

## Case Report

## A ruptured pulmonary arteriovenous fistula after laparoscopic operation

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## ABSTRACT

Pulmonary arteriovenous fistula (PAVF) is a rare anomaly in the lung, and hemothorax or massive hemoptysis due to spontaneous rupture of the fistula sac is even rarer. The patient described here was a 47-year-old woman who presented with massive hemoptysis resulting from the rupture of her PAVF just after laparoscopic operation. To our knowledge, this may be the first case ever reported that the rupture of PAVF may be correlated with a laparoscopic operation. The patient survived without adverse events after emergency pulmonary lobectomy.

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## Introduction

Pulmonary arteriovenous fistula (PAVF) is a rare abnormal vascular structure. The clinical manifestations and complications include hypoxemia, stroke, transient ischemic attack, massive hemoptysis, spontaneous hemothorax, dyspnea and cyanosis. PAVF hemorrhage leading to massive hemoptysis and hemothorax is a relatively rare feature but can be life-threatening. The two main exceptions are if the patient is pregnant or if the PAVF is perfused at systemic pressure.<sup>1</sup> Here we report a case that presented as massive hemoptysis resulting from the rupture of PAVF just after laparoscopic operation. To our knowledge, this may be the first case ever reported that the rupture of PAVF was correlated with laparoscopic operation.

## Case report

A 47-year-old woman, who had no symptoms in lung, was admitted to our hospital because of cholelithiasis and received laparoscopic cholecystectomy. An abnormal shadow was detected in the right lower lobe on her chest X-ray by routine test before operation. Chest computed tomography (CT) revealed a 30 mm × 25 mm nodule with defined margins and smooth contours in the lobe, and found the feeding artery and draining vein

connecting the nodule (Fig. 1). A continuous murmur was heard in the area near the medial margin of right subscapular angle. A diagnosis of PAVF was made before the operation but this abnormal finding unfortunately did not arouse great attention of the surgeons due to its benevolent features. Laparoscopic cholecystectomy was performed under general anesthesia. Sudden massive hemoptysis occurred about 5 h after the operation, and the amount of hemoptysis was about 700 ml in the first 3 hours. We took an emergency consultation for the patient. Bedside chest X-ray examination revealed total pulmonary atelectasis in the right lower lobe. Chest CT scan showed diffuse consolidation of the whole right lower lobe with uneven density in it (Fig. 2). The diagnosis of ruptured PAVF was confirmed immediately. An emergency right posterolateral thoracotomy was performed. During the operation, a dark red swollen right lower lobe was visible, which was full of blood and the tension of the visceral pleura increased significantly (Fig. 3A). There was no blood in the thoracic cavity. The right lower lobe was completely resected. When the resected specimen was dissected, a great deal of blood came out of the incision. There was a thin-walled cavity about 6 cm in diameter under the visceral pleura, which was filled with blood clots (Fig. 3B). It was estimated that the total amount of bleeding was about 2000 ml including the amount of hemoptysis. The cyst was disconnected from the hilus of the lung and its inside wall was not smooth. There were no normal pulmonary tissue found in the whole lobe. Microscopically, diffuse hemorrhaging and obvious tissue necrosis were observed in the alveolar space and within interstitial pulmonary tissue (Fig. 3C). The patient survived and postoperative recovery was uneventful.

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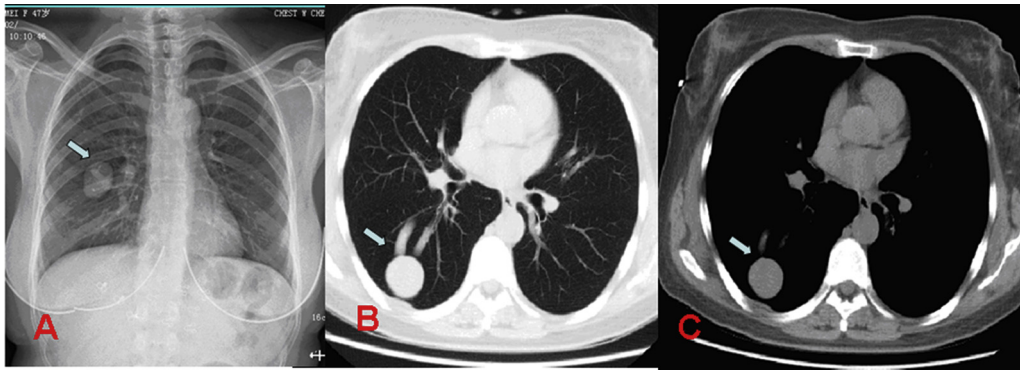


Fig. 1. Pulmonary arteriovenous fistula (PAVF) before rupture. A: Chest X-ray. B&C: Computed tomography.

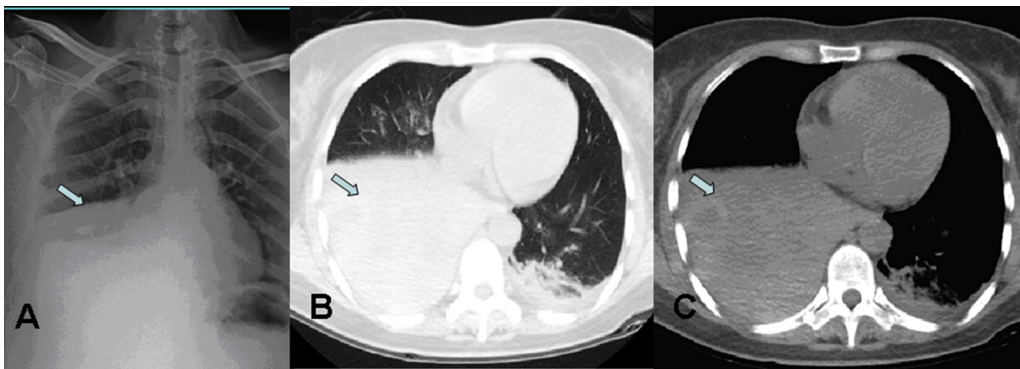


Fig. 2. Pulmonary arteriovenous fistula (PAVF) after rupture. A: Bedside chest X-ray. B&C: Computed tomography.

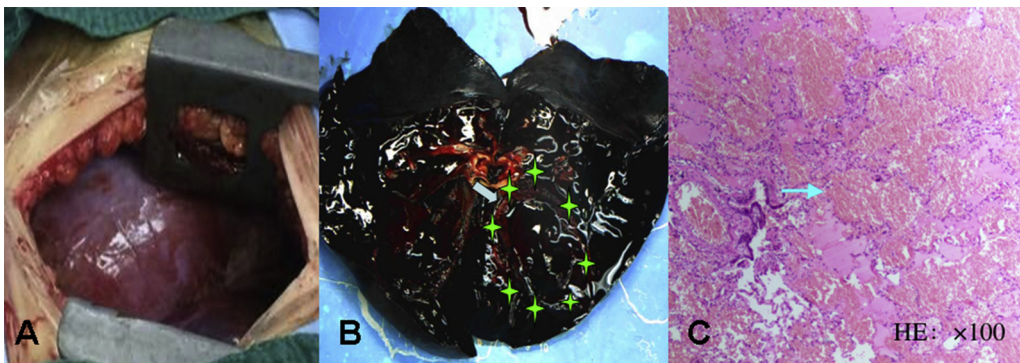


Fig. 3. A: Picture at operation. B: The cross section of resection specimen. PAVF sac indicated by arrow (the margin labeled by green stars). C: Microscopic examination of the resected tissue demonstrates tissue necrosis.

## Discussion

PAVF is a rare vascular malformation and represents abnormal communications between an artery and a pulmonary vein that allows blood to bypass the pulmonary capillary bed, resulting in an intrapulmonary right-to-left shunt. PAVF may be single or multiple, unilateral or bilateral, and simple or complex. Most PAVFs are congenital, and their precise etiology is unknown. A strong association is noted between hereditary hemorrhagic telangiectasia (HHT) and PAVF. Acquired PAVFs are less common, and the possible causes include infections, such as schistosomiasis and actinomycosis, trauma, and Fanconi syndrome.<sup>1,2</sup>

Clinical features found in PAVFs are not uniform. Signs and symptoms of patients with PAVFs vary depending on the size,

number, and flow through the PAVF. Patients may be completely asymptomatic or experience dyspnea on exertion. The most dangerous complications, especially the rupture of PAVF leading to massive hemoptysis and hemothorax, are sometimes life-threatening. Spontaneous rupture of fistula sac is rare and usually occurs in pregnancy because of the increased blood volume, cardiac output, and venous distensibility during this special period.<sup>3</sup> Worsening of PAVFs with pregnancy has been reported by multiple studies, and approximately 30% of massive hemorrhagic events occur during pregnancy.<sup>4</sup> The case we described here presented massive hemoptysis as its initial symptom. Pre-operative chest CT image showed a clear round nodule with feeding artery and draining vein in right lower lobe, but the nodule disappeared in the postoperative bedside chest X-ray film when

massive hemoptysis occurred and CT scan showed diffuse consolidation of the whole right lower lobe.

Thoracotomy demonstrated the rupture of PAVF just after laparoscopic cholecystectomy. It evoked our great interest to explore the possible causes of rupture in this case. Although we can not confirm the relationship between the rupture and laparoscopic operation, we still found some clues of the high-risk factors for the rupture of PAVF in this patient. Firstly, chest X-ray and CT scan showed us a smooth round nodule in right lower lobe, about 3 cm in diameter, clear feeding arteries and draining veins, without any notch on the surface. The appearance indicated that there was much higher tension inside the fistula cyst than those which have notches on the surface. The PAVF may rupture and bleed as its size increases quickly and its surface becomes smoother and with no notches, which are challenging to treat. Evidence is lacking on the rate of growth of PAVF and its determinants, but pregnancy and puberty have been described as potential factors that induce nodule enlargement.<sup>1,3</sup> In this case, the patient has experienced the pregnancy and puberty several years prior, and the size and features of nodule predicted an increased risk of rupture. Secondly, we note the potential influence of laparoscopic operation on the rupture of PAVF in this patient, although no such reports have been published previously. General anesthesia and endotracheal intubation must be performed during the operation, and mechanical ventilation and positive airway pressure will inevitably exert pressure on the PAVF sac. Furthermore, artificial pneumoperitoneum is necessary for laparoscopic surgery, which may put pressure on the diaphragm and decrease the volume of thoracic cavity. These two factors could lead to the variation of the pressure in the cavity and might result in the rupture of PAVF.

Therapeutic options of PAVF include percutaneous transcatheter embolization (TCE) and surgery. Surgical resection is

rarely necessary because the majority of PAVFs are amenable to TCE. Surgery is used as emergency procedure to control bleeding and when loss of lung is justified. Surgical techniques depend on the complexity of PAVFs and include local excision, segmental resection, lobectomy, ligation, and even pneumonectomy.<sup>1,5</sup> Thoracoscopic lung resection and fistula ligation are good therapeutic options for multiple small peripheral PAVFs.<sup>6</sup> In this case, we had to perform the lobectomy because the lobe was severely damaged.

In conclusion, the rupture of PAVF is rare and very dangerous. In regard to this, we should take full account of the risk factors of rupture when we face a similar situation, which might prevent the need for emergency surgical resection. Once the rupture of PAVF is suspected or confirmed, the emergency thoracotomy with wedge resection or lobectomy is the first choice to decrease mortality and recurrence rate.

## References

1. Cartin-Ceba R, Swanson KL, Krowka MJ. Pulmonary arteriovenous malformations. *Chest*. 2013;144:1033–1044. <http://dx.doi.org/10.1378/chest.12-0924>.
2. Ahn S, Han J, Kim HK, et al. Pulmonary arteriovenous fistula: clinical and histologic spectrum of four cases. *J Pathol Transl Med*. 2016;50:390–393. <http://dx.doi.org/10.4132/jptm.2016.04.18>.
3. Lin F, Mei J, Liu C, et al. A ruptured pulmonary arteriovenous fistula. *J Thorac Cardiovasc Surg*. 2015;150:716–717. <http://dx.doi.org/10.1016/j.jtcvs.2015.05.029>.
4. Ghelani SJ, Rathod RH. Pulmonary arteriovenous malformations: the consequences of bypassing the capillary bed. *J Thorac Cardiovasc Surg*. 2015;150:717–719. <http://dx.doi.org/10.1016/j.jtcvs.2015.05.055>.
5. Trerotola SO, Pyeritz RE. PAVM embolization: an update. *Am J Roentgenol*. 2010;195:837–845. <http://dx.doi.org/10.2214/AJR.10.5230>.
6. Sano A, Tsuchiya T. Thoracoscopic surgery for multiple peripheral pulmonary arteriovenous fistulas. *Ann Thorac Surg*. 2015;99:1808–1809. <http://dx.doi.org/10.1016/j.athoracsur.2014.08.047>.