

## Endovascular embolisation with Amplatzer vascular plug of ruptured pulmonary artery aneurysm in Behçet's disease

Sirs,

We present a case of pulmonary artery aneurysm (PAA) with Behçet's disease (BD) successfully treated with Amplatzer™ vascular plaque (AVP-AGA Medical Corp., Plymouth, MN, USA). A 34-year-old male patient with massive haemoptysis was admitted to our emergency department. He had been suffering from recurrent oral and genital aphtha and erythema nodosum for seven years. BD was diagnosed in 2016 when a thrombosed pulmonary artery aneurysm was detected in the segmental branch of the right pulmonary artery and he was treated with methylprednisolone (MP) and cyclophosphamide (CYP) pulse (1g x 3 days and 15mg/kg, respectively). However, the patient refused the treatment after the 5<sup>th</sup> dose of CYP and he was referred to our clinic with haemoptysis (150 cc/day) three months later. After three cycles of CYP pulses and high dose MP, azathioprine (AZA) was started at 2.5 mg/kg/day as a maintenance therapy. The disease was controlled by this treatment but after six months, the patient ceased taking his medication again. At that time, he applied to our clinic with massive haemoptysis.

On physical examination, he was pale, tachypneic and tachycardic. In laboratory studies, high acute phase was detected [erythrocyte sedimentation rate (ESR): 57 mm/h, C-reactive protein (CRP): 6.2 mg/dl]. Thorax computerised tomography (CT) revealed an aneurysm sized 66x52 mm in transverse diameter in the segmental branch of the right pulmonary artery (Fig. 1A). Despite immunosuppressive treatment with high dose of MP and CYP, massive haemoptysis continued, and it was decided to perform embolisation of the pulmonary artery aneurysm.

A 7F long sheath of 90 cm in length was introduced via right femoral vein to the right main pulmonary artery in a coaxial fashion. The diagnostic angiography revealed a partially thrombosed aneurysm of 43x46 mm in size (Fig. 1C). The sheath was placed at the neck of the aneurysm. An AVP II multi-layered and multi-segmented embolisation device of 18 mm in size was introduced through the long sheath and delivered at the neck of the aneurysm. The device was opened partially within the sac and at the neck of the aneurysm. The control angiography after the delivery of the device revealed near total cessation of blood flow into the aneurysm sac (Fig. 1D). After the procedure, haemoptysis was dramatically reduced and PAA was regressed. Three more cycles of CYP pulse followed by 2.5 mg/kg/day of AZA were given. The



**Fig. 1.** Thorax computerised tomography (CT) and conventional angiography images before and after endovascular embolisation. Thorax computerised tomography (CT) revealed pretreatment aneurysmatic dilatation in the segmental branch of the right pulmonary artery (A) and the 6<sup>th</sup> month control thorax CT examination reveal near total regression (B). The pulmonary aneurysm also seen on the image of angiogram before the procedure (C) seems to be embolised with Amplatzer™ (D).

6<sup>th</sup> month control thorax CT examination showed near total regression of the aneurysm sac (Fig. 1B).

Vascular involvement is characterised by the association of arterial and venous lesions such as thrombosis or aneurysm and affect about 40% of patients with BD (1). Pulmonary artery involvement may lead to PAA and/or pulmonary thrombosis with high mortality rate. High dose steroids and immunosuppressant drugs appear to be successful in achieving clinical remission (2). However, bleeding due to rupture of inflamed pulmonary artery wall may not be stopped with immunosuppressive treatment in some patients. Chemo-embolisation and/or surgical procedure such as lobectomy, aneurysmectomy are emergency treatment options for refractory patients with PAA (3). Surgical treatment requires invasive open thoracic procedure, depending on the location, and have a high risk of mortality and morbidity (4, 5). Endovascular therapies which include minimal invasive techniques is performed by embolisation using a variety of embolic agents (chemical embolisation) or mechanical stabilisers, such as AVP. The choice of the embolic agents depends on the location, size, number and width of the neck of the aneurysm (6). The chemical embolisation is a relatively well-known option in PAA related to BD compared with vascular plug embolisation. In

our case, the chemical embolisation was not feasible because the aneurysm size was too large. The main advantage of AVP is that it can resolve aneurism by occluding feeding vessel with a single device, thus being more rapid. The elasticity nature allows the AVP to become securely anchored to the artery wall. However, the possibility of migrating the device and rupture of vessel wall during procedure should be considered. According to the literature, AVP has previously been applied in a few cases with pulmonary aneurysm related different disease (7). This is the second report on endovascular embolisation with AVP of PAA in BD (8). Unlike previously reported patient with BD, the diameter of the pulmonary artery was very large, and the procedure was performed during massive hemoptysis in our patient. As a conclusion, we consider that endovascular embolization with AVP m be an alternative treatment option in selected patients with PAA related to BD.

N. ALPAY-KANITEZ<sup>1</sup>, Assoc Prof Dr

S. ÇELİK<sup>2</sup>, Assoc Prof Dr

F. BALTACIOĞLU<sup>3</sup>, Prof Dr

O.C. İÇAÇAN<sup>2</sup>, MD

C. BES<sup>2</sup>, Assoc Prof Dr

B. YILDIZELI<sup>4</sup>, Prof Dr

<sup>1</sup>Division of Rheumatology, Department of Internal Medicine, Koc University School of Medicine, Istanbul; <sup>2</sup>Division of Rheumatology, Department of Internal

# Letters to the Editors

Medicine, Health Science University, Bakırköy Dr. Sadi Konuk Research and Training Hospital, Istanbul; <sup>3</sup>Department of Radiology, Marmara University School of Medicine, Istanbul; <sup>4</sup>Department of Thoracic Surgery, Marmara University School of Medicine, Istanbul, Turkey.

Please address correspondence to:  
Dr. Nilüfer Alpay-Kanitez,  
Division of Rheumatology,  
Department of Internal Medicine,  
Koc University School of Medicine,  
Istanbul, Turkey.  
E-mail: nilalpay@gmail.com

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