A case report regarding general anesthesia management of a patient with pulmonary vein stenosis that underwent balloon dilatation and stent implantation

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Abstract
Rationale: Pulmonary vein stenosis (PVS) is a rare cardiovascular deformity that can lead to high mortality if left untreated. Patients frequently experience multiple complications such as hemoptysis, pulmonary hypertension, bronchial venous rupture and cardiac insufficiency. Currently, pulmonary vein stenosis balloon dilatation (stent implantation) is the only treatment, and this can be performed under local or general anesthesia. However, a case report on the general anesthesia management of PVS has not been previously reported. In this case report, we discuss anesthetic considerations in patients with PVS, focusing specifically on perioperative airway and circulatory management as well as the risk evaluation, and the appropriate effective management of all potential complications intraoperatively.

Patient concerns: A 58-year-old male patient was admitted because of coughing rusty sputum for during 2 years as well as experiencing dyspnea and chest distress after exertion or exercising. The difficulty breathing and chest distress had been going on for a year. This patient had undergone circumferential pulmonary vein isolation twice during the last 2 years.

Diagnoses: Based on the transthoracic echocardiography and computed tomography, this patient’s diagnosis was considered as pulmonary vein stenosis, pulmonary hypertension (secondary), and pulmonary arteriovenous thrombosis.

Interventions: We considered that such severe patients with PVS require respiratory and circulatory supports and perhaps emergency surgical interventions. Henceforth, we administered general anesthesia to the patient and had extracorporeal membrane oxygenation (ECMO) on standby.

Outcomes: The duration of the surgery was approximately 4 hours, the intraoperative vital signs were stable, no pericardial effusion was observed postoperatively, the blood flow in the pulmonary vein stent was smooth, and the patient was discharged 7 days later following rehabilitation.

Lessons: This surgical procedure involving respiratory and circulatory supports requires the involvement of different medical personnel such as interventionists, anesthesiologists, and surgeons. Therefore, multidisciplinary cooperation under general anesthesia will undoubtedly benefit such patients.

Abbreviations: CCU = cardiac care unit, CT = computed tomography, ECG = electrocardiogram, ECMO = extracorporeal membrane oxygenation, HR = heart rate, ID = inner diameter, NBP = noninvasive blood pressure, PCO2 = partial pressure of carbon dioxide, PG = pressure gradient, PVS = pulmonary vein stenosis, RR = Respiratory rate, TAPVC = total anomalous pulmonary venous connection.

Keywords: balloon dilatation, general anesthesia management, pulmonary vein stenosis, stent implantation

1. Introduction

The development of cardiology interventional techniques has led to atrial fibrillation radiofrequency ablation therapy becoming the alternative therapy of anti-arrhythmic drug therapy. It is recommended as the first-line treatment option because its efficacy is superior to that of any anti-arrhythmic drugs.[1,2] Pulmonary vein stenosis is one of the significant and most severe complications of early atrial fibrillation radiofrequency ablation. The improvement of technical skills and the increasingly updated radiofrequency devices has resulted in the gradual decrease of radio frequency-related complications. Specifically, the incidence of pulmonary vein stenosis after radiofrequency ablation has significantly decreased from 42% to 0.32% to 3.4%. [3-4] However, pulmonary vein stenosis after ablation has been reported to occur occasionally, which can be ascribed to the heterogeneities in technique and device in different centers.
Unfortunately, such patients are frequently misdiagnosed and grouped with other multiple complications such as hemothysis, pulmonary hypertension, bronchial venous rupture, and cardiac insufficiency, which are very easy to misdiagnose. These misdiagnosed patients are associated with numerous cardiopulmonary complications; therefore, they may encounter a variety of intervention-related complications during perioperative period posing an inevitable challenge to the perioperative anesthesia management. Consequently, sufficient preparation and favorable intraoperative anesthesia management will benefit such patients.

2. Methods

We report an observational case of general anesthesia management of a patient with pulmonary vein stenosis that underwent balloon dilatation and stent implantation. The committee waived the requirement for approval to conduct this single case study with access to medical records. Informed consent to publication has been obtained from the patient.

3. Case report

A 58-year-old male patient was admitted because of coughing rusty sputum for over 2 years as well as experiencing dyspnea upon exertion and chest distress after exercise for 1 year. The diagnosis included pulmonary vein stenosis, pulmonary hypertension (secondary), and pulmonary arteriovenous thrombosis. The patient received circumferential pulmonary vein isolation due to recurrent atrial fibrillation 2 years ago in a different hospital. He gradually developed coughing rusty sputum and exertional dyspnea after surgery. One year ago, he received circumferential pulmonary vein isolation again due to recurrent atrial fibrillation. However, the above symptoms were remarkably aggravated with heart tiredness and chest distress only after mild exercise 4 months ago, which was subsequently accompanied by hemoptysis 1 month ago. The patient was proposed to undergo pulmonary vein stenosis stent implantation under general anesthesia.

3.1. Physical examination

On physical examination temperature was 36.5°C, heart rate (HR) was 78 times/min, respiratory rate (RR) was 19 breaths/min, noninvasive blood pressure (NBP) was 96/59 mmHg, height was 165 cm, and body weight was 56 kg. The jugular vein and the heart border were normal, the heart rate was homogeneous, and no murmur was heard in all valve areas. No abnormality was seen in the thorax, and no abnormal sound was detected in both lungs percussion and auscultation. Also, nothing special was discovered during the abdominal physical examination. No swelling was detected in both lower limbs.

3.2. Auxiliary examinations

Transthoracic echocardiography revealed slightly enlarged right ventricle, tricuspid regurgitation (mild), pulmonary hypertension (moderate-severe), pressure gradient (PG) 71 mmHg, and the estimated pulmonary arterial systolic pressure was 74 mmHg. Chest computed tomography (CT) revealed scattered ground glass degeneration in both lungs and interstitial edema. CT pulmonary angiography: stenosis or occlusion in the initial segments of bilateral inferior pulmonary veins, which was more significant in the left side, with heterogeneous end luminal density and light distal vessel display. Meanwhile, stenosis in the initial segment of the left superior pulmonary vein was also seen, filling-defect seemed to be seen in the lumen, the possibility of embolism could not be excluded, and favorable distal vessel filling could also be observed. The preoperative electrocardiogram (ECG) displayed the suspicious pulmonary embolism ECG (S1Q3T3). No apparent abnormality was seen in laboratory examination.

3.3. Surgical experience and anesthesia management

Conventional ECG monitoring was given after the patient was sent to the operating room, the HR was 96 beats/min, NBP was 91/56 mmHg, RR was 20 breaths/min, and oxygen saturation (SpO2) was 92% (which could increase to 99% after 80% oxygen inhalation). Right-hand radial artery catheterization was completed under local anesthesia for invasive blood pressure monitoring, the body surface defibrillation electrodes were stuck, ECMO was standby, and drugs including epinephrine, noradrenaline, nitroglycerin, milrinone, and frusemide were prepared. After perfecting preoperative preparation, midazolam, sufentanil, propofol, and cisatracurium were administered for conventional induction. Meanwhile, noradrenaline was injected at an initial infusion of 0.03 µg/kg/min during induction to inhibit the peripheral vascular dilation induced by general anesthetics. Following a central venous catheterization (7F) in the right central vein, propofol and remifentanil were used for intravenous maintenance. Radiotherapy was conducted after completing preoperative preparation, which verified the apparent pressure gradient between the pulmonary vein and left atrium (Table 1). The pressure of each part before and after balloon dilatation, left inferior and superior pulmonary vein, and right inferior pulmonary vein stenosis. Before balloon dilatation, epinephrine (0.05 µg/kg/min) was added and noradrenaline was reduced. The left superior pulmonary vein was selected as the first site of intervention. It had the most stenosis and with almost complete occlusion as well as mild interference to the blood circulation. The furosemide (5 mg) was twice administrated intravenously before the procedure of left superior and left inferior pulmonary vein balloon dilatation was performed to prevent the preload of left heart failed increasing dramatically after pulmonary vein blood flow recovery. Three metal stents (I. D. =18 mm, 10 mm, 10 mm) were implanted respectively after each balloon dilatation. Vital signs were maintained stable intraoperatively, and the surgery had lasted for 4 hours. During the surgery, the fluid supplement was 900 mL, and the urine output was 1000 mL. Postoperative ultrasonic cardiomogram revealed no pericardial effusion, no accelerated pulmonary vein stent blood flow, mild tricuspid regurgitation, and normal left

| Pressure of each part before and after balloon dilatation. |
|------------------------------------------------------------|
| **Position** | **Pressure before balloon dilatation, mmHg** | **Pressure after balloon dilatation, mmHg** |
|--------------|-------------------------------------------|-------------------------------------------|
| LA           | 12/6/3                                     | 17/10/6                                   |
| RV           | 45/15/2                                    | 44/15/1                                   |
| SVC          | 10/7/5                                     | 9/6/4                                     |
| LSPV         | 22/17/12                                   | 19/13/10                                  |
| LSPA         | 37/19/10                                   | 35/19/12                                  |

LA = left atrium, LSPA = left superior pulmonary artery, LSPV = left superior pulmonary vein, PA = pulmonary artery, RV = right ventricle, SVC = superior vena cava.
ventricular systolic function. The vasoactive agents were gradually discontinued, and the tracheal tube was extubated after the patient awake. The patient was transferred to cardiac care unit (CCU) and discharged 7 days later after rehabilitation.

4. Discussion

4.1. Etiology and pathophysiological changes of pulmonary vein stenosis

Pulmonary vein stenosis is a rare cardiovascular-pulmonary deformity with extremely high mortality. A patient may develop stenosis in 1 or multiple vessels. The site of stenosis may be located in the vein–artery junction or left atrial pulmonary vein. Alternatively, it may also extend into the pulmonary parenchyma. The stenosis can be divided into primary and secondary types based on its etiology.\textsuperscript{31} The primary type refers to the simple congenital pulmonary vein stenosis (accounting for 0.4% of congenital heart disease). Meanwhile, it can be complicated with other cardiovascular abnormalities, such as septal defects, transposition of the great arteries, and total anomalous pulmonary venous connection (TAPVC). The secondary pulmonary vein stenosis is commonly observed as the pulmonary vein opening stenosis, which is induced by the atrial fibrillation radiofrequency ablation (early pulmonary vein isolation is circumferential ablation within the pulmonary vein opening; therefore, it is likely to form pulmonary vein stenosis when the ablation site is too deep),\textsuperscript{6,7} circumferential scarred stenosis of pulmonary vein anastomotic stoma after TAPVC, right pulmonary vein stenosis after Fonton surgery, and nodule disease. Moreover, the site of stenosis may be found in the pulmonary vein–artery junction and left atrial opening of the pulmonary veins. The pathophysiological manifestations include elevated local pulmonary wedge pressure and pulmonary venous pressure. The pulmonary capillary–pulmonary venous pressure (Pv) of 10 mmHg (1.3 kPa) is de
defined as the pulmonary hypertension; the pulmonary venous pressure of 18 mmHg is deemed as the pulmonary congestion, and that of 20 to 25 mmHg (3.3 kPa) may result in the plasma leakage, leading to the pulmonary interstitial edema. Furthermore, if the pressure continues increasing over 25 mmHg, the pulmonary alveolar edema may happen. In some severe cases, the pressure may even rise to 5 to 45 mmHg (4.6–6 kPa). In addition, without timely corrections, increased right heart afterload, the formation of pulmonary hypertension, and mismatched ventilation/perfusion may occur at the advanced stage, resulting in the refractory situation. Besides, long-term pulmonary vein stenosis will also induce less bloodstream and vital oxygen supplies in the left heart, which reduces the left heart preload and causes left heart disused atrophy.

4.2. Anesthesia preparation

Given that such patients may present a variety of cardiopulmonary-related complications and fierce circulatory fluctuations intraoperatively, some additional cautions and preparations should be done before the conventional general anesthesia, including vasoactive drugs (epinephrine, noradrenaline, nitroglycerin and milrinone), defibrillation electrode in vitro, and the ECMO.

4.3. Anesthesia management

4.3.1. Monitoring. Invasive blood pressure, central venous pressure, blood gas analysis, as well as trans-thoracic or transesophageal ultrasound monitoring, should be conducted during perioperative and intraoperative periods, other than the conventional monitoring.

4.3.2. Intraoperative management. Normal partial pressure of carbon dioxide (pCO\textsubscript{2}) level should be maintained, and hypoxemia should be corrected by modulating the administration of O\textsubscript{2} to avoid further increase in pulmonary arterial pressure. In addition, acute pulmonary arteriovenous rupture hemorrhage, acute increase in right heart after-load, acute right heart failure, transient ST segment elevation, blood clot detachment, pulmonary vein laceration-induced hemothorax, left atrial opening rupture of pulmonary vein-induced acute cardiac tamponade, and left heart failure may occur during and after balloon dilatation. Simultaneously, airway pressure should be monitored closely and prompt suction is necessary to remove the airway secretion, sustain the respiratory tract unobstructed and maintain the normal pCO\textsubscript{2} level. In the meanwhile, the ECMO should be immediately performed before the occurrence of any complications, such as pulmonary vein rupture and pericardial tamponade, and the patient should be transferred to the thoracotomy immediately.

Furthermore, long-term pulmonary vein stenosis will induce to the reduced left heart preload, which may be further reduced during the balloon dilatation, resulting in decreased cardiac output and hypotension. Therefore, intraoperative hypotension should be managed temporarily using vasoactive prescriptions. Pulmonary ventilation resistance should be closely monitored during and after the stent implantation, so as to prevent the sharp decreasing of the right heart after-load, which may result in the excessive pulmonary perfusion, leading to acute and severe pulmonary effusion. Meanwhile, the respiratory parameters should be well set, and the positive end-expiratory pressure should be properly added to reduce the possibilities of pulmonary effusion. The left heart function should be carefully monitored by using cardiaogram ultrasonic to avoid significant increasing of the left heart preload after dilatation, which may induce acute left heart failure and malignant arrhythmia. Specifically, small dose applications of epinephrine and furosemide have the ability to enhance myocardial contractility, reduce the blood volume, and prevent the occurrence of acute heart failure in this period. In addition, ECMO should be placed timely when respiratory circulation cannot be maintained intraoperatively and the regular medications failed to ameliorate the symptoms.

5. Conclusion

The key points in the anesthesia management for such patients reside in the perioperative airway, circulatory management, as well as the risk evaluation, and the effective administration of all potential complications intraoperatively. Therefore, multidisciplinary cooperation under general anesthesia will undoubtedly benefit such patients.

Author contributions

Conceptualization: Jin Liu.
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