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Massive hemoptysis in pregnancy treated by ECMO combined with electronic bronchoscopy: A case report

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ABSTRACT

Background: Massive hemoptysis during pregnancy is very rare. Dieulafoy's disease is one of the causes of massive hemoptysis. There are few reports of ECMO use to treat massive hemoptysis during pregnancy.

Findings: We report for the first time a patient with Dieulafoy's disease diagnosed at 29 weeks of pregnancy. The patient's hemoptysis occurred rapidly with large volumes. The bleeding amount reached 500 ml within half an hour, with the development of asphyxia and respiratory and cardiac arrest due to a blood clot blocking the airway. After successful cardiopulmonary resuscitation, the ventilator could not maintain effective ventilation. Emergency establishment of VV-ECMO was performed to maintain oxygen, and hemostasis was successfully achieved by performing bronchial artery embolization twice. We successfully cleaned blood clots in the airway four times by freezing and using a foreign body retrieval basket with an electronic bronchoscope. At the same time, small and smooth nodular lesions were found under bronchoscopy, and blood vessels with a diameter of 1.5 mm were found under Doppler mode with an ultrasonic bronchoscope, which was consistent with a diagnosis of Dieulafoy's disease. VV-ECMO was successfully stopped on the 3rd day of the disease course, tracheal intubation was successfully removed on the 5th day of the disease course, and the patient was discharged with no complications on the 16th day of the disease course.

1. Introduction

Massive hemoptysis during pregnancy is rare and potentially life-threatening condition [1]. Dieulafoy's disease, characterized by the rupture and hemorrhage of aneurysms in submucous membranes, is recognized as one of the underlying causes of massive hemoptysis [2]. We report for the first time a pregnant woman at 29 weeks gestation who was admitted to the hospital due to massive hemoptysis during pregnancy and was diagnosed with Dieulafoy's disease. This study aimd to enhance clinicians' understanding of this rare condition and contributes to the improvement of its diagnosis and treatment through an analysis of this case and a comprehensive review of the relevant literature.

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1.1. Case description

The patient was a 29-year-old pregnant chinese woman of Han nationality at 29 weeks of gestation. The patient had no obvious reason for blood loss, which reached 500 ml in half an hour. When the patient entered the emergency room, she suffered from hemoptysis and dyspnea. Past health. Physical examination showed the following: a temperature of $37.2 \,^{\circ}$ C, a pulse rate of 101 beats/min, a respiratory rate of 20 breaths/min, a blood pressure (BP) of 90/60 mmHg, and the presence of a few moist rales and dry crackles in both lungs ... There were no abnormalities in blood biochemistry or coagulation function. Routine blood tests showed the following: a WBC count of 10.82×109 /L; a Hb level of 81.0 g/L; a PLT count of 297×109 /L; and a D-Dimer level of 1318 ng/ml. Blood gas analysis showed the following: a pH of 7.40, a PaCO2 of 237 mmHg, and a PaO2 of 55 mmHg. Bronchial artery CTA showed multiple tortuous and thickened bronchial arteries in the trachea, beneath the carina and hilar area, and a large area of increased density in the right lung (Fig. 1).

1.2. Diagnosis and treatment process

On August 12, 2022, at 10:45 a.m., we administered hemostatic treatment, including Tranexamic acid (0.5g ivgt), Human fibrinogen (2.0g ivgtt) and Vasopressin (12u ivgtt). But the patient suffered from hemoptysis again, with approximately 200 ml of blood loss, blurred consciousness and restlessness, and a decrease in the SPO2 value to 50 %. Immediately at 10:58 a.m., endotracheal intubation was performed, connecting the patient to mechanical ventilation with settings of P-A/C, Pi 20 cmH2O, PEEP 8 cmH2O, and FiO2 1.0. The SPO2 value ranged between 70 and 80 %. Bronchoscopy showed that there were many fresh bleeding and blood clots in the airway, and the visual field under the microscope was blurred. Then, the fetal heart rate dropped to 50–60 beats/min, and the obstetrics department suggested terminating the pregnancy as soon as possible. Emergency multidisciplinary consultation was initiated to formulate the rescue process: first, cesarean section was performed to rescue the fetus, and bronchial Artery Angiography and bronchial artery embolization were established under the support of VV-ECMO.

After the cesarean section, the patients was urgently transferred to the compound operating room to be supported by V–V ECMO (blood flow 4–4.5 L/min, oxygen flow 4 L/min, FiO2 1.0) August 12, 2022, at 12:50 AM. Blood gas analysis was performed again and showed the following: a pH of 7.35, a PaO2 of 189 mmHg, and a PaCO2 of 234 mmHg. Then, bronchial arteriography showed that the right bronchial artery and the right superior intercostal artery shared a trunk from the thoracic aorta, which was obviously thickened and tortuous, with dense contrast medium staining at the distal end of the blood vessel. The left bronchial artery shared a trunk with the right inferior bronchial artery, with a thickened and disordered distal section and dense staining. A 2.7 F microcatheter was utilized to inject polyvinyl alcohol particles (PVA 300 µm and 500 µm), and both the left and right bronchial arteries were embolized as confirmed by posterior angiography (Fig. 2).

After bronchial artery embolization, a large number of blood clots were found under bronchoscopy on August 12, 2022, which completely blocked the airway and could not be removed by negative pressure suction. The frozen blood clots shown under bronchoscopy. The cryoprobe was placed on the blood clot, freezing was started, the lens body and probe was pulled out of the airway together with the frozen blood clot, and the operation was performed repeatedly to successfully dredge the central airway. However, it

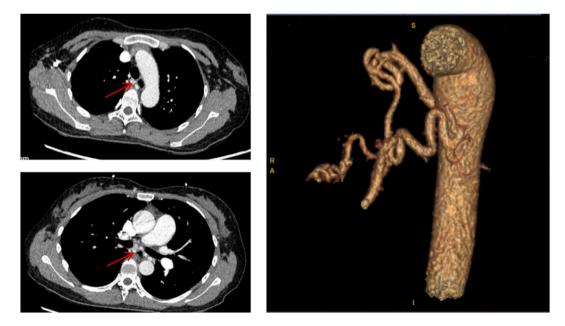


Fig. 1. Bronchial artery CTA showed multiple tortuous and thickened bronchial arteries in the trachea, beneath the carina and hilar area, and a large shadow with increased density in the right lung.

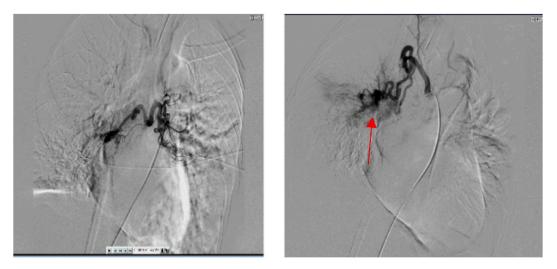


Fig. 2. Bronchial arteriography showed that the right bronchial artery and the right superior intercostal artery shared a trunk from the thoracic aorta, which was obviously thickened and tortuous, with dense contrast medium staining at the distal end of the blood vessel. The left bronchial artery shared a trunk with the right inferior bronchial artery, with a thickened and disordered distal section and dense staining.

was difficult to remove bronchial blood clots in each subsegment, so a disposable foreign body retrieval basket was used for thrombus removal. Blood clots and scabs weighing 120 g were removed (Fig. 3). After removal, the patient received treatment under bronchoscopy three times in the intensive care unit until there was no blood clot residue and no active bleeding in the airway.

On the 11th day of the disease course, bronchoscopy showed that there were no blood clots in the trachea. Microscopically, small and smooth nodular lesions were found. Under the Doppler mode of an ultrasonic bronchoscope, 1.5 mm diameter blood vessels were found under the submucosa, which was consistent with a diagnosis of Dieulafoy's disease (Fig. 4). On the 16th day, the patient was discharged with no complications, and the mother and child were safe. CTA for lung arteries reviewed in July 2023 showed that the right bronchial artery originated from the right side of the thoracic aorta (T6 level), the proximal lumen was slightly enlarged, and the distal part was not shown, while the left bronchial artery originated from the left side of the thoracic aorta (T7 level), it was located in the subcarina of trachea and hilum of lung.

2. Discussion

Although massive hemoptysis accounts for only 5–15 % of hemoptysis cases, it is a serious life-threatening respiratory disease [3,4]. Dieulafoy's disease is often one of the life-threatening causes of massive hemoptysis [5,6]. The patient was confirmed by bronchial arteriography and ultrasound bronchoscopy to have massive hemoptysis due to Dieulafoy's disease. Dieulafoy's disease occurs during pregnancy and is considered to be related to multiple factors. Peyrat E et al. believe that the increase in pulmonary blood volume during pregnancy [7], the sharp change in hormone levels, and the combined effects of respiratory mucosal edema caused by uterine enlargement and diaphragm elevation increase the risk of massive hemoptysis during pregnancy.

Acute respiratory failure due to Dieulafoy's disease with massive hemoptysis requiring VV-ECMO support has not been reported. Moore et al. summarized that 41 pregnant patients received VV-ECMO treatment, and the maternal and fetal survival rates reached 77.8 % and 65 % [8], respectively. Ramanathan et al. concluded that the survival rate of pregnant women and perinatal patients with refractory heart or respiratory failure receiving ECMO treatment was 70 % [9]. In this case, the patient's blood clot blocked the airway, which led to the ventilator being unable to ventilate effectively. Timely use of VV-ECMO is a very necessary choice [10]. The start of

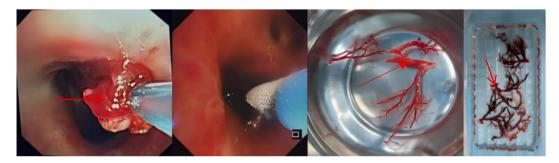


Fig. 3. The frozen probe and a disposable foreign body basket were used to remove the blood clot via bronchoscope.

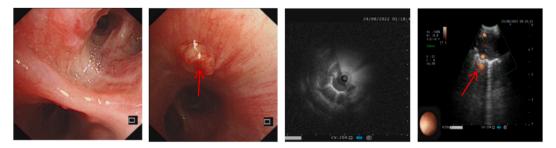


Fig. 4. Under the Doppler mode of an ultrasonic bronchoscope, 1.5 mm diameter blood vessels were found under the submucosa, which was consistent with a diagnosis of Dieulafoy's disease.

VV-ECMO is consistent with the management requirements of VV-ECMO reported by Levy et al. [11]. Moreover, it should be noted that due to physiological changes during pregnancy, approximately 1/3 of pregnant patients experience bleeding when using VV-ECMO [9, 12]. In this case, VV-ECMO-assisted airway interventional therapy can effectively ensure the safety of endoscopic interventional operations and reduce the risk of asphysia death caused by the recurrence of massive hemorrhage.

Interventional therapy under bronchoscopy is an important method to deal with massive hemoptysis [13,14]. Bronchoscopy can directly show the airway and allows for the removal of blood clots, the identification of bleeding sites, the performance of local hemostasis under bronchoscopy, and even the placement of balloon compression hemostasis. An experienced respiratory endoscopic intervention team can control and reduce the risk of bronchoscopic operations [15,16]. In this case, a large number of blood clots blocked the airway, and we tried to use a cryoprobe to remove the blood clots [17]. In the central airway, blood clots are easily cleared, but for blood clots in the airway below the subsegment, the cryoprobe is difficult to insert and easily sticks to the tube wall, damaging the airway mucosa and increasing the risk of bleeding. The method of removing blood clots with a foreign body retrieval basket was used instead [18], and a large number of subsegment bronchial blood clots were successfully removed with satisfactory curative effects. Our treatment method further confirms the feasibility of using bronchoscopy at the bedside to remove airway blood clots for critically ill patients with pulmonary hemorrhage, as proposed by Schmidt et al. [19]. It is hoped that the experience described above inspires endoscopic blood clot removal after massive hemoptysis. However, during the bronchial cryotherapy procedure, there is a potential risk of tearing and bleeding at the frozen site. Therefore, it is contraindicated for patients with coagulation disorders or a bleeding tendency, and its clinical use is limited. In this patient's case, the risk of rebleeding has been significantly reduced through bronchial artery intervention therapy before using cryotherapy to clear blood clots.

3. Conclusion

The rescue and treatment of patients with massive hemoptysis during pregnancy is extremely challenging. VV-ECMO support is a good choice when conventional mechanical ventilation is ineffective for acute respiratory failure caused by massive hemoptysis. Interventional therapy under bronchoscopy with the assistance of VV-ECMO is more efficient and safe. We recommend bronchoscopic cryosurgery as an effective technique for removing blood clots in critically ill patients.

Data availability statement

The original contributions presented in the study are included in the article/Supplementary material, further inquiries can be directed to the corresponding author.

Ethics statement

This study obtained ethical permission from the Science and Ethics Committee of Changsha First Hospital.

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CRediT authorship contribution statement

Keyu Li: Writing - original draft, Validation, Investigation. Long Wen: Writing - review & editing, Validation. Haibo Zhou: Writing - review & editing, Investigation.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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