Radiology Case Reports 16 (2021) 2853–2856

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journal homepage: www.elsevier.com/locate/radcr

Case Report

Arteriovenous malformation in the middle mediastinum with spontaneous hemoptysis and successful management by Endovascular embolization ☆

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ARTICLE INFO

Article history:
Received 21 June 2021
Revised 4 July 2021
Accepted 6 July 2021

Keywords:
Arteriovenous malformation
Middle mediastinum
Embolization

ABSTRACT

Arteriovenous malformations in the mediastinum are rare and extremely unusual in middle mediastinum. The present report describes a 26-year male with spontaneous massive hemoptysis due to arteriovenous malformation in middle mediastinum and successful management by endovascular embolization. Spontaneous massive hemoptysis can be a presenting feature of arteriovenous malformation in middle mediastinum and endovascular embolization can be curative.

Introduction

Arteriovenous malformations (AVM) are congenital abnormalities characterised by multiple direct communications between arteries and veins without intervening capillary network. AVMs located in the mediastinum of thoracic cavity, particularly in adults are exceedingly rare. Due to its wide spectrum of clinical presentations, AVMs are difficult to diagnose, and treat. The present study reported an extremely rare case of middle mediastinal AVM in 26-year-old man presented with massive haemoptysis and managed by endovascular AVM embolization to completely exclude it from the circulation.

Case report

A 26-year male presented with complaint of spontaneous massive hemoptysis (500-600 mL) and history of 2 episodes of small quantities (5-15 mL) of hemoptysis in last 2 weeks. There is no history of fever, prior infections like tuberculosis

☆ Competing Interests: The authors declared no conflict of interest.

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https://doi.org/10.1016/j.radcr.2021.07.015

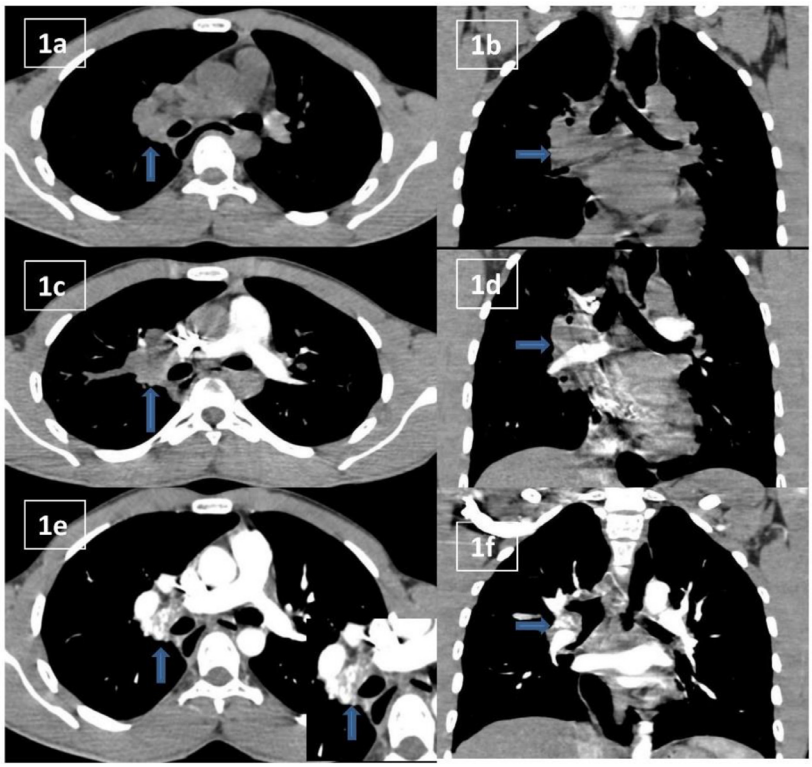

Fig. 1 – Contrast enhanced CT of chest.
Figs. 1A and B: NECT Axial (1A) and coronal (1B) showing isodense middle mediastinal AVM (block arrow). Fig. 1C and 1D: CECT Axial (1C) and coronal (1D) showing non–enhancing middle mediastinal AVM (block arrow) in pulmonary arterial phase. Figs. 1E and F: CECT Axial (1E) and coronal (1F) showing enhancing middle mediastinal AVM (block arrow) in the aortic phase. Inset image in the 1E is delineating the AVM (block arrow).

or chest trauma. His medical history, physical examination, routine blood tests, electrocardiogram were unremarkable. A Chest radiograph revealed a mildly bulky right hilum. Nonenhanced computed tomography (CT) of chest revealed abnormal isodense soft tissue in middle mediastinum (Figs. 1A and B). CT pulmonary angiogram phase revealed no obvious pulmonary arterial feeders (Figs. 1C and D). Contrast enhanced CT of chest in aortic phase revealed an irregular meshwork of intensely enhancing dilated serpiginous vessels in the middle mediastinum of size 3.5 × 3.0 cm located in the right hilar region, around the right main bronchus (Figs. 1E and F). A small component of the meshwork was noted in subcarinal region. These findings were in favor of AVM. The lesion was enhancing in aortic phase suggesting its supply from the bronchial artery. So, for better delineation of angioanatomy, digital substraction angiography (DSA) was considered. A finding of achalasia cardia noted. DSA of aorta and selective angiograms of right intercostobronchial trunk giving rise to first bronchial artery (Fig. 2A), second bronchial artery from aorta (Fig. 2C), left bronchial artery and posterior intercostal arteries confirmed the middle mediastinal AVM supplied by 2 right bronchial arteries and draining into the right superior pulmonary vein (Figs. 2A and C). The treatment options of surgery and embolization was put forward, out of which the patient gave consent for embolization.

Endovascular embolization was done with polyvinyl alcohol particles (PVA-500) and gelfoam, after selective cannulation of the first bronchial arterial feeder (Figs. 2A and B) and second bronchial arterial feeder (Figs. 2C and D). With complete exclusion of the AVM from the circulation at the end of the procedure with no complications. Patient was discharged and is on regular follow up for 4 years with no recurrence of hemoptysis.

Discussion

Arteriovenous malformations (AVM) of the mediastinum are extremely rare [1–8] lesions and more commonly present in childhood. This vascular anomaly was first described in the mediastinum by Lunde and colleagues in 1984 [6]. Since then, fewer than 10 cases of posterior mediastinal AVMs, 2 cases and 1 case of anterior and middle mediastinal AVMs in the adults have been documented [8]. To the best of our knowledge from literature, symptomatic middle mediastinal AVM with massive haemoptysis in adult is extremely rare. AVMs are reported in many organ systems of the body. In the thorax, pulmonary AVMs are more commonly encountered and may be associated with hereditary hemorrhagic telangiectasia

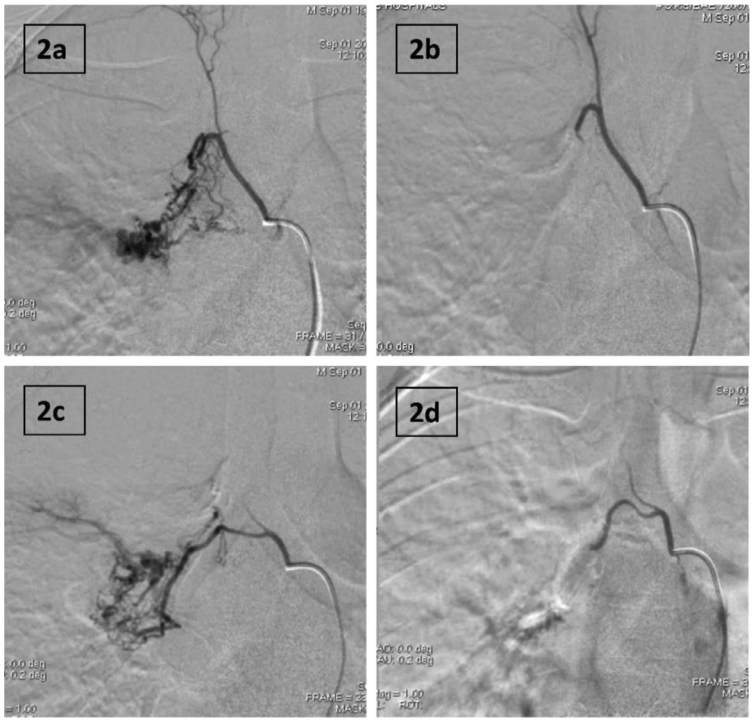

Fig. 2 – DSA-bronchial artery embolization.
Figs. 2A and B: DSA, A-P view showing Pre embolization (2A) and Post embolization (2B) images of right 1st bronchial arterial feeder of middle mediastinal AVM and its complete occlusion. Fig. 2C and D: DSA, A-P view showing Pre embolization (2C) and post embolization(2D) images of right 2nd bronchial arterial feeder of middle mediastinal AVM and its complete occlusion.

(osler-weber-rendu disease). Our case of middle mediastinal AVM is extremely rare and was supplied by 2 right bronchial arteries with no association with osler-weber-rendu disease. An AVM may be asymptomatic and becomes symptomatic if it gets infected or enlarges and exerts pressure on vital mediastinal structures, such as trachea or superior venacavae. Severe hemorrhage is also a risk, although to date, only 1 patient with spontaneous rupture of posterior mediastinal AVM with bilateral hemothoraces has been documented [4].Our case of middle mediastinal AVM with spontaneous rupture into the airway with massive hemoptysis is unusual and hasn't been addressed. Asymptomatic AVMs may be managed conservatively, using wait and watch approach. If the AVM is symptomatic as was our case, treatment options are embolization or surgery or preoperative embolization followed by surgery. Complete surgical removal of mediastinal AVMs may be complicated by encroachment, adherence of the lesion on adjacent vital mediastinal structures such as major airway and great vessels [8]. In our case there was encroachment of the right intermediate bronchus, so surgical resection was thought to be complicated and not feasible. The patient consented for endovascular embolization, which was carried out successfully with complete exclusion of middle mediastinal AVM from the circulation. There was no recurrence of hemoptysis on follows up.

Conclusion

Arteriovenous malformation in the middle mediastinum of an adult presenting with spontaneous hemoptysis is extremely rare and endovascular embolization can be curative. To the best of our knowledge from the literature, such a case is extremely rare.

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