

CASE REPORT

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

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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 woman 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 crucial in patients who present with suspected hemorrhagic shock attributable to AEF. The timely implementation of REBOA has shown promise and potential effectiveness in such cases.

Keywords Hemorrhagic shock, Aortoenteric fistula, REBOA

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 both cerebral and coronary circulation, providing a window for definitive care, including surgery and embolization, and preventing death.

Case presentation

A 51-year-old woman 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

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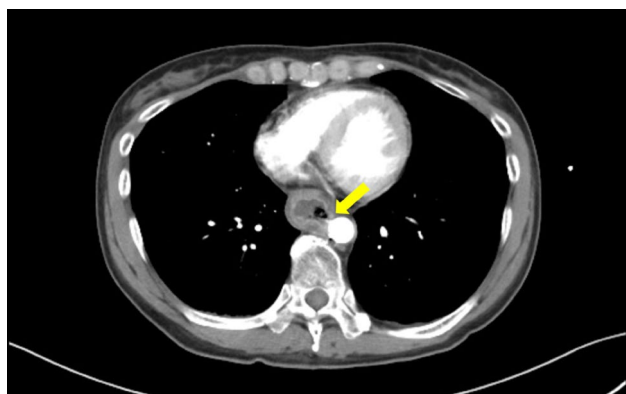


Fig. 1 Thoraco-esophageal cancer with aortic invasion and rupture: ← shows esophageal contrast medium extravasation

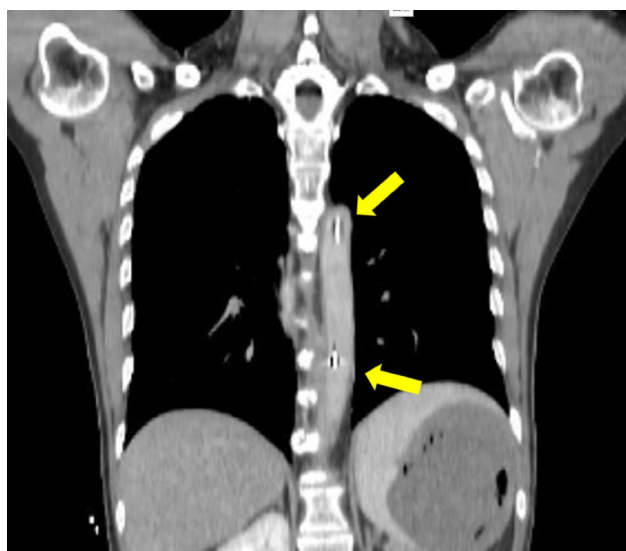


Fig. 2 REBOA inserted into the aorta; the high absorption area represents the top and bottom of the balloon

gastroenterology services and can perform 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 treatment of 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. A double lumen central venous catheter (CVC) and 4Fr arterial sheath were inserted from the right inguinal vein with ultrasound guidance, and high-dose fluid replacement

therapy was started through the CVC. The patient responded to fluid replacement and the blood pressure slightly improved. Blood tests showed a substantially decreased hemoglobin concentration of 6.8 g/dL. We upgraded the arterial sheath to 7Fr, and a REBOA device (Tokai Medical Products, Inc., Aichi, Japan) was kept on standby within the aorta. Computed tomography (CT) was performed with the REBOA deflated, which showed an aorto-esophageal perforation at the level of the eighth thoracic vertebra. 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. We confirmed the absence of endoleak through aortography. The total occlusion time with REBOA was 55 min, and external bleeding was minimal. The patient was admitted to the intensive care unit for further management. She developed sepsis due to stent infection and required multidisciplinary treatment in the intensive care unit for 7 days. The wound was in a good condition postoperatively, but the patient remained paraplegic. We diagnosed the paraplegia as related to spinal cord ischemia due to the REBOA-induced decrease in aortic blood flow. The patient initially required dialysis therapy, but she was weaned from the therapy after 27 days. One month later, selective esophagectomy was performed; 2 months later, aortic vascular replacement and gastrointestinal reconstruction were performed. After rehabilitation, the patient was able to navigate in a wheelchair without assistance. She was transferred to another hospital on day 89 for long-term rehabilitation. At the time of this writing, she was attending regular follow-up at our hospital and remained alive 2 years after the event.

Discussion and conclusions

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, 4]. Conservative medical treatment has a poor prognosis and invariably results in a fatal outcome [5]. To the best of our knowledge, no

patient has survived emergency surgery following hemorrhagic shock due to AFE bleeding.

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 the thoracic aortic vasculature [6]. Aortic occlusion preserves cerebral perfusion and coronary filling in patients exhibiting extremis with refractory hypotension. This leads to increased central aortic pressure, carotid flow, and brain oxygenation [7, 8]. In the present case, renal artery occlusion due to REBOA placement led to blood flow disturbances and worsening renal function. Additionally, the patient became paraplegic due to residual nerve damage caused by spinal cord ischemia, possibly attributed to the prolonged balloon occlusion time. Newer REBOA techniques, such as partial or intermittent REBOA, may be considered; however, they might result in uncontrolled bleeding as a compensatory effect [9].

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 [10]. 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 [11]. In this case, the patient was suspected to have a single point of upper gastrointestinal bleeding due to tumor invasion. Therefore, we performed standby REBOA in zone 1, identified the bleeding point by CT examination, and inflated the balloon. 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 [12]. After a certain amount of blood loss, sympathetic inhibition occurs, vascular resistance decreases, and bradycardia develops, which is rapidly followed by cardiocirculatory arrest [13]. 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. Ultrasound-guided placement enhances precision, minimizing the risk of multiple punctures and related complications such as hematomas, arterial wall injuries, thrombosis, and incorrect access to the superficial femoral artery. This technique thus reduces the risk of vascular injury and limb ischemia [9, 14]. Despite a prior CT scan, complete exclusion of aortic rupture due to tumor invasion remained challenging. Consequently, the balloon was inflated after CT evaluation to mitigate this risk in our patient.

The present case suggests that, even in cases of potentially fatal hemorrhagic shock, appropriate hemostasis with REBOA can be effective. Although REBOA offers the promise of additional time to address complex and severe perforations, optimal pressures above and below the balloon and the maximum duration of occlusion have not yet been established. Further robust studies are required to fully elucidate the role of this technology and technique in trauma patient management. Additionally, REBOA should be performed by experienced, well-trained providers who are proficient with the technology and understand its appropriate indications and usage. REBOA is not intended to provide definitive hemorrhage control; it should serve as an adjunctive tool that can provide time to mobilize resources and expand treatment options for hemorrhage management. Additional studies are needed to establish an optimal algorithm for deploying and managing REBOA in patients with upper gastrointestinal bleeding due to tumor invasion.

In conclusion, we have reported a case of successful rescue using REBOA for management of AEF in a patient 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

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Not applicable.

Author 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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Data availability

No datasets were generated or analysed during the current study.

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.

Competing interests

The authors declare no competing interests.

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