

## RECURRENT HEMOPTYSIS CAUSED BY ARTERIOVENOUS MALFORMATION

Ivana Meta Jevtovic, Romana Susa, Bojan Djokic  
University Clinical Center Kragujevac, Clinic for Pulmonology, Kragujevac, Serbia

## REKURENTNE HEMOPTIZIJE UZROKOVANE ARTERIOVENSKOM MALFORMACIJOM

Ivana Meta Jevtović, Romana Suša, Bojan Đokić  
Univerzitetski klinički centar Kragujevac, Klinika za pulmologiju, Kragujevac

### ABSTRACT

*Pulmonary arteriovenous malformation is a rare disorder of pulmonary vascularization. We are going to describe the case of recurring hemoptysis with a patient who was diagnosed late with pulmonary AV malformation. The 68-year-old patient was hospitalized for the fourth time in the Clinic for Pulmonology, Clinical Center of Kragujevac due to recurring hemoptysis. Bronchoscopy was conducted which showed no signs of bleeding nor new changes in the bronchial lumen. MSCT of the chest eliminated the possibility of bronchiectasis as the cause for hemoptysis. Chest MSCT was repeated, and it was without evolutionary aspect when compared to the previous one. In April 2019, at the Military Medical Academy in Belgrade, the patient had bronchoscopy performed, the results were normal, and the examination was completed with exploration, along with selective angiography, where AV shunt could be seen on the left side. Since the diameter of the left bronchial artery was less than 2 mm, it was not possible to place the micro-catheter and do embolization, but hemostasis was done by manual compression, which lasted for 10 minutes. After six months of follow-up examinations, no complications were registered with the patient. The method of choice for diagnosing PAV malformation is bronchial angiography, while other chest radiographic methods are not reliable. Embolization is the method of choice for treating this disorder.*

**Key words:** lung; hemoptysis; arteriovenous malformations

### INTRODUCTION

Hemoptysis occurs with the frequency of 0.1% with outpatients and 0.2% with inpatients (1). They are more frequent in cases of lung inflammation in relation to malignant lung tumors (25.8 vs. 17.4%) (2). Vascular blood vessels malformations occur in only 0.2% of patients with hemoptysis (2). Pulmonary arteriovenous malformation (PAVM) is a rare disorder of pulmonary vascularization (3). The disorder reflects in the abnormal communication between the pulmonary artery and veins that ultimately leads to the right-to-left shunt which causes the fall in arterial blood oxygenation (4). The etiology of this disease is usually congenital; however, they may be acquired in certain conditions, such as mitral stenosis, schistosomiasis, tuberculosis, trauma, and metastatic

### SAŽETAK

*Plućne arteriovenske malformacije redak su poremećaj plućne vaskularizacije. Opisacemo uzrok rekurentnih hemoptizija kod bolesnika kod kog je dijagnostikovana AV malformacija u kasnom životnom dobu. Bolesnik star 68 godina četiri puta je hospitalizovan u Klinici za pulmologiju Kliničkog centra Kragujevac zbog rekurentnih hemoptizija. Bronhoskopskim pregledom nisu uočeni znaci krvarenja, niti novotvorina u lumenu bronha. MSCT grudnog koša nije pokazao bronhiektazije kao mogući uzrok hemoptizija. Kontrolni MSCT grudnog koša bio je bez evolutivnosti u odnosu na prethodni. Aprila 2019. godine u Vojnomedicinskoj akademiji u Beogradu ponovljena je bronhoskopija, čiji je nalaz bio uredan, a pregled je završen eksploracijom. Učinjena je selektivna angiografija i uočen levostrani AV šant. S obzirom na to da je dijametar leve bronhijalne arterije bio manji od 2 mm, nije bilo moguće plasirati mikrokater radi embolizacije. Hemostaza je učinjena manualnom kompresijom u trajanju od 10 minuta. Nakon šestomesečnog praćenja nisu bile registrovane komplikacije kod bolesnika. Metod izbora za dijagnozu plućnih arteriovenskih malformacija je bronhijalna angiografija, dok druge radiografske metode nisu pouzdane. Embolizacija je metod izbora za lečenje ovih poremećaja.*

**Cljučne reči:** pluća, hemoptizije, arteriovenske malformacije

thyroid carcinoma. Clinically and radiologically, the disorder can be simple or complex and it is usually connected to genetic telangiectasia (3). We are going to describe a case of recurring hemoptysis with a patient who was diagnosed late with pulmonary AV malformation; however, in our case after a meticulous search, no cause was identified.

### THE CASE

The 68-year-old patient was hospitalized for the fourth time in the Clinic for Pulmonology, Clinical Center of Kragujevac due to recurring hemoptysis. She was first hospitalized due to similar problems in August 2014. Bronchoscopy was conducted which showed no signs of bleeding nor new changes in the bronchial lumen. MSCT

of the chest eliminated the possibility of bronchiectasis as the cause for hemoptysis. Following that, there were two more hospitalizations due to mild hemoptysis which were treated conservatively.

The previous hospitalization happened in January 2017 following the expectoration of 2 dc of bright red blood. MSCT of the chest was repeated which showed individual subpleural bullous changes, right on the level of the medial lobe subpleural two micronodular lesions and three more similar lesions on the level of lower lobe, two in the anterobasal segment and one laterobasal as well as one laterobasal to the left – the multiplanar reconstruction showed that all lesions had a striped form without a significant post-contrast density enhancement – featuring adhesions; while the liver examination showed individual cysts of up to 10 mm size. Other CT results were normal. Moreover, the gastroenterological examination was conducted, EGDS was normal. At the admission 28/03/2019, the patient claimed that one day prior to the hospitalization she had coughed up about 2dl of bright red blood.

Other problems she claimed to had had included coughing, the feeling of uneasiness in the chest; she denied febrility and problems with breathing. She was examined by a surgeon and an ENT specialist. The findings were normal. Since the problems continued, the patient was referred to a pulmonary specialist the next day and she was hospitalized. Personal history showed she had previously been treated for arterial hypertension and osteoporosis. She was allergic to penicillin.

At the admission, the following vital signs were documented: BP 120/80 mmHg, fr: 60/min. Laboratory test showed WBC  $7.11 \times 10^9/l$ ; Er  $4.31 \times 10^{12} /l$ ; Hgb 128 g/l; Hct 38%; MCV 88,5 fL; PLT  $238 \times 10^9/l$ ; INR 1,10; aPTT 26,9 s; D Dimer 0,20 ng/ml. The values of bilirubin, hepatogram, glycemia, nitrogenous substances, ionograms and inflammation parameters were within the reference range. When arterial blood was analyzed in room air, light hypoxemia was detected in gas analysis (pO<sub>2</sub>-9,2, kPa; pCO<sub>2</sub>-5,7 kPa; pH-7,44; HCO<sub>3</sub>-28,4; SaO<sub>2</sub>-94%). On PA lung radiography in the pulmonary parenchyma, there was a basally emphasized vascular drawing without other pathological changes. Echocardiographic finding was normal, without enlargement of the right heart cavities and without indirectly assessed elevated pressure in the pulmonary artery.

Chest MSCT was repeated, and it was without evolutionary aspect when compared to the previous one from February 2017. Having in mind the clinical picture and diagnostically excluded inflammatory, malignant and thromboembolic diseases, the patient was referred to selective bronchial angiography.

In April, at the Military Medical Academy in Belgrade, the patient had bronchoscopy performed, the results were

normal, and the examination was completed with exploration, along with selective angiography, where AV shunt could be seen on the left side (Figure 1). Since the diameter of the left bronchial artery was less than 2 mm, it was not possible to place the micro-catheter and do embolization, but hemostasis was done by manual compression, which lasted for 10 minutes. After six months of follow-up examinations, no complications were registered with the patient.

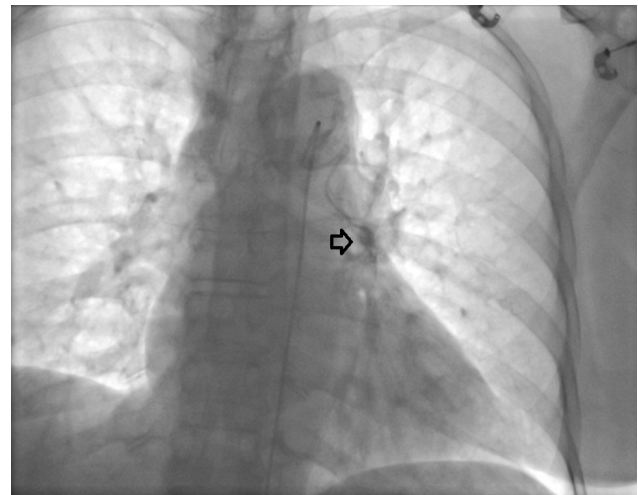


Figure 1. Selective bronchial angiography

## DISCUSSION

PAV malformation is a rare disorder with an incidence of 2-3 per 100,000 people (5), more common among female patients (5). The ratio of men and women varies from 1:1.5 to 1:1.8 in various reports (6). The first presentation varies from infant to 70 years of age, but it is most commonly diagnosed in the first 30 years of life (6). Our patient is female, and the first presentation of disease was manifested when she was 64 years old. In more than 80% of the cases, PAV malformation is of congenital origin and acquired PAV does not occur very often. The causes of acquired PAV are: chest trauma, surgery, cirrhosis of the liver, metastatic carcinomas and infections (7). Presentation is also possible with complications such as cerebral embolism and brain abscesses with an incidence of 10-19% (8) and up to 40% according to some authors (6). The risk of cerebrovascular complications is greater when the diameter of the pulmonary artery is  $\geq 3$  mm. Malformation can be manifested in a form of individual or multiple changes. Individual malformations occur with incidence of 42-74%, while multiple malformations occur in 8% out of 20% of the cases. The most common location is the lower right lobe, in individual changes.

Our patient had acquired PAV malformation which was most probably caused by earlier infections. This malformation was individual and it was localized in lower left lobe, and cerebral-vascular complications were not

recorded, most probably because of the small diameter of the pulmonary artery. PAV malformation can cause right-to-left shunt which, if it is small, goes without symptoms. Almost 50% of the patients are without symptoms or have hemoptysis as the first symptom (5). Symptoms develop when 20% of total blood volume goes through right-to-left shunt. The fall of oxygenation causes hypoxemia, cyanosis and dyspnea (5). With our patient, the first presentation of AV malformation was hemoptysis, while right-to-left shunt was not developed, due to small diameter of pulmonary artery. This malformation can cause severe complications with untreated patients, morbidity is in the range of 26-33%, whereas mortality is from 8 to 16% (9).

The method of choice for diagnosing PAV malformation is bronchial angiography, while other chest radiographic methods are not reliable (5). Embolization is the method of choice for treating this disorder.

### ABBREVIATIONS

aPTT-activated partial thromboplastin time  
AV-arteriovenous (malformation)  
BP-blood pressure  
EGDS-esophagogastroduodenoscopy  
Er-erythrocytes  
HCO<sub>3</sub>-bicarbonates  
Hct-hematocrit  
Hgb-hemoglobin  
INR-international normalized ratio  
MSCT-medium-multislice cell volume  
PAVM-pulmonary arteriovenous malformation  
pCO<sub>2</sub>-partial pressure of carbon dioxide  
pH-concentration of hydrogen ions  
PLT-platelets  
pO<sub>2</sub>-partial pressure of oxygen  
SaO<sub>2</sub>-blood oxygen saturation  
WBC- white blood cells

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