Medical Science

25(118), December, 2021

Pre-operative embolization of giant pulmonary arteriovenous malformation: Case report

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To Cite:
Al-Metlag MH, Al-Dhawi AF, Al-Ahmari KS, Al-Obaidi H, Al-Ammari SN, Alruhaimi A, Al-Ahmari F, Almat'hami A. Pre-operative embolization of giant pulmonary arteriovenous malformation: Case report. Medical Science, 2021, 25(118), 3140-3145

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Peer-Review History
Received: 25 October 2021
Reviewed & Revised: 27/October/2021 to 20/November/2021
Accepted: 22 November 2021
Published: December 2021

Peer-review Method
External peer-review was done through double-blind method.

ABSTRACT

Among pulmonary vascular anomalies are arteriovenous malformation (AVM) featured by miscommunication between pulmonary arteries and veins. These unusual communications allow the unoxygenated blood to escape the standard capillary beds within the lung. AVMs cause right-to-left shunts, paradoxical embolization with hypoxemia. Clinical signs and symptoms differ according to the size, number, and blood flow within the AVMs. Among of the treatment modalities, transcatheter embolization is the preferred for managing pulmonary AVMs. Although, this technique is ineffective in cases with large AVM or having multiple affected main arteries which are managed mainly by surgical resection. The current case report focuses on a patient with a giant pulmonary AVM measuring 4.8 x 5.6 x 6.2 cm with a single feeding artery that pre-operatively managed with vascular plug embolization.

Keywords: Pulmonary Arteriovenous Malformations, Embolization, Therapeutic, Pulmonary Artery, Pulmonary Veins.

1. INTRODUCTION

Pulmonary arteriovenous malformations (PAVMs) are one of the congenital vascular disorders which are uncommon (Martinez-Pitre & Khan, 2021). This pulmonary vascular anomaly featured by congenitally dilated vessels responsible for direct capillary-free linkage amongst the pulmonary and general circulation. This anomaly has three main clinical changes (Lakshminrusimha, 2021). First, Arterial blood within lung flow through these right-to-left shunts failed to be oxygenated, causing arterial hypoxia. Second, Paradoxical embolism (Thromboembolic material passes to the systemic circulation) because of the lack of the well-known filtering capillary bed (Ministro et al., 2008), this embolizationcan may ends with having a transient ischaemic attack, stroke, with brain abscess (due to bacterial embolization) (Sonneville et al., 2011). Third, cases with PAVMs may have hemoptysis or haemothorax, particularly during pregnancy as a result of upture of the thin

walled of these vascular malformations, as hormonal disturbances may enhance a rapid enlargement of PAVMs (Di Guardo et al., 2019).

In 1897, PAVMs were first described Churton after doing autopsy of a cyanotic young male who had recurrent episodes of hemoptysis. In 1938, Rodes assessed the linkage between PAVMs and HHT (Hereditary Hemorrhagic Telangiectasia) which is an autosomal dominant inherited vascular disorder (Rauh et al., 2017). The first successful surgical management of PAVMs including a pneumonectomy was in year 1942 (Rauh et al., 2017). Besides, pulmonary fistula in children was first described by Werner Porstmann who performed the first PAVM embolization in 1977. Embolization therapy approved to be the corner stone method in the management of PAVMs since 1983 13 (Carnevale et al., 2010).

In the present report, authors describe a case with a large (4.8 cm x5.6 cm ×6.2 cm) pulmonary AVM which effectively conserved through pre-operative embolization using an Amplatzer vascular plug.

2. CASE REPORT

Fifty-nine years old medically free lady, presented with right chest pain and decrease LOC, the patient was visiting her admitted mother at Prince Sultan Military Medical City (PSMMC). The patient shifted to the ER immediately, and a chest radiograph was performed following the standard emergency assessment. The radiography showed a large mass-like opacity project over the lower part in right lung with significant ipsilateral pleural effusion (Figure 1). Then, the patient was referred to a pulmonologist. Additional respirationalcomplaints as cough, sputum production, or hemoptysis were absent. On physical examination, her breathing sounds are diminished on the right side.

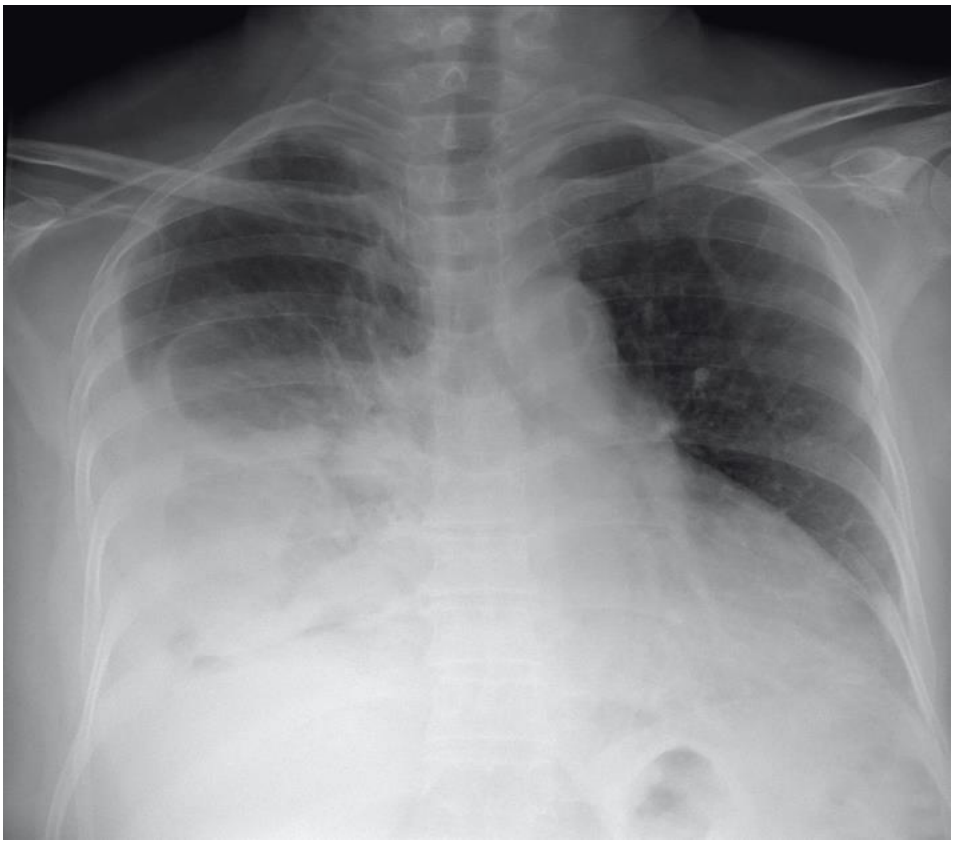

Figure 1 First chest radiograph revealing a sizeable lump-identical opaqueness in the right inferior lung arena with right side significant pleural effusion

On admission, the initial SpO2 value on area air was 83%; with normal ranges of all other vital signs. Arterial blood gas analysis showed hypoxemia with a PaO2 level of 21.3 mm Hg. The electrocardiography indicated standard sinus pulse lacking substantial ST-segment change. Laboratory studies showed normal range erythrocyte. Her white blood cell total number was 9 ×109/L, hemoglobin level was low at 9 g/dL, then platelet number was 375×109/L.

Contrast-enhanced chest computed tomography (CT) revealed a large (approximately 4.8 cm ×5.6 cm ×6.2 cm) lobulated non-calcified lesion showing similar contrast enhancement to the vascular structures. The lesion is supplied by a large branch from the

right lower lobe segmental artery and drained by the right lower lobe draining vein. Marked right-side pleural effusion with different densities suggestive of varying stages of hemorrhage is seen as well. Findings are matching with giant pulmonary AVM (Figure 2) the nourishing vessel with an all-out width of 7mm.

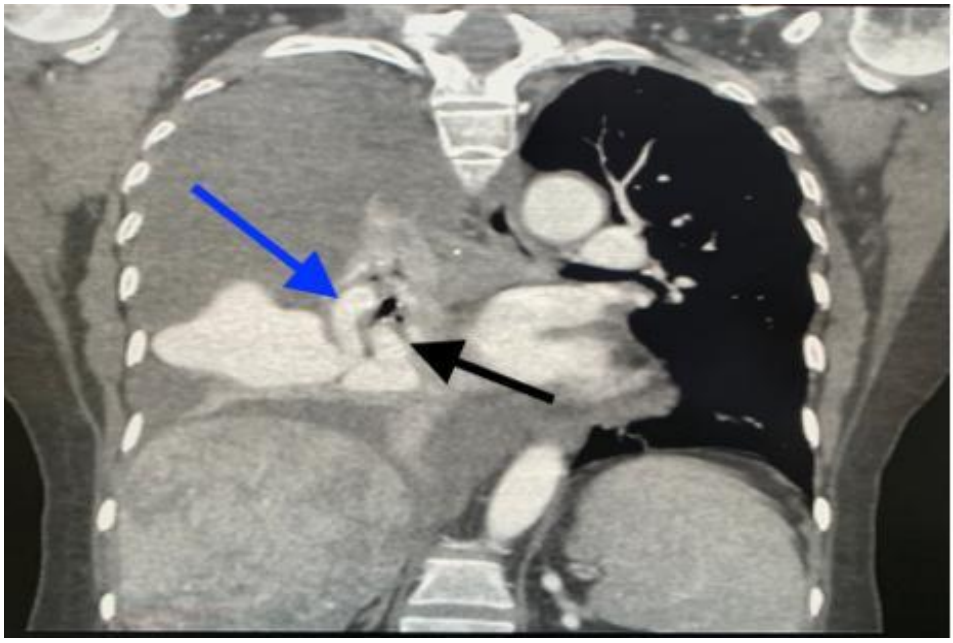

Figure 2 A large (approximately 4.8 cm ×5.6 cm ×6.2 cm) lobulated non-calcified lesion with similar contrast enhancement to the vascular structures supplied by the right lower lobe segmental artery (blue arrow) and drained by the right lower lobe draining vein (black arrow) seen within the right lower lobe of the lung. There is marked right-sided pleural effusion with different densities suggestive of varying stages of hemorrhage.

The patient's oral cavity, nasal cavity, and skin were perfectly examined, with no detected telangiectatic lesions. Family history regarding visceral AVMs was negative. As there was no evidence evidencing the diagnosis of HHT, this pulmonary AVM is decided as non-HHT-related. As the size of AVM was moderately large, transcatheter embolization elected as a first management step before the thoracoscopic surgical resection. The case experienced pulmonic angiography through the right femoral vein, besides a 62-mm pulmonic AVM was imagined in the right lower lobe. A single large feeding artery was detected. The feeding artery of the pulmonary AVM was selectively catheterized and embolized using 14 x 8 mm vascular plug. Post-embolization DSA confirmed complete obliteration of the arterial supply (figure 3, 4 & 5).

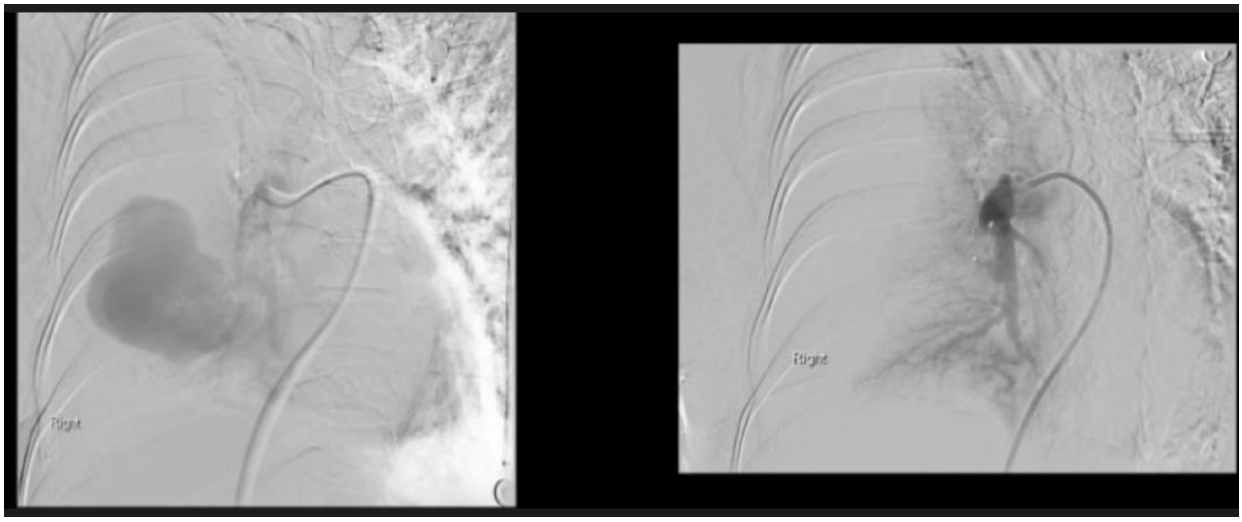

Figure 3 Pulmonary angiography through the right femoral vein access showing a 62-mm pulmonary AVM at the lower part of the right lobe.

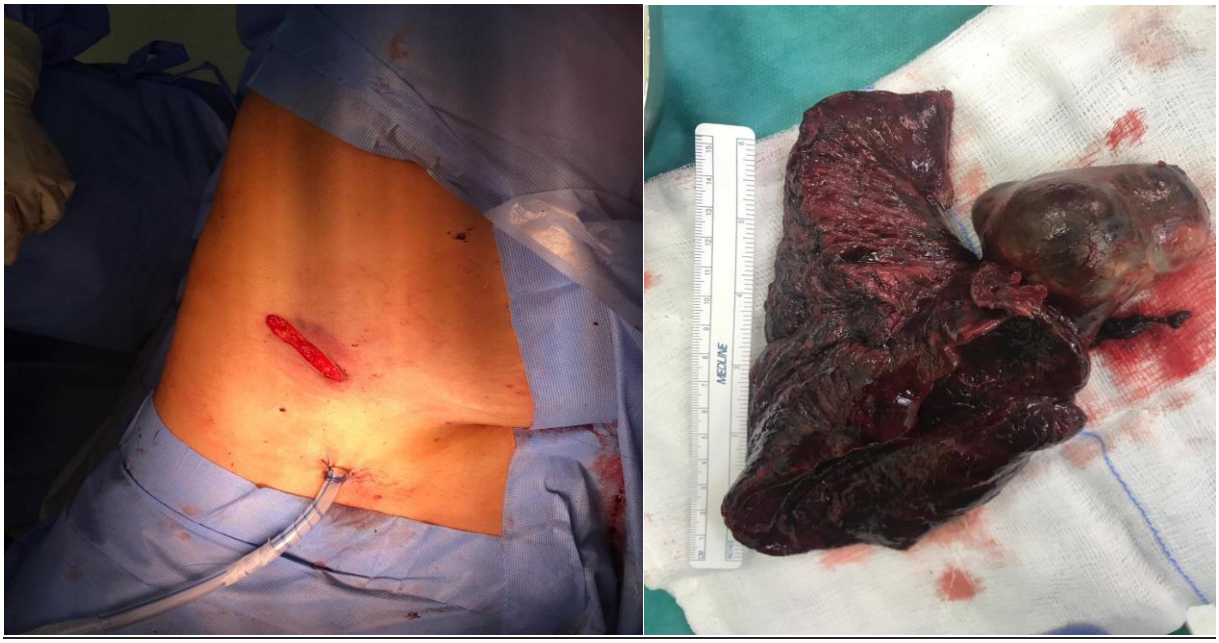

Figure 4 A & B Post-embolization thoracoscopic surgery with small surgical incision was performed instead of standard open thoracotomy for such pAVM size.

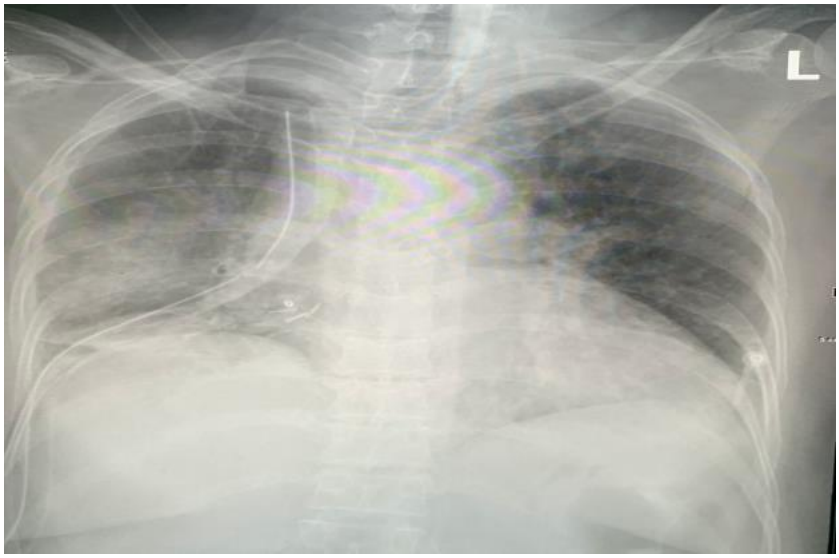

Figure 5 CXR (day 2 post-surgery) shows: excellent reminant right lung aeration with no effusion or abnormal opacity, Chest tube and surgical clips are seen as well.

3. DISCUSSION

Pulmonary AVM is an extraordinary disease category. Nevertheless, it's far crucial to keep in mind this disorder in the discrepancy diagnosis of well-known pulmonary disorders including hypoxia, pulmonary nodules, or hemoptysis. HHT is evident to be associated with PAVM that is an autosomal main hereditarycondition. Previous screening research found that approximately 70% to 90% of pulmonary AVMs detected in cases with HHT3. The mainstream of PAVMs with no HHT is idiopathic, as in the current case (Cartin-Ceba et al., 2013).

Pulmonary AVM may be asymptomatic among many cases but with hypoxia. This may be explained by what is called chronic recompense, comprising the subordinate erythrocytic response. In the current cases, patient similarly presented substantial hypoxia, nonetheless she denied subjective dyspnea. Lack of hypoxia related symptoms would not prevent developing significant complication among cases with PAVMs. The most frequent complications are neurological problemscomprising stroke, TIA

(transient ischemic attack), cerebral abscess, and migraine. Patients may progress till developing life-threatening hemorrhagic complications, but this is uncommon (Park et al., 2015).

Recently, it is advised that all pulmonary AVMs reliable to embolization evidenced to be treated with transcatheter embolization to minimize AVMs associated substantial morbidity rate especially with the advancement of angiographic techniques, With successful embolization, the obstruction of nourishing arteries resulting in reversion of pulmonary AVMs, causing the resolve of a right-to-left shunt, better oxygenation, and the inhibition of embolic complications (Hsu et al., 2018). Even though the latest improvements in angiographic interventions; some technical constraints to embolization still exist in some cases. The recanalization of pulmonary AVMs after embolization was proved to be a result of the increase in diameter of feeding artery, using of little coils, using of large coils, and proximal coil location (Park et al., 2015).

Large pulmonary AVMs with or without several large feeding arteries are difficult to be completely occluded. If cases with AVMs are not agreeable to undergo transcatheter embolization, surgical resection is advised (Milic et al., 2005). Nevertheless, there is no agreement regarding which cases of pulmonary AVMs must be managedby surgical resection as instead of transcatheter embolization. The decision repeatedly depends on the experience, skills, and preference of the management staff (Hart et al., 2010). The usefulness and possibility of trans-catheter embolization as treatment method for large pulmonary AVMs was assessed by many researches (Kucukay et al., 2014). An exciting and differentiating feature of the current report case is that researchers used a vascular plug for a single giant pulmonary AVM. The AVM was blocked compactly on post-embolization angiogram images. This favored thoracoscopic rather than open surgical resection for such huge pulmonary AVM size (Lee, 2016; Majumdar & McWilliams, 2020).

4. CONCLUSION

Though transcatheter embolization is the favorite management for most pulmonary AVMs, some cases where some technical problems with this approach had. To author's information, there is no strong suggestion or magnitude level for preferring surgical resection over embolization. Think About the risks of surgery-related disease and death, transcatheter embolization must be tried even for larger diameters PAVMs which was applied for the current case, or high flow rate and short supplying artery, AVPs are recommended to achieve a faster and precise procedure.

Informed consent

Written & Oral informed consent was obtained from all individual participants included in the study

Funding

This study has not received any external funding.

Conflict of Interest

The authors declare that there are no conflicts of interests.

Data and materials availability

All data associated with this study are presented in the paper.

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