Acute thrombus in pulmonary trunk after autologous pericardial revascularization: a case report

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Case Report

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Abstract

**Background:** the occurrence of thrombi after revascularization procedures in patients with primary pulmonary leiomyosarcoma (PPL) is rare.

**Case presentation:** We report the case of a patient with PPL who developed an acute thrombus in the pulmonary trunk after revascularization with autologous pericardium. The patient was a 39 year old woman diagnosed with PPL, in order to preserve the patient's more pulmonary function, the first surgery was performed with resection and revascularization of the upper lobe of the left lung, resulting in postoperative thrombosis of the pulmonary trunk. We consider that the appearance of thrombus is related to tumor, operation, materials and postoperative management measures. After that we had to resect the patient’s remaining lower lobe of the left lung with no significant event. The patient was discharged without postoperative complications.

**Conclusions:** We report a case of a patient with PPL who developed a thrombus after revascularization.

Introduction

Primary pulmonary leiomyosarcoma (PPL) is a rare malignancy. The revascularization procedure in our case was an effective treatment for lung cancer invading the pulmonary vasculature, trachea, and the pulmonary sleeve resection was performed in order to preserve more lung function. Artificial materials for vascular reconstruction have used autologous pericardium, compared with artificial blood vessels without efflux reaction, good histocompatibility, rarely hemolysis or thrombus occurs, but after surgery unexpectedly pulmonary thrombus appears, presumably related to the patient's tumor, surgical operation, the choice of materials, and the use of anticoagulant drugs. The left total lung resection was performed again after timely diagnosis, and the patient was converted to safety. In this report, we describe our pathological observations and review the literature in order to improve our understanding of such operations and provide evidence for its clinical treatment and prognosis.

Case Presentation

A 39-year-old woman visited the hospital because of cough and expectoration accompanied by throat discomfort. The pulmonary CT (computed tomography) showed a soft tissue density shadow about 41 * 56 mm in size in the left upper lung, and a small adjacent outer margin nodule was considered as tumor infiltration (Fig.1). After excluding relevant contraindications, we implemented percutaneous lung biopsy for the patient, and the postoperative pathological result was leiomyosarcoma. The patient refined abdominal CT, brain magnetic resonance imaging (MRI) as well as whole body bone imaging and other related examinations, which did not reveal the presence of distant metastasis. Based on the patient's medical history, testing as well as imaging studies, the patient was considered to have a diagnosis of left upper lung malignancy (leiomyosarcoma), cT3N0M0, IIB stage. After multidisciplinary discussion,
combined with the patient's own status, we planned to implement left upper pulmonary sleeve resection and pulmonary arterioplasty for the patient. If the left pulmonary trunk was involved and the left lower lung could not be preserved, a total left pneumonectomy was performed. Intraoperative exploration showed: left lung adhesive thorax, after separating adhesions see left lung upper lobe mass involving anterior chest wall, adventitia of aortic arch, phrenic nerve. After isolation of the lingual pulmonary artery ligation, the pericardium was opened, the affected and phrenic nerves were transected, the left pulmonary artery trunk and superior pulmonary vein were isolated, and the superior pulmonary vein was closed and transected. The root and interlobar trunk of the left pulmonary artery were clamped, and the vein was anticoagulated with half dose heparin. The left upper lobe bronchus was cut, the tumor involved part of the left pulmonary trunk, the adventitia of the aorta and the vagus nerve, and resection could not be preserved. The involved pulmonary trunk was resected and a vascular anastomosis was performed with autologous pericardium to connect the proximal and distal pulmonary trunk, and a heparinized saline flush was performed before knotting to exclude intravascular thrombus. The involved upper lobe bronchus was resected by sleeve dissection, and the left main bronchus was anastomosed to the lower lobe bronchus. Hilar and mediastinal lymph nodes were dissected, the chest was thoroughly irrigated by hemostasis, exudates were net aspirated, and the wound was closed by suturing after placement of a drain. The anticoagulant medication used by the patient after the procedure was dalteparin sodium injection [0.3ml: 7500AXaIU] 5000IU every 12 hours.

However, on the sixth postoperative day, the patient had dyspnea with fever, after undergoing emergency CT found that the left pulmonary artery and its branches were not shown, pulmonary artery embolism, left lower lobe infarction, pulmonary infection were considered (Fig.2). After urgent discussion, considering the patient had infarction in the lower lobe of the left lung and loss of lung function, the patient was given an emergency resection of the lower lobe of the left lung, and the thrombus was cleared. Intraoperatively, the left lung was found to be mildly adherent to the chest wall, left lower lung consolidation was visible after dissociation, the left lower pulmonary vein was cut by ligation, the free left pulmonary artery trunk, after root block cutting, the distal thrombosis was seen, the left proximal pulmonary artery was sutured, the bronchus was cut, and the proximal bronchus fracture was sutured, the left lower lung was completely removed. After the operation, after anti infection and nebulization treatment, the patient was breathing better, no fever, cough and other discomfort. The patient was finally diagnosed with left lung malignant tumor (leiomyosarcoma), pT3N0M0, IIB stage (Fig.3). Postoperative treatment was performed on an outpatient basis 3 weeks after discharge. On follow-up visit 4 months after discharge, the patient did not complain of special discomfort.

Discussion

PPL is a rare lung malignancy that can be asymptomatic at an early stage, and common symptoms that can appear as the mass increases are cough, hemoptysis, chest pain, and chest tightness[1], but generally mild. When causing atelectasis or secondary infection, they can have symptoms such as high
fever, chest pain, chest tightness, and dyspnea. Fiberoptic bronchoscopy and percutaneous lung biopsy allow a definite diagnosis of this disease.

Early surgery is an effective treatment for PPL, and the 5-year survival rate of early surgery can reach more than 50%[2]. Local invasion and hematogenous metastasis can occur in this disease, but hilar or mediastinal lymph node metastasis generally does not occur. The surgical procedure can be performed according to the tumor location and size by segmented, lobar, whole lung, or sleeve resection, and if necessary, wide excision of the locally invaded chest wall, large blood vessels, and so on.

In the present case, the tumor tissue invaded the main pulmonary artery trunk, and the main bronchus was also involved, and the patient's normal ventilation as well as pulmonary blood gas exchange would have been greatly limited if a total resection of the affected side had been performed. However, with effective pulmonary artery reconstruction, avoidance of total lung resection, and preservation of ventilatory function of the affected side's lungs as much as possible as well as the area of the lungs for blood gas exchange, can maximize patient vital capacity improvement, better postoperative pulmonary function recovery, and reduce the incidence of postoperative complications. The material connecting the proximal and distal ends of the main trunk of the left pulmonary artery during surgery was an autologous pericardium. Studies have shown that if patch transplantation is needed for pulmonary artery repair, pericardial patch is the first choice for left lung reconstruction, which has good histocompatibility and is easy to obtain, and can meet the needs of surgery[3]. But on the sixth postoperative day, the patient developed a thrombus in the left pulmonary artery and the left lower lobe was infarcted and could not be preserved, so a second surgery was performed to remove the left lower lobe of the lung. The occurrence of a thrombus was an unexpected situation, which undermined purposes such as preserving more lung function for the first surgery, so we undertake exploratory analyses in various aspects to identify possible causes of the thrombus formation:

1. Tumor. Firstly, tumor directly leads to activation of the coagulation system in tumor patients, while lung cancer belongs to the group of cancers with the highest rate of thrombosis[4]. Patients with lung cancer had higher WBC count, CRP level, and blood coagulation and fibrinolysis parameters, including platelet count, FDP, fibrinogen, thrombomodulin, TAT, and D-dimer, compared to healthy people[5]. The second is that the tumor patients are bed ridden for a long time with a reduced amount of mobility, which leads to hemagglutination and easily forms deep vein thrombus, and they can easily develop pulmonary embolism if shedding occurs. Compared with patients without malignancy, patients with malignancy had a 7-fold increased overall risk of venous thrombosis, with the highest risk in patients with hematologic malignancies, followed by lung and gastrointestinal cancers, with the highest risk of venous thrombosis in the first few months after diagnosis of malignancy[6]. It is important to note that the location of the thrombus seen in the patient in our case was distal to the left pulmonary trunk and not at the finer arterial branches, so deep vein thrombi are less likely. And the hypercoagulable state of the blood might be one of the reasons for the thrombus in this patient.
2. Surgery. It is inevitable to compress blood vessels, destroy blood vessel walls or endothelium during surgery, and the vessel wall after injury easily activates the endogenous and exogenous coagulation system, resulting in the hypercoagulable state of blood. Early surgery increases the cure rate for patients with early-stage tumors without contraindications to surgery, but the risk of developing fatal pulmonary thromboembolism (PTE) after surgery is 2-fold or even higher for patients with malignant tumors than for those with conventional surgery[7]. Revascularization procedure in the case, a vessel made using an autologous pericardium connected the proximal to the distal end of the pulmonary trunk, but since the caliber of the pulmonary trunk is thicker, the larger the radius of the flow tube and the more likely turbulence will occur according to Reynolds number formula. If turbulent flow occurs, the direction of fluid movement in adjacent flow layers is staggered longitudinally, then it generates a pulsatile high shear stress in the flow tube section, which makes the vessel more distensible than normal segments, thereby damaging the vascular endothelium, increasing the permeability of the tube wall, causing the necrosis, calcification, lipid infiltration and formation of partial accumulation, etc., and eventually the formation of a thrombus. If the suture technology is not perfect, the suture will be uneven and the interface with the blood vessel will be relatively rough, which will also lead to turbulence. If the reconstructed pulmonary artery is too long, it may lead to kinked vessels, impaired blood flow, and eventually thrombus formation[8].

3. Materials. Autologous pericardium used in surgery is a good vascular material and it has the following advantages: (i) It is so easily accessible that the corresponding pericardial patch was cut out according to the shape and size of the vascular defect; (ii) It has good plasticity and is easy to suture and anastomose. (iii) Due to good tissue compatibility, there is no tissue rejection. (iv) This technique maintains cell activity, involves minimal chances of infection[9]. In addition, shortly after implantation, grafts re epithelialize in humans with autologous epithelial cells, which is associated with a low risk of infection, reduced platelet deposition, and reduced thrombogenicity of flow surfaces[10]. However, fresh autologous pericardium also has some limitations because of its obvious shrinkage and curling, making the adaptation of the patch to vascular wall defects more difficult. In addition, it is also difficult to suture the patch on the vessel because of the potential for uneven bite and bleeding sites. This may lead to the formation of a thrombus[11].

4. Postoperative management measures. Low molecular weight heparin (LMWH) is one of the anticoagulants of choice for the prevention and treatment of venous thrombosis[12]. One of the major contributors to thrombosis is the activation of thrombin, and LMWH has a strong anti Xa effect, which can inhibit the activation of thrombin and then act as an antithrombotic agent[13]. LMWH has more potent antithrombotic and weaker anticoagulant effects than unfractionated heparin (UFH), largely reducing the risk of bleeding. A meta-analysis of randomized controlled trials showed similar efficacy between LMWH and UFH for acute deep venous thrombosis prevention, and no bleeding risk difference[14] with a reduced mortality rate in favor of LMWH. The major appealing feature of LMWH is the more predictable relationship betweendose and response with LMWH than UFH, which translates to weight-adjusted dosing without laboratory monitoring. A meta-analysis of randomized controlled trials showed similar efficacy between LMWH and UFH for acute deep venous thrombosis prevention, and no bleeding risk difference with a reduced mortality rate in favor of
LMWH[15]. However, a previous meta-analysis showed that there was no statistically significant difference in the incidence of bleeding events between the anti-Xa monitoring group and the control group, while the incidence of venous thromboembolism events in the anti-Xa monitoring group was significantly lower than that in the control group, suggesting that patients receiving LMWH anticoagulation treatment to prevent VTE could benefit from anti-Xa monitoring[16]. Therefore, the future clinical treatment should determine the dose of LMWH by anti-Xa monitoring based on the weight of patients.

In this case, the most likely cause of the thrombus formation distal to the pulmonary trunk was the surgical manipulation, but it was not the only cause of this, a combination of factors contributed to the thrombus appearance. In order to avoid the occurrence of postoperative thrombus, we should pay more attention to the tumor patients, pay attention to the operation of revascularization during the operation, strictly put the diameter of the control vessel, use postoperative anticoagulation drugs properly for the patients and always carry out the monitoring of FIB, D dimer and other markers. In order to avoid the occurrence of postoperative thrombosis, we should pay attention to tumor patients, pay attention to the operation of revascularization during operation, strictly control the diameter of blood vessels, use anticoagulants appropriately after operation, and often monitor FIB, D dimer and other indicators.

PTE is life-threatening, and approximately one-third of patients with undiagnosed and untreated pulmonary embolism do not survive. But when the condition is promptly diagnosed and treated, the survival rate rises. Therefore, prompt diagnosis is the key to the management of this disease. Patients presenting with unexplained chest tightness, cough, and even syncope need to think of the possibility of pulmonary embolism. Coagulation factor test, blood gas analysis, electrocardiogram, as well as cardiac color ultrasound can assist the diagnosis, and chest enhanced CT can detect thrombi in the pulmonary arteries above the segment visible as low-density filling defects in the pulmonary arteries. In addition, the recurrence rate of pulmonary embolism is higher than that of other diseases, and the recurrence of pulmonary embolism is mostly at 6 to 12 months after treatment, and is higher in men than in women, with the highest recurrence rate in the malignant tumor population.

**Conclusion**

In conclusion, PPL is a less common lung malignancy, and angioplasty with autogenous pericardium is an effective treatment for lung cancer invading the pulmonary vasculature, expanding the indications for surgery, avoiding left pneumonectomy, and achieving lung preservation. Improper handling about lung tumors, surgical procedures, vascular materials and postoperative handling are highly likely to generate pulmonary thrombi. The situation that can be prevented if adequate precautions are in place. The case is a profound lesson learned. Suspicious indications for pulmonary embolism should be made immediately related examinations, diagnosed early, and treated accordingly.

**Abbreviations**
PPL
Primary pulmonary leiomyosarcoma
CT
computed tomography
MRI
magnetic resonance imaging
WBC
White blood cell
CRP
C-reactive protein
FDP
Fibrinogen degradation products
TAT
ThrombinAT- complex
PTE
Pulmonary thromboembolism
LMHW
Low molecular weight heparin
UFH
Unfractionated heparin
VTE
Venous thromboembolism
FIB
Fibrinogen

Declarations

Ethical Approval and consent to participate:

The study was approved by the ethics committee of Zhongnan Hospital of Wuhan University. Written informed consent was obtained from the participants for publication of the details of their medical case and any accompanying images.

Competing interests:

The authors declare no competing interests.

Authors' contributions:

Ma Shenglong and Guo Zixin wrote the main manuscript text and Ma Shenglong prepared figures 1-3. All authors reviewed the manuscript.
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References


Figures

**Fig. 1**

The pulmonary CT (computed tomography) showed a soft tissue density shadow about 41 * 56 mm in size in the left upper lung, and a small adjacent outer margin nodule was considered as tumor infiltration.

- **a.** lung window.
- **b.** mediastinal window
Left pulmonary artery and its branches were not shown, pulmonary artery embolism, left lower lobe infarction, pulmonary infection were considered. **a.** pulmonary CT enhanced scan. **b.** pulmonary artery CT angiography

**Figure 2**

See image above for figure legend
Pathological stage pT3N0Mx, 16 lymph nodes in four groups showed no cancer metastasis

Figure 3

See image above for figure legend