

Case Report

Ruptured noncoronary sinus of Valsalva aneurysm into the right atrium: a rare cause of pulmonary thrombus with pneumonia

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Abstract: A 32-year-old woman received ineffective anti-infection therapy, due to left lower lung pneumonia caused by thrombus of left distal pulmonary. She further underwent transthoracic echocardiography, which showed a ruptured noncoronary sinus of Valsalva aneurysm into the right atrium. After repair of the ruptured noncoronary sinus of Valsalva aneurysm, enhanced chest CT revealed no thrombus in the left distal pulmonary artery, while consolidation and atelectasis in left lower lung were absorbed. Ruptured noncoronary sinus of Valsalva aneurysm into the right atrium was the main reason for pulmonary thrombus and pneumonia, and surgery the main treatment protocol showing certain effectiveness.

Keywords: Ruptured noncoronary sinus of Valsalva aneurysm into the right atrium, pulmonary thrombus, pneumonia, surgery

Introduction

There are three coronary sinuses in the aortic root. Deficiencies of muscle and elastic fibers in the middle layers, combined with continuous impact from high pressure of aortic blood flow, will cause disorders of linkage with fibrous tissue of aortic annulus, and generate a weak area, which gradually progresses into a sac structure, thereby leading to aneurysm. The incidence of Valsalva aneurysm is less than 0.15% [1]; it is more commonly found in men and Asians [2], and often associated with ventricular septal defect and other malformations. Of these, right coronary sinus aneurysm is the most common, followed by NCSA, left coronary sinus aneurysm is the least frequent [3]. This disease is often congenital, and only a small number of cases are caused by infective endocarditis, arteriosclerosis, rheumatic fever or syphilis. Other cases are also caused by rupture of degenerative or calcified noncoronary sinus of Valsalva aneurysm (NCSVA) [4, 5]. The majority of patients with non-ruptured aneu-

rysm show no symptoms, while a few cases present mechanical obstruction, regurgitation, and other anatomic and physiological changes as well as related symptoms, including abnormalities of conduction system, thromboembolism and myocardial ischemia, etc [6]. However, once the aneurysm ruptures into the atrium, ventricle or pericardial cavity, acute heart failure, or catastrophic bleeding may appear. Most ruptured NCSVA cases occur with acute onset into right ventricle or atrium, leading to altered right ventricular hemodynamics, acute pulmonary arterial hypertension, acute right heart failure, or even sudden death. However, pulmonary thrombus caused by ruptured NCSVA has not been reported previously. Definite diagnosis of this disease mainly relies on cardiac ultrasound and magnetic resonance imaging (MRI) as well as other examinations. Its main treatment protocol is surgery, while transcatheter occlusion is also applicable for patients with surgery contraindications or hemodynamic instability [7]. This study described a case suffering from left distal pulmonary thrombus and left

A rare case ruptured noncoronary sinus aneurysm

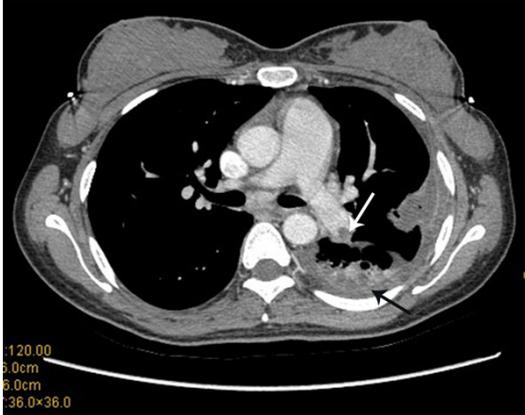


Figure 1. Preoperative enhanced chest CT showing thrombus in left distal pulmonary artery (thin arrow), and consolidation and atelectasis in left lower lung (thick arrow).

lower pneumonia caused by ruptured NCSVA into the right atrium. The patient gained fast recovery from pneumonia and pulmonary thrombus after surgical repair of ruptured non-coronary sinus aneurysm.

Case presentation

The patient was a 32-year-old woman, of Han nationality, farmer. She was admitted to respiratory department due to recurrent fever for 20 days, in addition to cough with yellow and bloody sputum. She had shortness of breath, but did not present chest pain, hemoptysis, hectic fever, night sweat, erythra, arthralgia and other symptoms. She had a history of hypothyroidism, which was healed, with no previous heart and lung diseases, syphilis, and autoimmune diseases. Her family had no specific disease history, and her parents and elder brother were in good health. Physical examination showed: body temperature, 38.9°C; pulse, 118 beats/min; flushed face; mild dark lip; crackles in the left lower lung. Blood routine examination revealed: leucocyte count, $38.65 \times 10^9/L$; neutrophil percentage, 88.10%; erythrocyte sedimentation rate, 58 mm/h; high-sensitivity C-reactive protein, 100.64 mg/L; procalcitonin, 0.114 $\mu\text{g/L}$. Enhanced chest CT revealed thrombus in left distal pulmonary artery as well as consolidation and atelectasis in left lower lung (**Figure 1**). Bronchoscopy showed congestion and edema of bronchial mucosa in the lower lobe of left lung, as well as luminal stenosis (**Figure 2A**). Relatively more neutrophils

were visible (**Figure 2B**) and acid-fast bacilli absent in liquid-based cell smears; bacterial culture was negative in bronchoalveolar lavage fluid (BALF). Tracheal-bronchial biopsy of the lower lobe of left lung revealed chronic mucosal inflammation and epithelial proliferation in the focal area (**Figure 2C**), with absence of thrombus in deep and superficial veins of lower limbs. After admission, the patient was administered successively intravenous Cefmetazole injection (2.0 Q12 h) and Moxifloxacin injection (0.4 QD) for 3 days, intravenous Vancomycin injection (1.0 Q8 h) for 1 week, and Voriconazole injection (200 mg Q12 h) (300 mg Q12 h for the first day) for 1 week. However, she still suffered from recurrent fever. The patient underwent a bronchoscopy one week later, and results similar to the previous ones were obtained: congestion with edema in left lower lobe bronchial mucosa. At this time, the patient underwent physical examination again, which revealed continuous systolic-diastolic murmur of grade III/IV in left margin of precordial sternum, and further transthoracic echocardiography demonstrated a ruptured NCSVA into the right atrium (**Figure 3**). Then, the patient was referred for thoracic surgery, where intraoperative observation showed: thinner NCSVA, protruding into the right atrium: basement, 3.6 mm; tumor size, 8.8 mm; rupture on wall, 2.9 mm. The left and right coronary arteries were in normal shape. The patient underwent a repair of ruptured NCSVA. After surgery, she was administered intravenous Piperacillin tazobactam (3.375 Q8H) for 1 week, and body temperature was restored to normal; the bloody sputum disappeared, and cough and expectoration were alleviated. One week after surgery, transthoracic echocardiography review showed no residual shunt in the repair site (**Figure 4A**), and the patient was discharged. During follow-up, the patient did not present other complications or adverse reactions. One month later, transthoracic echocardiography showed no residual shunt in the repair site (**Figure 4B**); meanwhile, enhanced chest CT revealed no thrombus in left distal pulmonary artery, and consolidation and atelectasis in left lower lung were almost completely absorbed (**Figure 5**).

Discussion

The patient presented here complained of fever, and cough with yellow and bloody spu-

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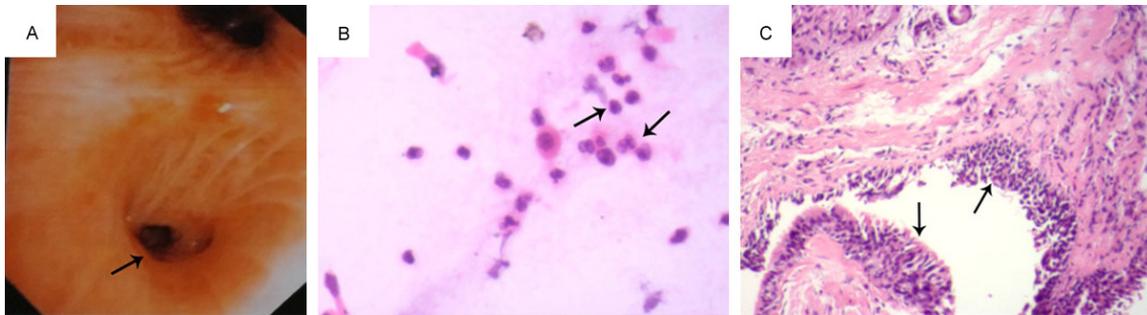


Figure 2. A: Congestion and edema of bronchial mucosa in the lower lobe of left lung, as well as luminal stenosis; B: Neutrophils are visible in liquid-based cell smear with BALF (arrows) (magnification: 40×); C: Tracheal-bronchial biopsy of the lower lobe of left lung reveals: chronic mucosal inflammation and epithelial proliferation in focal area (arrows) (magnification: 10×).

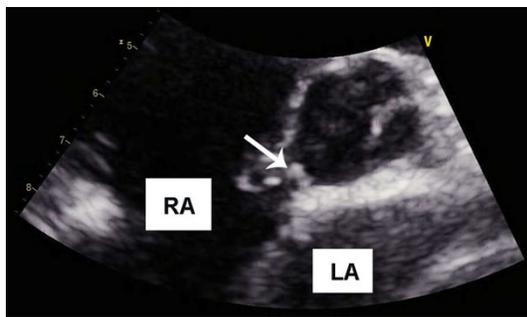


Figure 3. Preoperative transthoracic echocardiography: congenital heart disease-ruptured NCSVA into the right atrium (arrows); LA: left atrium; RA: right atrium.

tum. Crackles were heard in left lung. Leucocyte count and neutrophil percentage were elevated. Consolidation in left lower lung was observed, with more neutrophils found in the BALF. There was no evidence of fungal infection or tuberculosis. Relevant examinations ruled out the possibility of pulmonary lesions from autoimmune diseases or anti-neutrophil cytoplasmic antibody (ANCA) associated vasculitis. Venous ultrasound excluded deep venous thrombosis of lower limbs. Clinical diagnosis provided sufficient proof of bacterial pneumonia. However, the neglected heart murmur led to poor treatment with focus on adjusting antibiotic regimens. Immediate actions were taken for thoracic surgery based on auscultation and echocardiographic examination. This case revealed that pneumonia and pulmonary thrombus could be well treated after the operation. In that case described above, treatment was postponed due to the negligence of heart murmur in the early stage.

NCSA is an aortic sinus aneurysm. It is common that no acute onset happens in patients without its rupture. However, one case was reported by Mahmood [8] with unruptured coronary sinus aneurysm (a maximum diameter of 4.5 cm) and progressive exertional dyspnea. Tomonobu Abe [6] also reported a similar situation: a patient without-rupture left coronary sinus aneurysm causing exertional dyspnea, due to mitral valve obstruction; subsequently, the patient died of secondary pneumonia after three years, due to surgery refusal. It is well known that once the NCSA ruptures into the right atrium or right ventricle, it causes hemodynamic disorders, acute heart failure, and catastrophic bleeding. Generali [5] reported a 77-year-old male patient with breathing difficulties and congestive heart failure for only a week, who died from secondary acute respiratory distress syndrome after surgery in the intensive care unit (ICU). His death was confirmed by echocardiography to be due to NCSA ruptured into the right atrium. To date, no NCSA ruptured into the right atrium has been reported to cause pulmonary thrombus, except a female patient that had chronic right heart failure due to NCSA rupture, described by Cronin [9]. Nabati M [10] reported a case with blood reflux into the pulmonary vein found by echocardiography; this was due to noncoronary sinus broken into the left atrium, which caused respiratory distress and chest pain. Consequently, this disease would happen chronically, triggering atrial or ventricular hemodynamics, or pulmonary vascular disorders and secondary pneumonia as well as respiratory distress syndrome.

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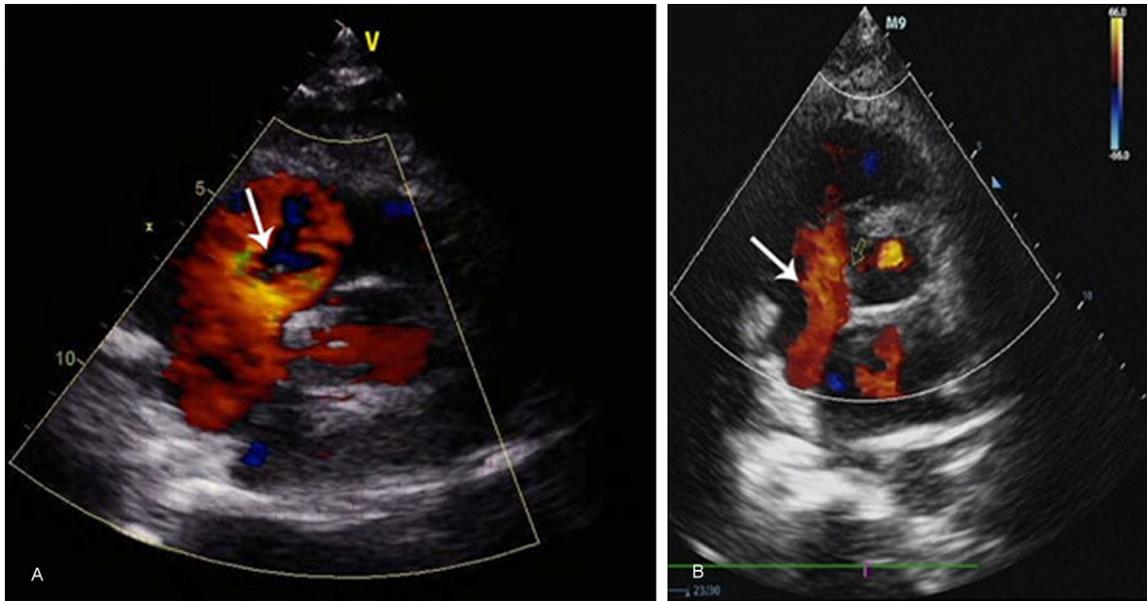


Figure 4. Transthoracic echocardiography at one week (A) and one month (B) after surgery: absence of residual shunt in the repair site (arrows).

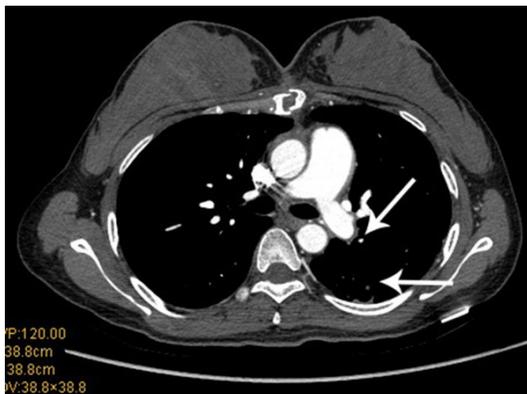


Figure 5. Enhanced chest CT at 1 month after surgery: absence of thrombus in left pulmonary artery, with consolidation and atelectasis in left lower lung almost completely absorbed (arrows indicate the original lesions).

The case presented here is clinically rare, with an unclear pathological mechanism. The patient showed symptoms of left distal pulmonary thrombus and pneumonia without acute right heart failure and congestion. It was speculated that this case is closely linked to small and slow breakage factors (around 2.99 mm) into the arteries. Although this patient had no acute pulmonary hypertension, she still showed artery distal embolization in left lung due to

hemodynamic instability within the right ventricle and pulmonary artery, followed by vascular endothelial injury and coagulation system activation. This further affected left pulmonary blood supply, which triggered secondary infections and pneumonia. Therefore, timely operation was carried out according to the diagnosis of heart murmur. Pulmonary thrombus and pneumonia in this patient were well treated. It is worth mentioning that underlying heart or vascular diseases could be alleviated after proper and timely treatment of pneumonia.

This case showed pulmonary symptoms in the early stage, and no relative remission was obtained after anti-infection treatment. Final treatment was decided after auscultation of heart murmurs due to NCSVA ruptured into the right atrium. Subsequently, the operation was performed for pulmonary symptoms. This case revealed that in the event of unexplained pneumonia and poor treatment, heart assessment should be undertaken immediately.

Conclusions

Ruptured noncoronary sinus of Valsalva aneurysm into the right atrium was the main reason for pulmonary thrombus and pneumonia, and

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surgery the main treatment protocol showing certain effectiveness, and anti-infection therapy was ineffective.

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Written informed consent was obtained from the patient for publication of this report and any accompanying images.

Disclosure of conflict of interest

None.

Authors' contribution

RD wrote the draft of the manuscript and obtained the written consent. XL performed the literature review. YJ participated in the manuscript writing. TZ and ZD helped to the final writing of the paper. ZL gave final approval of the manuscript. All authors have read and approved the final manuscript.

Abbreviations

NCSA, Noncoronary sinus aneurysm; NCSVA, Noncoronary sinus of Valsalva aneurysm; CT, computed tomography; MRI, magnetic resonance imaging; BALF, bronchoalveolar lavage fluid; ANCA, anti-neutrophil cytoplasmic antibody; ICU, intensive care unit.

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