafted the report. YG supervised medical management, interpreted clinical data and contributed the writing of the report.

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Tadayuki Hirai is a cardiologist working at Department of Emergency, Kanazawa University Hospital, Japan. He is also an MD at the Graduate School of Medicine, University of Kanazawa, Japan. His main interests are structural heart disease and the role of sympathetic nerve activity in heart failure, especially in the context of intensive care and emergency medical care.

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Figures

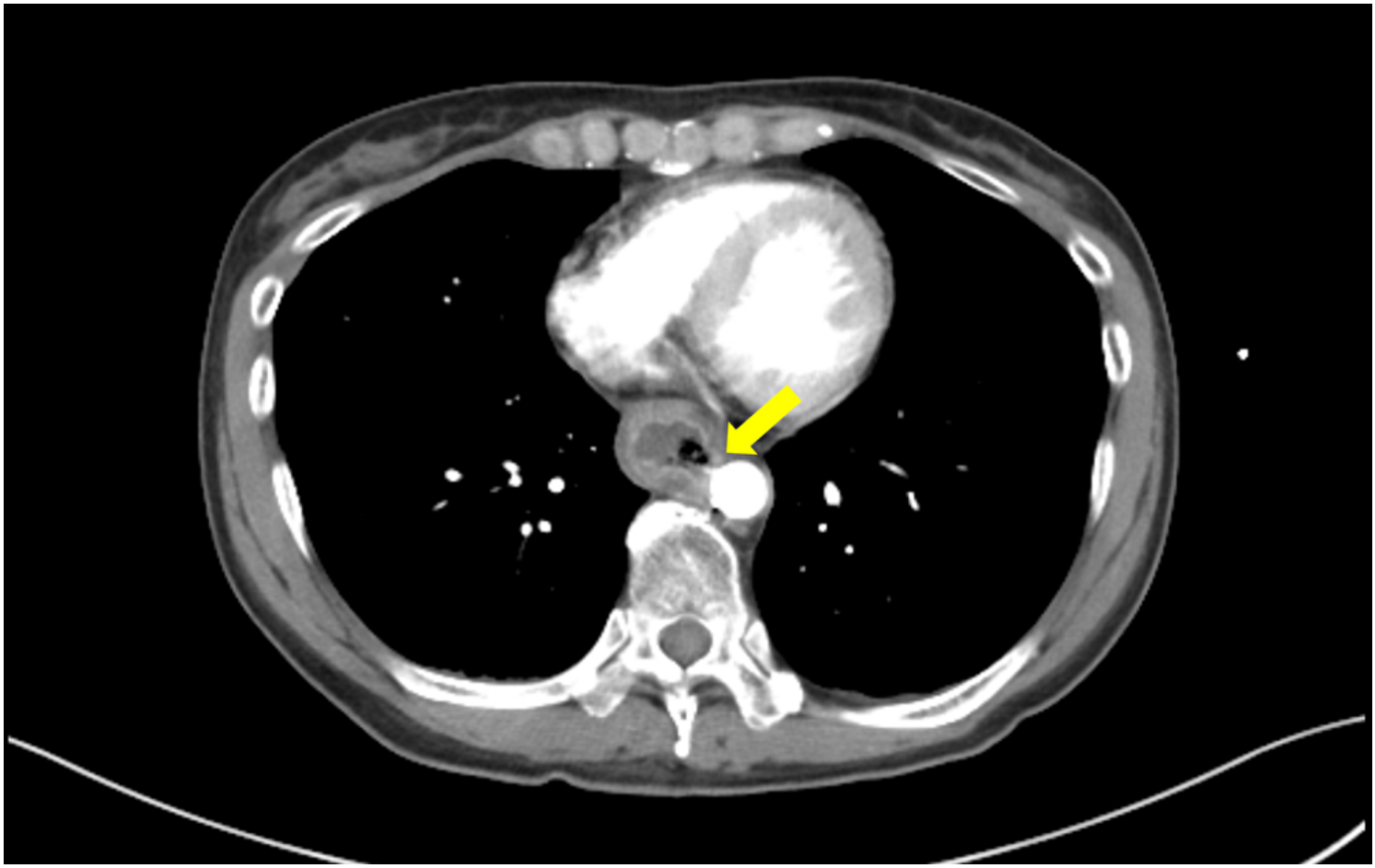

Figure 1

Thoracoesophageal cancer with aortic invasion and rupture: ← shows esophageal contrast medium extravasation.

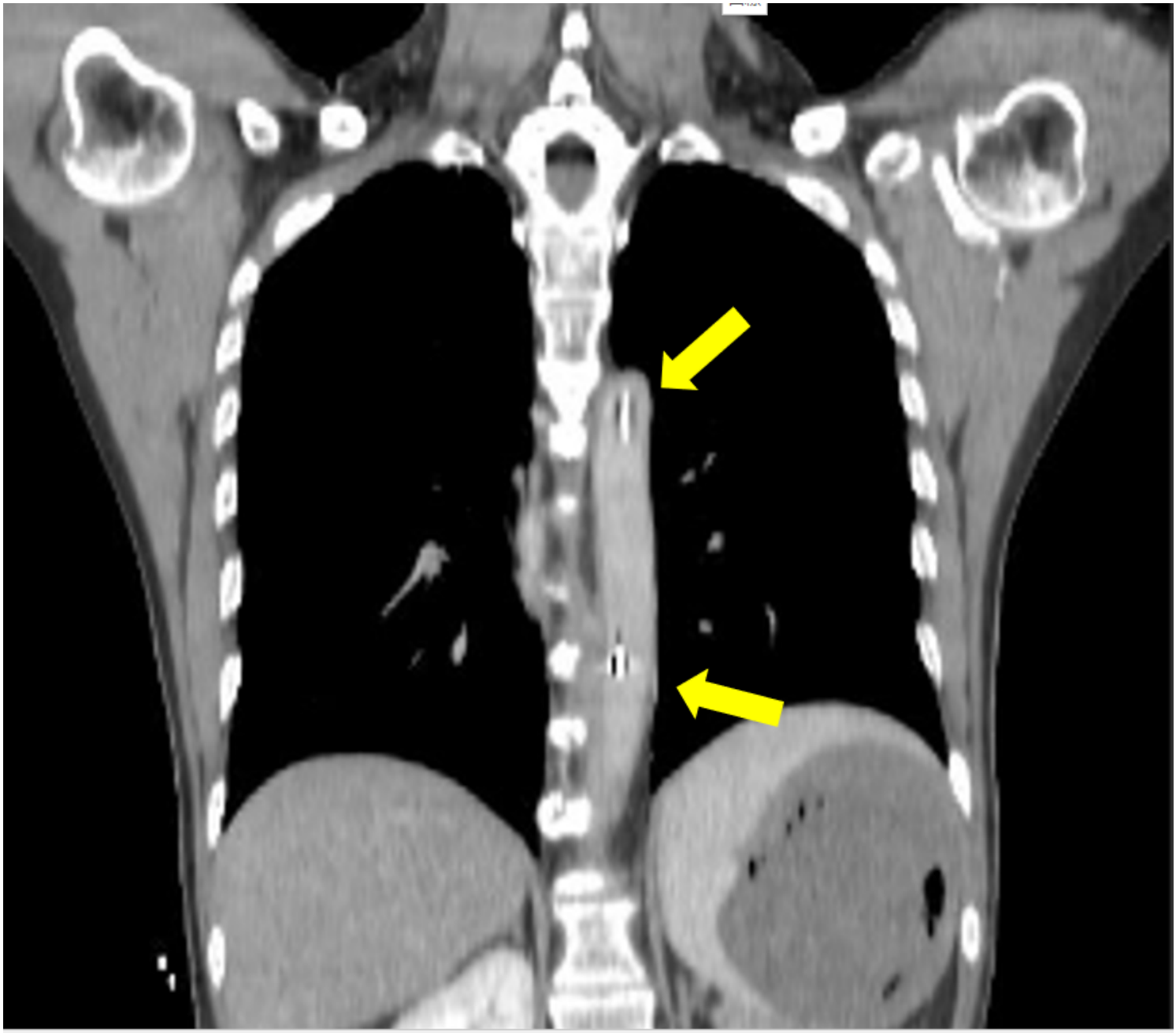

Figure 2

REBOA inserted into the aorta; the high absorption area represents the top and bottom of the balloon.