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Case Report

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Multiple pulmonary arteriovenous malformation in post Tuberculosis patient, a rare incident or an unknown complication

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A B S T R A C T

We report a case of pulmonary arteriovenous malformation (PAVM) and associated pulmonary tuberculosis in a 20 years old male who presented with shortness of breath on exertion mainly walking on steep up surfaces, bluish discoloration of nails, fingers and lips, platypnea and orthodeoxia. He was treated with anti-tuberculous therapy (ATT) for pulmonary tuberculosis 4 years ago. He also he had plethoric face with grade 4 clubbing. During his stay at hospital he underwent x-ray chest, CT chest with contrast and CT pulmonary angiogram that confirmed multiple pulmonary arteriovenous malformation with aneurysmal dilatation. Trans-catheter coil embolization was planned by interventional radiologist. AVM on left size, due to large size, was difficult to occlude while AVMs on the right were planned if patient later on present with right sided AVMs complication. Due to peculiar nature and sizes of pulmonary AMVs, Patient was not willing for such procedure involving high risk consent, and also due to his financial issues, he opted to be treated conservatively and was discharge on will with home oxygen.

Keywords: Pulmonary arteriovenous malformation (PAVM); Pulmonary tuberculosis

Introduction

Pulmonary arteriovenous malformation (PAVM) is a rare and abnormal communicating network between pulmonary artery and pulmonary vein with reported prevalence of 1 in 2630 with female predominance.[1] Pulmonary AVM are mostly located in lower lobes of the lungs and are unilateral. If they are multiple then they tends to be bilateral. These can be simply called pulmonary arteriovenous fistula. Although majority of cases occur congenitally but tuberculosis is one of the rare acquired cause of formation of PAVM. Our case primarily focuses on this fact that tuberculosis, even if completely treated, can lead to such serious but rare complication like pulmonary arteriovenous malformation.

Case Presentation

20 years old male, a college student from lower Dir, Khyber Pakhtunkhwa, was admitted in our unit from emergency floor with chief complaint of shortness of breath on exertion mainly walking on steep up surfaces and he also noticed gradual bluish discoloration of nails, fingers and lips for last few month.

Patient suffered from pulmonary tuberculosis 4 years ago after which he observed dyspnea. He had characteristic finding on CXR (ill define radiological shadows on right zone of the lungs) and diagnosis was confirmed by sputum acid fast bacilli (AFB) analysis. He completed course of anti-tuberculous therapy (ATT) for 8 months, taking 3 tablets each day before meal along with vitamin B6 tablets, well complaint to medications and was cured. He felt improvement in his weight and respiratory symptoms.

According to the patient, shortness of breath was gradual in onset, was grade 1 according to MMRC dyspnea scale which progressed over the course of couple of months. It aggravated with running or walking on an inclined surface while relieved upon resting and was associated with bluish discoloration of lips, fingers and nails. On systemic review it was found that patient had palpitations, double vision, occasional tinnitus and syncopewhile running or walking on a steep up surface.

On examination, he became short of breath while standing (platypnea) with oxygen saturation drop to 70 percent on standing as well (bed side test for orthodeoxia) and his resting oxygen saturation fluctuated between 80-90 percent during supine position and standing. Furthermore, he had plethoric face with grade 4 clubbing in all his fingers. He had obvious cyanosis in his fingers, nails and lips when sitting up from lying position and walking with extra pace. He had mild bilateral bi-basal fine crepitation on chest auscultation but no murmurs were audible on precordial auscultation. JVP was within normal range.

For couple of months, he was taking bronchodilators via DPI and was started on long term oxygen therapy by some physician in remote area without proper workup. Upon further questioning and examination there was no significant history or physical finding with supported any evidence of hereditary haemorr hagic talangieactasia (HHT).

During his stay in the hospital, he had undergone x-ray chest that showed homogeneous shadow at left middle zone of the lung and two irregular shape white shadow at middle and lower zone of the lungs (Picture - 1).

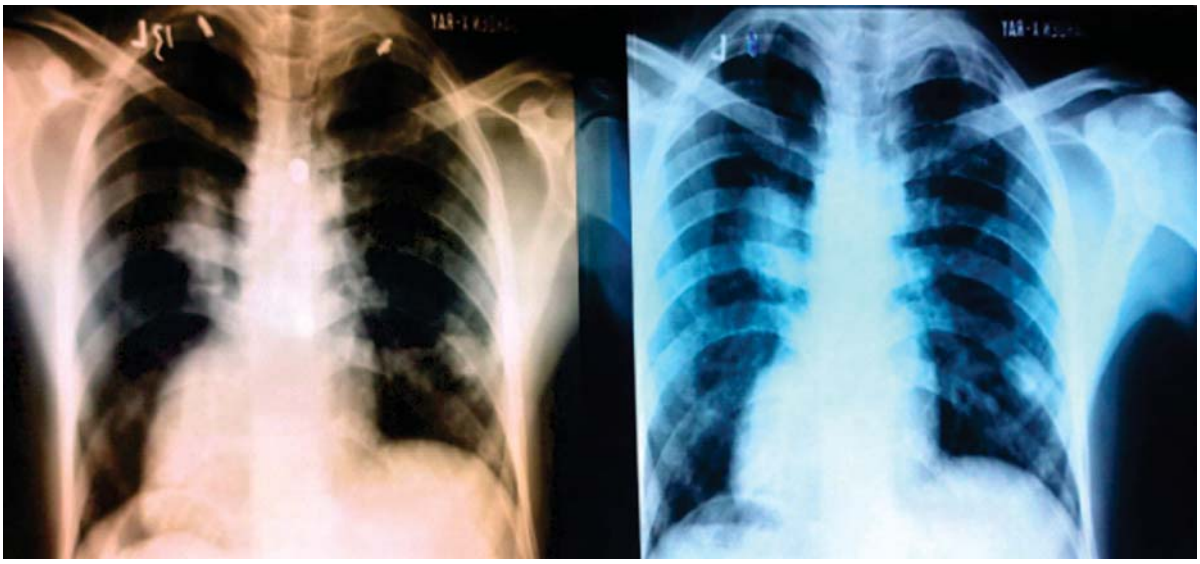

Figure 1: Chest X-Rays

CT chest with contrast was done, which initially suggested bilateral soft tissue density lesion having a dilated vascular channel close to it (Picture - 2)

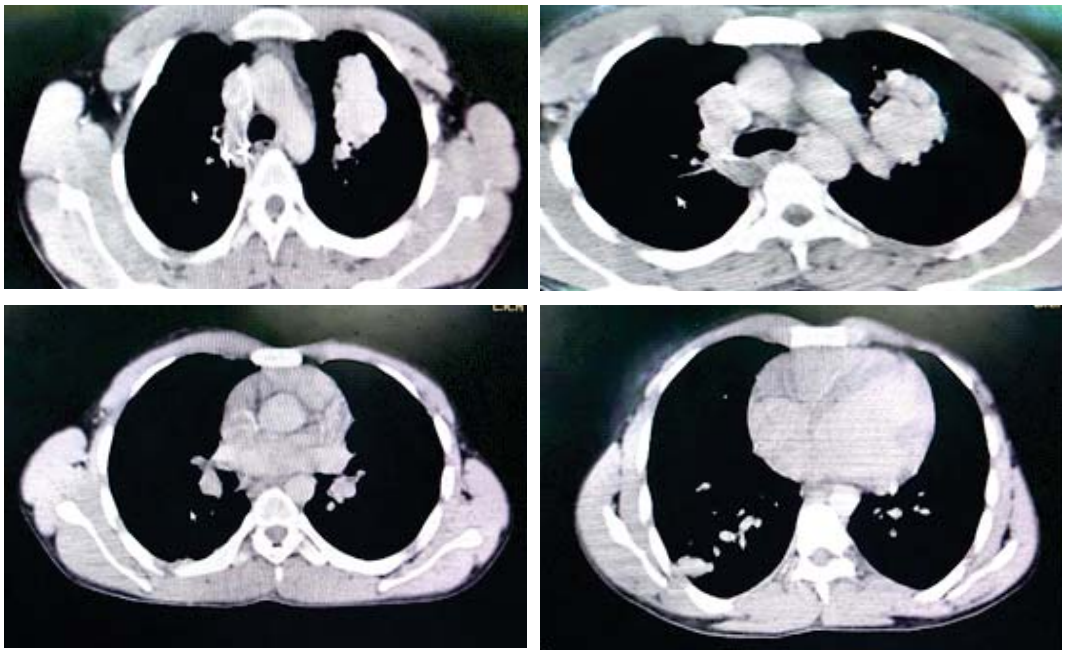

Figure: 2 (A, B, C & D): Ct Chest

Further CT pulmonary angiogram confirmed multiple pulmonary arteriovenous malformation with aneurysmal dilatation (Picture - 3).

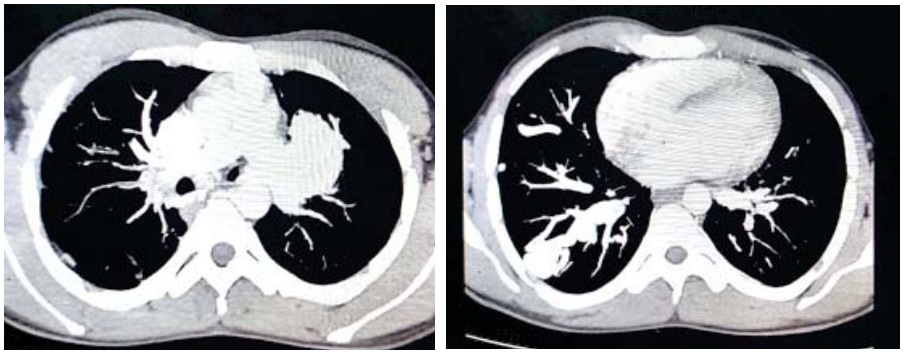

Figure: 3 (A & B): CTPA

Larger PAMV on left side in upper lobe with its nidus measuring 5.2 x 5.3cm fed by single artery from left pulmonary artery and draining into left pulmonary vein. Largest AVM on right side was noted in lower lobe with its nidus measuring 2.9 x 2.6cm fed by right pulmonary artery and draining into right pulmonary vein. Two moderate size PAVM also noticed in right lower zone of the lungs. Due to significant evidence of CTPA and limited financial resources contrast bubble echocardiography was not performed. Echocardiography and ECG were normal. PFTs showed mild obstructive pattern. ABGs showed type 1 respiratory failure with pO2 of 30% on room air. Complete blood picture show hemoglobin of 20g/dl with hematocrit of 72 which explained his plethoric face and hyper-viscosity syndrome like symptoms such as tinnitus and syncope attacks. It also explained his normal breathing during normal daily work as his high hemoglobin provide oxygenation to the body despite having large left to right shunts in his pulmonary circulatory system. Rest of biochemical profile analysis were in normal range.

He received O2 inhalation via nasal prongs as per need,

empirical antibiotics, bronchodilators and venesection was done thrice during his stay. Trans-catheter coil embolization was planned involving interventional radiologist and anesthesia team as a part of multi disciplinary approach. Due to large size of the AVM on left size it was difficult to occlude the entire AVM of such size which may be needing more than 10 coils and there was also minimal chances of stroke as coil may dislodge during procedure and may end up in brain. At the same time other AVMs on the right were also was also planned during same procedure by interventional radiologist. It was planned that, if not possible by any complication, he should be considered for surgical intervention which may include lung transplantation. Anesthesiology team recommended that a high risk consent should be taken due to peculiar nature of the procedure and possible mortality risk during operation. Patient was not willing for such procedure involving high risk consent, and also due to his financial issues, he opted to be treated conservatively and was discharge on will on home oxygen to avoid complication associated with high hemoglobin due to chronic hypoxia and partly contributed by large pulmonary left to right shunt in his pulmonary circulatory system.

Discussion

Pulmonary arteriovenous malformation (PAVM) are abnormal communicating network between pulmonary artery and pulmonary vein that are mostly located in lower lobes of the lungs. These are mostly unilateral but can be bilateral. These range from small size, usually in millimeters, but are reported to be more than 10cm in size. Most of the arteriovenous malformation are located in brain, lung (second common place) and other organs. They are mostly linked as either congenital or syndrome such as hereditary hemorrhagic telangiectasia 2 (approximately 70 percent of pulmonary arteriovenous malformation). An acquired isolated pulmonary arteriovenous malformation (PAVM) is a rare entity. One of the most common causes of acquired pulmonary arteriovenous malformation (PAVM) is liver disease mostly in liver cirrhosis. Some of the rare causes of acquired (PAVM) are being report due to chronic infection such tuberculosis, schistosomiasis, actinomycosis, metastatic thyroid cancer.[1]

The exact pathogenesis is unknown. Abnormal commutation is created due to incomplete resorption vascular septum that separate arterial and venous plexus during fetal development. It is capillary developmental failure during growth of the fetal. Later in life due to abnormality it lead formation of torturous loops and multi-loculated sacs.

One of the rare cause that was also found in our case study was tuberculosis. Tuberculosis is chronic granolumatous disease caused mycobacterium tuberculosis. It is one of the most common worldwide multi-systemic disease.[3] Mycobacterium is high immunogenic which causes activation and migration of immune cell such as langerhans cells, lymphocytes, monocytes, leukocytes which in term lead to chronic inflammation leading to distortion of the bronchus and bronchial artery, bronchiectasis and parenchymal fibrosis. Due to complex nature of the pathophysiology of the tuberculosis it is possible that it may cause pulmonary arteriovenous malformation. Previously a small number of cases of PAVM have been reported in relation to tuberculosis. Thomas et al.[4] reported a 14 year old girl with a large PAVM that occurred in association with pulmonary tuberculosis and was treated with combination of embolization and resection. Similarly, Tenget al.[5] also reported a case of a 23 years old male patient with repeated history of TB who was initially treated as a case of TB and polycythemia vera but was found to have a left lingual lobe PAVM and was eventually treated with lobectomy.

Therapeutic intervention such trans-catheter coil embolization of the feeding vessel in attempt to successfully occlude the PAVM is considered now gold standard treatment in the new modern era of interventional radiology.[6] Possible complication are rupture of blood vessel, arrhythmia and paradoxical stroke in large PAVM. In selected case surgical resection is potential option.[7]

Conclusion

Tuberculosis should be considered as cause of isolated PAVM.Patient treated previously with tuberculosis presenting with short of breath should be evaluated for symptoms and sign of PAVM along with bed side orthodexnia and radiological investigation involving contrast echocardiography and CT Pulmonary angiography. Moreover, trans-catheter coil embolization and surgical resection are suitable treatment options.

Abbreviations

AFB - Acid fast bacilli

ATT - Anti-tuberculous therapy

CTPA - CT pulmonary angiogram

CXR - Chest X-ray

ECG - Electro Cardiogram

HHT - Hereditary haemorrhagictalangieactasia

JVP - Jugular venous pressure

MMRC - Modified Medical Research Council

PAVM - Pulmonary arteriovenous malformation

PFT - Pulmonary function tests

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Publication Consent

Obtained from patient in writing.

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Availability of data and materials

Author Muhammad Noman can be contacted for data and materials.

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