

Asian Journal of Medicine and Health

Volume 21, Issue 9, Page 73-79, 2023; Article no.AJMAH.98692 ISSN: 2456-8414

Hemoptysis Revealing a Pulmonary Arteriovenous Fistula: A Case Report

Ghizlane Jaabouti ^{a*}, Achraf Cherrat ^b, Soumia Bencchakroun ^a, Chafiq Mahraoui ^a and Naima El Hafidi ^a

^a Division of Pediatric Immuno Allergology and Infectious Diseases, Ibn Sina University Hospital, Rabat, Morocco.

^b Children University Hospital, Ibn Sina University Hospital, Rabat, Morocco.

Authors' contributions

This work was carried out in collaboration among all authors. All authors read and approved the final manuscript.

Article Information

DOI: 10.9734/AJMAH/2023/v21i9859

Open Peer Review History:

This journal follows the Advanced Open Peer Review policy. Identity of the Reviewers, Editor(s) and additional Reviewers, peer review comments, different versions of the manuscript, comments of the editors, etc are available here:

https://www.sdiarticle5.com/review-history/98692

Received: 05/02/2023 Accepted: 08/04/2023 Published: 23/06/2023

Case Study

ABSTRACT

Pulmonary arteriovenous malformations are a rare type of vascular anomaly, which can be congenital or acquired. These malformations can present as single or multiple lesions and are often associated with Rendu-Osler disease. Diagnosis is typically made through thoracic CT scanning, and the standard treatment is percutaneous embolization.

We present a clinical cases that can help raise awareness about rare diseases such as Pulmonary arteriovenous malformations and improve early diagnosis and management. It is important for healthcare providers to be familiar with the clinical and etiological characteristics of these conditions to ensure that patients receive appropriate care and treatment.

In this case report, we describe a 7-year-old girl who presented with hemoptysis in the context of pneumopathy. Despite having no significant medical history, the patient's condition was successfully treated with antibiotic therapy and percutaneous embolization of the pulmonary arteriovenous malformation.

*Corresponding author: E-mail: ghiz.jaabouti@gmail.com;

Keywords: Pulmonary arteriovenous malformations; hemoptysis; digital clubbing; embolization.

1. INTRODUCTION

Pulmonary arteriovenous malformations are a rare type of vascular anomaly characterized by an abnormal connection between the arterial and venous pulmonary circulations, this results in a right-to-left shunt [1,2], bypassing the lungs' normal oxygenation process and leading to chronic hypoxia. While these malformations are typically congenital, they can also be acquired. Diffuse forms are often associated with Rendu-Osler disease.

Symptoms can appear before the age of 15 and the diagnosis is often made based on abnormal radiological images. A thoracic CT scan with vascular injection is necessary to confirm the diagnosis. The primary treatment for pulmonary arteriovenous malformations is typically percutaneous embolization.

2. PRESENTATION OF CASE

The patient is a 08-year-old child, female sex, no consanguinity or notion of tuberculosis contagion, nor of repeated infection, she presented 5 months ago hemoptic sputum

without cough or chest pain, and without exertional dyspnea, the evolution was marked by the aggravation of hemoptisis who becomes medium abundance, which motivated the consultation.

Clinical examination finds an afebrile patient, without mucocutaneous telangiectasia, normocolored conjunctiva, eupneic, SO2 at 92% on room air, no auscultatory rales, or a thoracic deformity, no staturoposterior delay, with a continuous left subcostal murmur and a digital clubbing.

The chest X-ray shows a left paracardiac opacity without cardiomegaly [Fig. 1], the thoracic CT scan with injection of contrast medium shows a large focus of left posterior basal condensation, measuring 8. 2 cm long, associated with a central excavation and a bullous images, it encompassing the left inferior lobar branch, with an arterio-cavernous fistula [Fig. 2].

The thoracic angioscan confirms the arteriocavernous fistula [Fig. 3], the echocardiography was normal.



Fig. 1. The chest X-ray with a left paracardiac opacity without cardiomegaly

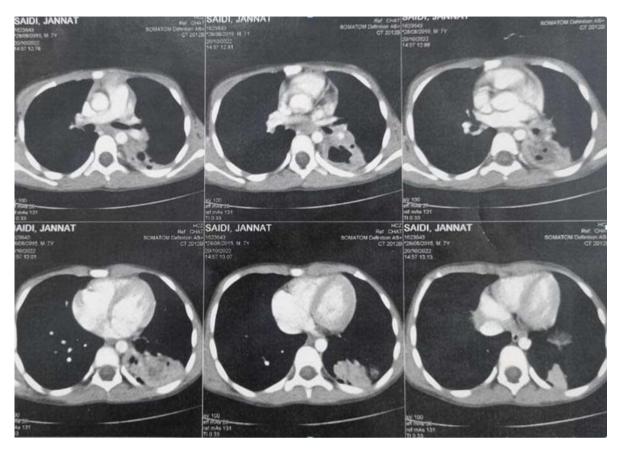


Fig. 2. The thoracic scan shows a left posterior basal condensation, with an arterio-cavernous fistula



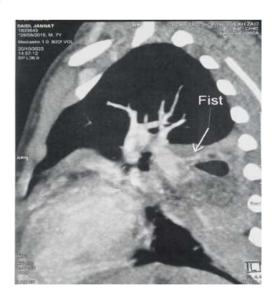


Fig. 3. The thoracic angioscan confirms the arterio-cavernous fistula

Plethysmography was normal, the nocturnal oximetry on room air confirms the permanent decrease of SpO2, and its average value was 92%, with a value below 90% during 25% of the recording time.

The blood count found a hemoglobin at 12.8 g/dL with a hematocrit at 55%, the C-reactive protein at 64 mg/l, and the sputum cytobacteriological examination (ECBC) found a Haemophilus influenzae, and Moraxella catarrhalis.

Aspergillary serology is negative with an average IgE level, the phtysiological assessment (GeneXpert MTB/Rif with quantiferon) is negative.

A bronchoscopy performed with broncholaveolar enema where the cytobacteriological examination also found a Haemophilus influenzae, and Moraxella catarrhalis.

The patient was put on amoxicillin protected for one month with an antifibrinolytic (tranexamic acid), due to persistent hemoptysis, a thoracic arteriography was performed with percutaneous embolization [Fig. 4].

The patient had a good clinical evolution with a saturation of 98% on room air and cessation of hemoptysis, the control x-ray after a month of amoxicillin protected and the thoracic arteriography with percutaneous embolization finds the disappearance of the pulmonary focus [Fig. 5].

3. DISCUSSION

Pulmonary arteriovenous fistulas, are rare vascular anomalies characterized by abnormally dilated vessels that create a right-to-left shunt between the pulmonary artery and vein [1,2]. The medical literature has used different terms to describe this condition, including PAVF, arteriovenous aneurysm, and arteriovenous hemangioma [1-3].

PAVFs can be classified into two types: simple and complex. Simple PAVFs are more common [1,4] and typically present as a well-defined peripheral nodule that can be rounded or multi-lobulated. Histologically, the simple type is characterized by a single aneurysmal sac containing a single segmental artery feeding the malformation. On the other hand, complex PAVFs involve multiple abnormal vessels and consist of one or more lobulated venous sacs of variable size supplied by more than one feeding artery, often arising from adjacent segmental pulmonary artery branches [1].



Fig. 4. The thoracic arteriography performed with percutaneous embolization



Fig. 5. The control x-ray after a month of amoxicillin protected and the thoracic arteriography with percutaneous embolization finds the disappearance of the pulmonary focus

The patient in this case has a simple form of pulmonary arteriovenous fistula (PAVF). Complex PAVFs, on the other hand, involve multiple abnormal vessels and can consist of one or more lobulated venous sacs of variable size supplied by more than one feeding artery. These feeding arteries often arise from adjacent segmental pulmonary artery branches, and complex PAVFs may involve whole lung segments or an entire lobe [1].

The age of onset for arteriovenous fistulas can vary widely, from the neonatal period to adulthood. Women are more commonly affected, with a two-fold higher incidence than men, except in the neonatal period where males are more frequently affected [5].

Pulmonary arteriovenous malformations can manifest as single lesions, multiple lesions, or diffuse forms. They can occur at any age, and it is believed that 75% of cases are associated with Rendu-Osler disease or hereditary hemorrhagic telangiectasia. Localized forms of PAVMs can also be acquired, resulting from trauma or surgical correction of cyanotic congenital heart

disease, gestational trophoblastic disease, or hepatopulmonary syndrome.

Pulmonary arteriovenous malformations can lead to serious complications such as chronic hypoxemia, cerebral vascular accidents, cerebral abscesses, hemoptysis, hemothorax, or endocarditis. These complications can arise from the abnormal communication between the arterial and venous pulmonary circulations, leading to a right-to-left shunt and bypassing the lungs' normal oxygenation process. Early detection and treatment are crucial to prevent these complications and improve the patient's prognosis [5].

However, clinical signs are often mild in cases where PAVFs are small (less than 3 mm) or involve a single vessel (simple form), with exertional dyspnea being the main symptom [6]. In the case of our patient, digital clubbing and polycythemia were also present, in addition to hemoptysis.

Pulmonary arteriovenous fistulas can be difficult to diagnose due to their rarity and non-specific

symptoms. Routine examinations may not detect them, but chest X-rays and thoracic CT scans can help in diagnosing the condition. In fact, chest X-rays show abnormalities in 95 to 98% of PAVF cases [7,8]. However, CT scans are considered the most reliable diagnostic tool for PAVFs [1,9].

It is important to note that while angiography may be used for small or thrombosed PAVFs, the primary treatment for PAVFs is typically percutaneous embolization, which involves the injection of a substance to block the abnormal connection and restore normal blood flow [10], the cardiac echography remains a sensitive screening examination.

Congenital fistulas may not require treatment unless there are significant complications. Asymptomatic forms can be managed with prophylactic antibiotic therapy in case of invasive procedures or dental care. The primary treatment option for PAVFs is percutaneous embolization during arteriography. Surgical removal of the arteriovenous fistula may only be considered for very large forms or if embolization is not successful [11,12].

4. CONCLUSION

Pulmonary arteriovenous fistulas are vascular anomalies that may be discovered incidentally or present with serious complications. They should be considered in patients with chronic hypoxemia and abnormal radiological findings. Percutaneous embolization is the preferred treatment option, with surgical removal reserved for cases where embolization fails or for very large fistulas. Asymptomatic forms may not require treatment, but prophylactic antibiotics are recommended for invasive procedures or dental care.

CONSENT

We present a clinical case with the written consent of the parent.

ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

COMPETING INTERESTS

Authors have declared that no competing interests exist.

REFERENCES

- 1. Gill SS, Roddie ME, Shovlin CL, Jackson JE. Pulmonary arteriovenous malformations and their mimics. Clin Radiol. 2015;70:96–110.
- 2. Gossage JR, Kanj G. Pulmonary arteriovenous malformations: A state of the art review. Am J Respir Crit Care Med. 1998;158:643–61.
- 3. Ahn S, Jung S, Cho JH, Kim TS, Han J. A rare case of pulmonary arteriovenous hemangioma presenting as a peribronchial Mass. J Pathol Transl Med; 2015.
- 4. White RI Jr, Mitchell SE, Barth KH, et al. Angioarchitecture of pulmonary arteriovenous malformations: An important consideration before embolotherapy. AJR Am J Roentgenol. 1983;140:681–6.
- Losay J, Lambert V, Sigal A: Malforamtions artérioveineuses pulmonaires congénitales et acquises diagnostic et traitement. Archives de Pédiatrie. 2008;15:518-519.
- 6. Gupta S, Faughnan ME, Bayoumi AM. Embolization for pulmonary arteriovenous malformation in hereditary hemorrhagic telangiectasia: A decision analysis. Chest. 2009;136:849–58.
- 7. Georghiou GP, Berman M, Vidne BA, et al. Pulmonary arteriovenous malformation treated by lobectomy. Eur J Cardiothorac Surg. 2003;24:328–30.
- 8. Pejhan S, Rahmanijoo N, Farzanegan R, et al. Surgically treatable pulmonary arteriovenous fistula. Ann Thorac Cardiovasc Surg. 2012;18:36–8.
- Nakayama M, Nawa T, Chonan T, et al. Prevalence of pulmonary arteriovenous malformations as estimated by low-dose thoracic CT screening. Intern Med. 2012; 51:1677–81.
- Remy-Jardin M, Remy J, Wattinne L, Giraud F: Central pulmonary thromboembolism: Diagnosis with spiral volumetric CT wih the single-breathbreath-hold technique- comparison with pulmonary angiography. Radiology. 1992; 185:381-7.
- Terry PB, White RI Jr, Barth KH, Kaufman SL, Mitchell SE. Pulmonary arteriovenous malformations. Physiologic observations and results of therapeutic balloon embolization. N Engl J Med. 1983;308: 1197–200.

 Gupta P, Mordin C, Curtis J, Hughes JM, Shovlin CL, Jackson JE. Pulmonary arteriovenous malformations: Effect of embolization on right-to-left shunt, hypoxemia, and exercise tolerance in 66 patients. AJR Am J Roentgenol. 2002;179: 347–55.

© 2023 Jaabouti et al.; This is an Open Access article distributed under the terms of the Creative Commons Attribution License (http://creativecommons.org/licenses/by/4.0), which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited.

Peer-review history:
The peer review history for this paper can be accessed here:
https://www.sdiarticle5.com/review-history/98692