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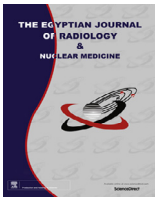

Case Report

Utility of endovascular embolisation in management of peripheral pulmonary artery aneurysms

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ABSTRACT

There are no large scale studies exclusive for pulmonary artery aneurysms as a cause of massive hemoptysis, only small studies are there. No randomised control trial is there to suggest efficacy of surgical excision or endovascular treatment over each other. It's well known that definite treatment by surgical excision carries high mortality when done in emergency sitting. Endovascular approach in such a patient is a less utilised treatment modality and that too with coil embolisation is not much practiced.

This article emphasises the role of CTA in diagnosing and embolisation as emergency management of these aneurysms.

1. Introduction

Pulmonary artery aneurysms are uncommon causes of hemoptysis; however, they should be kept in the differential diagnosis and must be recognised at the earliest to prevent life threatening haemorrhage. Patients of ruptured pulmonary artery aneurysms present with intermittent hemoptysis, with the amount of expectorated blood varying from mild to massive. Endovascular embolisation is an effective and relatively non-invasive technique compared to surgical procedures which involve lobectomy or pneumonectomy of the involved lung leading to prolonged patient recovery, hospital stay and decreasing pulmonary reserve.

We discuss a case series of three patients who came with complaints of massive hemoptysis. Computed Tomography Angiography (CTA) of thorax revealed a peripheral pulmonary artery aneurysm as the cause of hemoptysis. These patients were managed by endovascular coil embolisation.

2. CASE 1

A 60 year old female, non-smoker, non-alcoholic, non-diabetic, hypertensive patient presented with massive hemoptysis. She had history of pulmonary tuberculosis 10 years back for which she took incomplete treatment. The patient complained of cough with expectoration and breathlessness for last two months, and occasional bouts of hemoptysis, about 100–150 ml sputum mixed fresh blood. She came to emergency with increasing frequency and amount of blood in sputum. Her chest X-ray revealed fibrobronchiectatic changes with an ill defined inhomogeneous opacity in left upper zone. CTA thorax was done in both pulmonary arterial and aortic phases of contrast enhancement to look for pulmonary and bronchial arteries. CTA revealed a lobulated, 10 × 10 mm aneurysmal lesion in apicoposterior segment of left upper lobe (Fig. 1a axial

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section and Fig. 1b coronal section), surrounded by a thick walled cavity and fibrobronchiectatic changes. There were areas of ground glass opacities in the same lobe and also in bilateral basal lung segments on account of aspiration of blood. Bronchial arteries were not enlarged and no other aberrant systemic vessels supplying the lesion were observed. After reviewing all the images a diagnosis of Rasmussen aneurysm was made likely due to pulmonary tuberculosis.

Patient was taken up for emergency Digital Substraction Angiography (DSA) under General Anaesthesia (GA). On DSA there was a pseudo aneurysm filling from apicoposterior branches of left ascending pulmonary artery (Fig. 1c). On super selective cannulation, single feeder leading to aneurysm was not seen and instead multiple small vessels leading to a pseudoaneurysmal cavity were seen (Fig. 1c). These feeding vessels were selectively cannulated and embolised (Fig. 1d) using 0.018 coils (Micronester coils, Cook, USA). There was no fresh bout of hemoptysis and patient was discharged after a week. However mild hemoptysis recurred after two months. CTA thorax this time did not reveal any dilated feeding vessel or filling of the pseudoaneurysm. A well defined cavity was visualised surrounded by fibrotic parenchyma. The patient was referred for lobectomy.

3. CASE 2

A 35 year old male, non-smoker and non-alcoholic presented with two episodes of massive hemoptysis within a gap of 15 days. Patient gave prior history of off and on fever and Deep Vein Thrombosis (DVT) involving right common and superficial femoral vein. Autoimmune causes of hypercoaguable states such as IgG and IgM anticardiolipin antibodies and anti nuclear antibodies were negative and tested for protein C and S deficiency, hyperhomocysteinemia, AntiThrombin-III deficiency and factor V leiden mutation were also negative. Patient had cough and breathlessness for last 2 weeks and in between he had two episodes of massive hemoptysis. Chest X-ray PA view revealed a nodular opacity in left infrahilar region (Fig. 2e). CTA chest revealed a large aneurysm measuring approximately 2×2 cms, arising from anterior basal segmental branch of left descending pulmonary artery (Fig. 2a volume rendered image and Fig. 2b axial section). Emergency coils'

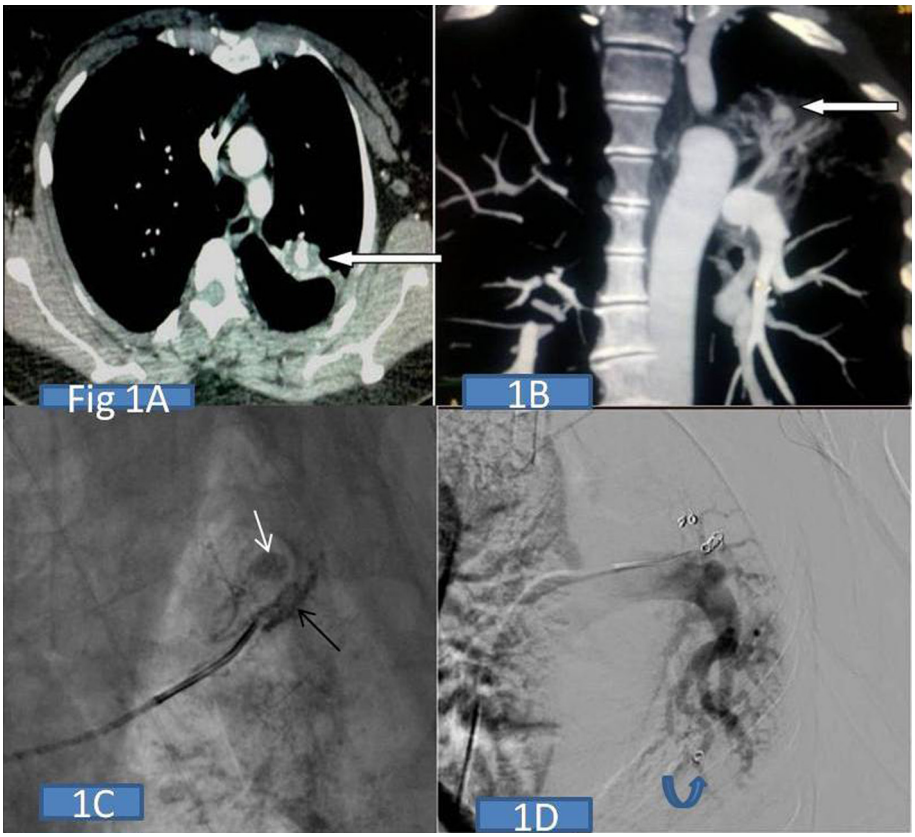

Fig. 1. A. Contrast enhanced CT axial image showing a small Rasmussen aneurysm in the apicoposterior segment of left lung with surrounding cavity with hypervascular wall (arrow). B. coronal MIP image showing the aneurysm in relation to left upper lobe segmental branches. C. DSA shows catheter in left upper lobe segmental branch with filling up of aneurysm (white arrow) and contrast blush (black arrow) in the surrounding lung. D. DSA post coil embolisation in the subsegmental branches of left upper lobe showing no filling of aneurysm and the absence of hypervascularity and blush in the surrounding lung. One of the coils (curved arrow) has accidently migrated into one of the segmental branch of left descending pulmonary artery.

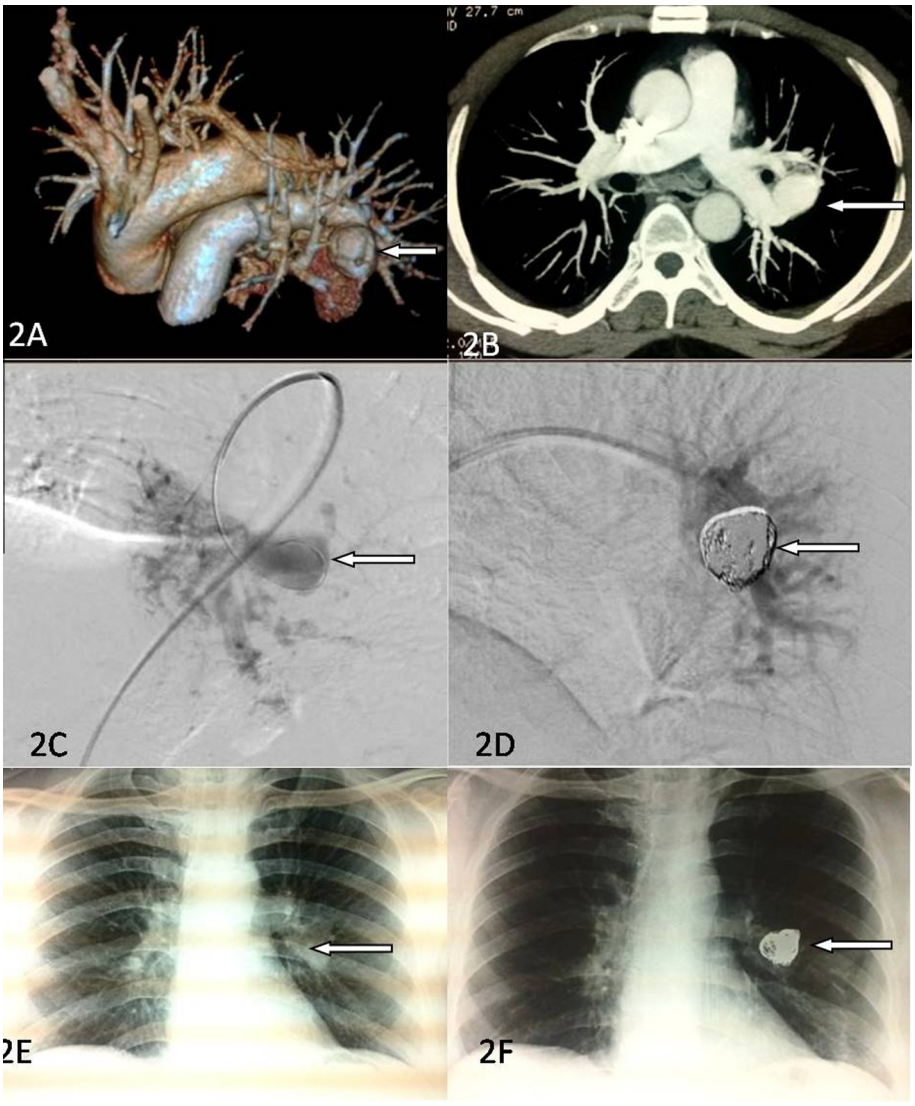

Fig. 2. A. 3D Volume Rendered image showing a globular aneurysm arising from descending branch of left pulmonary artery (arrows). B. Axial MIP image showing the aneurysm (arrows). C. Oblique DSA image showing angiogram taken selectively from left descending pulmonary artery opacifying the aneurysm (arrow). D. DSA image showing coil mass with obliteration of aneurysmal sac (arrow). E and F – chest X-ray PA view showing left infrahilar opacity which was aneurysm from left descending pulmonary artery before and after embolisation with coils (arrow).

embolisation of the aneurysm was planned under General Anaesthesia. As Colour Doppler revealed sub acute stage of thrombus in right superficial and common femoral vein with partial recanalisation, hence pulmonary angiogram was performed through left femoral vein. Catheter angiograms revealed filling up of aneurysm sac from the anterior basal segmental branch (Fig. 2c) of left pulmonary artery. Double micro catheter technique is used i.e. two micro catheters were placed into aneurysm sac one after the other. Packing of the aneurysm with 0.010 detachable coils (Axium, Ev3 Covidien, USA) was done, initially by 3D framing coils followed by smaller coils till no contrast filling was seen within aneurysm (Fig. 2d). Patient was started on low molecular weight heparin and warfarin to maintain INR between 2 and 3 for DVT prophylaxis. There were no fresh bouts of hemoptysis and the patient was discharged satisfactorily after antibiotic course and his subsequent chest X-ray showed coil mass in place (Fig. 2f). On follow-up his ESR was always raised and he had few episodes of blood tinged sputum in next six months. In follow-up all of a sudden patient had massive hemoptysis after one year of previous endovascular coiling and landed in emergency. Chest X-ray showed right infrahilar nodular opacity (Fig. 3a is scanogram showing the same finding). CT Angiography of the thorax was done which revealed a new aneurysm (not seen on previous CT angiogram Fig. 3b) in the right lower lobe posterior segmental branch of pulmonary artery with surrounding ground glass and alveolar haemorrhage (Fig. 3c and d). Patient was taken up for endovascular embolisation but this time as there was DVT which had progressed to involve bilateral external iliac veins so right internal jugular approach was taken

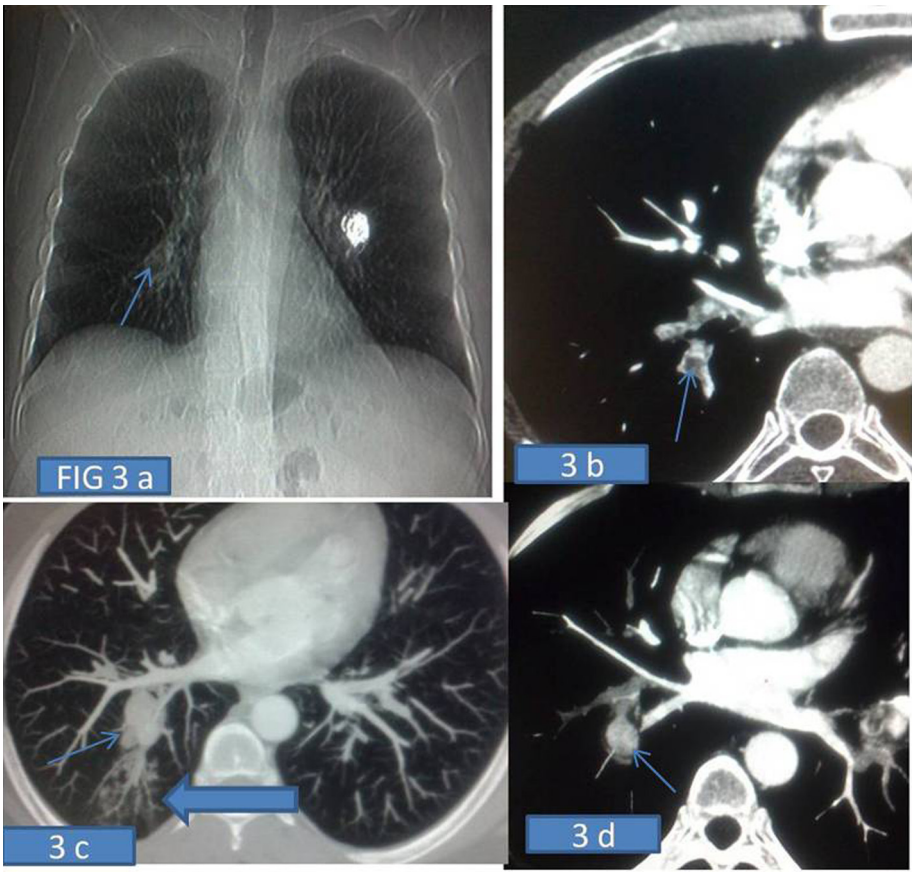

Fig. 3. a. CT scanogram showing right infrahilar opacity (arrow) suggesting some vascular pathology. b. CT angiogram image at the time of first episode showed no aneurysm in the right pulmonary artery branch (arrow). c. CT Angiogram Lung window image showing right descending pulmonary artery branch aneurysm (arrow) with surrounding parenchyma showing groundglass and alveolar haemorrhage (thick arrow). 3d. CT Angiogram showing right descending pulmonary artery aneurysm (arrow) which was not there in earlier CT as shown in Fig. 3b by arrow.

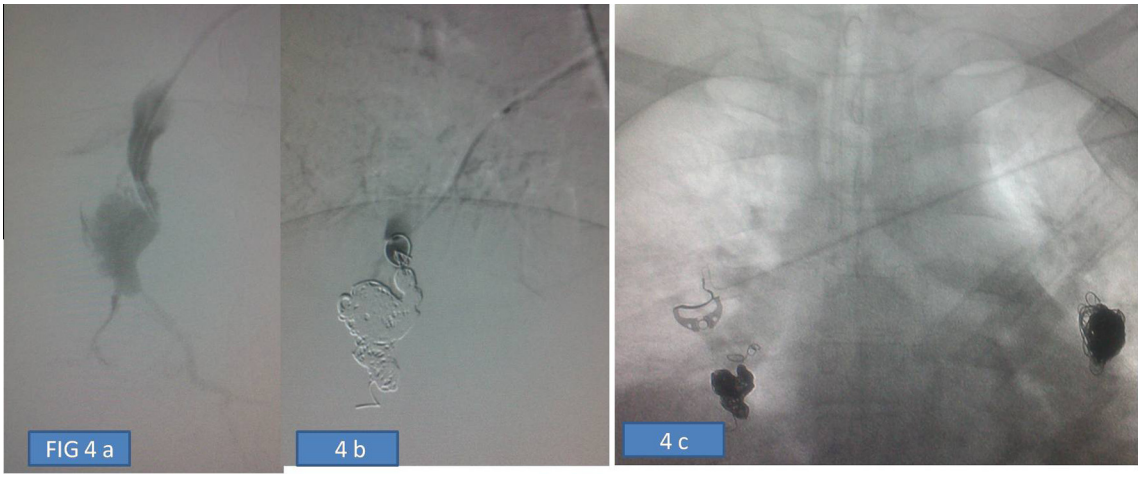

Fig. 4. a. DSA image showing selective angiogram in right descending pulmonary artery branch with opacification of the aneurysm. b. DSA image showing coil mass with obliteration of the aneurismal sac. c. A fluoroscopy image showing coil masses in both lung fields used to obliterate the aneurysms.

and coil embolisation of the aneurysm was done (Fig. 4a and b) using multiple 0.018″ detachable microplex coils (Microvention, USA) (Fig. 4c). Post embolisation his clinical status was again reviewed and was started on immunosuppressant and since more than six months have passed there are no any other hemoptysis episodes. Ini-

tially labelled as Behcet's disease he was finally retrospectively diagnosed as Huges Stovin syndrome.

4. CASE 3

A 17 year old male patient presented with recurrent bouts of massive hemoptysis (more than 300 ml/episode) for last 2 years with no history of cough, fever or weight loss. On examination, patient was absolutely normal except for mild pallor. Chest X-ray showed two rounded opacities in bilateral lower lung fields near pulmonary hila. CTA was done and it showed aneurysms arising from pulmonary arteries on both sides (Fig. 5a and b). Patient underwent left sided lobectomy. Eight days after lobectomy patient was taken for endovascular embolisation of right sided pulmonary artery aneurysm under general anaesthesia. Angiogram revealed an aneurysm from right descending branch of pulmonary artery (Fig. 5c). Swan ganz balloon catheter was used for flow control and multiple coils (.035 Nester, Cook, USA) were used to pack the aneurysm (Fig. 5d). Post embolisation angiogram did not reveal any significant filling of the aneurysm. Patient was discharged after antibiotic course and on follow-up patient did not complain of further episodes of hemoptysis.

5. Discussion

Massive hemoptysis almost always arises from systemic circulation i.e. supplied by bronchial arteries; however, pulmonary artery aneurysms can also account for a minority of cases. Pulmonary artery aneurysms were first described by Churton [1] and constitute less than 1% of aneurysms occurring in thoracic cavity. Majority of them are in main pulmonary artery with few cases described in peripheral pulmonary circulation.

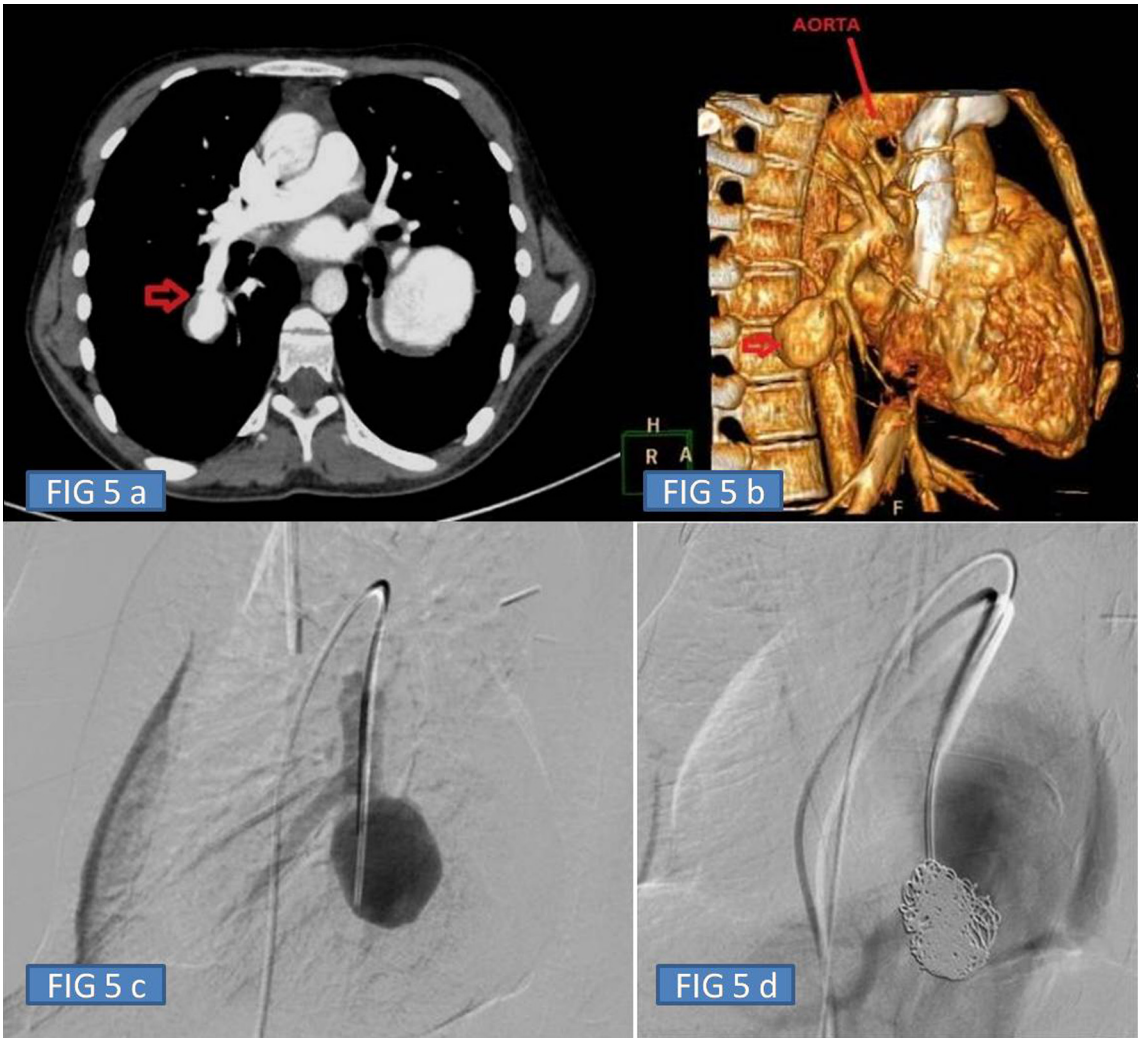

Fig. 5. a. CT axial MIP image showing a rounded aneurysm from right descending pulmonary artery (arrow). Aneurysm is also seen on left side which was managed by lobectomy. b. Volume Rendered CT image showing the right descending pulmonary artery aneurysm (arrow) and one arrow showing the aorta. c. DSA image showing catheter tip in aneurysmal sac which is filled with contrast. d. DSA image showing coil mass within the aneurysm which is obliterating the aneurismal sac.

Pulmonary artery aneurysm can be broadly classified into congenital and acquired. Congenital causes can be due to vessel wall weakness as in diseases such as Ehler Danlos Syndrome, Marfans syndrome [2,3]. Conditions that cause left to right shunt and pulmonary artery stenosis can cause enlargement of main pulmonary arterial trunk or proximal parts of right/left pulmonary artery.

Acquired causes include infective, which may be bacterial, fungal or mycobacterial as was in our first case. A Rasmussen aneurysm is an uncommon complication of pulmonary tuberculosis and it presents as a pulmonary artery aneurysm adjacent or within a tubercular cavity [2,3]. It is usually distributed peripherally and beyond the branches of main pulmonary arteries. Other mycotic aneurysms in the pulmonary arteries include multiple aneurysms from septic emboli (associated with bacterial endocarditis), which lodge in the segmental pulmonary arteries. Inflammatory causes include vasculitis, and most common vasculitis associated with pulmonary artery aneurysms are Behcet's disease [2,3]. Hughes Stovin syndrome is another entity comprising bilateral or unilateral pulmonary artery aneurysms with associated deep venous thrombosis. Hughes-Stovin syndrome is a rare clinical condition characterised by multiple pulmonary artery aneurysms and peripheral venous thrombosis, first described in 1959 [4]. It usually affects young males who present with haemoptysis, cough, dyspnoea, chest pain, and signs of pulmonary hypertension. These patients also have recurrent phlebitis largely affecting the peripheral veins. All these findings were present in the second case. Both of our patients whether Behcet's or with Hughes Stovin had bilateral pulmonary artery aneurysm. Takayasu arteritis can also can pulmonary artery aneurysm but mainly central in location [2].

Traumatic causes, often iatrogenic, are due to Swan Ganz catheter placement and chest tube insertions [2,3]. Lung biopsies and conventional pulmonary angiographies also account in few cases. Non-iatrogenic causes include stab/gunshot injuries. Neoplastic lung lesions can also erode pulmonary arterial wall and form aneurysm or pseudoaneurysm [3].

Multi Detector CT pulmonary angiography is the modality of choice to diagnose pulmonary artery aneurysms. It gives detailed information about the presence, number, size, shape and origin of peripheral pulmonary artery aneurysms. It allows detailed assessment of the orientation and size of aneurismal sac and size of neck on multiplanar reformation. Volume rendered (VR) reformation gives 3D imaging of the aneurysmal anatomy (Figs. 2a and 5b) which is very important for endovascular treatment planning. At the same time we can look for associated lung abnormalities such as fibrotic bands, cavities, bronchiolar wall thickening and associated signs of pulmonary embolism elsewhere.

Medical and surgical management of massive hemoptysis is associated with a mortality rate ranging from 35–100%. Endovascular embolisation has an initial success rate of 95% and surgery is reserved for those patients where multiple sittings of embolisation have failed [5].

Multiple endovascular embolisation procedures have been reported in the literature [5,6]. Various agents that have been used include coils, glue, covered stents, detachable balloons, vascular plug and PDA closure devices, depending upon the anatomy of the aneurysm embolic agents such as coils which pack the aneurysm and spare as many distal pulmonary artery branches as possible and preserve residual pulmonary function distal to the aneurysm. Placing coils within an aneurysm carries a potential risk of rupture. Care must be exercised during coiling procedure and over packing of the sac should be avoided [6].

Successful use of glue for embolisation has also been reported by some authors but needs experience hands and always carries a risk of inadvertent embolisation and catheter adhesion. Although glue embolisation has proved to be permanent few reports of recanalisation are there [7,8].

There are case reports [9] where percutaneous injection of thrombin or N-butyl cyanoacrylate is done to treat Rasmussen aneurysms that are not amenable to coil embolisation.

Vascular plugs such as Amplatzer vascular plug (AVP) have been used for pulmonary artery aneurysm [10]. And few authors have reported superiority of AVP over coil embolisation in endovascular procedures owing to its property of occluding feeding and draining vessel by the same device as opposed to coil embolisation which may need multiple coils thus increasing the cost of treatment. Another advantage is that the delivery system enables increased precision and control during deployment. The elasticity of nitinol allows the device to become firmly anchored to the vessel wall by outward radial force, which prevents migration and allows lengthening to be predicted. As a result, selection of the diameter of the AVP does not need to be as precise as selection of the correct diameter, length, and type when using coils [11].

Likewise PDA closure device has also been used preferably in cases where aneurysm neck was broad and short and not amenable to coil embolisation [12].

Choice of embolisation material depends upon the site, type and anatomy of aneurysm and also upon the comfort ability and experience of the operator. Complications of endovascular embolisation are less frequent and include contrast induced nephropathy, arterial dissection and pulmonary infarct. Mortality associated with surgical procedures or conservative management is high. Recurrence or development of a new aneurysm in cases of Behcet's and Hughes Stovin syndrome is known like in our second case. Surgical resection in this case would have led to decreased and insufficient pulmonary reserve. Consequently, transcatheter embolisation has become the therapy of choice for massive hemoptysis, with surgical resection reserved for failed embolisation or for recurrent massive hemoptysis following multiple previous embolisations [5].

6. Conclusion

Peripheral artery aneurysm is a potentially life threatening condition. Embolisation allows effective emergency management and helps to tide over the haemorrhagic crisis. In many cases it provides a stable occlusion of the

aneurysm. Recurrences may be dealt with a repeat procedure if feasible.

Conflict of interest

Authors hereby declare that there is no conflict of interest and no financial support has been taken from any source.

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