

World Journal of *Clinical Cases*

World J Clin Cases 2023 October 6; 11(28): 6670-6973



MINIREVIEWS

- 6670 Neurotransmitters regulate β cells insulin secretion: A neglected factor
Kong CC, Cheng JD, Wang W

ORIGINAL ARTICLE

Case Control Study

- 6680 Factors influencing the surveillance of re-emerging intracranial infections in elective neurosurgical patients: A single-center retrospective study
Wang JL, Wu XW, Wang SN, Liu X, Xiao B, Wang Y, Yu J

Retrospective Study

- 6688 Clinical value of chemiluminescence method for detection of antinuclear antibody profiles
Xiang HY, Xiang XY, Ten TB, Ding X, Liu YW, Luo CH
- 6698 Value of ultrasound guided biopsy combined with Xpert *Mycobacterium tuberculosis*/resistance to rifampin assay in the diagnosis of chest wall tuberculosis
Yan QH, Chi JY, Zhang L, Xue F, Cui J, Kong HL
- 6707 Research on the intelligent internet nursing model based on the child respiratory and asthma control test scale for asthma management of preschool children
Pei CF, Zhang L, Xu XY, Qin Z, Liang HM
- 6715 Effects of different doses of long-acting growth hormone in treating children with growth hormone deficiency
Xia W, Wang T, Pan JY
- 6725 Efficacy and anti-inflammatory analysis of glucocorticoid, antihistamine and leukotriene receptor antagonist in the treatment of allergic rhinitis
Qiu C, Feng D
- 6733 Subchondral fatigue fracture of the femoral head in young military recruits: Potential risk factors
Yang JZ, Chen P, Chen BH, Zhao B
- 6744 Anemia status of infants and young children aged six to thirty-six months in Ma'an Shan City: A retrospective study
Wang XM, Wang QY, Huang J
- Observational Study
- 6754 Impact of coronary artery bypass grafting surgery on the chorioretinal biomicroscopic characteristics
Shahriari M, Nikkhah H, Mahjoob MP, Behnaz N, Barkhordari S, Cheraqpour K

Prospective Study

- 6763** Effects of humanized nursing care on negative emotions and complications in patients undergoing hysteromyoma surgery
Liu L, Xiao YH, Zhou XH

Randomized Controlled Trial

- 6774** Randomized controlled trial on the efficacy and safety of autologous serum eye drops in dry eye syndrome
Zheng N, Zhu SQ

SYSTEMATIC REVIEWS

- 6782** Primary adrenal Ewing sarcoma: A systematic review of the literature
Manatakis DK, Tsouknidas I, Mylonakis E, Tasis NP, Antonopoulou MI, Acheimastos V, Mastoropoulou A, Korkolis DP

CASE REPORT

- 6792** Pulmonary artery aneurysm protruding into the bronchus as an endobronchial mass: A case report
Li M, Zhu WY, Wu RR, Wang L, Mo MT, Liu SN, Zhu DY, Luo Z
- 6797** Rare rectal gastrointestinal stromal tumor case: A case report and review of the literature
Dong RX, Wang C, Zhou H, Yin HQ, Liu Y, Liang HT, Pan YB, Wang JW, Cao YQ
- 6806** Bilateral retinal nerve fiber layer thickness reduction in a 9-year-old myopic boy suffering from unilateral optic neuritis: A case report
Zhao FF, Yao SQ, Wang Y, Li TP, Yang JF, Pang CP, Cen LP
- 6812** Application of negative pressure wound therapy after skin grafting in the treatment of skin cancer: A case report
Huang GS, Xu KC
- 6817** Diagnosis and treatment of McCune-Albright syndrome: A case report
Lin X, Feng NY, Lei YJ
- 6823** Paraneoplastic myopathy-related rhabdomyolysis and pancreatic cancer: A case report and review of the literature
Costantini A, Moletta L, Pierobon ES, Serafini S, Valmasoni M, Sperti C
- 6831** Multi-organ hereditary hemorrhagic telangiectasia: A case report
Chen YL, Jiang HY, Li DP, Lin J, Chen Y, Xu LL, Gao H
- 6841** Hyperprogression after anti-programmed death-1 therapy in a patient with urothelial bladder carcinoma: A case report
Yang HY, Du YX, Hou YJ, Lu DR, Xue P
- 6850** Effectiveness of antidepressant repetitive transcranial magnetic stimulation in a patient with refractory psychogenic dysphagia: A case report and review of literature
Woo CG, Kim JH, Lee JH, Kim HJ

- 6857** Entrapment neuropathy of common peroneal nerve by fabella: A case report
Lin JC, Tsai MH, Lin WP, Kuan TS, Lien WC
- 6864** Importance of accurate diagnosis of congenital agenesis of the gallbladder from atypical gallbladder stone presentations: A case report
Sun HJ, Ge F, Si Y, Wang Z, Sun HB
- 6871** Dorsal approach for isolated volar fracture-dislocation of the base of the second metacarpal: A case report
Kurozumi T, Saito M, Odachi K, Masui F
- 6877** Rotationplasty type BIIIb as an effective alternative to limb salvage procedure in adults: Two case reports
Chen ZX, Guo XW, Hong HS, Zhang C, Xie W, Sha M, Ding ZQ
- 6889** Primary cutaneous anaplastic large cell lymphoma with over-expressed Ki-67 transitioning into systemic anaplastic large cell lymphoma: A case report
Mu HX, Tang XQ
- 6895** Confusing finding of quantitative fluorescent polymerase chain reaction analysis in invasive prenatal genetic diagnosis: A case report
Chen C, Tang T, Song QL, He YJ, Cai Y
- 6902** Testicular mixed germ cell tumor: A case report
Xiao QF, Li J, Tang B, Zhu YQ
- 6908** Leukemic transformation during anti-tuberculosis treatment in aplastic anemia-paroxysmal nocturnal hemoglobinuria syndrome: A case report and review of literature
Xiu NN, Yang XD, Xu J, Ju B, Sun XY, Zhao XC
- 6920** Pancreatic arteriovenous malformation treated with transcatheter arterial embolization: Two case reports and review of literature
Shin SH, Cho CK, Yu SY
- 6931** Cecal duplication cyst in an infant presenting as shock: A case report
Kim SM, Lee SH, Park GY, Kim SS, Lee CG, Jin SJ
- 6938** Pulmonary reversed halo cycles and consolidations after immunotherapy: A case report
Suo H, Shi YJ, Huang ZD, Xu K, Huang H
- 6943** Unusual case of emphysematous cystitis mimicking intestinal perforation: A case report
Kang HY, Lee DS, Lee D
- 6949** Malignant proliferative ependymoma of the neck with lymph node metastasis: A case report
Wang K, Wen JZ, Zhou SX, Ye LF, Fang C, Chen Y, Wang HX, Luo X
- 6955** Wandering spleen torsion with portal vein thrombosis: A case report
Zhu XY, Ji DX, Shi WZ, Fu YW, Zhang DK

- 6961** Intracranial infection and sepsis in infants caused by *Salmonella derby*: A case report
Yu JL, Jiang LL, Dong R, Liu SY
- 6967** Large gastric hamartomatous inverted polyp accompanied by advanced gastric cancer: A case report
Park G, Kim J, Lee SH, Kim Y

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Pulmonary artery aneurysm protruding into the bronchus as an endobronchial mass: A case report

Min Li, Wen-Ye Zhu, Rong-Rong Wu, Le Wang, Meng-Ting Mo, Shi-Nan Liu, Dong-Yi Zhu, Zhuang Luo

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Abstract

BACKGROUND

Pulmonary artery (PA) aneurysms are usually diagnosed radiographically and present as small or large lesions resembling inflammation or a neoplasm on chest radiography. It has rarely been reported as an endobronchial mass.

CASE SUMMARY

We report the case of a 64-year-old man who presented with recurrent hemoptysis. Bronchoscopy revealed a tumorous protrusion blocking the right middle lobe bronchus, which was confirmed to be a PA aneurysm using endobronchial ultrasound bronchoscopy and computed tomography angiography.

CONCLUSION

Although endobronchial PA aneurysms are rare, bronchoscopists need to add this lesion to the list of endobronchial masses for which a biopsy is to be assiduously avoided.

Key Words: Pulmonary artery aneurysm; Endobronchial mass; Endobronchial ultrasound bronchoscopy; Computed tomography angiography; Case report

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Core Tip: Pulmonary artery (PA) aneurysms usually present as small or large lesions resembling inflammation or a neoplasm on chest radiography. It is rarely reported as an endobronchial mass. We report a case of a tumorous protrusion blocking the right middle lobe bronchus, which was confirmed to be a PA aneurysm by endobronchial ultrasound bronchoscopy and computed tomography angiography. Although endobronchial pulmonary arterial aneurysm is rare, bronchoscopists need to enhance their awareness and vigilance of this lesion in order to avoid the disastrous consequences caused by improper biopsy.

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INTRODUCTION

Pulmonary artery (PA) aneurysms are uncommon but often lethal abnormalities of the pulmonary vasculature. The clinical presentation of a PA aneurysm ranges from a silent incidental finding on imaging to severe hemoptysis[1]. Generally, most cases of PA aneurysms are diagnosed radiographically and present as small or large lesions resembling inflammation or neoplasms on chest radiography[2]. Here, we report a rare case of a PA aneurysm diagnosed using a combination of endobronchial ultrasound (EBUS) bronchoscopy and chest computed tomography angiography (CTA), in which the PA aneurysm protruded into the bronchus as an endobronchial mass that blocked the airway lumen.

CASE PRESENTATION

Chief complaints

A 64-year old male patient was admitted to our hospital due to intermittent hemoptysis for 2 wk.

History of present illness

The patient experienced severe episodes of massive hemoptysis without any premonitory symptoms, with a total blood loss of approximately 400-500 mL. Except for a slight cough, no other symptoms, such as fever, sputum production, or chest pain were observed. Before admission, the patient was treated with antibiotics and hemostatics at another hospital. However, the patient's clinical symptoms were not completely resolved.

History of past illness

The patient had no significant past medical or surgical history.

Personal and family history

The patient had no significant personal or family history.

Physical examination

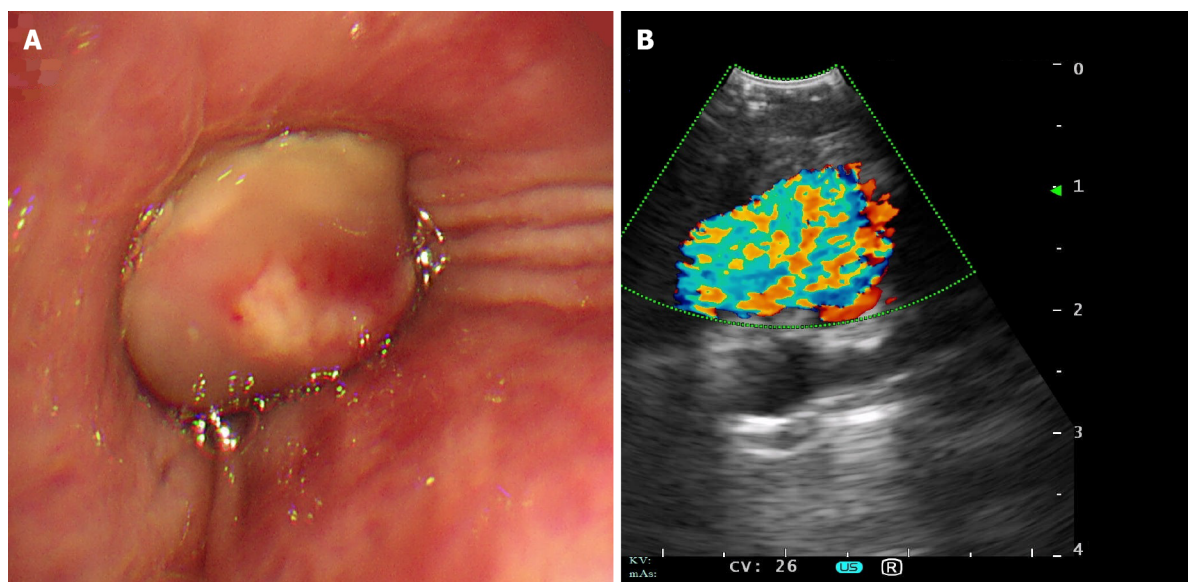
At our hospital, the patient's vital signs were normal and stable. Pulmonary auscultation revealed moist rales in the middle and lower field of the right lung.

Laboratory examinations

Laboratory tests were unremarkable except for a hemoglobin concentration of 109 g/L (normal 115-150 g/L) and D-dimer 0.69 µg/mL (normal 0-0.5 µg/mL).

Imaging examinations

High-resolution computed tomography of the chest revealed partial exudation and consolidation of the right lung. Flexible bronchoscopy was performed to confirm the diagnosis, which showed a smooth-surfaced, flesh colored and tumorous protrusion completely blocking the right middle lobe of the bronchus (Figure 1A). Subsequently, EBUS bronchoscopy was performed to detect the mass, only to find a hypoechoic vascular lesion with a color Doppler flow signal (Figure 1B), suggesting a vascular abnormality of the bronchus. Therefore, the endobronchial biopsy was discontinued because the irritation caused by the biopsy procedure may have caused massive bleeding. CTA of the chest was also performed and revealed a right middle lobe PA aneurysm protruding into the right middle lobe bronchus (Figure 2).



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Figure 1 Bronchial examination. A: Endobronchial lesion visualized under conventional bronchoscopy. It revealed a round, smooth-surfaced mass and flesh colored tumorous protrusion completely blocking the right middle lobe bronchus; B: Endobronchial ultrasonography revealing a vascular lesion.



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Figure 2 Computed tomography angiography confirming a pulmonary artery aneurysm in the right middle lobe.

FINAL DIAGNOSIS

PA aneurysm.

TREATMENT

The patient underwent a right middle lobectomy.

OUTCOME AND FOLLOW-UP

The postoperative course was uneventful, and hemoptysis did not recur after the surgery. The patient was discharged on the seventh postoperative day.

DISCUSSION

PA aneurysms are rare and their prevalence has been estimated to be 1 in 14000 based on the findings from 109571 autopsies conducted at the Mayo Clinic[3]. Currently, PA aneurysms are categorized based on their etiology. PA aneurysms secondary to congenital heart disease have been most frequently reported, with patent ductus arteriosus, ventricular septal defect and atrial septal defects being the most frequently described causes. In addition, PA aneurysms caused by infectious diseases, such as mucormycosis, tuberculosis, and syphilis, have also been reported. PA aneurysms may also occur secondary to pulmonary hypertension, autoimmune diseases or vasculitis. Finally, PA aneurysms with no clear cause were considered idiopathic[4-6]. Generally, PA aneurysms are less often clinically evident and are usually diagnosed based on chest radiographs or at autopsies[7]. Some patients with PA aneurysms may experience shortness of breath, coughing or chest pain[8]. Hemoptysis may imply that an unstable PA aneurysm erodes the airway mucosa[9]. Spontaneous PA aneurysm rupture is rare but is, when it occurs, often a catastrophic complication of massive hemoptysis. Most PA aneurysms appear to rupture into the alveolar parenchyma and do not impinge directly on the bronchus, and they are rarely detected by bronchoscopy[4]. Here, we report a rare condition in which a PA aneurysm protruded into the bronchus as an endobronchial mass that blocked the airway lumen, providing clinical implications for the differential diagnosis and management of endobronchial lesions.

Although the incidence of tumor-like vascular malformations of the bronchus is low, it is highly risky and making the differential diagnosis from a solid endobronchial mass is extremely important. Inappropriate interventions, such as biopsy or fine-needle aspiration of vascular lesions, can have devastating consequences, and in some cases, lead to massive intra-pulmonary bleeding and even death[7,10]. Among the vascular malformations of the bronchus, there are relatively more reports of Dieulafoy's disease[11]. There are even fewer reports of PA aneurysms presenting as endobronchial masses, which rarely protrude into the airway lumen. Two studies described such a condition and both cases were initially misdiagnosed as solid lung masses on conventional imaging[7,12]. Conventional bronchoscopy alone was ineffective. In one case, the lesion was described as a smooth, orange-red intrabronchial mass that protruded into the airway lumen. The signs suggestive of a vascular region, such as a pulsating sensation, and tortuous blood vessels within the submucosa, were absent. Fine needle aspiration biopsy was subsequently performed, resulting in brisk massive hemorrhage[7]. In another case, an endobronchial mass was diagnosed as a PA aneurysm using a convex probe EBUS bronchoscope. Invasive interventions, such as endobronchial biopsies, have been avoided[12]. In the present case, the endobronchial mass was diagnosed as a PA aneurysm using EBUS. Thus, as a useful tool to differentiate vascular from solid endobronchial masses, the EBUS bronchoscope helps to avoid potentially disastrous interventions.

However, we must also mention that intervention techniques like EBUS may not be available in all health care settings. Therefore, it is important to distinguish vascular lesions from solid masses using conventional methods, such as conventional bronchoscopy. In the case reported by Lerner and Riker[12], the endobronchial PA aneurysm presented as a flesh colored and hypervascular lesion. Following continued examination for several circulatory cycles, the lesion was observed to be pulsatile, and its size changed in synchrony with the patient's pulse[12]. In another case, the PA aneurysm presented as a smooth, orange-red endobronchial mass with a plexiform red pattern, protruding into the airway lumen [7]. In our case, the endobronchial mass had a smooth-surface, was, flesh colored and hypervascularized. Fluoroscopy may show a pulsation within the lesion, known as Pezzi's sign[3]. All the bronchoscopic appearances could provide bronchoscopists with implications for the endobronchial vascular lesions, in which a biopsy should be assiduously avoided.

Endobronchial vascular malformations, such as endobronchial PA aneurysms, are so rare that they are often not included in the differential diagnoses and are usually missed on plain CT scan. In cases of recurrent unexplained hemoptysis, where chest CT shows no abnormalities, these types of diseases should be considered, and it is necessary to perform CTA. CTA is relatively non-invasive and can effectively identify abnormalities in the vasculature. This case report was confirmed using CTA. Our case emphasizes the importance of CTA for unexplained hemoptysis.

CONCLUSION

In conclusion, we report a rare endobronchial PA aneurysm, notable for its mimicry of an endobronchial solid mass. Although pathological vascular conditions are extremely rare, they may result in massive hemorrhage with a possibly fatal outcome. Therefore, bronchoscopists must enhance their awareness and vigilance of this lesion to avoid the disastrous consequences of improper biopsy. CTA and EBUS are useful tools for confirming the diagnosis of PA aneurysms and may help avoid inappropriate invasive interventions.

FOOTNOTES

Author contributions: Li M and Zhu WY contributed equally to this work; Li M and Zhu WY contributed to design of the study; Li M and Luo Z contributed to manuscript writing; Wu RR, Mo MT, Wang L, Liu SN and Zhu DY contributed to data collection, analysis, and interpretation; Luo Z reviewed the manuscript and approved the final version of the manuscript; All authors have read and approved the final manuscript.

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