

Case report:

Treating pulmonary arteriovenous malformation with coil embolization: a case report

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Abstract

Pulmonary arteriovenous malformation is an uncommon entity. Here we present the story of a young male who presented with cough, breathlessness, haemoptysis and epistaxis with the chest x-ray showing an opacity in the left mid zone. He was subsequently diagnosed to suffer from pulmonary arteriovenous malformation using contrast enhanced computed tomography demonstrating an elliptical lesion in the lingular area with a dilated and lobulated serpiginous vessel. The diagnosis was confirmed by selective pulmonary angiography and the patient was successfully treated with coil embolisation.

Ke Words: Pulmonary arteriovenous malformation, computed tomography, coil embolisation.

(The Pulmo - Face; 14:2, 52-54)

ABBREVIATIONS:

PAVM: pulmonary arteriovenous malformation

HHT: hereditary haemorrhagic telangiectasia

INTRODUCTION:

Pulmonary arteriovenous malformations (PAVM) are abnormal vascular structures that provide a direct communication between the pulmonary arteries and veins and thus between the pulmonary and systemic circulations.⁽¹⁾ Thus, they allow an effective bypass of the normal pulmonary capillary bed. PAVM are rare.⁽²⁾ They are more common in women than men. Majority of the lesions occur as single but multiple malformations are formed frequently in patient with hereditary haemorrhagic telangiectasia (HHT).⁽³⁾ Here We present one case of PAVM diagnosed and successfully treated at our centre with coil embolisation.

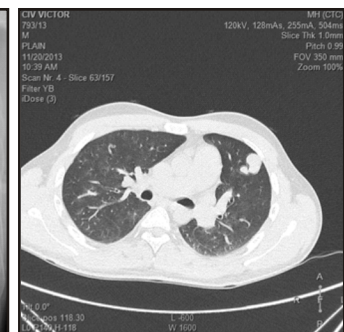
CASE REPORT

A 27 year old non smoker male presented with history of breathlessness on exertion and cough with mucoid expectoration for one month. During this period, he had of two episodes of haemoptysis each amounting approximately 10 ml. There was no other significant history but the story of having multiple episodes of epistaxis since childhood. Clinically, his vital signs were stable without any cyanosis and clubbing; the ENT (ear, nose, and throat) examination was unremarkable. With a SpO₂ of 98% in room air, his systemic examination

revealed no abnormality. The routine hemogram and the blood biochemistry were unremarkable. The x-ray chest (PA view) revealed opacity in the mid zone of left lung with the left hilum looking prominent (picture 1). contrast enhanced computerized tomography of chest revealed a smooth, sharply defined, elliptical lesion formed by a lobulated serpiginous vessel in the lingular segment (picture - 2).



Picture - 1



Picture - 2

Picture 1: X-ray chest reveals opacity in left mid zone of lung

Picture 2: CECT chest reveals lesion with a vessel involving lingular segment

Based on the radiological picture, a diagnosis of pulmonary arteriovenous fistula (PAVF) was made and in view of the future risk of hemorrhage, embolisation was planned. The job was subsequently accomplished using the right transfemoral route under local anaesthesia. A 6 French sheath was placed in the right common femoral vein. Through that, a 5 French Grollman pigtail catheter was advanced and the tip placed in the common pulmonary artery to do a pulmonary angiogram. The Angiogram revealed an arterio-venous malformation in the lingular segment of left lung. Thereafter the feeding artery was catheterized super selectively with a 5 French Counard catheter and embolisation was done with three coils (10 x 80, 8 x 80, 8

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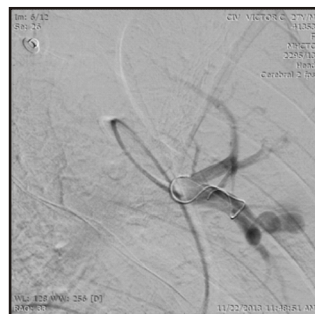
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x 80 (mm) (picture 3, 4). Post procedure, the check angiogram showed no residual filling of the fistula. The patient was kept on complete bed rest under observation for 8 hours and on finding no complication related to the procedure, he was discharged on the next day. At the next follow up after one month he was completely asymptomatic.



Picture - 3



Picture - 4

Picture 3: Feeding artery angiography

Picture 4: Feeding artery embolisation done with three coils

DISCUSSION

Pulmonary arteriovenous malformation (PAVM), also known as pulmonary arteriovenous fistula, results from deficient formation or abnormal dilatation of pulmonary capillaries because of developmental defect in capillary wall. It slowly enlarges over time and is usually diagnosed in the third or fourth decade. PAVMs are commonly found in lower lobes, typically in sub pleural location. There may be bilateral fistulae in 10 % of cases and multiple fistulae are found in 35% of the cases of PAVM. A simple PAVM has a single dilated vascular sac connecting one feeding artery with one vein as seen in the angiography of our case. Complex PAVMs have more than one feeding artery.

Twenty-five to fifty patients with PAVM are asymptomatic. Pulmonary symptoms include dyspnoea on exertion, platypnoea, orthodeoxia and haemoptysis.⁽⁴⁾ There may be chest pain, epistaxis and features of transient ischaemic attacks. Rarely, there may be preceding history of trauma in some of the cases of pulmonary AV malformations.⁽⁵⁾ The symptomatic patients may have cyanosis, clubbing and secondary polycythaemia or they may present with complications like massive haemoptysis and haemothorax from rupture, or brain abscess from infective embolism. Complications like pulmonary hypertension, migraine, gastrointestinal bleeding, cirrhosis of liver and high output heart failure are also seen.⁽⁶⁾ In some cases, PAVM is observed along with hereditary haemorrhagic telangiectasia (HHT) or Osler-Rendu-Weber syndrome.⁽³⁾ Telangiectasia in the form of small, red vascular blemishes may be seen on face, lips, tongue, ears, chest and feet.

Diagnosis of PAVM is made through investigations following clinical suspicion in the appropriate settings. Abnormalities in chest x-ray or CT chest with history of

haemoptysis provide strong diagnostic clues. CT scan of chest with contrast enhancement usually delineates the lesion with feeding artery/arteries and draining vein / veins.⁽⁷⁾ Transoesophageal echocardiography with agitated saline as contrast is useful in the diagnosis of intrapulmonary shunt (transthoracic echocardiography may not be able to detect small PAVMs). The air bubbles offer a snow storm appearance in the left side of heart by the delay of 4 - 5 cardiac cycles suggesting a pulmonary shunt.⁽⁸⁾ This does not happen in normal air-contrast echocardiography that rules out PAVM. If it is positive, pulmonary angiography, the gold standard test, has to be done to confirm the diagnosis.⁽⁶⁾

Earlier the treatment consisted of resection of the lesion by thoracotomy⁽⁹⁾. Later video assisted surgeries were used to be carried out. Presently, embolisation with coils⁽¹⁰⁻¹¹⁾ or balloons is the procedure of choice, as has been done in our case. The procedure is much less invasive than surgery and is highly successful.^(12,13) Rarely, the recanalisation of embolised vessel (occurring in 2 - 8% of cases) may demand repeat embolotherapy. The procedure is relatively safe but, complications of embolotherapy can rarely occur even in expert hands; they include migration of embolic device or perforation of PAVM. There are reports of using polyvinyl alcohol (PVA), wool coils, and Amplatzer duct occluder (designed for the occlusion of patent ductus arteriosus) being successfully used in treating PAVM.^(11,16) Rarely surgery is performed in patients in selected difficult situations when vascular ligation and / or local excision is performed.⁽¹⁵⁾ Though glue is popular in closure of various fistulae and varices, after doing a PUBMED search, we could not find any report of the use of glue in a case of PAVM.

Since the frequency of AVM is high (36-96% and usually multiple) in hereditary hemorrhagic telangiectasia, one should screen the patients of PAVM for the presence of this autosomal dominant disease. Alternatively, all cases of this hereditary syndrome should be screened for the presence of PAVM. Recurrent haemoptysis, multiple telangiectasia, and several visceral lesions in organs as lungs, gastrointestinal tract, liver, brain etc with family history of similar illness in the first degree relatives suggest the presence of hereditary hemorrhagic telangiectasia.

The case elaborates the success of coil embolization to treat PAVM.

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